Summer 2017

A parent-mediated habit reversal intervention for chronic tic disorders in children

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A PARENT-MEDIATED HABIT REVERSAL INTERVENTION FOR CHRONIC TIC DISORDERS IN CHILDREN

by

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A thesis submitted in partial fulfillment
of the requirements for the Doctor of Philosophy
degree in Psychological and Quantitative Foundations in the
Graduate College of
The University of Iowa

August 2017

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Iowa City, Iowa

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PH.D. THESIS

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ACKNOWLEDGEMENTS

My academic journey began 15 years ago when I answered the fearful, "What do you want to be when you grow up?" question in high school. It is impossible to thank everyone who has helped me get to this point in my career and there are not enough words to fully express my gratitude. However, there are many individuals who must receive thanks through the written word.

First, I thank my committee: Matt O'Brien, Kit Gerken, Stewart Ehly, Youjia Hua, and Samuel Kuperman. Your assistance with recruitment, edits, and/or feedback have positively shaped the way I view research and have been immensely helpful throughout the process. I am grateful for the time you have dedicated to this project and your support throughout. This dissertation would not have been completed without you.

To Kit Gerken, my co-chair, previous advisor, and all around incredible human: something wonderful was in the cards that allowed me to be your advisee. I will always cherish our discussions and truly felt like someone had my back throughout "graduate school part 2." I have learned so much from you and feel blessed to have gotten to know and spend time with you. From listening to my stress, providing answers to questions, discussing shared experience in Illinois, to making sure I met crucial timelines, to finding me employment – you have had a great impact on my life. Thank you from the bottom of my heart.

To Matt O'Brien, my other co-chair, current advisor, mentor, and clinician extraordinaire: thank you for letting me learn from you. Through you, I learned what it means to be truly compassionate as a psychologist. You helped me better understand and shape behavioral skills, which have been and will be completely useful in my professional world. Most of all, you introduced me to habit reversal, which can never be repaid. This dissertation would not have been

even an idea without you. In the future, I hope to be at least 10% of the clinician you are. Thank you for your continued support.

To Robert and Susanne Duffin, my parents (a.k.a. parental units 1 and 2): From the day I was born (and probably before), you always set high expectations. I can officially say that I understand it now. Without your guidance and gentle pushing to try my best and be better, I would not be at this final stage of a doctorate. Thank you for your unconditional support and for providing 100% of the love 100% of the time. While I did not attend the University of Notre Dame as originally intended, I do think that you have helped me achieve something just as awesome.

To Matthew, my husband and permanent roommate: I love you. These are three simple words that you have not heard enough for the past few years, especially as this document was in process, but that you hopefully know. If anyone were to ask me what sacrifice looks like, I would point them immediately in your direction. Thank you for agreeing with my plan to go *back* to graduate school. Thank you for following me around the country when the plan to go back to graduate school required it. Thank you for providing quiet space when I needed it. Thank you for tolerating me when things were rough and helping me celebrate when things were great. I am thankful every day for saying "Yes" and cannot wait to see what the next step of this journey brings for us.

To Jim and Dawn Henning, the world's best in laws: Besides being able to marry my best friend, I think the greatest thing that came out of my marriage to your son was that I get to have you as my parents-in-law. This academic process has not always allowed me to be where I want to be. Thank you for loving me as your daughter, providing constant support, and for understanding when I needed to prioritize work.

To my friends (a.k.a. the siblings I never had): I appreciate all of the hours spent providing support and empathy when it got overwhelming, providing distraction when I needed it, and providing praise when it was going well. I do not know how I got to have the support network that I do, but I am not going to waste time questioning it.

To Katie Schabilion: thank you for your direct assistance with implementing the procedures, data collection, and coding. I am in awe of your positive attitude and am excited to see what great things the future has in store for you. In addition, thank you for letting me interrupt you at work when I exhibited attention-seeking behaviors. I greatly appreciate that you did not put my behavior on extinction.

To Deva Carrion: I might have gone through this round of graduate school without getting to know you as well as I do now, which would have been dreadful. I am glad we got to sit next to each other during practicum seminar during second year, which led to an incredible friendship. I think I owe you approximately 32 meals, give or take 200. Thank you for always listening when I needed it and not judging me for going down the rabbit hole of anxiety. Also, a big thank you for helping with implementing the procedures, data collection, and coding. This dissertation was made possible by you and Katie and did not go unnoticed. I am excited to support you through graduation and beyond.

To Nicole Hendrix: You are an amazing friend/researcher/student/person. With regard to the dissertation, thank you for offering to help with IOA. With regard to life, just thank you for being you. I am excited to say, "I know her," when people talk of your accomplishments in the future. I am always proud of you and am lucky to have you in my life.

ABSTRACT

Chronic tic disorders (CTDs), including Tourette's disorder and persistent motor or vocal tic disorder, are neurobiological conditions affecting an estimated 3 to 4 percent of children and adolescents. These disorders include the presence of motor and/or phonic tics, which can range in number, frequency, and severity. Although CTDs are typically treated through medications, the available medications have the potential of adverse side effects, do not result in long-term coping strategies, and may not be effective or preferable for all individuals. Habit reversal training (HRT), a behavioral intervention for tics, has been identified as a well-established treatment. The purpose of habit reversal is to build an individual's awareness of his or her tics and disrupt tics through developing a competing behavioral response.

One avenue or service delivery that has not yet been explored for individuals with CTDs is use of a parent-mediated approach to habit reversal. Parent-mediated interventions have been used successfully with children with challenging behavior and autism. They are based on a triadic model, in which a therapist works directly with a parent to teach the therapeutic techniques. Parents, in turn, work directly with their children while receiving feedback from the therapist.

The current study investigated a parent-mediated habit reversal intervention for the treatment of chronic tic disorders in two children. A delayed multiple baseline design was used. Baseline data were collected for three sessions. Intervention was delivered over six sessions, during which time coaching was faded. Follow up data collection occurred one month after the end of treatment. Based on changes in parental fidelity of implementation from baseline to intervention, this study provides preliminary evidence for parents being trained as therapists and providing habit reversal training strategies to their children. This treatment was reported as

acceptable by both parent participants and by one of the child participants. Treatment motivation remained high and stable throughout baseline, intervention, and follow up. Child motivation was more variable during baseline, intervention, and follow up. Tic severity was also variable and more research would be needed to determine the impact of treatment for tics. Limitations and implications for future research are provided.

PUBLIC ABSTRACT

Chronic tic disorders (CTDs) affect about 3 to 4 percent of children. Although CTDs are usually treated through medication, they may have negative side effects and individuals may not respond positively. Other behavioral treatments have emerged, of which habit reversal training (HRT) has been identified as effective. HRT has resulted in decreased tics and increased coping skills for children with tics.

One potential method of delivering this treatment not yet been explored is a parent-mediated, or parent-led, approach to HRT. The purpose of the current study was to teach parents HRT techniques to use with their children with a CTD. Training parents to be co-therapists may help with early difficulties experiences by children with CTDs and may result in more intervention time, more generalization of skills, fewer symptoms of CTDs, and decreased family stress. In rural areas, it may also address concerns with access to trained therapists.

The current study used a delayed multiple baseline design with two families. Baseline data were collected for three weeks, followed by 6 weeks of intervention and follow up after one month. Data were collected on parents' ability to implement the strategies, child tic frequency, motivation to participate in treatment, and treatment acceptability. Overall, this study provides initial evidence for a parent-led version of habit reversal based on parents' ability to provide treatment. Parents found the treatment acceptable and were motivated to participate. Child treatment acceptability, motivation, and tic frequency were more variable. Limitations of this study and future research studies are discussed.

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CHAPTER ONE

INTRODUCTION

Chronic tic disorders (CTDs), including Tourette's disorder and persistent motor or vocal tic disorder, are neurobiological conditions that affect an estimated 3 to 4 percent of children and adolescents (Roessner, Hoekstra, & Rothenberger, 2011a). These disorders include the presence of motor and/or phonic tics, which can be simple or complex and which can range in number, frequency, and severity (Jankovic & Kurlan, 2011; Piacentini, Chang, Snorrason, & Woods, 2014; Robertson, 2012; Singer, 2005). CTDs often develop in childhood, become worse in adolescence, and tend to decrease in severity by adulthood (Robertson 2008a). While the specific etiology is yet undefined, available evidence suggests a combination of genetic and environmental factors plays a role in the expression of these disorders (Cavanna & Termine, 2012; Swain, Scahill, Lombroso, King, & Leckman, 2007). Although CTDs are typically treated through medications, behavioral interventions have gained support in the literature as a well-established treatment modality for persons with CTDs (Cook & Blacher, 2007). One potential avenue that has not yet been explored for individuals with CTDs is use of a parent-mediated behavioral intervention. Parent-mediated interventions have been used successfully with children with challenging behavior (Eyberg, Nelson, & Boggs, 2008) and autism (McConachie & Diggle, 2007). The purpose of this study was to expand the available research regarding interventions for CTDs by implementing a parent-mediated version of habit reversal therapy. This chapter includes an historical background of tic disorders, a discussion of diagnostic criteria and how they have changed over time, a brief review of available treatments for CTDs, an overview of habit reversal raining, a rationale for parent-mediated interventions, and the specific purpose for the current study.

History of CTDs

Georges Gilles de la Tourette is broadly recognized as the first person to describe the symptoms for the syndrome that would eventually be associated with his name; however, the history of tic disorders extends to a few cases prior to Gilles de la Tourette's seminal description (Goetz, Chmura, & Lanska, 2001; Rickards, Woolf, & Cavanna, 2010). The first such mention of tics may have been in 1489 by Jacob Sprenger and Heinrich Kramer, Dominican clergymen who discussed motor and phonic tics in a fellow priest (Goetz et al., 2001). Later in the 17th century, written reports were created regarding the Prince de Conde in France who would put objects in his mouth in an attempt to stifle his involuntary noises (Goetz et al., 2001). The first medical case description was published by Jean Marc Gaspard Itard, a French physician, in the *Archives Generales de Medecine* in 1825 (Goetz et al., 2001; Kushner, 1999). In his description, he detailed a woman, Marquise de Dampierre, who displayed what is now known as coprolalia, or unintentional profanity or inappropriate language, which she would shout at others. She also experienced involuntary motor movements. Itard attributed these movements to an "underdeveloped will" as her cognitive abilities appeared to be average (Kushner, 1999). Itard's case would eventually be sent to Jean-Martin Charcot, the leading neurologist of the time and the mentor of Gilles de la Tourette (Kushner, 1999).

Before the publication of Gilles de la Tourette's work, Armand Trousseau also described similar symptoms, which were published after his death in 1873 (Rickards et al., 2010). He described a chronic, hereditary disorder of "nonpainful tics," or sudden involuntary movements and vocalizations. He believed these tics were located primarily in the facial region, but also recognized that they could affect other parts of the body. His descriptions were similar to the current presentation of tic disorders; however, his publication was based on a small sample of

anecdotal observations. While Gilles de la Tourette acknowledged Trousseau in his publication, he also disagreed with some of the descriptions, which may have contributed to Trousseau's work not reaching the significance of Tourette's (Rickards et al., 2010).

In the late 1880s and after receiving case reports from around the world and treating individuals with tics, Jean-Martin Charcot instructed his student, Georges Gilles de la Tourette, to describe his collection of case studies for publication (Kushner, 1999). These case descriptions that would eventually be associated with Gilles de la Tourette's name were published in an article in the *Archives de Neurologie* in 1885 (Goetz & Klawans, 1982; Piacentini et al., 2014; Lajonchere, Nortz, & Finger, 1996). His article outlined nine case studies of French men and women who demonstrated similar features. Marquise de Dampierre, described previously, was the first case study included. Gilles de la Tourette described a variety of symptoms of involuntary, rapid, and sometimes inappropriate movements and vocalizations, as well as repetitive speech and motor movements. He noted that each individual was affected in different ways, but that the "disease" typically manifested during childhood. He also discussed the potential for the symptoms to have a hereditary basis. He reported the disease had almost a degenerative nature, in that tics became worse over time; however, the patients appeared to have no disruptions in their cognitive abilities. In addition, he described what would now be considered the waxing and waning nature of tics (Goetz & Klawans, 1982; Kushner, 1999). Much of his description still holds true in the current diagnostic criteria and in our current understanding of tic disorders. At the time, he also described the disorder as being completely resistant to intervention, a conclusion which has changed over time (Goetz & Klawans, 1982).

Tourette's description of the cases as representative of a separate disorder was challenged after publication (Kushner, 1999). Very few patients outside of those described by Tourette

seemed to "fit" his description. Other physicians believed the symptoms to be part of a diagnosis of hysteria or a result of infection from rheumatic fever (i.e., chorea). These beliefs prevailed and the disorder described by Tourette was attributed to these other diagnoses (Kushner, 1999).

The original mode of treatment for Tourette's disorder and other tic disorders was primarily psychoanalysis, due to the growing popularity of this treatment orientation in the early 20th century, as well as the general lack of effective neurological treatments at the time (Piacentini et al., 2014). One implication of a psychoanalytic model for tic disorders was the view of the individual as having a character flaw or as lacking willpower for change. For example, in the first psychoanalytic description of tics, Ferenczi (1921) described the sensations and thoughts surrounding tics as "pathological expressions of the patient's sexuality and of hypochondriacal narcissism in particular" (p. 9). He described tic patients as "of a mentally infantile character" (p. 10) based on their emotional descriptions of feeling tics. Ferenczi advocated for long-term psychoanalysis for individuals experiencing tics, despite the fact that he had never worked with a patient exhibiting tics (Kushner, 1999). This point of view became the prevalent model, which further resulted in a complete misunderstanding of symptoms, lack of advocacy on behalf of the individuals experiencing tics, lack of relief from tics, and increased self-blame for individuals with tics (Chang & Piacentini, 2007). Because of the popularity of the psychoanalytic view of tics, the conversations shifted from biological models to a psychological model.

A few events occurred beginning in the 1940s that would challenge the psychoanalytic model for CTDs. First, in the 1940s, multiple outbreaks of infectious encephalitis occurred, which led to surgery to remove sinuses or tonsils. It was discovered that removal of sinuses or tonsils led to remission of tics, leading some physicians to believe that the psychoanalytic view

was not completely accurate (Kushner, 1999). However, psychoanalysts had a stronghold on treatment at the time. Based on the "post-infectious theories" and to assuage both sides of the argument (i.e., psychoanalytic versus medical), an agreement was reached by psychologists and medical professionals that individuals with a clear history of infection prior to tics would receive surgery. Without this history, individuals would receive psychoanalysis (Kushner, 1999).

Medical advances continued in the 1950s, while psychoanalysis still remained the prevailing treatment (Kushner, 1999). In 1954, Clauss and Balthasar, German neurologists, discovered lesions in the striatum of individuals with tic disorders in post-mortem studies. In addition, lobotomies and leucotomies were becoming popular in America, and it was discovered that by severing or disconnecting parts of the brain associated with motor functioning, tics abated. Despite this evidence, it was not until the 1960s, after success with the dopamine antagonists chlorpromazine and haloperidol, that the treatment started shifting from psychoanalysis to medical treatment based on a neurobiological model (Chang, Piacentini, & Walkup, 2007; Kushner, 1999).

The shift to a neurobiological model resulted in consequences for the field of psychology in general (Chang et al., 2007). In the 1950s, behavioral treatments for tics began to appear in the literature; however, these were primarily overlooked due to the popularity of psychoanalysis at the time. With the shift to a neurobiological model and the lack of success with psychoanalysis, all psychological interventions were rejected. Medical practitioners had observed the stigma that rose out of psychoanalysis, namely that individuals were blamed for having tics. They strongly recommended against all psychological interventions for fear that the stigma would return, leading to further harm for individuals with CTDs.

Despite warnings regarding psychological treatments, a behavioral procedure for the reduction of tics and nervous habits was developed and initially described in a study by Nathan Azrin and Gregory Nunn (1973). Even with promising results for tics, this procedure, called habit reversal training (HRT) has only relatively recently been more accepted as a treatment modality for tic disorders, thanks in part to the rise of a biopsychosocial model of tic disorders in the late 1990s and early 2000s. A more comprehensive behavioral model will follow later in this chapter, but the biopsychosocial model recognizes that CTDs have a neurological basis while also recognizing that factors within an individual's environment play a role in evoking, reducing, or maintaining tics (Chang & Piacentini, 2007).

Diagnostic Information

A medical "test" for tic disorders does not currently exist (Swain et al., 2007). Diagnoses of tic disorders are most often made by neurologists, primary care physicians, or psychiatrists, and less frequently by psychologists and therapists (Woods et al., 2010). Neurologists, psychiatrists, and others in the mental health field may not be the first point of contact for individuals with possible tics and tend to become more involved when comorbidity is a concern (Murphy, Lewin, Storch, Stock, and the AACAP Psychiatry Committee on Quality Issues, 2013). Recommendations for best practice include comprehensive assessment, consisting of a clinical interview focused on a thorough family and personal history, clinical observation, symptom checklists, and neurological exam (Murphy et al., 2013; Robertson, 2012; Scahill et al., 2006). Diagnostic assessment should be comprehensive given the potential for differential diagnosis and comorbidity (Robertson, 2012). The following is a discussion of the diagnostic criteria and general profile of CTDs.

Core Symptoms: Tics and the Premonitory Urge

Inherent in the name, a common aspect for all individuals with CTDs are the presence of tics. Tics are sudden, quick, repetitive, nonrhythmic movements or vocalizations which tend to be involuntary (Jankovic & Kurlan, 2011; Piacentini et al., 2014; Robertson, 2012; Singer, 2005). Tics often resemble typical behavior but are typically more intense and/or repetitive. They tend to wax and wane over time and vary in their frequency and intensity (Jankovic & Kurlan, 2011). In addition to frequency and intensity, tics can be clinically assessed based on their bodily location, number, and complexity (Leckman, 2003). Tics tend to vary in their topography such that one individual's clinical expression of tics can change over time (Cavanna & Termine, 2012; Jankovic, 1997). Tics are also suppressible and suggestible (Piacentini et al., 2014). Suppressibility refers to an individual's ability to stop tics from occurring for brief periods of time, which is usually uncomfortable and effortful (Banaschewski, Woerner, & Rothenberger, 2003; Baym, Corbett, Wright, & Bunge, 2008). This feature suggests that tics may be under partial voluntary control (Swain et al., 2007), but it may be misunderstood as the individual "choosing" to tic or that tics are easy to stop (Murphy et al., 2013). Suggestibility refers to demonstrating tics while talking about an individual's tic history, which usually includes expression of older tics that have waned (Leckman, 2003).

Tics are typically classified in two ways (Jankovic, 2001; Piacentini et al., 2014). First, tics can be classified as motor or phonic. Motor tics involve an involuntary movement of the body. They can further be described by the type of movements as clonic, dystonic, or tonic (Jancovic & Kurlan, 2011). Clonic tics are brief and appear jerky. Dystonic tics are slower and involve a brief pause in movement, resulting in an abnormal sustained posture. Tonic tics involve isometric muscle tensing. Motor tics tend to follow a cephalocaudal trend, meaning that

they tend to start with the face or head and move downwards. A common first motor tic is eye blinking (Grados & Mathews, 2009). Phonic tics, also referred to as vocal tics, involve the production of involuntary sounds. A common first phonic tic is throat clearing (Grados & Mathews, 2009). Arguments have been made regarding whether or not the separation of motor and phonic tics is necessary, as many phonic tics involve the contraction of muscles instead of solely vocal cords; however, diagnostic criteria still divide tics using motor and phonic descriptors (APA, 2013; Jancovic, 1997; Jancovic & Kurlan, 2011).

Next, tics can be categorized as simple or complex (Jankovic, 2001; Piacentini et al., 2014). Simple motor tics involve one muscle group in one area of the body, which tends to yield a sudden, quick, and jerky movement. Examples of simple motor tics include eye blinks, shoulder shrugs, head jerks, and facial grimaces. Simple phonic tics tend to result in a quick meaningless sound, such as single sounds (e.g., /ah/ or /ee/), throat clearing, sniffing, or coughing. Complex motor tics involve a coordinated series of actions by different muscle groups of the body. These tics are often slower than simple motor tics and are more likely to resemble typical actions. Examples of complex motor tics include arching the back, jumping, touching objects, hand and arm gestures, and leg movements. Complex vocal tics include meaningful utterances or verbalizations, such as repetitive words or phrases, echolalia (i.e., immediate repetition of others' speech), palilalia (i.e., delayed repetition of others' speech), or coprolalia (i.e., repetitive use of obscene language; Jankovic, 2001; Piacentini et al., 2014). In general, both motor and phonic tics progress from simple to complex over time (Bornstein, King, & Carroll, 1983).

Tics often need to be sdifferentially diagnosed from other disorders with repetitive movements (e.g., autism, Sydenham's and Huntington's chorea, stereotyped movement disorder,

Wilson's disease) through the presence of a premonitory urge, although age of onset, history of simple tics, and history of phonic tics are also used to help differentiate disorders (Jankovic & Kurlan, 2011; McCracken, 2000; Swain et al., 2007). Joseph Bliss (1980) helped to build awareness to the internal sensory experiences of tic disorders through description of his own journey with Tourette syndrome, which eventually led to a better understanding of tics and the premonitory urge. A premonitory urge is a sensation that occurs prior to engaging in a tic that may build if a tic is not performed (Himle, Woods, Conelea, Bauer, & Rice, 2007; Jankovic, 2001). It is usually unpleasant or uncomfortable, and may present as feelings of tension, tightness, pressure, itchiness, or energy (Banaschewski et al., 2003; Jankovic & Kurlan, 2011; Singer, 2005). The feelings may be localized to a specific anatomical area of the body or may be more general (Banaschewski et al., 2003). These urges may sometimes be more bothersome or unbearable than the tic itself (Himle, Woods, Conelea, Bauer, & Rice, 2007; Leckman, 2003). While not every individual reports experiencing a premonitory urge, it has been estimated that up to 90 percent of individuals with a CTD report some type of somatic sensation prior to engaging in a tic (Kwak, Vuong, & Jankovic, 2003). Awareness of the premonitory urge also seems to increase with age, which has been hypothesized to be associated with cognitive development rather than CTDs themselves (Banaschewski et al., 2003). Children under the age of 10 may have particular difficulty describing the premonitory urge without assistance (Leckman, King, & Cohen, 1999).

Some evidence suggests a potential functional relationship between the premonitory urge and the occurrence of a tic (Himle et al., 2007). Based on a reported increase in the premonitory urge when tics are suppressed, it is believed that the relationship between tics and the urge involves an automatic negative reinforcement loop, during which a tic occurs to alleviate the

uncomfortable feeling of the premonitory urge (Himle et al., 2007; Singer, 2005). Kwak and colleagues (2003) found that 72 percent of individuals in their sample reported relief from the premonitory urge. The premonitory urge may help an individual to recognize that a tic is going to occur, which may lead to an attempt to suppress the tic (Mink, 2001). This urge has become the primary target for many behavioral treatments (Piacentini & Chang, 2006).

Diagnostic and Statistical Manual of Mental Disorders (DSM) Criteria

Tic disorders were first included in the third edition of the *DSM-III* (APA, 1980) and included Tourette's disorder, chronic motor tic disorder, transient tic disorder, and atypical tic disorder. Tourette's disorder required both motor and phonic tics and had to be present between the ages of 2 and 15 years. Symptoms had to be present for over a year. Chronic motor tic disorder was characterized by an absence of phonic tics and also had to be present for at least a year, with no specific indication of age of onset. Transient tic disorder was diagnosed for tic symptoms that persisted for at least one month but less than a year. Atypical tic disorder was used when tics were present, but did not meet symptom criteria for the other disorders.

Over time, the diagnostic criteria and nomenclature of some tic disorders have changed (Robertson, 2008a). In the *DSM-III-R* (APA, 1987), tic disorders included Tourette's disorder, chronic motor or vocal tic disorder, transient tic disorder, and tic disorder not otherwise specified. A diagnosis of Tourette's disorder still required multiple motor tics and at least one phonic tic. It also required that tics occur frequently throughout the day and be present for over a year with an age of onset before 21. In addition, substance use and the presence of other medical causes of tics had to be ruled out. The diagnosis of chronic motor tic disorder was expanded to include chronic vocal tic disorder. The criteria were similar in that individuals had to exhibit motor or phonic tics for more than one year. Like Tourette's disorder, the onset had to be before

21. Substance use and other medical disorders also had to be ruled out. Transient tic disorder evolved in that tics had to occur for at least two weeks but not longer than one year and had to occur prior to age 21 as well. Tic disorder not otherwise specified was similar to atypical tic disorder in that it was used for individuals who had tics but did not meet criteria for any of the above tic disorders.

When the *DSM-IV* (APA, 1994) was developed, the names of the tic disorders remained the same; however, two significant changes were made. First, the age of onset for Tourette's disorder, chronic motor or vocal tic disorder, and transient tic disorder was moved to before age 18. Also, a statement about the requirement for "clinically significant distress or impairment" was added to all the tic disorders. With the publication of the *DSM-IV-TR* (APA, 2000), the statement about distress or impairment was removed, as it was noted that this statement limited clinicians' diagnostic ability as many children with tic disorders do not experience significant distress or impairment (APA, 2000).

With the change to the *DSM-5* (APA, 2013), CTDs have slightly changed again. First, tic disorders are now classified under the broad category of Neurodevelopmental Disorders, as the category of Disorders Usually First Diagnosed in Infancy, Childhood, or Adolescence in the *DSM-IV* was discontinued. They are also under a broader subcategory of Motor Disorders, which includes other diagnoses such as developmental coordination disorder and stereotypic movement disorder. This category also did not exist in the *DSM-IV*. Further, the names of some of the disorders have changed. Chronic motor or vocal tic disorder is now persistent motor or vocal tic disorder. Transient tic disorder is now provisional tic disorder. Tic disorder not otherwise specified has been removed and has been replaced with other specified tic disorder and unspecified tic disorder. Both of the latter diagnoses can be given if an individual demonstrates

symptoms of tic disorders but does not meet the full criteria. They are differentiated based on whether the diagnosing clinician chooses to state the reason that an individual does not meet criteria for another tic disorder. Lastly, in the description of tics, the word "stereotyped" was removed in the hopes of preventing confusion between tics and stereotyped behaviors. Overall, even though there have been some changes to the names, the criteria from the *DSM-IV-TR* to the *DSM-5* remain largely unchanged.

Over time, while there have been relatively few changes to the diagnostic criteria and classifications, there has been considerable disagreement among diagnosing professionals regarding which symptoms to include (Kushner, 1999). Examples of symptoms that are not included but are often cited as "belonging" to CTDs include obsessive-compulsive symptoms and explosive behavioral outbursts. Other disagreements have surrounded the age of onset criteria, as well as the length of time symptoms must be present to diagnose the CTDs (Kushner, 1999). In addition, concerns have arisen regarding the actual diagnosis of CTDs. While the diagnostic criteria for CTDs appear straightforward, considerable variation in expression exists, which may lead to misdiagnosis. Thus, comprehensive assessment by individuals familiar with CTDs prior to diagnosis is warranted (Robertson, 2008a).

Prevalence and Epidemiology

CTDs used to be considered as very rare; however, with the increase in tic disorder research in the 1980s and 1990s, which included more prevalence and family studies, this view changed (Jancovic & Kurlan, 2011; Robertson, 2003; Robertson, 2008a). Prevalence rates for simple and transient tics during childhood are estimated to be between 6 to 20% of all children (Robertson, 2003). These statistics suggest that tics are a fairly common occurrence in childhood; however, the true prevalence rate for transient tics is unknown because they tend to

be brief and do not persist over time (APA, 2013; Bruun & Budman, 1997). Most individuals experiencing tics have transient tics and will not eventually meet criteria for a CTD (Mink, 2001). In childhood populations (i.e., children between the ages of 6 and 17), Tourette's disorder, usually considered as more severe than other tic disorders, is estimated to occur in about 0.5 to 1 percent (Robertson, 2008a), though there is variability in the literature. Other tic disorders are more common, with prevalence rates between 1 to 29 percent reported in the literature (Robertson, 2008a), and a rate of up to 3 to 4 percent most commonly reported (Roessner et al., 2011).

Males are more likely than females to be diagnosed with Tourette's disorder by a mean ratio of about 4 to 1 (APA, 2013; CDC, 2009; Freeman et al., 2000). The reason for the disparity in Tourette's disorder diagnosis based on gender is unclear; however, it is likely due to come combination of genes, environment, and potentially hormones (CDC, 2009). In addition to diagnosis, males are more likely to demonstrate a comorbid condition (e.g., AD/HD, OCD, learning disorders, speech difficulties, etc.) and are more likely to have taken a medication for tics than females (Freeman et al., 2000).

Based on the available literature, tic disorders occur in almost all countries, though at different rates (Robertson, 2008a). Internationally, Tourette syndrome has been reported to occur in 0.4 to 3.8 percent of individuals, and in 1 percent of childhood populations. It appears to be more common in individuals from European descent and significantly less common in individual from sub-Saharan black Africa. Worldwide, the phenomenology and presentation of tic disorders is similar. CTDs also occur across social classes. Additionally, the male to female ratio remains relatively constant across cultures (Robertson, 2008a).

Natural Course of Symptoms

Based on the available literature, the onset of tics ranges from age 2 to 21 years, with a mean age of onset for motor tics between 5 to 7 years (CDC, 2009; Freeman et al., 2000; Robertson, 2008a). The onset of phonic tics specifically tends to be slightly later, with age 11 years being most commonly reported (Robertson, 2008a). Tics tend to increase in intensity over time with worst-ever tics reported on average at age 10 years (Leckman et al., 1998; Robertson, 2003). A diagnosis of Tourette's disorder is twice as likely in individuals between the ages of 12 and 17 years compared to children between the ages of 6 and 11 years (CDC, 2009).

After an increase and peak in adolescence, tics tend to decrease in severity by early adulthood (CDC, 2009; Leckman et al., 1998). Initial estimates suggested that two thirds of individuals with CTDs would experience amelioration of tic symptoms by adulthood (Bruun & Budman, 1997), with one third of individuals essentially tic free as adults and with one third only experiencing mild symptoms as adults (Bloch et al., 2005b; Roessner et al., 2011). Other estimates suggest that by age 18, roughly half of individuals with CTDs in childhood no longer demonstrate tics as adults (Jankovic & Kurlan, 2011; Leckman et al., 1998). Even though tics tend to decrease over time, they still have the potential to continue into adulthood. Most often tics observed in adulthood tend to involve the face, neck, or trunk (Jankovic, Gelineau-Kattner, & Davidson, 2010; Jankovic & Kurlan, 2011). They also tend to be a "reemergence" of tics experienced in childhood as opposed to newly developed tics, and tic persistence into adulthood is partially based on severity of tics during childhood (Jankovic et al., 2010). More severe symptoms, such as tics that result in self-injury, or tics involving coprolalia or copropraxia, may be more common in adulthood (Swain et al., 2007). Additionally, when compared with childhood samples, adults with CTDs may be more likely to have substance abuse or mood

disorders, whereas children are more likely to demonstrate AD/HD and oppositional behavior and to be on medication for the treatment of tics (Jankovic et al., 2010; Pappert, Goetz, Louis, Blasucci, & Leurgans, 2003).

While it is less common to be diagnosed in adulthood, Freeman and colleagues (2000) found that 16% received a diagnosis after age 18 years based on analysis of their database of 3500 individuals with Tourette's disorder from 22 different countries. A diagnosis of Tourette's disorder may be delayed or missed if the symptoms are misidentified (e.g., viewing constant sniffling as "just allergies"), if the symptoms are mild or atypical, or if the family has lack of access to healthcare specialists (Center for Disease Control and Prevention [CDC], 2009). If tics first appear in adulthood without a history of tics in childhood, it is suggested to examine other potential causes other than tic disorders, such as drug use, brain injury, infection, or trauma (Jankovic & Kurlan, 2011).

Comorbidity

Individuals diagnosed with Tourette's disorder frequently have a comorbid condition (Termine et al., 2006). According to the CDC (2009), 79 percent of children with Tourette's disorder were reported by their parents to have at least one co-occurring neurodevelopmental or mental illness diagnosis. Other investigations have estimated that as many as 85 to 90 percent of individuals with CTDs have another comorbid condition (Cavanna et al., 2011; Cavanna & Rickards, 2013; Hirschtritt et al., 2015). On average, individuals with CTDs tend to have at least two other comorbid conditions (Freeman et al., 2000) with some estimates suggesting more than half of individuals will have two comorbid disorders (Hirschtritt et al., 2015). The most common comorbidity is attention deficit-hyperactivity disorder (AD/HD), followed by OCD, behavioral issues or conduct problems, anxiety and mood disorders, and developmental disabilities,

particularly autism (Baron-Cohen, Scahill, Izaguirre, Hornsey, & Robertson, 1999; CDC, 2009). It is estimated that approximately 30 to 90 percent of individuals with CTDs have a co-occurring diagnosis of AD/HD (CDC, 2009; Freeman et al., 2000). Individuals with diagnoses of CTDs and AD/HD are rated as more likely to have disruptive behavior than individuals with either diagnosis alone (Kraft et al., 2012; Sukhodolsky et al., 2003). Obsessive compulsive disorder (OCD) is also a common comorbidity and is estimated to co-occur with CTDs in 20 to 60 percent of cases (CDC, 2009; Freeman et al., 2000). Severity of tics in early childhood has been found to predict OCD symptoms in late adolescence and early adulthood. OCD symptoms arising in late adolescence were also found to predict tics in adulthood (Peterson, Pine, Cohen, & Brook, 2001). Some individuals will have a combination of Tourette's disorder, AD/HD, and OCD. These three diagnoses all have difficulties inhibiting inappropriate behavior as a common feature (Mink, 2001). Individuals diagnosed with all three diagnoses tend to have the highest rates of difficulty regulating anger, as well as difficulty sleeping (Freeman et al., 2000).

Anxiety and depression also tend to co-occur with tic disorders (Robertson, 2000). Anxiety symptoms for this population may include difficulty sleeping, worries or phobias, motor restlessness, or habits arising from tension. Depression may occur due to adjustment with having a chronic illness or as a result of bullying. Robertson (2000) highlighted how anxiety and depression are common in general, and it is possible that higher rates of these mood disorders with individuals with tic disorders may occur by chance.

Most children with CTDs are functioning within the average range of intelligence. However, individuals with CTDs have also been found to have elevated rates of learning disabilities, especially with a co-occurring diagnosis of AD/HD (Burd, Freeman, Klug, & Kerbeshian, 2005; Singer, 2010). Even without a specific learning disability diagnosis, an

estimated 16 to 68 percent of individuals with CTDs will demonstrate academic underachievement. This difficulty in school may be due to cognitive difficulties, as some children with CTDs may struggle with executive functioning, difficulties with visual motor integration, and impairment in visual-spatial abilities (Singer, 2010).

Etiology of Tic Disorders

Genetics

Tourette's disorder is widely recognized as a heritable disorder with a significant genetic influence; however, no single gene has been identified (Cavanna & Termine, 2012; Robertson, 2003). Some evidence has suggested genetic and chromosomal abnormalities occurring in tic disorders; however, these abnormalities have not been consistently found in the literature for all individuals with tics (Cavanna & Termine, 2012; Swain et al., 2007). At least 10 chromosomes have been implicated in the expression of Tourette's disorder and further research may discover more (Robertson, 2008c). A few hypotheses exist for why a single gene has not been identified, including the heterogeneity of the disorder and the potential for different responsible genes in different families (Robertson, 2003). In addition, interaction with the environment has also been proposed as a requirement for expression of tic disorders (Robertson, 2012). Currently, the heritability of tic disorders is considered to be complex and involve multiple genes (Jankovic & Kurlan, 2011).

Family history of psychiatric disorders has also been associated with CTDs. Khalifa and Von Knorring (2005) found that over 80 percent of individuals with a tic disorder had a first-degree family member with a known psychiatric diagnosis, the most common of which were tic disorders, AD/HD, OCD, or depression. For individuals with a first degree relative with Tourette's disorder, there is a higher risk (i.e., approximately 35 percent) of these individuals

developing CTDs or OCD (Pauls, Alsobrook, Gelernter, & Leckman, 1999). In addition, twin studies investigating hereditary transmission have also yielded a high concordance rate (i.e., 77 percent) of CTDs in monozygotic twins versus dizygotic twins (i.e., 23 percent), suggesting significant but not complete genetic influence (Price, Kidd, Cohen, Pauls, & Leckman, 1985; Robertson, 2012; Swain et al., 2007).

Neurobiology

The relatively common occurrence of tics in childhood versus adulthood suggests that the developing nervous system may have different features contributing to tics compared to a matured nervous system. In addition, the increase in tics that commonly occurs during adolescence as well as the disparity in diagnosis between males and females suggests hormones, specifically androgen, may play a role in tic expression (Mink, 2001).

Discussions in the literature of brain structures involved in tic disorders tend to include the basal ganglia, a collection of connected subcortical nuclei located in the forebrain and midbrain (Mink, 2001). The basal ganglia receive input from all parts of the cortex and have direct connection with the thalamus and other limbic structures. These structures form the cortico-striato-thalamo-cortical (CSTC) circuits, regions of the brain associated with motor function (Yoon, Gause, Leckman, & Singer, 2007). In individuals with tic disorders, parts of the basal ganglia, specifically the caudate nucleus and lenticular nucleus, have been found to have less volume when compared to neurotypical controls (McCracken, 2000; Peterson et al., 2003). Reduced volume of the caudate nucleus in childhood has been found to be predictive of higher severity of tics in adulthood (Bloch, Leckman, Zhu, & Peterson, 2005a).

A few explanations have been proposed for what brain processes might lead to the production of tics. First, within the CSTC circuit, particularly in the basal ganglia and frontal

lobes, there may be some abnormality of neurotransmitter release (Harris & Singer, 2006). Abnormalities in dopamine production in these areas are most often cited. This explanation is supported by clinical observations of blocking dopamine receptors, as well as a decrease in tics after taking medications that stimulate dopamine production; however, studies to examine this explanation have yielded inconsistent results (Harris & Singer, 2006; Mink, 2001; Yoon et al., 2007). It is likely that dopamine interacts with other neurotransmitters (e.g., acetylcholine, GABA, glutamate) in cases of CTDs (Mink, 2006)

A second explanation involves the output of the basal ganglia. In individuals with tic disorders, the basal ganglia may have abnormalities in inhibitory output to the cortex and other brain structures responsible for movement and vocalizations. These abnormalities may lead to a failure to inhibit unwanted movements or vocalizations (Cavanna & Termine, 2012; Mink, 2001). This model has been used to explain the high comorbidities and potential commonalities in neural circuitry for tic disorders, AD/HD, and OCD (Cavanna & Termine, 2012; Mink, 2001).

In addition to the basal ganglia, other structural abnormalities in the brain have been reported for tic disorders. Specifically, sensory and motor neuron thinning, enlarged limbic structures and prefrontal cortex, reduced cerebellar hemispheres, and a reduced corpus callosum have been observed in individuals with tic disorders (Robertson, 2012). These differences in brain structure may play a role in symptom expression and severity. As Rickards (2009) explains, the neurobiology of CTDs encompasses many different neural structures which interact in complex ways to result in the expression of tics.

Environmental Factors

Evidence from the literature suggests a stress diathesis model for tics involving interaction of genes and factors in the environment to contribute to the expression of tic disorders

(Cavanna & Termine, 2012; Swain et al., 2007). Specific environmental factors contributing to the expression of tic disorders have been studied less than genetic and neurobiological underpinnings, and have yielded somewhat inconsistent results (Mathews et al., 2006). Tic disorders have been associated with infections (e.g., streptococcus, Lyme disease, viral infection), prenatal issues (e.g., maternal smoking, paternal age, severe maternal emesis, maternal stress), postnatal issues (e.g., inducing labor, delivery by C-section or use of forceps, prematurity, nuchal cord, jaundice) and androgen hormones (Cavanna & Termine, 2012; Hoekstra, Dietrich, Edwards, Elamin, & Martino, 2013; Robertson, 2000; Swain et al., 2007). Prenatal maternal smoking, increased paternal age, and family history of OCD were found to contribute to comorbid OCD for individuals with tic disorders (Mathews et al., 2006). Prenatal maternal smoking and low birth weight have been found to contribute to comorbid AD/HD (Pringsheim, Sandor, Lang, Shah, & O'Connor, 2009).

Variables within an individual's environment may serve to exacerbate or attenuate tics (Conelea & Woods, 2008). Setting and antecedent events have been linked with increased tic severity, even after controlling for age (Lin et al., 2007). Tics often increase when an individual is stressed, anxious, or frustrated, and often decrease during periods in which an individual is relaxed (Conelea & Woods, 2008; Robertson, 2000, Silva, Munoz, Barickman, & Friedhoff, 1995). Common stressors noted in the literature have included starting a new school year, family conflicts, and moving (Conelea & Woods, 2008; Silva et al., 1995). Increases in tics have also been observed in times of fatigue, during social events (e.g., holidays, birthday parties), when bored, during academic tasks, and in the presence of specific individuals (Conelea & Woods, 2008). They may also increase during periods of relaxation after stressful situations (Mink, 2001) and when talking about tics (Woods, Watson, Wolfe, Twohig, & Friman, 2001).

Decreases in tics have been observed when individuals are specifically asked to suppress tics, during times of concentration, during leisure activities, and during clinic or doctor visits (Conelea & Woods, 2008). Tics also tend to decrease, but not abate, during sleep, though they may increase during REM sleep (Singer, 2005).

Consequence variables, which are environmental events or changes that occur after tics, also have the potential to influence tic expression, though have been studied less in the literature than setting events (Conelea & Woods, 2008). Increases in the likelihood of future tics have been associated with being removed from a setting or activity and attention surrounding tic occurrence from others. Decreases in the likelihood of future tics have included rewarding tic-free periods and rewarding compliance with habit reversal procedures (Conelea & Woods, 2008).

While a variety of environmental variables have been identified in relation to tics, the results have been inconsistent in that not every individual with tics will experience them in the settings mentioned above. It is possible that it is not tics themselves that are exacerbated or attenuated, but tic suppression or inhibition (Conelea & Woods, 2008).

Impact of Tic Disorders

While some individuals with tic disorders may not report any associated impairment from tics (Coffey et al., 2004; Erenberg, Cruse, & Rothner, 1987), research suggests that tic disorders can be associated with impairments in a variety of life domains such as school, home, work, and social situations (Conelea et al., 2011; Storch et al., 2007a). Socially, individuals may experience embarrassment from the presentation of tics, which may also be linked with lower self-esteem (Leckman, 2003). Similarly, they may receive negative social feedback (e.g., criticism) from others who want them to control their behavior. Having difficulty controlling the tics in general may also be frustrating (Swain et al., 2007). Some children with CTDs may be

victims of teasing or bullying, which may be upsetting and impact self-esteem (Lombroso et al., 1995; Swain et al., 2007). The likelihood of peer victimization of children with CTDs increased with tic severity, complexity, and frequency and with the presence of comorbid disorders, especially AD/HD or disruptive behavior disorders (Zinner, Conelea, Glew, Woods, & Budman, 2012). Victimization of individuals with CTDs by peers has been found to be positively correlated with increased parent and self-reported loneliness and anxiety, as well as self-reported lower quality of life (Storch et al., 2007b; Zinner et al., 2012). Children with tic disorders may have particular difficulty making friends (Storch et al., 2007a). Overall, children with tic disorders have reported being treated differently and experiencing discrimination (Conelea et al., 2011).

Individuals with tics are more likely to receive special education services in schools than to be fully in general education classes, despite typically falling in the average to above average range on tests of intelligence (Kurlan et al., 2001; Robertson, 2008a). However, children in general education classes may still experience tics or be diagnosed with tic disorders, but may experience milder symptoms (Robertson, 2000). In some children, tics may be so severe or frequent that they interfere with completing schoolwork (Carter et al., 2000; Leckman et al., 1998). Storch and colleagues (2007a) also found that tics may interfere in schoolwork, with challenges at school noted during writing, completing homework, concentrating on academic tasks, and being prepared for class. Difficulties completing academic tasks may increase with tic severity (Baym et al., 2008).

Children with CTDs rated themselves as having more internalizing symptoms than neurotypical controls (Carter et al., 2000). The presence of comorbid AD/HD was associated with more impairment in the realm of psychosocial functioning, social adaptation, and

externalizing symptoms (Carter et al., 2000; Dykens et al., 1990 Pringsheim, Lang, Kurlan, Pearce, & Sandor, 2008). Gorman and colleagues (2010) also found that older adolescents diagnosed in childhood with Tourette's disorder continue to have significantly poorer psychosocial and global functioning, and tend to have lifetime prevalence of comorbidities, including AD/HD, OCD, major depression, learning difficulty, and significant behavioral challenges when compared to community controls. Ratings of anxiety, depression, conflict within the family, and dissatisfaction with life have been found to be significantly different from neurotypical controls, but similar to other individuals with psychiatric diagnoses (Conelea et al., 2011). With the increase in comorbid conditions, there is an increase in the risk of behavioral issues including difficulty sleeping, social skill deficits, sexually inappropriate behaviors, difficulty regulating anger, and self-injurious behavior (Freeman et al., 2000). Additionally, the presence of comorbid conditions contributes to lower ratings for quality of life, higher ratings of impairment, and higher likelihood of avoiding settings (Conelea et al., 2011).

In rare cases, tics may be more severe and lead to dangerous outcomes (Cheung, Shahed, & Jankovic, 2007). These cases have been called "malignant Tourette syndrome," which is estimated to occur in 5 percent of cases of Tourette syndrome (Cheung et al., 2007). These cases have been associated with higher rates of self-injury and suicidal ideation, which have resulted in increased hospital admissions. Physical injuries can vary widely and include blindness from retinal detachment (Leckman, 2003), damage to oral muscles due to biting, fracturing of arms or legs, and spinal cord injuries, concussions, or even death from neck jerking or head banging (Cheung et al., 2007).

In addition to individuals diagnosed with CTDs, significant others in their lives may also be impacted by CTDs. Schoeder and Remer (2007) found that in families with a child diagnosed

with a CTD, caregivers tend to experience stress. This stress was predicted by higher tic severity and by age, with older caregivers reporting more stress. Caregiver stress was mediated by perceived social support, with more perceived social support being associated with less stress.

Treatments for CTDs

Many children and adolescents with tics do not seek treatment for tics (Bloch, State, & Pittenger, 2011, Robertson, 2008b; Woods et al., 2010). A number of reasons exist for why some individuals do not seek treatment, such as having mild tics, having transient tics, or having tics that may not result in significant impairment in daily living (Roessner et al., 2013). For others, psychoeducation about tics and use of a "watch and wait" strategy may be enough, especially when symptoms are mild (Roessner et al., 2011b). Regardless of tic severity, psychoeducation has been suggested as the recommended first step before making any decisions for more intensive treatment (Singer, 2010). In addition, if comorbid disorders are present, attention should be given to the most impairing symptoms and combination treatment can be considered (Kurlan, 2014). Suggestions have been made regarding when consideration of more intensive treatment for tics would be appropriate (Roessner et al., 2011b). These suggestions include when tics lead to pain, injury, social or emotional difficulties, or impairment in daily life activities (e.g., at school, home, or work).

When treatment for tics is deemed appropriate, medications are often the first line treatment. However, a neurobehavioral model has emerged which combines the neurobiological model with behavior analytic principles, which has begun to allow for the inclusion of behavioral treatments as an adjunct or alternative to medication (Himle, Woods, Piacentini, & Walkup, 2006). To date, no "cure" for tic disorders has been found. The various methods that have been identified for treatment of tic disorders involve reductions in tics. The following sections

describe the current status of treatments for CTDs, with a discussion of the behavioral model of treatment.

Biomedical Interventions

Psychopharmacology. Prescription medications are the most common treatment approach for tics (Piacentini & Chang, 2006), which have been associated with moderate reduction of tics by 30 to 70 percent, depending on the type of medication (Scahill et al., 2006). The current first line medications are alpha-2-adrenoreceptor agonists, including guanfacine and clonidine (Roessner et al., 2013). For moderate to severe tics, or for individuals who do not respond to the alpha-2-adrenoreceptor agonists, antipsychotics are considered, which are associated with a more reliable response for tics (Bloch, 2008; Roessner et al., 2013; Weisman et al., 2013). They are also associated with a higher risk for negative metabolic side effects (Bloch, 2008). Traditional, or typical, antipsychotics used for tics include haloperidol and pimozide (Roessner et al., 2013). Atypical antipsychotics, such as risperidone, ziprasidone, and aripiprazole, have also been used, resulting in fewer significant negative side effects compared to typical antipsychotics (Hartmann & Worbe, 2013).

Botulinum toxin (Botox) injections. For the treatment of tics which are isolated to specific muscle groups (e.g., phonic tics such as throat clearing, simple isometric muscle tensing, eye blinking, etc.), Botox injections have been used (Hartmann & Worbe, 2013; Kurlan, 2014). Botox is injected into the affected muscle group, which removes control of the muscle by the nerve, weakening the muscle and preventing the tic from occurring (Awaad, 1999; Bloch, 2008). The most common tics for which this approach has been used include eye blinking, neck and shoulder tics, and phonic tics (Kurlan, 2014). These injections have been utilized effectively for the reduction of tics and the premonitory urge; however, the benefits are temporary and multiple

injections are often required (Kurlan, 2014; Porta et al., 2003). In addition, negative side effects have been reported such as infection at the site of injection and temporary paralysis of the muscle group (Awaad, 1999; Porta et al., 2003).

Deep brain stimulation. When individuals do not respond to the above medical treatments and tics are severe, deep brain stimulation (DBS) is considered (Muller-Vahl, 2013). DBS involves planting electrodes into the brain centers believed to be responsible for tics and providing electrical stimulation to these centers. It has since been used for over 120 patients worldwide and has demonstrated evidence for significant tic reduction in the majority of individuals (Schrock et al., 2015). However, much of the evidence for this treatment is based on small sample sizes (Mink, 2009) and there is no consensus regarding which locations in the brain to target, nor the strength of the dosage (Bloch, 2008). This treatment also has the highest risk of significant adverse side effects, so careful consideration of its use is required (Muller-Vahl, 2013; Schrock et al., 2015). It is also not currently supported for use with children and adolescents (Murphy et al., 2013).

Behavioral Interventions for CTDs

Behavioral interventions can be considered as a standalone treatment option or as an adjunct to medical treatment, particularly medications (Himle et al., 2006). While medications have been shown to lead to reductions in tics, they may not be the preferred route of intervention for a few reasons. First, they have the potential to lead to short or long-term negative side effects (Waldon, Hill, Termine, Balottin, & Cavanna, 2013). Second, some individuals may not respond to medications, leading to the potential need for more intensive treatments such as DBS (Schrock et al., 2015; Waldon et al., 2013). Third, they do not lead to the development of any strategies to cope with tics (Cook & Blacher, 2007; van de Griendt, Verdellen, van Dijk, & Verbraak, 2013).

A neurobehavioral model for treatment of CTDs. Behavioral interventions are based on a neurobehavioral model of tics (Himle et al., 2006; Franklin, Walther, & Woods, 2010). This model adopts a similar initial premise as the neurobiological model in that it recognizes that CTDs have a neurological basis. However, it adds to the neurobiological model by considering the influence of the environment on tics (Himle et al., 2006; Woods et al., 2008b).

Once tics are exhibited by individuals with CTDs, they can be evoked or strengthened by contextual factors in the environment (Conelea & Woods, 2008; Silva et al., 1995). These factors can be external or internal (Capriotti, Himle & Woods, 2014b). External factors include antecedents (i.e., events that occur prior to tics) and consequences (i.e., events that occur after tics). Internal factors include the premonitory urge as well as strong emotions (Franklin & Woods, 2010). By identifying, evaluating, and adjusting environmental factors where possible, it is hoped that individuals with CTDs can learn tic management and coping strategies to maximally improve quality of life (Himle et al., 2006). Keeping in mind the history of CTDs and their treatment, it is important to note that the neurobehavioral model does not assume that tics are volitional, nor does it assume CTDs are the fault of the diagnosed individuals or their significant others (Capriotti et al., 2014b).

Generally, the neurobehavioral model is not an attempt to override the neurobiological model. It appears to be more worthwhile to consider medication and behavioral interventions as two treatment modalities targeting different aspects of CTDs that can work in conjunction with one another. Medication targets the biological dysfunction, while behavioral intervention focuses on internal and external events in the environment that impact tics (Franklin & Himle, 2007). Together, both interventions can provide comprehensive treatment for tics.

A variety of behavioral approaches have been examined as treatments for CTDs. These approaches include massed negative practice, self-monitoring, contingency management, relaxation training, supportive psychotherapy, cognitive behavioral therapy, exposure and response prevention, and habit reversal training (Cook & Blacher, 2007; Frank & Cavanna, 2013; Franklin, Walther, & Woods, 2010; Woods & Miltenberger, 1995). Of these approaches, only habit reversal training is currently considered as a well-established treatment for CTDs based on the American Psychological Association's (APA) criteria for effective treatments; however, exposure and response prevention is also emerging as a potential treatment option and is considered probably efficacious per the same criteria (Cook & Blacher, 2007).

Habit reversal training. Habit reversal training (HRT) is the most studied behavioral intervention for the treatment of persons with CTDs to date (Franklin et al., 2010; van de Griendt et al., 2013). It was first developed in the 1970s as a multicomponent behavioral treatment to address a range of concerns including tics, trichotillomania (i.e., hair pulling), excoriation (i.e., skin picking), and other habits (e.g., nail biting; Piacentini & Chang, 2006; Woods & Miltenberger, 1995). Azrin and Nunn (1973) developed the original treatment methods based on the concept that tics are typical behaviors that have increased in frequency either due to biology, injury, or psychological trauma. They believed the behaviors became so frequent that they were automatic or habitual, hence some individuals are not always aware of their tics. Therefore, the purpose of habit reversal is to build the individual's awareness of the occurrence of the tics and disrupt him or her from ticcing through the development of a competing response (CR). Azrin and Nunn (1973) described five major components of the original procedures including awareness training (AT), development of a CR, relaxation skills training, contingency management, and generalization procedures. AT and CR are considered to be the main

therapeutic components and have been successfully used alone in a simplified version of the intervention (Miltenberger, Fuqua, & McKinley, 1985); however, it is common to see all of the components used.

AT is implemented to build awareness of the urge to tic, with the goal of making it possible to engage in another action to disrupt tics (Franklin & Himle, 2007). AT involves five separate tasks: response description, response detection, early warning detection, self-monitoring, and functional assessment and intervention (Piacentini & Chang, 2006). In response description, an individual is asked to describe a "target tic" with great detail to a therapist, which may include demonstrating or practicing the tic in person, in front of a mirror, or through use of a video recording. The target tic is often the most distressing to the individual or the most frequent. Next, the individual moves into response detection and is asked to signal every occurrence of the target tic. The therapist may provide prompts during this time to point out when tics occur if the individual fails to acknowledge a tic occurrence. Before moving to the next stage, the goal is that the individual will be able to identify at least 80 percent of tic occurrences. During the next stage of early warning detection, the individual is asked to signal whenever s/he notices the premonitory urge or the first part of the tic's occurrence (Woods et al., 2008b). While this step is not absolutely necessary for HRT to be effective, it may lead to better treatment outcomes ((Woods et al., 2008b). The last two tasks include the individual self-monitoring tics outside of the clinic setting and a functional assessment of situations in which tics are likely to occur and in which treatment exercises are most likely going to be used.

After completing AT, the individual moves into competing response training (CRT). During this phase of treatment, the individual is asked to use a different behavioral response once s/he notices the premonitory urge (Piacentini & Chang, 2006). In developing a competing

response (CR) for the tic, four suggestions have been made regarding the characteristics of the alternative behavior (Piacentini & Chang, 2006; Woods et al., 2008b). First, the CR should be incompatible with the tic, at least initially. Most often, the CR tends to involve isometric tensing of muscles opposite of the muscles involved in the production of the tic (e.g., for arm movements that involve flexing the bicep muscle, focus would be on extending the arm), though the CR may involve performing the tic at a slower pace. Some research has suggested that any focused behavior may be able to be used as a competing response, and that it does not necessarily have to be physically incompatible with the tic (Sharenow, Fuqua, & Miltenberger, 1989; Woods et al., 1999). However, it is still recommended to attempt to find an incompatible response first (Woods et al., 2008b). Next, the CR should be able to be performed for at least one minute or until the urge goes away, whichever is longer. Third, the CR should be less socially noticeable than the tic. Finally, the CR should be able to be integrated into normal activity, such that an individual's life is not completely disrupted by engaging in the competing response. In addition to the above "rules," the CR needs to be performed contingent on the premonitory urge or after a tic has occurred to be maximally effective (Miltenberger & Fuqua, 1985). Once the individual masters use of a competing response, the next most distressing or frequent tic is targeted for treatment.

Contingency management involves strategies that help an individual stay motivated for treatment (Piacentini & Chang, 2006). These strategies include social support training, inconvenience reviews, and reward systems. Social support training involves teaching significant others (e.g., parents, spouses, etc.) to praise an individual's use of the HRT strategies as well as to provide gentle reminders to use the strategies. Social support may have the added benefit of increasing awareness (Franklin & Himle, 2007). Inconvenience reviews involve

creating a list of how tics have been distressing or have led to impairment, as well as why an individual may benefit from doing HRT (Piacentini & Chang, 2006). Finally, reward systems are often developed with children to reinforce participation in HRT and for continued use of the strategies (Woods et al., 2008b).

Relaxation training is also often used in conjunction with HRT as an antecedent-based or preventative strategy. Individuals are taught strategies such as deep breathing or progressive muscle relaxation to use during times of stress, when tics tend to be exacerbated (Piacentini & Chang, 2006). Use of relaxation techniques alone has not been shown to be effective for reducing tics over time (Bergin, Waranch, Brown, Carson & Singer, 1998; Canavan & Powell, 1981; Crawley & Powell, 1986); however, use of relaxation techniques with HRT has been reported as being useful as tics are exacerbated during times of stress (Piacentini & Chang, 2001; Frank & Cavanna, 2013).

The final element of HRT is generalization training. Since HRT is usually conducted in a clinic setting, care is taken to extend the strategies learned in clinic to more real world settings (Piacentini & Chang, 2006). It usually involves the individual practicing the techniques outside of the clinic, reviewing all the competing responses developed during treatment, and developing competing responses for tics that have not occurred in clinic but occur outside the clinic setting. The primary goal of generalization is independence in using the HRT strategies (Piacentini & Chang, 2006).

Multiple reviews of HRT have been conducted and it is now considered a well-established treatment for CTDs per APA standards (Cook & Blacher, 2007). Use of HRT can help children and adolescents to develop coping strategies leading to less life impairment and potentially to avoid negative side effects of medication (Hwang, Tillberg, & Scahill, 2012).

European and Canadian guidelines for the treatment of tics have been proposed and have suggested the use of HRT as a first line treatment for tics before medication (Verdellen, van de Griendt, Hartmann, Murphy & ESSTS Guidelines Group, 2011; Steeves et al., 2012). Since its first inception, HRT has been evaluated as a full treatment package and by its separate components; it has been modified for use in school and as a telehealth model; and it has been enhanced with a focus on function-based interventions to address situations in the environment which may provoke tics through the Comprehensive Behavior Intervention for Tics (CBIT). In over forty years, at least 27 studies have been published specifically investigating the use of HRT strategies with individuals with CTDs, of which 8 were randomized control trials. HRT has been associated with a reduction in tics between 39 and 100 percent (Bate, Malouff, Thorsteinsson, & Bhullar, 2011; Franklin et al., 2010). In addition, two meta-analysis studies have been conducted, which indicated that HRT was also effective in significantly reducing tic severity (Wile & Pringsheim, 2013) and had a large effect size (d = 0.80; Bate et al., 2011).

Parent-Mediated Interventions

While HRT has been established as an efficacious treatment for CTDs, one avenue that has not been explored is the delivery of HRT through a parent-mediated model. Parent-mediated interventions, or interventions focused on training parents to use therapeutic skills, have been found to be an effective mode of delivery for a variety of child and adolescent populations, including children with externalizing and challenging behaviors (Conroy, Dunlap, Clarke, & Alter, 2005; Dunlap, Ester, Langhans, & Fox, 2006; Eyberg, Nelson, & Boggs, 2008; Pelham & Fabiano, 2008), language impairments (Roberts & Kaiser, 2011), and autism (McConachie & Diggle, 2007; Moes & Frea, 2002; National Autism Center, 2009). Positive child outcomes as a result of parent-mediated intervention have included increases in cognitive functioning,

communication skills, social skills, and adaptive skills (Strauss, Mancini, the SPC Group, & Fava, 2013; Strauss et al., 2012), increases in positive interactions between parents and children (McNeil & Hembree-Kigin, 2010), decreases in autism symptom severity (Strauss et al., 2012), and decreases in challenging behaviors such as noncompliance and aggression (Eisenstadt, Eyberg, McNeil, Newcomb, & Funderburk, 1993; McNeil & Hembree-Kigin, 2010). In addition to positive child outcomes, parent-mediated interventions have been associated with positive parent outcomes. Examples include decreases in parent stress, increases in parents' positive verbalizations toward their children, increased positive affect, increased parental knowledge, and increases in positive parenting skills (Baker, Landen, & Kashima, 1991; Brookman-Frazee, 2004; Eisenstadt, Eyberg, McNeil, Newcomb, & Funderburk, 1993; Koegel, Bimbela, & Schreibman, 1996; Ingersoll & Dvortcsak, 2010; McConachie & Diggle, 2007; McNeil & Hembree-Kigin, 2010).

History

Parent training interventions first emerged in the 1960s and were primarily used for intervening with challenging behavior exhibited by children and adolescents (Kaminski, Valle, Filene, & Boyle, 2008). The development of these interventions represented a shift from clinicians focusing on changing child behavior to helping shape positive parenting practices. Practitioners discovered that parents, not just clinicians, could be trained to become behavioral change agents for their children. Over time, parent-mediated interventions have evolved and expanded to include different concerns (e.g., behavior management, increasing communication skills, using effective discipline practices) in a variety of settings (i.e., home, clinics, community centers). In addition, variety of techniques have been employed including direct coaching and feedback from a clinician, homework, role play, group training, collaborative goal setting and

progress monitoring, in vivo practicing of skills, contingency management, and problem solving (Barton & Fettig, 2013).

Model for Parent-Mediated Interventions

The main goal of parent-mediated interventions is to transfer skills from clinicians to parents, such that parents use these skills to become the primary behavior change agents within their children's lives (Barton & Fettig, 2013; Garbacz, Brown, Spee, Polo, & Budd, 2014). This goal is accomplished through a triadic model of intervention delivery (Barton & Fettig, 2013; Lieberman-Betz, 2015; Roberts & Kaiser, 2011; Salisbury & Kushing, 2013). First, parents are trained in the specific skills of the intervention by experienced clinicians. Next, the parents execute these skills with their children. Finally, through implementation of the learned skills, children hopefully benefit and demonstrate improvement on the targeted outcome. The triadic model is considered indirect, as the clinician is providing direct intervention to the parent, who provides intervention to the child (Barton & Fettig, 2013). In this type of model, both the clinician and the parent are considered experts (Brookman-Frazee, 2004). Clinicians enter the intervention context as experts in the specific techniques, whereas parents enter the intervention as experts in knowledge of their children. Although clinicians have the role of teaching specific skills to parents, parents will ultimately decide how to interpret and employ this information within their family (Turnbull, Blue-Banning, Turbiville, & Park, 1999).

The Current Study

To date, no known studies have examined a parent-mediated version of habit reversal for the treatment of CTDs. The Comprehensive Behavioral Intervention for Tics (CBIT) manual expresses that parents should be involved in treatment, but most of the focus is on the clinician providing the intervention to the child (Woods et al., 2008b). Training parents to implement

HRT may assist with its dissemination. As of now, it is expected HRT will be provided by trained professionals (Kurlan, 2014); however, there are a limited number of therapists to provide CBIT (Woods et al., 2010) and a wide range of potential benefits for training parents to act as therapists (Baker, Landen, & Kashima, 1991; Brookman-Frazee, 2004; Eisenstadt, Eyberg, McNeil, Newcomb, & Funderburk, 1993; Koegel, Bimbela, & Schreibman, 1996; Ingersoll & Dvortcsak, 2010; McConachie & Diggle, 2007; McNeil & Hembree-Kigin, 2010). Training parents to be habit reversal co-therapists may also help with early difficulties experienced by children with CTDs. Well-meaning parents sometimes have inappropriate expectations and may view tics as voluntary behaviors of their children. This view may lead to demands or punitive consequences to control tics (Lombroso et al., 1995). Thus directly teaching habit reversal strategies to parents could help avoid early negative experiences for children with CTDs. Training parents to provide HRT strategies may also result in more intervention time for children, more generalization and maintenance of skills, fewer symptoms associated with CTDs, and decreased family stress (Ingersoll & Dvorcsak, 2010). Finally, in rural areas, parent-mediated interventions may provide a reduced need for access to therapists. The current study was the first known attempt to combine the evidence-based strategies of HRT with the evidence-based delivery system of parent-mediated interventions with the goal of improved outcomes for both children with CTDs and their parents.

Research Questions

Based on the available literature surrounding interventions for CTDs and parent-mediated interventions, the following research questions were the foci of study:

1. Will parents of children with CTDs participating in a parent-mediated version of HRT learn to implement the strategies with adequate fidelity?

2. Will children with CTDs receiving a parent-mediated version of HRT demonstrate decreases in number, frequency, and severity of tics as measured through direct observations, parent questionnaires, and self-report questionnaires?
3. Will children with CTDs receiving a parent-mediated version of HRT demonstrate decreases in premonitory urge sensations as measured through self-report questionnaires?
4. Will parents and children participating in a parent-mediated version of HRT find the intervention acceptable as measured through self-report questionnaires?
5. Will treatment motivation on the part of the children with CTDs impact parents' fidelity of implementing the strategies of a parent-mediated version of HRT as measured by self-report questionnaires on the part of the children and clinician observation and rating of parents?
6. Will parent motivation to participate in treatment be related to increased skill acquisition and adequate fidelity as measured by self-report and direct observation?

CHAPTER TWO

LITERATURE REVIEW

The following is a review of the literature to provide the reader a more in-depth understanding of the available research on habit reversal training (HRT) and to further discuss the current study in the context of this information. In addition, parent-mediated interventions are discussed in more depth to provide the rationale for the application of HRT through a parent-mediated model.

To collect studies of HRT for inclusion, a systematic review of the literature was conducted. The primary databases used were EBSCOhost, PubMed, and Google Scholar. Search terms used associated with CTDs included the following: "Tourette's disorder," "Tourette's syndrome," "chronic tic disorder," "chronic motor tics," "chronic phonic tics," "chronic vocal tics," "persistent motor tic disorder," "persistent vocal tic disorder," and "tic disorders." To identify studies that used HRT for tics, the following search terms were used: "habit reversal," "habit reversal training," and "habit reversal therapy." From this search, 27 studies were identified that involved implementation of HRT for the treatment of chronic tics. These studies are reviewed in the next section.

Habit Reversal Training

HRT has been the most studied behavioral intervention for tics. It is also considered the most effective behavioral intervention for the treatment of tics (Cook & Blacher, 2007; Himle et al., 2006). While it has been used successfully for "nervous habits," (e.g., nail biting, thumb sucking), trichotillomania, and stuttering (Woods & Miltenberger, 1995), studies specific to treating tics are presented below.

The first study published using HRT methods was by Azrin and Nunn (1973). Twelve individuals between the ages of 5 and 64 years participated in this original study. Of these participants, 8 were female and 4 were male. They presented with a variety of motor tics including head jerking or shaking, shoulder jerking, elbow flapping, eyebrow plucking, nail biting, thumb sucking, and tongue pushing on teeth. One participant also presented with lisping for treatment. One participant demonstrated more than one tic; the rest only presented with one.

Regarding methods, one week of baseline data were collected using self-report and validation by a significant other of the participants (Azrin & Nunn, 1973). After this week, Azrin and Nunn (1973) applied the methods of habit reversal, including awareness training (AT; i.e., response description, response detection, early warning detection, situation awareness), competing response training (CRT), motivation techniques (i.e., inconvenience review and social support), and generalization training (i.e., symbolic rehearsal). These methods were implemented in one or two sessions. By the third week post-intervention, all of the participants demonstrated an average 90 percent reduction of habits and tics. The reductions were reportedly maintained 5 months after the end of treatment for 11 of the participants.

Azrin, and colleagues (1980) used a larger sample size and compared HRT to massed negative practice. Participants in this study included 17 males and 5 females between the ages of 11 and 62 who had motor and/or phonic tics. They were randomly assigned to the HRT (n=10) and massed negative (n = 12) conditions. The HRT procedures were identical to the procedures in Azrin and Nunn's (1973) original study and were implemented in one or two sessions of about two and a half hours. The massed negative practice condition involved a pattern of the participants performing the tic repeatedly for 30 seconds followed by a brief rest for an hour. They performed the tic in front of a mirror and said, "This is what I'm not supposed to do"

(Azrin et al., 1980). This procedure was continued daily until four days after the tics stopped occurring, at which point the procedures were faded for a period of two weeks. Post-treatment, participants were encouraged to continue practicing the massed negative practice procedures for 30 seconds each hour. Data were collected via self-monitoring techniques. Both groups demonstrated a reduction in tics; however, at an 18-month follow-up session, participants in the HRT condition were still demonstrating an average 97 percent reduction in their tics from baseline. The massed negative practice participants who only showed an average 33% reduction in tics from baseline at a four week follow up (Azrin et al., 1980).

Finney, Rapoff, Hall, and Christophersen (1983) studied the use of HRT for simple and complex tics. Participants in this study were two boys aged 11 and 12. Techniques were slightly adapted from the original Azrin and Nunn (1973) study. AT still included response description and response detection, but Finney and colleagues (1983) also added a self-monitoring component using paper and pencil or a manual counter. Self-monitoring was not used after one week of treatment. Treatment also included situation awareness and an inconvenience review. Before moving into a CR, Finney and colleagues introduced deep breathing as a relaxation technique. CR and social support training were similar to the original procedures. Treatment was conducted in three sessions of approximately an hour to an hour and a half.

Finney and colleagues (1983) used a multiple baseline across subjects design. They also employed multiple baseline across behaviors design with one of the participants. The behaviors of concern were pairs of tics (i.e., two tics were treated at a time before an additional two tics would be treated). For the first participant, tics reduced to low or zero rates during treatment and were not observed at follow-up sessions at 2, 6, and 12 months. For the other participant, the first pairs of tics reduced to low or zero rates during HRT procedures; however, the second pair

of tics were more variable during HRT. A HRT booster session occurred during the ninth week focused on these last two tics. One tic reduced to zero quickly; the other decreased more gradually. At the 12 month follow-up, none of the tics were observed for the second participant.

Miltenberger and colleagues (1985) were primarily interested in the role of the CR and compared all of the components of HRT as described by Finney and colleagues (1983) to AT and CRT alone. Participants were nine individuals with motor tics between the ages of 12 and 60. Four individuals were assigned to the HRT condition and five individuals were assigned to the AT and CRT condition. A multiple baseline across subjects design was used in each group. The dependent variable of tic frequency was measured via direct observation and videorecording by the study's authors. Overall, three out of four individuals in the HRT group and all of the individuals in the CR group demonstrated significant reductions in tics from baseline, which were maintained at 7 and 15 week follow-ups. Also, the participants generally rated each treatment favorably on an acceptability rating scale post-treatment. This study suggested that AT and CRT had the same effects as the full HRT treatment package (Miltenberger et al., 1985).

Azrin and Peterson (1988) also investigated the effectiveness of HRT for Tourette syndrome. Three male adults aged 28, 37, and 42 years participated in this study. Two were formally diagnosed with Tourette syndrome, whereas the other met criteria but had never been formally diagnosed. An AB single case design was utilized and frequency of tics were recorded at the beginning and end of sessions for 10 minutes each; however, only the data at the beginning of the sessions were reported to represent tics that had not been influenced by the intervention that day. Frequency data were also recorded in the home setting. If tics were low frequency, the participants were instructed to record data all day; if tics were high frequency, participants were instructed to record data for a daily 10-minute session. Habit reversal was implemented

according to the procedures in the original Azrin and Nunn (1973) study; however, relaxation techniques (i.e., progressive muscle relaxation, deep breathing, visual imagery, and relaxing self-statements) were also included. Procedures were implemented weekly in one hour sessions. Total number of sessions ranged from 10 to 27, depending on the participant. Data analysis revealed an average decrease in all tics, both motor and phonic, by at least 57% for all three participants in home and in clinic settings.

Azrin and Peterson (1990) conducted another group design comparing the effects of HRT to a waitlist control group. This study included 10 participants between the ages of 6 to 36 who all had diagnoses of Tourette's disorder. Seven individuals were male and three were female. Three individuals were on medication during the study, but were asked not to change dosage to maintain stability throughout the study. Participants were matched based on frequency of tics and medication usage, then randomly assigned to the HRT group or waitlist control. HRT consisted of AT, self-monitoring, relaxation training, CRT, contingency management, and generalization training. Participants received between 13 to 30 sessions over the course of 8 to 11 months. The sessions occurred weekly for the first four weeks before decreasing to monthly sessions. Frequency data were collected across home and clinic settings via direct observation of video recordings. Significant reductions in tics occurred for individuals in the immediate treatment group across settings when compared to the waitlist control group, suggesting that HRT was successful at reducing tics. In addition, when looking at within-group comparisons, significant reductions in tics occurred for all participants pre- to post- treatment. Specifically, during the first month of treatment, a 50 percent reduction in tics was observed in either clinic or home settings for all participants. During the second month, the 50 percent reduction was observed in both settings for all participants. In the last month 9 out of 10 participants

demonstrated reductions of at least 88 percent across settings. The one participant who did not reach this level of reduction still demonstrated a 67 percent reduction in tics. HRT's effects were found for both motor and phonic tics.

O'Connor, Gareau, and Borgeat (1997) compared HRT to a cognitive behavioral therapy (CBT) approach for treating tics. Thirteen adult subjects, of which seven were male and six were female, who were between the ages of 23 and 49 years and who experienced chronic motor tics participated in the study. Three participants were on antipsychotics during the study, with no change in dosage. Seven participants were randomly assigned to receive traditional HRT (Azrin & Nunn, 1973), while six participants were assigned to a CBT-enhanced HRT group, which included more emphasis on cognitions and affect. Treatment occurred over the course of 10 sessions. The first five sessions involved baseline data collection and observation. The last five sessions involved either HRT or CBT. Data were collected via participants' self-reported frequency. Results indicated a significant reduction in tics posttreatment with no difference between groups. These results were maintained at 3-month and 2-year follow ups.

Wilhelm and colleagues (2003) conducted a randomized controlled trial to compare HRT to supportive psychotherapy in 29 individuals with Tourette's disorder. Of these 29 participants, 13 were female and 16 were male. Fourteen were on medications during treatment; however, for inclusion in the study, they had to have been taking a stable dosage of the medication for three months prior to treatment and maintain a stable dosage throughout treatment. The participants were randomly assigned to the HRT group (n = 16) or the supportive psychotherapy group (n = 13). The average age of participants in the HRT group was 36 and the average age of participants in the supportive psychotherapy group was 33. HRT consisted of AT, self-monitoring, relaxation training, CRT, and contingency management. Supportive psychotherapy

included patient-directed discussion of topics with encouragement to reflect and express feelings and with problem-solving. Each treatment was provided in 14 sessions. The first 8 sessions occurred weekly, whereas the remaining 6 sessions occurred bi-weekly. Data on tic severity were collected pre- and post-treatment and at a 10 month follow up using the Yale Global Tic Severity Scale (YGTSS). Results indicated a significant reduction in YGTSS scores (i.e., significant reduction in tic severity) pre- to post-treatment in the HRT group, with similar scores at the follow up. The supportive psychotherapy group did not have significantly different scores pre- to post-treatment, nor at the follow up.

Deckersbach, Rauch, Buhlmann, & Wilhelm (2005) conducted another randomized controlled trial comparing HRT to supportive psychotherapy in 28 individuals with Tourette's disorder. Of these participants, 15 were male and 13 were female. The average age of participants was 35 and all participants were adults. Sixteen of the participants were on medications; however, they had been taking the medications for at least three months and the dosages remained stable throughout the study. Participants were randomly assigned to an HRT group (n = 14) or a supportive psychotherapy group (n = 14). Each group received 14 sessions, each 50 minutes in length. The first 8 sessions were delivered weekly, whereas the last 6 sessions were delivered bi-weekly. HRT included the full treatment package as described by Azrin and Peterson (1990). Supportive psychotherapy included patient-directed topics, therapist encouragement, reassurance, and assistance with reframing and problem-solving. Tic severity was measured through the YGTSS. Other measures were included to assess psychosocial functioning, life satisfaction, depression, OCD, and AD/HD. Results of the study indicated that participants in the HRT group had significantly lower tic severity scores mid- and post-treatment. Both groups had significantly higher scores on measures of psychosocial functioning and life

satisfaction, as well as lower scores on measures of depression and OCD post-treatment. At a six month follow-up, individuals in the HRT condition continued to have lower ratings of tic severity; all other results were maintained.

All of the studies mentioned in this section suggest that HRT can successfully lead to reductions in tic severity. However, a few limitations should be noted. First, in three of the studies (Azrin & Nunn, 1973; Azrin et al., 1980; O'Connor et al., 1997), changes in tic severity were based on self-report from participants instead of direct observations of tics. Second, in three of the studies (Azrin & Nunn, 1973; Azrin et al., 1980; Azrin & Peterson, 1988), an AB single case design was limited. While a reversal would likely not have been possible, these results could have been made stronger by use of another single case design (e.g., multiple baseline design). Third, another three studies (e.g., Azrin et al., 1980; Azrin & Peterson, 1988; O'Connor et al., 1997) were limited by potential treatment interference. In the studies by Azrin and colleagues (1980) and Azrin and Peterson (1988), some of the participants were prescribed and taking antipsychotic medications and/or were involved in other therapy for the treatment of tics. Regarding medication, it was not reported that dosage was controlled for during the studies. In the O'Connor and colleages (1997) study, the CBT group still received components of HRT. It is possible that the reduction in tics observed in this group may have been influenced by HRT components. Fourth, in another three studies (Azrin et al., 1973; Azrin & Peterson, 1990; Miltenberger et al., 1985), researchers included participants with a broad range of demographic characteristics (e.g., tic disorder, comorbid disorders, age ranges). It would be difficult to generalize these results to a larger population of individuals with chronic tic disorders. Finally, the randomized control trials (Azrin et al., 1980; Azrin & Peterson, 1990; O'Connor et al., 1997; Wilhelm et al., 2003; Deckersbach et al., 2005) included had small sample sizes, which limits the

overall potential power of these studies. However, given the number of studies conducted, all of the studies included provided preliminary evidence for the effectiveness of HRT for chronic tic disorders.

Comprehensive Behavioral Intervention for Tics (CBIT)

In an effort to increase the evidence for HRT as an effective treatment, the Tourette Syndrome Association Behavioral Sciences Consortium, a multidisciplinary group of CTD researchers, developed and conducted two large-scale, multi-site randomized control trials comparing CBIT to supportive psychotherapy with children and with adults (Chang et al., 2007). The first study, conducted by Piacentini and colleagues (2010), involved a randomized control trial of CBIT, an enhanced HRT intervention, compared to supportive psychotherapy and education in 126 children between the ages of 9 and 17 who were diagnosed with a CTD. Participants included 99 males and 27 females. During the study, 36 percent were on psychotropic medication; however, inclusion criteria required them to be on it for at least six weeks with no expected change in dosage during the study. Participants were randomly assigned to receive CBIT (n = 61) or supportive therapy and education (n = 65). CBIT's main component was HRT (i.e., AT and CRT), with added psychoeducation about tics as well as the inclusion of relaxation training and functional interventions to address tic-provoking situations. Supportive psychotherapy and education included psychoeducation about tics and was patient-directed. Each group received 8 sessions over the course of 10 weeks. The first two sessions lasted 90 minutes. The remaining sessions were each 60 minutes. The first six sessions occurred weekly, whereas the last two sessions occurred bi-weekly. Data were collected regarding tic severity using the YGTSS. Treatment benefit was also measured using the Clinical Global Impressions – Improvement scale. Parents also completed the Parent Tic Questionnaire regarding number,

frequency, and intensity of their children's tics. Attrition for the study was low overall (i.e., 9 percent). Results indicated a 51% decrease in tic severity (i.e., average 7.6 point decrease on the YGTSS) in the CBIT condition compared to a 30% decrease (i.e., average 3.5 point decrease on the YGTSS) in the control group. The change in tic severity had an effect size of 0.68, which was considered clinically significant. Global improvement after treatment and parent ratings of tic severity were significantly better in the CBIT group posttreatment compared to the control group. Follow up at 3 and 6 months indicated treatment effect durability.

To date, the Piacentini and colleagues (2010) study is the largest study utilizing HRT in a child population for the treatment of CTDs. It provided the strongest support for the use of HRT for this population. This study demonstrated high levels of experimental control, as many variables were examined for a possible relation to the results of the study (e.g., use of medication by participants, differences across the three different clinical sites, adverse events in the families' lives, etc.). Two limitations were found regarding the follow-up data. Only the children who were identified as "acute responders" to the two different treatments were included for the follow-up. Inclusion of all children may have changed the follow-up results. In addition, Piacentini and colleagues (2010) included treatment integrity for the therapists, but it was not clear if this data also included the integrity of the participating children (i.e., whether or not they were using the strategies correctly).

The same procedures in the Piacentini and colleagues study (2010) were employed with 122 adults with CTDs by Wilhelm and colleagues (2012). Participants in this study included 78 males and 44 females with an age range of 16 to 69. They were randomly assigned to the CBIT group (n = 63) or the supportive psychotherapy and education group (n = 59), identical in description to the procedures of Piacentini and colleagues study (2010). Wilhelm and colleagues

(2012) found slightly different results. CBIT still resulted in a significant decrease in tic severity when compared to the supportive psychotherapy group; however, this decrease was less than in the previous study. Individuals in the CBIT group demonstrated a 25 percent decrease on the YGTSS compared to 11 percent decrease in the supportive psychotherapy group, associated with an effect size of 0.57. Wilhelm and colleagues (2012) separated out the tic severity score based on motor and phonic tics and found an effect size of 0.63 for motor tics and 0.35 for phonic tics. Participants in the CBIT group also were rated as having a significantly better positive treatment response pre- to posttreatment. Treatment durability was measured similarly to the previous study and demonstrated that effects were durable over time (i.e., at three and six month follow-ups). Limitations to the Wilhelm and colleagues (2012) study were identical to the Piacentini and colleagues (2010) study, with the exception that the Wilhelm and colleagues (2012) study included an adult sample.

Rowe, Yuen, and Dure (2013) sought to replicate the findings of the Piacentini and colleagues (2010) study. They delivered CBIT to 30 children between the ages of 7 and 19 diagnosed with CTDs. Similar to other studies, CBIT was delivered in 8 sessions over 10 weeks. Data were collected regarding number of tics and tic severity. Results indicated a significant reduction in the number of tics exhibited as well as in tic severity pre- to posttreatment. This study by Rowe and colleagues (2013) provides additional evidence for the utility of CBIT for the treatment of CTDs; however, multiple limitations were present. First, no waitlist control group was included to control for the natural course of CTDs. Additionally, the authors did not include any follow-up data past posttreatment, so no conclusions about long-term outcomes can be made. Finally, all of the measures used to measure tic number and tic frequency were self-report measures.

Simplified Habit Reversal (SHR) and Component Analyses

While recent studies are often using the full CBIT protocol to determine its efficacy, a variety of studies have been conducted to evaluate a simplified version of HRT, labeled SHR. In addition, component analyses have been conducted to determine which portions of HRT are most effective for treatment gains. The consensus that has arisen based on analysis of HRT components are that AT, CRT, and social support training are the necessary components of HRT.

Ollendick (1981) was the first to investigate components of HRT with CTDs, specifically self-monitoring and the competing response. Participants in his study included two boys aged 9 and 11. A multiple baseline across settings (i.e., home and school) design was used. In addition to frequency that was self-monitored by the boys, parents and teachers were also instructed in how to collect frequency data. After five days of baseline at school, both boys were instructed to self-monitor their tics. For both boys, self-monitoring led to a reduction in tics; however, for one of the boys, the tics dropped to zero. For this individual, CR methods were not used and the methods were employed at home with similar results. The other boy started implementing CR procedures with self-monitoring after five days of self-monitoring alone. With the addition of the CR procedures, his tics also dropped to zero. These results were replicated in the home as well.

While this study suggested that self-monitoring with CR might be beneficial for tic reduction, it was limited by having two participants who each received two different interventions. One of the participants did not receive CR procedures, which was the intent of the study. In addition, for the participants who did receive CR procedures, CR was never used alone; thus, no conclusions can be drawn about its effectiveness as a standalone intervention.

Similar to previous studies, self-report data was also used at the primary dependent variable, which may have influenced the results.

Wright and Miltenberger (1987) examined AT alone with a 19-year-old adolescent male with motor tics. They used a multiple baseline across tics design. Data were collected during baseline and intervention via direct observation, which provided frequency counts during 25-minute periods. AT was delivered in 11 sessions over 5 weeks for the first tic. An additional 6 sessions of AT were delivered for the second tic once the first tic was stable and at low levels. Results indicated an average decrease in tics of 39 percent. These results were maintained at follow-up sessions at 1, 2, 4, and 8 months. This study by Wright and Miltenberger (1987) suggested that AT might be a crucial component to HRT and can lead to reduction in tics when used alone. It was limited in the sense that the individual was not fully capable of identifying when his tics had occurred or were going to occur, which is the purpose of AT. Also, the data collected during AT represented variable amounts of time; however, data were still reported as frequency instead of as a rate per minute.

Azrin and Peterson (1989) also published a case study of a 9-year-old girl with frequent and noticeable eye blinking tics. They used habit reversal methods over the course of four months in three different experiments. In the first experiment, the child was taught to use a noncontingent CR (i.e., she was asked to blink softly every 5 seconds regardless of tic occurrence). An ABAB reversal design was used, with the baseline phase (A) lasting 5 minutes and the intervention phase (B) lasting 3 minutes. Frequency data were collected via direct observation by the authors. Tics decreased by 90 to 95 percent during the intervention phases. The authors noted a potential confound in that the child was asked to watch a clock during the intervention phase to know when to blink, which may have led to the reduction of tics. In the

second experiment, the authors attempted to remove the potential confound by having the child look at a picture on the wall in all conditions. In this second experiment, an ABCDDCBA alternating treatment design was used, with each phase lasting 2 minutes. A represented baesline; B represented facial relaxation only; C represented noncontingent CR only; and D represented a combination of facial relaxation and noncontingent CR. Facial relaxation led to an average 54 percent reduction in tics. CR led to an average 97 percent reduction in tics. The combination of facial relaxation and CR led to an average 77% reduction in tics. The authors concluded that use of the CR technique alone was superior to either intervention using facial relaxation. The final experiment included seven weeks of training of using contingent (i.e., in response to tics) and noncontingent (i.e., soft blinks every 5 seconds for short periods of time) CRs and social support training for parents. Data in this experiment were based on recordings in the home setting. Tics reduced to zero during the final two months of treatment, with a gradual decrease of 52 to 100 percent over time. At a two-year follow-up, no tics were observed by the clinicians.

While use of an ABAB reversal design and alternating treatments design were appropriate, the intervention periods were very short, making it difficult to determine response stability. Also, aggregate data instead of individual data were presented for the second experiment, which may have influenced the conclusions. The last experiment utilized an AB approach, making generalizations about potential replication challenging due to lack of a reversal during this phase. Also, it is possible that during the four months, a period of waning could have occurred, which might explain the decrease in tics; however, this issue was not addressed by the authors. They also did not discuss the differences in and potential confounds of using clinic and home settings, the two settings in which data were recorded.

Sharenow, Fuqua, and Miltenberger (1989) investigated the need for the CR to be physically incompatible with the tic with one female and two adult individuals who were aged 24, 32, and 66 and who experienced motor tics. A multiple baseline across subjects design was used for all participants, as well as a multiple baseline across tics for one participant. Data were collected using direct observation through video recording. Following baseline, an intervention phase consisting of AT, situation awareness training, and CRT using a dissimilar, but not incompatible CR was undertaken. The first two subjects demonstrated a decrease in tics from baseline, and the third subject demonstrated a decrease in tics to near zero levels using the dissimilar CR. For the first two subjects, an additional phase of intervention was added using the traditional incompatible CRT for comparison. The first subject's tics remained steady and at low rates during the incompatible CR condition. The second subject, for which the multiple baseline across tics was also used, demonstrated similar rates of tics for both tics examined during both the dissimilar and incompatible CRs. These rates were less than baseline, but not as significantly reduced as the other two participants. The authors concluded that dissimilar CRs resulted in reduction of tics across participants, suggesting that CRs do not necessarily have to be physically incompatible with the tic to be successful. This study was limited by potential sequencing effects, as all subjects were introduced to HRT using a dissimilar CR first before the incompatible CR. Self-monitoring was also included during CRT, which may confound the effects of the intervention alone. Sharenow and colleagues also noted difficulty in recording use of the CRs by the participants through data coding using video recording; thus, the results may have been influenced by this limitation.

Peterson and Azrin (1992) conducted another study which compared self-monitoring, relaxation training (i.e., progressive muscle relaxation), and HRT (CRT only). Six male subjects

with Tourette's disorder participated in the study, of which four were adults between the ages of 32 to 40 and two were children aged 10 and 13. One participant was on antipsychotic medication for the treatment of tics at the time of the study. In order to compare the three treatments, an alternating treatments design was used. The order in which treatments were presented was randomized for each participant for counterbalancing. The study was conducted in three sessions spaced two weeks apart. The first two sessions involved collection of baseline data. The third session also included baseline, but also evaluated the different treatments. Before each treatment, the participants received 30 minutes of instruction about the technique, 5 minutes of practice using the technique, and then 10 minutes of self-guided treatment in which they were video recorded. Results from this study indicated an average reduction in tics of 55 percent in the HRT group, 44 percent in the self-monitoring group, and 32 percent in the relaxation training group. Peterson and Azrin (1992) further analyzed the data using the Friedman test, which resulted in a statistically significant difference between the treatments and baseline. Post hoc analyses were also conducted which confirmed that each treatment was statistically significant from baseline, but not from each other. Peterson and Azrin (1992) reported that they did not anticipate multiple treatment interference and that the effects of each intervention would be reversible. However, even with counterbalancing and the instruction to only use the techniques described in the 30-minute training session during the 10-minute observation, it might be possible for carryover effects between treatments, especially because all treatments were conducted on the same day. Regarding the data collection, Peterson and Azrin (1992) collected 10 minutes of data for each baseline and for each treatment; however, these 10 minutes were broken down and reported as 2.5 minute segments without any rationale. This separation made the graphs difficult to interpret. If this decision was made because the tics were

occurring too frequently, consideration for other data collection techniques (e.g., partial interval recording) could have been made. This generalization of the results of this study is limited by inclusion of four adults and two children. In addition, for the individual who was on an antipsychotic medication, no report of strategies to keep the dosage stable during treatment were made and it could be an additional example of a multiple treatment effect. Finally, Peterson and Azrin (1992) noted being surprised by the results of the HRT group, since reduction in tics was much more modest than in previous studies; however, they did not discuss how only CRT was used, instead of the full treatment package. Use of CRT without AT may provide an additional explanation for the modest results.

Woods, Miltenberger, and Lumley (1996) examined the separate components of HRT to determine the necessity of the different components. Four children, three male and one female, between the ages of 8 and 12 with chronic motor tics participated in this study. The study was conducted in the homes of three participants and at the school of the fourth participant. The authors used a mixed multiple baseline across participants and tics design. After a 20-minute baseline, the different intervention components were implemented in the following order: AT alone, AT with self-monitoring, AT with self-monitoring and social support, and AT with CRT and social support. Woods and colleagues (1996) only implemented subsequent phases of the intervention if tics did not decrease to near zero frequency. Data on the frequency of tics were collected using a 10-second partial interval schedule. Results indicated that tics significantly decreased for all participants; however, these decreases were observed in different intervention phases for each participant. After implementation of AT alone, one participant's tics reduced to near zero. After implementation of AT with self-monitoring, a second participant's tics reduced to near zero. When CRT was added in the last phase, the last two participants' tics reduced to

near zero. As a result, only two participants received all four phases of the intervention. The results of this study were maintained at follow up 10 to 17 weeks after treatment ended. Overall, parents reported this treatment as acceptable on treatment acceptability measures.

The Woods and colleagues (1996) study revealed the possibility of the importance of the AT component; however, a few limitations must be noted. First, all treatments and data collection occurred in participants' homes or school. The variability across the different settings may have been a confound, as experimental control would be difficult. In addition, each child received different treatment components, which makes comparisons among the data and generalizations difficult. Last, Woods and colleagues noted compliance difficulties with the introduction of each new phase and hypothesized how it may have been related to increased effort required. Treatment acceptability could have been collected from the children to help support this hypothesis.

Carr and Bailey (1996) also conducted a comparison study, comparing the effectiveness of self-monitoring, CRT, and dissimilar response training on nasal tics of a 9-year-old male participant diagnosed with Tourette's disorder. Carr and Bailey (1996) utilized an alternating treatment design. Frequency data were collected in 5-minute sessions in the participant's home. After three baseline periods of 30 minutes each, all three treatments were implemented for 15 minutes each in the following order: self-monitoring, CRT, dissimilar response training. These interventions were repeated. Following a fourth baseline, CRT was repeated based on its higher benefit compared to the dissimilar response and based on parent preference. Overall, Carr and Bailey (1996) observed an almost 70 percent reduction in tics with similar effects noted in all three intervention phases. The reduction in tics was maintained at a follow up session one month posttreatment. In addition, the child found the treatment acceptable based on an acceptability

measure. This study again provided some evidence for the utility of HRT strategies; however, Carr and Bailey (1996) noted that during CRT and dissimilar response training, fidelity of implementing the two responses (i.e., breathing through the mouth in CRT and making a fist in dissimilar response training) was low. This low fidelity may suggest that some other variable could have led to the reductions in tics during these intervention phases.

Using a nonconcurrent multiple baseline design, Woods and Twohig (2002) investigated the use of simplified habit reversal (SHR) for treatment of chronic vocal tic disorder for three male children exhibiting vocal tics. The children were aged 7, 9, and 16 and all had at least two vocal tics. SHR included three weekly sessions. During the first session, AT, CRT, and social support training were implemented for one hour. The following two sessions lasted for 30 minutes and required the participants to describe and demonstrate SHR procedures with feedback from the researchers. Data were collected using a partial interval schedule via videotapes either in home or in clinic for 15 minutes during situations identified by the parents in which tics were most likely to occur, which for all participants involved being alone. Overall, SHR resulted in immediate reduction in tic frequency for two of the participants; however, only one participant continued to demonstrate reductions at a three month follow-up. Woods and Twohig (2002) discussed difficulties with compliance for one of the participants, who did not immediately improve with SHR and whose symptoms remained stable over time. A reinforcement contingency (i.e., token economy) was implemented, but still did not improve symptoms.

The Woods and Twohig (2002) study provided some support for the use of SHR with vocal tics; however, factors limiting the conclusions should be noted. Extraneous variables (e.g., compliance issues, development of other tics) influenced the performance of the participants. In addition, data were collected in the participants' homes or in a clinic setting, which impacts

experimental control. Also, a nonconcurrent multiple baseline design was used. It could be that starting at nonoverlapping times could also influence the conclusions drawn.

Woods, Twohig, Flessner, and Roloff (2003) replicated the procedures of the Woods and Twohig (2002) study, using HRT to treat vocal tics in five male children between the ages of 10 and 13. All of the children demonstrated motor and vocal tics; however, only the vocal tics were targeted for treatment. HRT in the Woods and colleagues (2003) study consisted of the SHR in the previous study. Following this session, two additional weekly sessions were conducted, during which time treatment methods were reviewed and practiced. Data were collected in home via videotape during a 15 minute period identified by the parents as a time when tics would be most likely to occur. Data were scored using 10-second or 30-second partial interval recording. Results suggested an 82% reduction in vocal tics across four participants, with three of the four children maintaining this reduction at a three month follow-up. One participant's tics did not appear to decrease directly after the implementation of treatment, and one participant did not have good compliance with the treatment.

This study also provided evidence for the use of SHR for children with vocal tics. However, similar to the Woods and Twohig (2002) article, a noncurrent multiple baseline design was used for the Woods and colleagues (2003) study. In addition, the authors did not report measuring treatment compliance, but reported issues with compliance. Use of a measure of compliance would potentially allow for conclusions to be drawn about the need for compliance to observe treatment benefit. Last, data were collected in participants' homes, which impacts experimental control.

Verdellen and colleagues (2004) conducted a randomized controlled trial investigating exposure and response prevention compared to HRT for the treatment of tics in 43 individuals

with Tourette's disorder. Participants ranged in age from 7 to 55. Nine participants were female and 34 were male. Seventeen of the participants were taking medication for the treatment of tics during the study; however, the dosage remained stable throughout the study. Participants were randomly assigned to a group receiving exposure and response prevention (n = 21) or HRT (n = 22). Exposure and response prevention consisted of 12 weekly sessions of 120 minutes. The first two sessions included training in tic suppression, whereas the remaining 10 sessions involved having the participant suppress tics for as long as they could while focused on bodily sensations and tic provoking situations. HRT consisted of 10 weekly 60 minute sessions which included AT and CRT. Data collected included YGTSS scores for tic severity and tic frequency during 15 minute periods in clinic and home settings. Results indicated a significant difference (i.e., decrease) from pre- to post-treatment for both groups on all measures. Post hoc analyses did not reveal any significant differences between the two groups, suggesting both were equally effective for treating tics. Scores were maintained at a three month follow-up.

The study conducted by Verdellen and colleagues (2004) was the first to investigate exposure and response prevention for the treatment of Tourette's disorder, while also comparing it to HRT. A number of limitations exist for this study. First, Verdellen and colleagues (2004) did not include a waitlist control. Given the nature of waxing and waning tics in Tourette's disorder, this comparison group would have given more credibility to the results as the common course of Tourette syndrome would have been controlled. Next, the two conditions differed in two ways. The exposure and response condition included two sessions of training in techniques, which were not included in the HRT session. In addition, the HRT sessions were half the time of the exposure and response prevention sessions. While both of these differences were prescribed in the treatment manuals, these differences may have influenced the results. Next, the HRT

condition only included AT and CRT, which is the most basic form of HRT and may have influenced the results. Regarding participants, a wide range of ages were included, which may limit generalizability of results to adult or childhood populations. The effects for different age groups were not considered in the results. Also, the authors reported 14 percent attrition. The participants who did not complete the study might have been significantly different from the individuals who completed the study. With regard to results, Verdellen and colleagues (2004) reported difficulties with their video recordings. Specifically, they had technical difficulties, resulting in a loss of 4 percent of the data. They also reported difficulty rating tic frequency based on the recordings, which may have impacted the results. Finally, the authors reported the results of a three month follow up; however, 14 percent of the individuals did not complete follow up and 68 percent of the individuals who did complete follow up had also received additional intervention for tics post-treatment. Thus, follow up data may not be truly representative of the effects of the interventions used in their study.

Carr, Sidener, Sidener, and Cummings (2005) also investigated HRT; however, they were primarily interested in initially assessing the functions of tics through functional analysis. Three boys aged 8, 9, and 12 participated in the functional analysis. All had diagnoses of Tourette's disorder. The functional analysis revealed that all participants' tics had non-social functions. Carr and colleagues (2005) provided explanations that this finding could possibly be a result of the tics having a function of automatic negative reinforcement, of the tics being an unconditioned response, or of insufficient condition manipulation. After the functional analysis, two of the participants continued on to receive simplified HRT (SHR) consisting of AT and CRT for both and additional social support training for one (Carr et al., 2005). The authors used a nonconcurrent multiple baseline design. Participation in SHR led to reductions in the tics

targeted through therapy. One of the participants did not demonstrate an immediate reduction in tics, which is why the social support training was added. For the participant only receiving AT and CRT, his other tics not targeted for treatment reduced to near zero levels after SHR was implemented. For the individual with additional social support training, the non-targeted tics remained elevated post-treatment; however, his parents did not want to pursue treatment for these tics.

The Carr and colleagues (2005) was one of the few studies to include a functional analysis of tics before implementing treatment. It also provided some support for the use of SHR with individuals with Tourette's disorder. However, this study was limited by a few factors. First, similar to Woods and Twohig (2002; 2003), a nonconcurrent multiple baseline design was used. Next, the participant who did not continue on to treatment may have yielded different results than the two who participated in SHR. Regarding the two who participated, different versions of SHR were provided (i.e., one received social support training), thus limiting the conclusions that can be drawn. Finally, Carr and colleagues (2005) did not collect follow-up data, thus conclusions cannot be made about long-term effectiveness.

Taken together, all of these studies varied with regard to interventions employed (i.e., components of HRT used). They also varied with regard to limitations. Despite the variability, HRT methods were still supported for reducing tic severity. Also, the critical elements of HRT, namely AT, CRT, and social support training, were supported.

School-Based HRT

Two studies have modified HRT to be delivered in a school setting. Both studies resulted in positive effects for participants; however, much more study in this area would be needed to conclude that HRT can be used effectively in school settings.

Clarke, Bray, Kehle, and Truscott (2001) investigated the use of HRT and HRT with self-modeling with four male students in public school settings. The students ranged in age from 11 to 16 and had been diagnosed with Tourette's disorder. HRT included six sessions and involved AT and CRT. After HRT was completed, self-modeling was introduced for another six or seven sessions. During the self-modeling component, the participants watched videos of themselves in the classroom that had been edited to show them not engaging in tics. The participants were also encouraged to keep using the strategies taught during HRT. Frequency data were collected in the classroom during 18-minute observations. The authors utilized a multiple baseline across participants as the design. Use of the procedures resulted in significant decreases in tics for three out of the four participants. Post-treatment, the reductions ranged from 16 to 71 percent. For the three who responded favorably to the intervention, the reductions in tics were somewhat maintained at a 5 to 10 weeks follow-up, with a range of 43 to 69 percent. The participants with the most significant reductions (e.g., two of the four students) were the ones who initially displayed more severe tics. For the student who appeared to benefit less from the intervention, the authors noted that he had developed a streptococcal infection, which could have interfered.

While the results of Clarke and colleagues (2001) study suggested that HRT and HRT with self-modeling might be beneficial for students with Tourette's disorder in a school setting, a few limitations should be noted. First, the authors attempted to determine whether traditional HRT or HRT plus self-modeling would be more effective; however, they implemented the procedures in the same sequence for all of the participants, thus making it impossible to determine if one or the other yielded more significant reductions. In addition, participants and schools self-selected to be included in the study, which could result in differences in motivation between participants and others with Tourette's disorder in the general population. Finally,

related to the school setting, experimental control was affected as each school would probably have a different climate.

Gilman, Connor, and Haney (2005) used SHR with a 14-year-old adolescent male with Tourette's disorder, specifically focused on motor tics. He was taking an antipsychotic medication during the treatment, which had been started six weeks prior. An AB single case design was used and frequency data were collected via direct observation. SHR for this individual included AT, CRT, contingency management through a reward program, and social support training of the teacher. It was delivered in a school setting. Four sessions were delivered over the course of three weeks, with each session lasting 45 to 60 minutes. The participant had emigrated from Mexico, and an interpreter was used throughout the treatment. Results indicated a decrease in tics to near zero levels after the introduction of SHR. Follow-up at three months indicated that tics were reduced from baseline, but higher than observed during the intervention phase.

Although the study by Gilman and colleagues (2005) provided some evidence to support the use of SHR in a school setting, as well as the potential utility for English Language Learners through use of an interpreter, limitations must be discussed. First, the study was limited by the use of an AB design, as there was not a reversal phase nor other baselines for comparison. The results may have occurred due to the natural waxing and waning course of Tourette's disorder. While the participant was on a stable dosage of antipsychotic medication, it may have introduced a multiple treatment effect. Finally, the four data points of baseline were collected during one week, with a gap between baseline and intervention of three weeks. It is possible that something occurred during the three weeks which could also explain the reduction in tics.

Telehealth and HRT

The use of telehealth for the provision of CBIT has also been investigated. Himle, Olufs, Himle, Tucker, and Woods (2010) provided CBIT to three children with CTDs who were aged 11, 13, and 17 using videoconferencing. Similar to the previous two studies, CBIT was delivered in 8 sessions over 10 weeks. The design utilized was a multiple baseline across participants. Frequency data were collected for tics using frequency counts or a 10-second partial interval recording. Acceptability data was also collected from the children and their parents. The children's percentage of nonoverlapping data after the intervention was initiated was between 69 to 100 percent, suggesting a moderate to strong treatment effect. Effects were maintained at a two week follow-up. In addition, children and their parents rated the treatment as acceptable. Limitations for the Himle and colleagues (2010) study include diversity within the sample and different settings. The three participants differed in terms of their tic presentation and severity, of diagnosed comorbid conditions, and of medication use, making generalizations difficult. Also, this study was based out of participants' homes, from which differences may have influenced the results. Finally, the follow up was conducted only two weeks after the study was completed, thus conclusions about long-term efficacy cannot be made.

Himle and colleagues (2012) replicated the procedures in a randomized controlled trial comparing CBIT delivered via videoconference to CBIT delivered face-to-face. Eighteen children between the ages of 8 and 17 with diagnoses of CTDs participated in the study. CBIT in both conditions was delivered in 8 sessions over 10 weeks. Data were collected regarding tic severity using the YGTSS and Parent Tic Questionnaire, treatment response using the Clinician Global Severity & Improvement Scales, and treatment acceptability using the Working Alliance Inventory and Treatment Acceptability Questionnaire. Results indicated significant reduction in

tic severity over time on both the YGTSS and Parent Tic Questionnaire. Effect sizes for the change on the YGTSS were 0.39 for the videoconference group and 0.41 for the face-to-face group. Seventy-eight percent of the participants had a significant treatment response on the Clinician Global Severity & Improvement Scales. Parents and children rated the treatment as highly acceptable. In summary, CBIT had similar effectiveness whether delivered via videoconference or face-to-face.

The Himle and colleagues (2012) study provided additional evidence for a telehealth model of CBIT. However, the authors did not include a waitlist control condition to control for the waxing and waning nature of tics, as well as other possible extraneous events that could explain the reduction in tic severity. Also, the individuals in the videoconference condition still attended sessions in a clinic setting. While the clinic setting was the same for all participants and demonstrated experimental control, the purpose of videoconferencing is to allow greater flexibility in terms of where therapy is delivered. The results may not reflect the provision of CBIT through videoconferencing in other settings (e.g., participants' homes).

More research regarding the use of telehealth as a delivery method for CBIT is needed. However, the two studies described above provide initial support for use of a telehealth model.

Mechanisms of Change in HRT

Most of the available research has investigated the efficacy of HRT, either by examining the traditional or enhanced forms, or by examining the components. The exact mechanisms which lead to changes in tics still remain unknown (Himlet et al., 2006; Franklin & Himle, 2007). A variety of hypotheses have been proposed.

Azrin and Nunn (1973) initially believed that HRT was effective because the incompatible response strengthened muscles opposite of the muscles used during the tic. This

hypothesis has not been supportive in the literature, especially with the discovery that responses that are dissimilar but not incompatible with the tic can be effective CRs (Sharenow et al., 1989; Woods et al., 2006).

Hypotheses of positive punishment and of positive reinforcement have been proposed by Miltenberger, Fuqua, and Woods (1998). It is possible that use of the CR is aversive and its use effectively punishes the tic. The same hypothesis has been proposed for self-monitoring of tics (Ollendick, 1981); however, it has not been empirically tested. It is also possible that HRT might be effective because it involves positive reinforcement of the CR by social praise and prompts made by the therapist during treatment and through social support training. Through this positive reinforcement, it is possible that the tic is replaced by the CR (Miltenberger, Fuqua, & Woods, 1998). While possible, social support training has not been found to be necessary with adults, which limits this hypothesis (Anthony, 1978; Woods & Miltenberger, 1995). This hypothesis has also not been empirically tested.

Another hypothesis has been proposed based on a model of automatic negative reinforcement (Capriotti et al., 2014a; Evers & Van de Wetering, 1994; Himle et al., 2006). This model suggests that tics occur as a result of the premonitory urge, leading to relief from this urge. By engaging in HRT, it is possible that individuals are able to habituate to the premonitory urge, which occurs through use of the CR and which is characterized by the urge becoming less noticeable and less bothersome over time. This urge habituation hypothesis has also been used to explain the effects of exposure and response prevention, as individuals are asked to suppress their tics for longer and longer periods of time (Hoogduin et al., 1997; Woods et al., 2000; Woods et al., 2001). However, not all individuals report experiencing a premonitory urge prior to tics; therefore, this hypothesis is not fully supported (Woods et al., 2005).

Criticism of HRT and Barriers to Its Use

Even though HRT is considered a well-established, evidence-based treatment for CTDs (Cook & Blacher, 2007), it is still underutilized for a variety of reasons (Capriotti et al., 2014b; Woods et al., 2010). In the community-based sample of the study by Woods and colleagues (2010), behavioral interventions were the fourth most utilized intervention with children (23.6 percent), following medication (82.9 percent), changes to diet (29.6 percent), and taking fatty acid supplements (24 percent). In adults, behavioral interventions were the eighth most utilized treatment (17.2 percent), following medication (92.7 percent), diet changes (26.5 percent), sensory integration therapy (26 percent), meditation (23 percent), talk therapy (21.9 percent), "other" (21.7 percent), and vitamin supplements (17.4 percent). These findings are somewhat disheartening as many of the other strategies utilized more often than behavioral interventions do not have empirical support (Woods et al., 2010). In addition, psychologists, who are often the providers of HRT, are not accessed as often as other clinicians (Woods et al., 2010). This section will discuss barriers to using HRT and will address common criticisms associated with the use of HRT.

For individuals who sought treatment in the Woods and colleagues (2010) study, the most frequently reported barriers included challenges finding a practitioner familiar with CTDs and their treatment, cost of treatment, the time commitment needed for treatment, and the potential negative side effects of treatment. When specifically asked about why they had not sought out behavioral interventions, the most frequent reasons for children included lack of knowledge about the interventions and concern about negative side effects (e.g., rebound effect, symptom substitution, concern about interference with other tasks). The adult sample most commonly reported lack of knowledge about the interventions and the cost of treatment, both in terms of

finance and of the time commitment required (Woods et al. 2010). In addition to individuals with CTDs, medical and mental professionals may also be less informed about HRT. Marcks, Woods, Teng, and Twohig (2004) administered a questionnaire to physicians (n = 35) and psychologists (n = 32) and found that 35 percent of psychologists and 14 percent of physicians were familiar with HRT. Fewer than 10 percent of either type of practitioner reported being familiar enough with HRT to implement it.

With regard to lack of knowledge about behavioral interventions, concerted efforts are being made to circulate information through larger-scale studies as well as through support groups such as the Tourette Syndrome Association (TSA; Woods et al., 2010). In addition, consideration should be given to specific outreach to clinicians. The Marcks and colleagues (2004) study indicated that 46 percent of physicians and 62.5 percent of psychologists would be interested in learning more about HRT. The TSA and CDC have joined together with the goal of increasing awareness of HRT, as well as increasing the number of providers of HRT (Woods et al., 2010).

Limited numbers of providers of HRT was another barrier endorsed by the adults and children in the Woods and colleagues (2010) study. This barrier has been reported elsewhere (Cook & Blacher, 2007). Through the training programs supported through the TSA and CDC, over 1000 clinicians have been trained in HRT. This number is expected to grow over time (Woods et al., 2010). In addition to increasing the number of providers, telehealth has been used effectively for the delivery of CBIT (Himle et al., 2010; Himle et al., 2012). This mode of delivery may allow for more individuals with CTDs to be reached for HRT, especially in rural areas; however, challenges are currently being faced with regard to clinician reimbursement and

security of technology. Regardless, this mode of delivery holds much promise (Woods et al., 2010).

The biggest criticisms surrounding HRT relate to the belief of potential negative side effects. The three most common negative side effects cited in the literature are the possibility of a "rebound effect," symptom substitution, and increases in tics due to specifically focusing on tics (Woods et al., 2010). None of these side effects have been empirically supported; however, the belief in the side effects remains strong (Marcks et al., 2004; Woods et al., 2010). Each of these criticisms will be addressed individually.

First, HRT has been criticized for leading to a "rebound effect." This belief is based on reports by significant others about a period of increased tics that occurs after a period of volitional suppression (Burd & Kerbeshian, 1987; Murphy et al., 2013). For example, parents might blame increased tics right when a child returns home from school as a release of pent up tics that were suppressed during the day (Woods et al., 2010). The potential for a rebound effect has been empirically tested and has not resulted in confirmation of this belief (Himle & Woods, 2005; Himle et al., 2007; Meidinger et al., 2005; Specht et al., 2014; Woods et al., 2008a). It has also not been a result of any of the studies mentioned above that specifically investigated the efficacy of HRT, which have all demonstrated either decrease in tics or tic stability pre- to posttreatment.

Next, concern has been expressed regarding symptom substitution. Symptom substitution is the fear that treating tics through behavioral methods will increase tics that are not currently being treated, will lead to new tics, or will lead to higher rates of anxiety or irritation (Burd & Kerbeshian, 1987; Burd & Kerbeshian, 1988). In the study conducted by Woods and colleagues (2003), all tics were monitored (i.e., targeted tics and untargeted tics). The untargeted

tics did not increase after the introduction of HRT. In fact, in a few of the participants, the untargeted tics decreased. In the other participants, the untargeted tics remained stable. Further evidence to suggest that symptom substitution does not happen was found the CBIT study by Piacentini and colleagues (2010). Information on a variety of adverse events, including illness, physical injury, personal conflicts, and symptom substitution were collected from the participants. This information did not provide evidence for symptom substitution.

Related to the rebound effect and symptom substitution, another common concern is that talking about or focusing on tics will make them worse (Woods et al., 2010). This concern is actually partially valid. Some studies suggest that talking about tics may lead to a brief increase in anxiety and tics (Dufrene, Watson, Echevarria, & Weaver, 2013; O'Connor, Brisebois, Brault, Robillard, & Loiselle, 2003; Silva et al., 1995; Storch et al., 2007a; Woods et al., 2001); however, this increase appears to be temporary. In addition, in studies that have investigated self-monitoring or AT alone (Ollendick, 1981; Woods et al., 1996; Wright & Miltenberger, 1987) as well as all of the studies that have investigated HRT, which includes AT, increases in tics after treating or talking about them were not observed.

Conclusions about HRT

All of the studies investigating HRT have limitations, the most frequent of which are small sample sizes and variable methodology. Multiple randomized controlled trials have been conducted in addition to studies employing single case designs. Despite the limitations, the available research base for HRT is impressive. Taken together, the research suggests that HRT methods are an effective behavioral treatment for CTDs.

Based on the available evidence, the common criticisms of HRTs, while understandable based on the history of psychological interventions, have not been supported in the literature. In

addition, while barriers to behavioral intervention have been identified, efforts are being taken to increase the number of practitioners trained in HRT as well as to provide information to medical and mental health care professionals and individuals diagnosed with CTDs. Additional research is being conducted regarding different delivery systems for HRT, the most recent of which being use of telehealth. One area that has not yet been explored is the possibility of a parent-mediated version of HRT. The next sections will outline the rationale for this type of intervention delivery with children with CTDs.

Parent-Mediated Interventions

As mentioned in the first chapter, parent mediated interventions were first developed in the 1960s and have been used for a variety of child and adolescent populations (Kaminski, Valle, Filene, & Boyle, 2008). The following sections discuss the important aspects of parent-mediated interventions, with a specific focus on parent coaching. Advantages and potential barriers are discussed. Finally, the potential application to children with CTDs is presented.

Important Components of Parent Coaching

Studies evaluating the effectiveness of parent-mediated interventions have indicated a wide variety of intervention techniques are used (Barton & Fettig, 2013). Kaminski and colleagues (2008) describe components that were associated with larger effect sizes for parent-mediated interventions. The two components resulting in the largest effect sizes included focusing on increasing positive interactions between parents and children and requiring parents to directly practice the new skills with their children. Similarly, Dunst and Trivette (2012) investigated use of practices that yield more effective adult learning. They found that strategies that involved active involvement and in vivo practice on the part of the learners led to the largest impact on knowledge, skills, and attitudes. With regard to use of strategies outside of

intervention sessions and practice at home, McConnell, Parakkal, Savage, and Rempel (2015) found that an important factor to parents' intervention adherence was how much change to their daily life was required. Interventions requiring the parents to set aside time which did not already exist in their schedule resulted in lower adherence to the intervention plan. These factors are important to consider when developing parent-mediated interventions.

Using a family-centered approach in parent-mediated interventions, clinicians are often referred to as coaches rather than therapists (Rush & Sheldon, 2005). The use of this specific language is important as it reflects a collaborative approach versus a top-down, directive approach. Important to the coaching role, five characteristics have been identified as leading to positive outcomes: joint planning, observation, action/practice, reflection, and feedback (Rush & Sheldon, 2005). Joint planning refers to parents playing an active role in the development of goals and provision of intervention strategies. Having parents select treatment goals directly ties the intervention to their particular concerns, which may result in better buy-in for the intervention. Observation refers to the coach's monitoring of the skills being used by the parents. Action refers to the opportunities to directly implement the skills in clinic and in real-world settings. Reflection refers to evaluating the strategies in terms of what went well and what could be improved to better refine skills. Feedback refers to positive and critical statements made by the coach to assist in refining the parents' skills. In order for an intervention to be considered as using a coaching model, these elements must be included (Rush & Sheldon, 2005).

Examples of two evidence-based interventions which utilize a parent coaching model are Parent Child Interaction Therapy (PCIT; McNeil & Hembree-Kigin, 2010) for young children with disruptive behavior and Project **Im**proving **P**arents **a**s **C**ommunication **T**eachers (Project ImPACT; Ingersoll & Dvortcsak, 2010) for young children with autism spectrum disorder. In

PCIT, coaching is provided from a different room through use of a one-way mirror and a "bug-in-the-ear" device. It typically is provided in 14 weekly sessions. Parents learn child-directed play interaction skills and parent-directed skills, including giving effective commands, praise for compliance to instructions, and discipline techniques (McNeil & Hembree-Kigin, 2010). In Project ImPACT, coaching is typically provided in the same room as the parent and child dyads. It occurs over the course of 12 weeks and can be delivered once or twice per week. It has also been modified as a group model and for use in schools. During Project ImPACT, parents learn how to help their children build social communication skills through interactive, child-directed play techniques and through direct, parent-directed teaching techniques. Both interventions have demonstrated positive outcomes for children. In addition, parents have been able to implement both with fidelity (Ingersoll & Wainer, 2013; Rae & Zimmer-Gembeck, 2007).

Advantages to Parent-Mediated Interventions

Many advantages exist for using parent-mediated interventions (Ingersoll & Dvortcsak, 2010). First, in addition to positive child outcomes, this type of delivery-model can increase the intervention hours received. Parents specifically trained in the strategies can use these strategies both during the coaching sessions and also at home. Next, parents have been able to increase skills during these interventions and implement them with fidelity, which may lead to increased feelings of self-efficacy and feelings of optimism for their children's future (Koegel, Schreibman, Britten, Burke, & O'Neill, 1982). This type of delivery model also can lead to better generalization and maintenance of parents' skills, and has been viewed by parents as more effective than provider-led models (Koegel et al., 1982).

There are also advantages to use of a parent coaching model specifically, instead of relying on parent education only (McNeil & Hembree-Kigin, 2010). First, direct coaching

allows for immediate correction of any errors instead of allowing incorrect practice in the home setting. Next, coaching clinicians can model problem-solving techniques that arise during intervention, which helps the parents learn how to individualize the intervention for their children. In addition to learning problem-solving, parents also have the opportunity to build confidence in their own skills. Direct coaching may also help parents learn skills faster.

Potential Barriers to Parent-Mediated Interventions

As with any treatment, potential barriers to use of a parent-mediated intervention model may arise (Ingersoll & Dvortcsak, 2010). First, most interventions have obstacles with respect to logistics. Family schedules are busy, so finding time to incorporate treatment may be difficult. Attendance to sessions are key for learning skills, so it is important to consider flexible scheduling and discussion of commitment with parents ahead of time. Along with scheduling conflicts, low motivation may be a barrier to treatment. Parents may not be motivated or may be uncomfortable with learning the strategies and are looking more for a "quick fix" (Ingersoll & Dvortcsak, 2010; McNeil & Hembree-Kigin, 2010). Rapport building at the outset is important to establishing a positive relationship. Including parents in goal setting and discussing techniques ahead of time while also providing opportunity for feedback and time to answer questions may also help with motivation (Rush & Sheldon, 2005). On the other hand, parents may also be more motivated than the child for intervention, so it is important to screen for motivation on the part of the child ahead of time.

In addition to logistics, the belief in the superiority of provider-led interventions may also be a barrier (Ingersoll & Dvortcsak, 2010). Despite the evidence in support of parent-mediated interventions, some practitioners do not think that parent-mediated interventions are appropriate. This opinion may occur for a variety of reasons. For example, practitioners have specific

training and competencies with particular training approaches, which could lead to bias against use of other approaches or the belief that only individuals trained in the approach should deliver the strategies. Additionally, some practitioners may believe that having parents trained as co-therapists could result in role confusion on the part of the parent. Finally, there is some belief that parent-mediated interventions place more burden on parents who already lead busy lives, or that parents may feel that they are being blamed for their children's difficulties. While these concerns are understandable, they have not been supported in the literature. Parents tend to rate these types of interventions as acceptable, tend to benefit from the interventions themselves, and may actually want to develop the skills (Hume, Bellini, & Pratt, 2005; Ingersoll & Dvortcsak, 2010; Mahoney et al., 1999; Whitaker, 2002). In general, the potential benefits for parent-mediated intervention appear to outweigh the potential barriers.

Application to CTDs

Delivery of HRT through a parent-mediated system has not yet been established in the literature. Freeman and Duke (2013) published a case report of a 3-year-old girl with a complex motor stereotypy, during which they taught the parents' the strategies for HRT who then implemented them with their daughter. They did not provide much detail in terms of how the therapy sessions were structured, but they did report that it was successful in reducing the child's stereotypy from occurring during 85 percent of intervals in a partial interval schedule to 2 percent of intervals. Freeman and Duke (2013) noted that parents can be taught the strategies. Unfortunately, no fidelity of implementation data were presented to further support use of a parent-model. Clearly this case study is limited in terms of methods and the conclusions that can be drawn; however, this case study does provide promise that it can be a useful treatment delivery system for HRT.

CHAPTER THREE

METHOD

Participants

All study methods were initially approved by the University of Iowa Institutional Review Board. Written informed consent and assent was collected from all participating parents and children, respectively. Child participants were three children referred from two hospital-based specialty clinics focused on treatment of tic disorders. For each child participant, only one parent served as a participant for all sessions. Each child participant was included based on the following selection criteria:

1. The participant was a child or adolescent between the ages of 7 years, 0 months and 15 years, 11 months.
2. The participant had a prior diagnosis of a chronic tic disorder (i.e., Tourette's disorder, persistent motor tic disorder, or persistent vocal tic disorder).
3. The participant had the following cutoff scores on the Yale Global Tic Severity Scale:
    a. For a diagnosis of Tourette's disorder, a score of greater than or equal to 13 was required.
    b. For a diagnosis of persistent motor tic disorder or persistent vocal tic disorder, a score of greater than or equal to 9 was required.
4. The participant had at least one motor or vocal tic occurring, on average, more than once within a 10-minute period.
5. The participant could have had a comorbid diagnosis of attention-deficit/hyperactivity disorder (AD/HD), obsessive compulsive disorder (OCD), learning disorder, or speech disorder, which was true for two of the participants.

6. The participant may be prescribed non-antipsychotic medication (e.g., clonidine or guanfacine) for the treatment of tics, provided that they were taking the medication for at least 6 weeks prior to participation in the study.

Child participants were excluded for the following reasons:

1. The child was below 7 years, 0 months of age or above 15 years, 11 months of age.
2. The child had a comorbid diagnosis of autism spectrum disorder, intellectual disability, oppositional defiant disorder, or conduct disorder.
3. The child was newly (i.e., within 6 weeks) prescribed non-antipsychotic medication for the treatment of tics.
4. The child was prescribed an antipsychotic medication (e.g., Risperdal) for the treatment of tics.
5. The child previously participated in HRT for the treatment of tics.

Each parent participant met the following inclusion criteria:

1. The participant was fluent with the English language.
2. The participant lived within 130 miles of the treatment setting.
3. The participant agreed to attend all sessions.

One of the participants elected to drop out of the study after the first intervention session due to cited personal reasons. Two parent-child dyads completed the study. First, Tom was an 8-year-old Caucasian boy in second grade with prior diagnoses of persistent vocal tic disorder and AD/HD. At the time of the study, he was experiencing one vocal tic: an /m/ sound. He also had a history of throat clearing. Per his mother, the frequency of the /m/ sound varied considerably from once every day to multiple times per hour. Tom was prescribed Focalin for AD/HD, but no other medications. He lived with his biological parents and four siblings. His mother, Mary,

participated in all sessions. She was also Caucasian, 41 years old, and a stay-at-home mother. She had a college education. Family history was significant for tics (i.e., Tom's grandfather reportedly exhibited a facial grimace and his older brother exhibited an eye twitch). Setting and antecedent variables for Tom's tics included feeling anxious, playing video games, going into new public places, the morning routine, and school. Consequences prior to participation were reported as ignoring from parents and siblings, parents asking him to change activities, parent providing comfort, negative attention from peers (e.g., "Stop!" or "That's annoying."), his teacher asking him to control it, and infrequent comments from strangers.

Nate was a 10-year-old multiracial boy in fourth grade and diagnosed with Tourette's disorder. At the time of the study, he was experiencing five motor tics: eye blinking, facial twitching, shoulder rolling, arm tensing, and hand flexing. He also had a history of a humming noise. Per parent report, these tics occurred daily up to multiple times an hour. Nate was prescribed medication for allergies only. He lived with his biological parents. His father, David, participated during all sessions. He was Caucasian, 57 years old, and retired. He completed high school. Family history was significant for tics (i.e., Nate's father reported experiencing body tensing when younger). Setting and antecedent variables for Nate's tics included exercise and physical activity, morning hours, evening hours (i.e., when feeling tired before bed), excitement, school, public settings, and meals. Consequences included ignoring and sometimes parental requests to stop or calm down.

Setting and Materials

All sessions were conducted in a clinic therapy room at the Center for Disabilities and Development at the University of Iowa Hospital and Clinics. These clinic rooms were equipped with video recording equipment and microphones, which were used to record all sessions for

data collection purposes. The video recording equipment fed into computer monitors in an adjacent room, allowing for direct observation as well as recording. This software was maintained by the hospital and the server on which videos are kept were secure. Videos on the server are deleted after 60 days, so videos were downloaded onto an external hard drive, which was kept in a locked file cabinet at the Center for Disabilities and Development when not being evaluated by the research team.

The therapy rooms contained a table and chairs. Toys and activities were provided by the researchers to the parent-child dyads during all sessions. These toys and activities were based on each child's preference as stated during the initial intake interview. Tom opted to play video games on a tablet when allowed during sessions. Nate chose to play basketball, board games, video games on a tablet, and games from home when allowed during sessions.

During coaching, a "bug in the ear" microphone system as described in the PCIT manual (McNeil & Hembree-Kigin, 2010) was used. In this system, the parent was provided an earpiece and two-way radio through which the research team provided feedback during applicable intervention sessions. The research team member providing the feedback watched the sessions via live, closed-circuit video in an adjacent room.

Parents were provided with training materials for the first four sessions of the intervention. These materials were developed by the researcher, but included adapted materials from the CBIT Parent Workbook (Woods et al., 2008c). The materials provided an outline and rationale for strategies to be learned in the following intervention session. In addition to these materials, PowerPoint presentations were created for each session to provide an outline prior to each session.

Measures

Clinical Interview

Prior to treatment, both children and their families attended an intake session to determine eligibility for the study through a clinical interview provided in Appendix A. This interview included the following information: ages of the children and their caregivers, race and ethnicity of children and caregivers, caregiver highest level of education obtained, caregiver occupation, prior diagnostic history of the children, tic history, tic severity, presence of comorbid disorders, and level of psychosocial functioning or impairment.

Yale Global Tic Severity Scale (YGTSS)

To assess tic severity, the YGTSS (Leckman et al. 1989) was conducted pre-, mid-, and post-intervention (i.e., during the last intervention session and at the one month follow up session). The YGTSS is a semi-structured interview completed by a clinician that includes a checklist of motor and phonic tics, severity ratings, and impairment ratings. Informants were asked about tics that have occurred during the past week and historically to attain information about the topography and anatomical location of tics. Following this checklist, clinicians ask additional questions to rate the tics based on number, frequency, intensity, complexity, and interference. Each of these dimensions were rated on a 0 to 5 scale. These scores were added together to attain severity scores for motor tics and phonic tics separately with a range of 0 to 25 each. The motor and phonic tic severity scores are added together to yield a total tic severity score with a range of 0 to 50. Higher scores on the YGTSS indicate higher tic severity and impairment from tics. This measure has acceptable internal consistency ($\alpha = .92$; Storch et al., 2005) and discriminant validity compared to the AD/HD Clinical Global Impression (CGI) Scales ($r = .11$, N.S.; Leckman et al., 1989). It was found to have good convergent validity with

the CGI scale (r = .90, p < .0001; Leckman et al., 1989). It has been shown to have adequate interrater reliability (r = .85, p < .0001; Leckman et al., 1989; Storch et al., 2005). In addition, it has been found to have good stability over time (ICC = .89; Storch et al., 2005).

Parent Tic Questionnaire (PTQ)

An additional measure of tic severity, the PTQ, was administered to parents weekly during baseline, intervention, and at the one month follow up sessions to assess their perception of the number, frequency, duration, intensity, and complexity of tics (Chang, Himle, Tucker, Woods, & Piacentini, 2009). The PTQ is a paper and pencil self-report questionnaire. Parents first filled out a tic checklist for their children, which included a list of 28 motor and phonic tics similar to the YGTSS. After identifying tics that occurred during the past week, parents rated each tic based on how frequently it occurred on a 0 (absent) to 4 (constantly) scale. They also rated how intense (i.e., noticeable) the tics were on a 1 to 4 scale (higher numbers on this scale indicate greater intensity). The frequency and intensity scores were added together to yield a total score ranging from 0 to 8 score for each tic. Scores for each tic were then added together to yield subscale scores for each class of tics. These subscale scores were added together to yield a total severity score between 0 to 79, where higher scores indicated greater tic severity. This measure has been demonstrated to have adequate internal consistency (α = .86 and .90 for two administrations) and test-retest reliability (ICC = .84; Chang et al., 2009). It has also been demonstrated to have high concurrent validity with the YGTSS (r = .72 for both administrations; Chang et al., 2009).

Premonitory Urge for Tics Scale (PUTS)

To assess presence and intensity of potential premonitory urges before intervention, mid-way through intervention, and after intervention, child participants filled out questions from the

PUTS. The PUTS is a 10-item questionnaire that examines sensory feelings prior to engaging in tics (Woods, Piacentini, Himle, & Chang, 2005). The items were rated on a 1 ("not at all true") to 4 ("very much true") scale. It has been found to have adequate internal consistency ($\alpha = .81$) and test-retest reliability ($r = .86$, $p < .01$; Woods et al., 2005). It has also been demonstrated to have moderate concurrent validity with the YGTSS ($r = .31$, $p < .05$; Woods et al., 2005).

Subjective Units of Distress Scale (SUDS)

Children were asked to rate their subjective level of experienced distress from their tics every session, including during pre-intervention baseline, using the SUDS. Originally developed by Wolpe (1969), SUDS usually has a scale of 0 to 100, which individuals use to gauge their subjective feelings of intensity of psychological symptoms. Woods and colleagues (2008b) adjusted the scale for children so that it ranges from 0 ("no distress") to 10 ("maximum distress"). Use of SUDS as a global measure of experienced discomfort has been supported in the literature. It appears to have moderate concurrent validity with the MMPI-2 ($r = .35$, $p < .05$ when compared with scale A; $r = .37$, $p < .01$ when compared with scales 1 to 3), as well as sensitivity to change over time (Tanner, 2012).

Motivation for Youth's Treatment Scale (MYTS)

To assess children's motivation for treatment, the MYTS (Breda & Riemer, 2012) was administered. This 8-item measure has questions that pertain to current level of impairment, desire for help, and readiness for treatment. It has two forms: one for children and one for the adult caregivers. Each question is rated on a 5-point Likert scale from "strongly disagree" to "strongly agree." This measure has been found to have adequate internal consistency ($\alpha = .82$ for the youth MYTS; $\alpha = .86$ for the caregiver MYTS) and sensitivity to change over time (Breda & Riemer, 2012). In the current study, this measure was administered before intervention, mid-

way through intervention, and after intervention to both children and their caregivers; however, the wording was modified so that it was directly applicable to children experiencing tics.

Parent Motivation Inventory (PMI)

To assess parents' motivation to participate in treatment and to gauge their readiness to learn the skills of habit reversal, the PMI was administered before intervention, mid-way through intervention, and after intervention. This 25-item paper and pencil self-report measure includes questions in three domains: desire for change in child's symptoms, readiness to change personal behavior, and perceived ability to change personal behavior (Nock & Photos, 2006). Each question is rated on a 1 (strongly disagree) to 5 (strongly agree) scale. This measure has been demonstrated to have strong internal consistency ($\alpha = .96$) and test-retest reliability ($r = .76$, $p < .001$; Nock & Photos, 2006).

Treatment Evaluation Inventory – Short Form (TEI-SF)

The Treatment Evaluation Inventory (TEI) was first developed by Kazdin (1980) as a measure of parents' acceptability of treatment for their children with challenging behavior. It has since been adapted into a shorter form, the TEI-SF (Kelly, Heffer, Gresham, & Elliott, 1989), and has been used with children with CTDs (Himle et al., 2010). The TEI-SF is a 9-item paper and pencil self-report questionnaire that assesses respondents' perception of treatment acceptability and discomfort with treatment procedures. Questions are rated on a 5-point Likert scale ranging from "strongly disagree" to "strongly agree." The TEI-SF has been demonstrated to have adequate internal consistency ($\alpha = .85$) and to be able to discriminate between treatments (Kelly et al., 1989). To assess social validity, this measure was administered to children and their parents before intervention after the procedures of HRT are explained, mid-way through the

intervention, and post-intervention. Slight wording adjustments were made to make it directly applicable to individuals with CTDs.

Treatment Acceptability Questionnaire (TAQ)

In addition to the TEI-SF, social validity data were collected from the children and their parents before, mid-way, and after intervention using the TAQ. The TAQ is a 6-item paper and pencil self-report measure (Hunsley, 1992). It includes questions pertaining to acceptability of treatment procedures as well as acceptability of the therapist. Each question is rated on a 7-point Likert scale, with higher scores indicated higher acceptability. It has been demonstrated to have good internal consistency (range of Cronbach's $\alpha = .74$ - .81 across four studies) and temporal stability ($r = .78$, $p < .001$; Hunsley, 1992). It also had good concurrent validity with the TEI ($r = .87$, $p < .001$; Hunsley, 1992).

Independent and Dependent Variables

The independent variable for this study was the parent-mediated format of HRT. Besides the measures listed above, the dependent variable of primary interest was parent fidelity of implementation. Parent fidelity of implementation was rated through percentage of steps completed based on the CBIT Practitioner Manual (Woods et al., 2008b). The list of steps is provided in Appendix B. Parent fidelity was rated during the direct practice portion of each session. Additional dependent variables included tic severity, child premonitory urge ratings, parent motivation, child motivation, and parent and child treatment acceptability. Tic severity was measured by direct observation and rating scales. For direct observation, tic frequency was measured using 10-second partial interval recording during 30 minutes of the sessions in which parent fidelity was observed. The other dependent variables were measured via the rating scales discussed in the Measures section.

General Procedures

Baseline

During baseline, each parent and child were provided with developmentally appropriate toys and activities. The parent was instructed to play with his or her child and respond to his or her child's tics as s/he typically would at home. This play period lasted for 30 minutes and parents' fidelity was scored using the rating form in Appendix B. In addition to parents' fidelity of implementation, data were collected regarding the frequency of tics exhibited by the child using 10-second partial interval recording during 30 minutes of the observation period.

Intervention

A parent-mediated version of HRT was implemented after parent fidelity data were stable for three consecutive baseline sessions. Data were considered stable if they occurred at low rates (i.e., 0 to 15 percent) with no increasing trend. The parent and child to first receive intervention were chosen based on when they were referred (i.e., there was a two week delay between the referral for the first patient and the referral for the second patient; the intervention for the first patient started soon after their referral to and consent to participate in the study). Intervention consisted of six sessions. Each session included psychoeducation about procedures delivered to the parent participants by the researchers for 15 to 20 minutes. Following this instruction, parents delivered the applicable habit reversal strategies for 30 to 40 minutes with their child. An outline of the intervention sequence is provided in Appendix C. Each intervention session followed a developmental trajectory as each session built upon skills learned during the previous session. As mentioned previously, these sessions were developed based on the CBIT Practitioner Manual and adapted for use with parents (Woods et al., 2008b). The parent was coached during the intervention by one of three doctoral students in School Psychology including the principal

investigator, trained in habit reversal procedures, all under the supervision of a licensed psychologist. Both the principal investigator and the licensed psychologist previously attended training in CBIT techniques through the Tourette Syndrome Behavior Therapy Institute.

Each intervention session began with the parent and/or child filling out questionnaires. The parent received didactic training from a research team member through a PowerPoint presentation highlighting the specific topics and skill areas assigned for that week (e.g., tic disorders, functional assessment, awareness training, competing response training, etc.). Throughout the presentation, the parents were allowed opportunities to ask questions of the research team member. The presentations included a model of the skills by the research team member, as well as role play with the parents. The child participants engaged in free play or another activity of choice in an adjacent room.

During one-on-one coaching sessions, the parent participants entered the treatment room with their child. The research team member observed from an adjacent room and provided coaching through a "bug in the ear" system. Parents' behavior was scored for fidelity of implementation. Data were also collected regarding the child's frequency of tics.

Follow-up Session

Parent and child participants returned to the same treatment room one month after the completion of the intervention. A research team member conducted the YGTSS. The child filled out the PUTS. The parents completed the PMI and PTQ. Parents were asked to implement the HRT strategies and were observed using the intervention techniques with their child. Data were collected on their fidelity of implementation, as well as the child's tic frequency.

Research Questions

This study sought to answer the following research questions:

1. Will parents of children with CTDs participating in a parent-mediated version of HRT learn to implement the strategies with adequate fidelity?
2. Will children with CTDs receiving a parent-mediated version of HRT demonstrate decreases in number, frequency, and severity of tics as measured through direct observations, parent questionnaires, and self-report questionnaires?
3. Will children with CTDs receiving a parent-mediated version of HRT demonstrate decreases in premonitory urge sensations as measured through self-report questionnaires?
4. Will parents and children participating in a parent-mediated version of HRT find the intervention acceptable as measured through self-report questionnaires?
5. Will treatment motivation on the part of the children with CTDs impact parents' fidelity of implementing the strategies of a parent-mediated version of HRT as measured by self-report questionnaires on the part of the children and clinician observation and rating of parents?
6. Will parent motivation to participate in treatment be related to increased skill acquisition and adequate fidelity as measured by self-report and direct observation?

Experimental Design

The design for this study was a nonconcurrent multiple baseline across participants design. The primary strength of this design was that it did not require that the intervention to be withdrawn for a reversal phase (Kazdin, 2011). Habit reversal therapy is an intervention that should result in skills that will not return to baseline levels of performance, although fidelity may

deteriorate over time. Additionally, it is a 6-week intervention with each week building on the skills learned during the previous week. Based on these factors, removing the intervention to observe whether parental fidelity of implementation returns to baseline levels would not produce meaningful results. In addition to lack of need for a reversal, the nonconcurrent multiple baseline design also allowed the intervention to start relatively soon after participants were referred to and consented to participate in the study.

CHAPTER FOUR

RESULTS

Parent Participants

Parent Fidelity (Research Question 1)

Research question 1 stated: Will parents of children with CTDs participating in a parent-mediated version of HRT learn to implement the strategies with adequate fidelity? Parent fidelity was measured via a step-by-step checklist (see Appendix B). Results from the direct observations of parent fidelity are presented in Figure D1 in Appendix D. Overall, both parent participants who completed the study demonstrated an increase in skills learned and adequate fidelity after the intervention was implemented. Each parent participant and their results are described below.

Mary. During baseline, Mary did not demonstrate any intervention skills (i.e., during each baseline session, she obtained a stable score of 0% of steps implemented independently). Immediately following the first intervention session, Mary displayed 19.05% of steps independently. Mary's skills continued to increase across sessions (M = 67.35% steps completed independently during the treatment and follow-up phases), with her highest fidelity rating achieved near the end of the intervention (i.e., 90.48% of steps completed independently during the fifth treatment session). During the final intervention session, coaching was completely faded. Mary completed 76.19% of intervention steps independently. At the one-month follow up, she once again achieved 90.48% of steps independently. The percentage of non-overlapping data (PND; (Scruggs & Mastropieri, 1994; Scruggs, Mastropieri, & Casto, 1987) was 100% from baseline to intervention and continued to be 100% after coaching was faded out, suggesting a strong treatment effect for Mary.

David. David did not complete any steps of the intervention independently during all baseline sessions (i.e., a stable baseline of 0% of steps completed independently was observed). Following introduction of the intervention, he independently implemented 14.29% of intervention steps. Data on David's skill acquisition demonstrated an increasing trend across sessions with his highest fidelity rating achieved mid-intervention (i.e., 95.24% of steps completed independently during the third and fourth treatment sessions). Towards the end of the intervention and during the follow-up phase, a slight decrease in fidelity was observed which coincided with the fading of coaching. David's overall average during treatment and follow-up phases was higher than baseline (M = 74.15% steps completed independently). At the one month follow up, David completed 76.19% of steps independently. PND for David was 100% from baseline to intervention and continued to be 100% after coaching was faded out, suggesting a strong treatment effect for David.

Parent Treatment Acceptability (Research Question 4)

Research question 4 stated: Will parents and children participating in a parent-mediated version of HRT find the intervention acceptable as measured through self-report questionnaires? Treatment acceptability for parent participants was measured via the TEI-SF and TAQ pre-intervention, mid-intervention, and twice post-intervention (i.e., during the last intervention session and at a one month follow up session). Results from both measures are presented in Table D1 in Appendix D. Across all data collection points, both parent participants rated the intervention as highly acceptable (TEI-SF overall M = 36.25, range 32 – 38); TAQ overall M = 40.25, range 39 - 42). In addition, these ratings appeared to remain stable over time.

Parent Motivation for Treatment (Research Question 6)

Research question 6 stated: Will parent motivation to participate in treatment be related to increased skill acquisition and adequate fidelity as measured by self-report and direct observation? Parent skills were measured by direct observation and results from these observations are presented in Figure 1. Parent motivation to participate in treatment was measured via the PMI and MYTS. Results from these measures are presented in Table D2 and Table D3, respectively, in Appendix D.

On the PMI, David consistently demonstrated higher ratings on the Readiness to Change scale (overall scale M = 69.75, range = 69 - 70) than Mary (overall scale M = 58, range = 55 – 60). David also consistently demonstrated slightly higher ratings on the Perceived Ability to Change scale (overall scale M = 17.75, range = 16 – 19) than Mary (overall scale M = 14.75, range = 14 – 15). Both parents' ratings on the Desire for Change scale (overall M = 22.5, range = 22 – 26) and were comparable. David's Total scale ratings (overall scale M = 112, range = 111 - 113) were higher than Mary's ratings (overall scale M = 95.25, range = 91 - 97). Despite the difference in ratings for Readiness to Change and Perceived Ability to Change, both parents' ratings would still be considered high for motivation as they were closer to the maximum value for this scale (maximum value for Readiness to Change = 70; maximum value for Perceived Ability to Change = 20). Ratings on this measure for both parent participants remained stable over time. In combination, parents' ratings indicated stable and high levels of motivation to participate in treatment (overall M = 103.63, range = 91 – 113; maximum possible Total score = 125).

On the MYTS, both parents' ratings fell in the "Medium" range (overall M across sessions = 3.4, range 3.3 – 3.8) overall. For the Treatment Readiness scale, David's ratings

consistently fell in the "High" range (overall M = 5, range = 5), whereas Mary's ratings varied between "High" and "Medium" (overall M = 4, range = 3.8 – 4.5). For the Problem Recognition scale, Mary's ratings varied between "Medium" and "Low" (overall M = 2.8, range = 2.5 – 3) and David's ratings were consistently "Low" (overall M = 2.2, range = 2 – 2.5). Ratings for both parents remained stable across scales and time.

As mentioned previously, both parent participants demonstrated an initial increasing trend in parent fidelity. Mary's data consistently demonstrated this increasing trend over time, whereas David's data indicated a slight decreasing trend from mid-intervention to the one month follow up session. Changes in parent skill acquisition and fidelity were not consistent with the stable ratings from the PMI and the MYTS.

Child Participants

Child Tic Severity (Research Question 2)

Research question 2 stated: Will children with CTDs receiving a parent-mediated version of HRT demonstrate decreases in number, frequency, and severity of tics as measured through direct observations, clinical interviews, child ratings, and parent-report questionnaires? Results from observations of tic frequency are presented in Figure D2 in Appendix D. Parent participant ratings by session on the PTQ are presented in Table D4 and Figure D3 in Appendix D. Parent and child participants' ratings about tic severity using the clinician-administered YGTSS are displayed in Table D5 in Appendix D. These ratings include pre-intervention and post-intervention (i.e., during the last intervention session and at the one month follow up) data. Child participants also provided SUDS ratings for each current tic during each session as a subjective measure of their experienced level of distress from tics. The results of child participants' SUDS ratings are available in Table D6 and Figure D4 in Appendix D.

Child tic frequency varied across the two child participants. During baseline, Tom exhibited an increase in tics prior to starting intervention (M = 24.08% of intervals, range = 10.56 – 51.11%). Nate's baseline data were more variable, with an overall decreasing trend (M = 8.89% of intervals, range = 2.22 – 19.44%). When the intervention was initiated, Tom's tic frequency demonstrated an immediate decrease from the baseline data, which remained consistent throughout the intervention and at the one month follow up session (M = 0.87% of intervals, range = 0 – 2.78%). Data for Nate did not exhibit the same change when the intervention was introduced; however, this data demonstrated a decreasing trend over time (M = 3.57% of intervals, range = 0 – 10.56%). PND from data for Tom during intervention and follow up was 100%, suggesting a strong treatment effect. PND from data for Nate during intervention and follow up was 57%, suggesting a minimal treatment effect.

Perception of tic severity, as measured by the PTQ, did not closely align with the direct observations of tic frequency in session. Mary's ratings for Tom's tics during baseline indicated a slight increasing trend (M = 8.67, range = 7 – 11), suggesting that tics increased in severity during this time. David's ratings for Nate's tics during baseline demonstrated a variable and increasing trend (M = 11.67, range = 3 – 23). When the intervention was initiated, Mary's ratings decreased compared to baseline; however, over time, her data indicated an increasing trend throughout intervention and follow up (M = 6.86, range = 2 – 17), with the highest score obtained at the one month follow up. David's ratings of Nate's tics, on the other hand, remained similar to the last data point until mid-intervention, after which time, a general decreasing trend was observed (M = 5.14, range = 0 – 9). PND for Mary's ratings of Tom's tics was 57%, suggesting a minimal treatment effect. PND for David's ratings of Nate's tics was 29%, suggesting no treatment effect.

Tic severity, as measured by the YGTSS, was not consistent with the ratings of the PTQ nor the direct observation of tics. Tom had vocal tics only, and his score for both vocal tics (M = 11, range = 11) and total tic severity (M = 21, range = 21) remained stable throughout all measurement points (i.e., pre-intervention, post-intervention, and at the one month follow up). Nate experienced both motor and vocal tics. His motor tic severity score decreased from pre-intervention to post intervention and maintained at the one month follow up (M = 8, range = 6 – 12). Nate's vocal tic severity remained stable throughout the study (M = 1, range = 1). Overall, Nate's total tic severity score decreased from pre-intervention to post-intervention and was maintained at the one month follow up (M = 12.33, range = 7 – 23).

Distress from tics as measured by SUDS ratings varied across participants. Tom identified and rated one tic (i.e., /m/ sound) for baseline and the first four intervention sessions. During the fifth intervention session, he identified another tic, throat clearing, and began rating it. During the one month follow up, he also identified a sniffing tic. Tom initially rated his tics as very distressing (i.e., rating of 10 on a 10-point scale) for the first two baseline appointments. After these ratings, his ratings dropped to 0 for the last baseline and first intervention session, suggesting no distress from tics. At the second intervention session, his rating was again 10, suggesting a high level of distress. For the rest of the intervention and at follow up, he rated his tics as not distressing (i.e., score of 0) or minimally distressing (i.e., score of 1). Nate rated four tics (i.e., arm tensing, finger flexing, facial grimacing, and eye blinking) throughout baseline, intervention, and follow up. During baseline, all tics were rated as not distressing to minimally distressing (i.e., scores of 0 to 2). When the intervention was initiated, he rated arm tensing as moderately distressing (i.e., scores of 4 to 6) for the first two sessions. All other tics were rated as not distressing to minimally distressing (i.e., scores of 0 to 3). During the third intervention

session and subsequent session, all tics, including arm tensing, were rated as not distressing to minimally distressing (i.e., scores of 0 to 2) with a slight decreasing trend.

Child Premonitory Urge Ratings (Research Question 3)

Research question 3 stated: Will children with CTDs receiving a parent-mediated version of HRT demonstrate decreases in premonitory urge sensations as measured through self-report questionnaires? Child participants provided information about their premonitory urges via the PUTS pre-intervention, mid-intervention, and post-intervention (i.e., during the last intervention session and at the one month follow up). The results from this measure are presented in Table D7 in Appendix D.

Tom's scores on the PUTS indicated a decreasing trend over time (M = 16.25, range = 13 – 20). He appeared to have difficulty understanding a few of the questions, so it is difficult to interpret whether or not his premonitory urges actually decreased over time. Nate's scores remained fairly consistent throughout intervention (M = 29, range = 27 – 31).

Child Treatment Acceptability (Research Question 4)

Research question 4 stated: Will parents and children participating in a parent-mediated version of HRT find the intervention acceptable as measured through self-report questionnaires? Similar to parent participants, treatment acceptability for child participants was measured via the TEI-SF and TAQ pre-intervention, mid-intervention, and twice post-intervention (i.e., during the last intervention session and at a one month follow up session). Results from this measure are presented in Table D1 in Appendix D.

Tom's ratings for the TEI-SF are difficult to interpret. His answers appeared to be patterned (e.g., answering the questions in a countdown such that the first question was 7, second question was 6, third question was 5, etc.). Patterned responding did not change following

prompting from the research team members. Similarly, his TAQ scores are difficult to interpret as he may not have understood the word "ethical" and answered in patterns as described above. Nate's ratings on the TEI-SF remained consistently high throughout intervention (M = 40, range = 37 – 42). His ratings on the TAQ were also consistently high throughout intervention (M = 39, range = 35 – 42). Overall, Nate's ratings on both measures suggest that he found the treatment to be acceptable.

Child Motivation for Treatment (Research Question 5)

Research question 5 stated: Will treatment motivation on the part of the children with CTDs impact parents' fidelity of implementing the strategies of a parent-mediated version of HRT as measured by self-report questionnaires on the part of the children and clinician observation and rating of parents? Parent fidelity was measured by direct observation and a step-by-step checklist. Results from these observations are presented in Figure D1 in Appendix D. Child motivation to participate in treatment was measured via the MYTS. Results from these measures are presented in Table D3 in Appendix D.

Tom's scores for the Problem Recognition scale on the MYTS had a decreasing trend over time (M = 2.8, range = 2 – 5), moving from "high" to "low" over time. Nate's scores on the same scale consistently fell in the "low" range (M = 1.5, range = 1.3 – 1.8). On the Treatment Readiness scale, Tom's scores were initially in the medium range at pre-intervention and mid-intervention, but decreased to the "low" range at post-intervention and at the one month follow up (M = 2.7, range = 2.3 – 3.3). Nate's scores on the same scale consistently fell in the "high" range (M = 4.6, range = 4 – 5). Overall scores on this measure for Tom were initially in the "high" range during pre-intervention; scores at all other time periods were in the "low" range (M = 2.8, range = 2.1 – 4.1). Overall scores for Nate were initially in the "low" range during pre-

intervention; scores at all other time periods were in the "medium" range (M = 3, range = 2.6 – 3.3).

As mentioned previously, both parent participants demonstrated an initial increasing trend in parent fidelity. Mary's data consistently demonstrated this increasing trend over time, whereas David's data showed a slight decreasing trend from mid-intervention to the one month follow up session. Given the variability of the child participant ratings from the MYTS, it does not appear that child motivation directly impacted parental fidelity of implementation.

Interobserver Agreement

Inter-observer agreement was collected for 40% of baseline and intervention sessions by either trained observers or the principal investigator, depending on the primary therapist of the observed session. Point-by-point agreement was used for parent fidelity and tic frequency during baseline and intervention sessions. Interobserver agreement ranged from 86% to 100% for fidelity of implementation and from 97% to 100% for tic severity.

CHAPTER FIVE

DISCUSSION

The primary purpose of this study was to determine if parents trained in habit reversal techniques could implement the intervention with integrity while receiving coaching from a therapist trained in HRT and subsequently independently. A second purpose was to investigate whether a parent-mediated version of habit reversal training would result in decreases in tic severity and premonitory urge ratings for the child participants. Additionally, parent and child motivation were assessed to determine whether motivation may have impacted treatment results. Finally, treatment acceptability was measured for both parent and child participants. A discussion of results obtained across these factors, limitations of the current study, and implications for future research follows.

General Discussion

HRT is an effective therapeutic intervention for the treatment of CTDs (Cook & Blacher, 2007); however, it has not been widely disseminated to clinicians (Himle et al., 2010). A variety of approaches have been in development to address concerns with access, such as manualizing CBIT, creation of an HRT-focused website, training programs for practitioners, DVDs for practitioners, and use of telehealth (Himle et al., 2010). To date, a parent-mediated version of HRT has not been published. Similar to other parent-mediated interventions, delivery of HRT through a parent-mediated model may assist with dissemination, increase the intervention hours received (Ingersoll & Dvortcsak, 2010), and increase parental feelings of self-efficacy and optimism about their children's future (Koegel, Schreibman, Britten, Burke, & O'Neill, 1982). The following is a discussion of the results of the current study by research question.

Research Question 1: Will Parents of Children with CTDs Participating in a Parent-Mediated Version of HRT Learn to Implement the Strategies with Adequate Fidelity?

The current study provides preliminary evidence that HRT can be successfully implemented via a parent-mediated model. With coaching, both parent participants were able to fully implement HRT steps, increasing from a baseline where no steps were completed. Acquisition of skills was immediately observed upon the initiation of the intervention and both parent participants continued to increase their skills over time until at least mid-intervention. When coaching was faded, both parent participants demonstrated a decrease in steps completed independently. This decrease may indicate a need for continued coaching beyond the number of sessions provided in this study. It is also possible that this decrease in parental fidelity may be due to leaving out steps that may be unnecessary. In the current study, both parent participants were using the habit reversal strategies on the same tic that had been identified in a previous session. The steps that parents were not completing independently included tasks such as providing a rationale for awareness training, providing a rationale for competing response training, and providing the guidelines for choosing a competing response, all of which would have been explained when the child first started awareness training and competing response training for the individual tic. These tasks may not be necessary to repeat if the child seems to understand this information already. It may be that the list of items included on the parent fidelity checklist may need to be further evaluated for their relative importance during intervention. During the one month follow up, Mary demonstrated a higher percentage of steps completed independently (i.e., 90.48%) than did David (i.e., 76.19%). It is possible that some parents may require more coaching than others; however, it is also important to note that Tom was reported to be experiencing more frequent and more intense tics during the one month

follow up than was Nate. It is possible that Mary would have had to practice the skills of HRT more often post-intervention than did David, resulting in better long-term maintenance.

Research Question 2: Will Children with CTDs Receiving a Parent-Mediated Version of HRT Demonstrate Decreases in Number, Frequency, and Severity of Tics as Measured through Direct Observations, Parent Questionnaires, and Self-Report Questionnaires?

Unlike most other studies investigating HRT, the primary dependent variable of the current study was not tic severity; however, this variable was evaluated throughout. Based on direct observation, there were differences between the two child participants with regard to tic frequency. Tom's tics were higher in baseline than during the intervention and follow up phases. Tics appeared to decrease after the initiation of the intervention and remain consistently low throughout intervention and follow up. Nate's tics, on the other hand, were variable during baseline and then stable after implementation of the intervention, with a decreasing trend over time. It is possible that Nate's tics were related to more of a gradual waning period. It is also possible that there was variability in practice of habit reversal strategies outside of the clinic setting. While review of homework was completed during each intervention session and parents and children reported that homework had been completed, there was no other measure that the strategies were being used consistently. It is also possible that the intervention helped Nate to build awareness to more consistently, but gradually implement the habit reversal strategies.

Variability was also observed for parent ratings on the PTQ and parent and child ratings on the YGTSS. On the PTQ, Mary's data suggested an increasing trend over time, suggesting that tics increased in severity over time. This trend is in direct contrast to what was observed in session. It could be that by participating in treatment, Mary was made more aware of tics and therefore was more observant of Tom's tics. It is also possible that Tom was exhibiting reactivity

within the clinic setting (Kazdin, 2011). David's ratings of Nate's tics demonstrated a similar trend (i.e., variable during baseline, stable after intervention was initiated with a decreasing trend over time) as the observations. On the YGTSS, Tom's ratings indicated stability and low rates over time, suggesting that tic severity remained stable. These ratings not directly align with the PTQ. It was also not directly aligned with baseline data during the observations; however, it was aligned with intervention data. Nate's ratings indicated a decrease over time, which was similar to the PTQ and direct observations. The PTQ and YGTSS scores should be highly correlated with one another (Chang et al., 2009), and it is difficult to explain the large discrepancies for the ratings for Tom. The discrepancies could be due to differences in rater perceptions, as parent participants completed the PTQ and a research team member completed the YGTSS. It could also be that Mary's anecdotal report of being bothered by the volume of Tom's tics. In addition to the volume of tics, Tom exhibited high motor activity in general, which may be associated with his diagnosis of AD/HD. Mary may have also been more sensitive to Tom's behaviors at home due to their disruptive nature, which might have led to overestimation on the PTQ.

Distress from tics as measured by SUDS was also variable for each participant. During baseline, Tom initially indicated no distress followed by high distress. His ratings, with the exception of one session early in the intervention in which he rated high distress, consistently indicated no to low distress. Nate, on the other hand, indicated zero to low distress from tics during baseline and early intervention Toward mid-intervention, he rated moderate distress, which then decreased over time. Neither of the child participants' ratings appeared to directly align with observation of tics or with PTQ and YGTSS data; however, SUDS is solely a subjective measure of distress that does not specifically take into account factors like number of

tics experienced or frequency of tics. Therefore, it is possible that it would not be consistent with the other measures.

Based on the variability in tic frequency and tic severity between the two child participants, it is difficult to draw conclusions regarding the intervention's effectiveness for decreasing tic severity. In the randomized clinical trial of CBIT conducted by Piacentini and colleagues (2010), 52.5% of participants demonstrated a positive response to treatment. With this statistic in mind, it is possible that the rates in the current study are similar; however, more participants would provide more information on this factor. In addition, perception of tics as a problem was low for both child participants through much of this study. It is possible that the perception of tics as a problem was related to lower motivation, which could be a mediating factor for tic severity.

Research Question 3: Will Children with CTDs Receiving a Parent-Mediated Version of HRT Demonstrate Decreases in Premonitory Urge Sensations as Measured through Self-Report Questionnaires?

As mentioned in the previous chapter, Tom's ratings on the PUTS were difficult to interpret as he reported having difficulty understanding a few of the questions. Therefore, while his ratings suggest a decreasing trend over time, it may not be representative of his actual experience of the premonitory urge. It is also possible that he did not have a "true" sense of the premonitory urge. Nate's ratings remained stable over times of administration, which may be more accurate given his older age (i.e., Nate was 10 years old versus Tom who was 8 years old). In both cases, the PUTS may not be the best measure, as it is recommended for children above the age of 10 years(Woods et al., 2005). Without another tool available for children 10 years of age and under, it is the only current option in an attempt to measure premonitory urges.

Research Question 4: Will Parents and Children Participating in a Parent-Mediated Version of HRT Find the Intervention Acceptable as Measured through Self-Report Questionnaires?

Both parent participants rated the parent-mediated HRT intervention as highly acceptable across measures and across times of administration. Data from the child participants were more difficult to interpret. Tom's responding on the self-report measure appeared invalid as he demonstrated patterned responding, which may be related to impulsivity and/or younger age. While his scores suggested adequate acceptability, they cannot be interpreted. Nate consistently rated the parent-mediated HRT intervention as highly acceptable across measures and times of administration. He did not appear to exhibit impulsive responding and asked questions regarding the wording of questions to better understand the question. Based on these observations, it is likely that Nate's scores were more likely to be accurate. In general, child and parent ratings of treatment acceptability in the current study are similar to those obtained in other HRT studies, which found HRT to be a highly acceptable intervention (Himle et al., 2010; Himle et al., 2012; Woods et all., 1996; Woods & Twohig, 2002).

Research Question 5: Will Treatment Motivation on the Part of the Children with CTDs Impact Parents' Fidelity of Implementing the Strategies of a Parent-Mediated Version of HRT as Measured by Self-Report Questionnaires on the Part of the Children and Clinician Observation and Rating of Parents?

Child motivation was assessed as a potential mediating factor for parents' skill acquisition and fidelity of implementation. Child motivation to engage in treatment may impact change in behavior, which may impact parental attributions made about their child's behavior and their child's treatment (Morrissey-Kane & Prinz, 1999). Child ratings on the MYTS in the

current study resulted in different patterns across scales. Tom's ratings decreased over time on the Problem Recognition scales, suggesting that his tics became less bothersome over time. Nate's ratings on this same scale were consistently low, suggesting mild impairment overall from tics. On the Treatment Readiness scale, Tom's scores initially indicated a "medium" level of motivation, which decreased to low motivation over time. Tom's motivation may have been impacted by his younger age and difficulty paying attention and sitting still during sessions. Conversely, Nate's scores on the same scale consistently indicated high motivation to engage in treatment. Despite these differences in patterns, they did not appear to coincide with parental fidelity (i.e., parent fidelity consistently increased for Mary working with Tom and declined slightly for David working with Nate). Therefore, it does not appear that child motivation as measured through the MYTS impacted parental participation and fidelity.

Research Question 6: Will Parent Motivation to Participate in Treatment Be Related to Increased Skill Acquisition and Adequate Fidelity as Measured by Self-Report and Direct Observation?

Both parent participants indicated stable and high levels of motivation to participate in treatment on the PMI across all times of administration. On the MYTS, differences between the two scales were noted. Specifically, both Mary and David indicated medium to high levels of motivation on the Treatment Readiness scale; however, on the Problem recognition scale, scores were low to medium. Results from this measure indicate that there was parent motivation to engage in treatment; however, the impairment caused by tics was rated as low to moderate, suggesting that the parent participants did not identify tics as severely impairing. Similar to the PMI, scores on the MYTS were consistent across times of administration. Given consistency of ratings on both measures of parental motivation and the increasing trend for both parent

participants on the fidelity measure, it is possible that parent motivation had an impact on fidelity of implementation (i.e., high motivation may have contributed to skill acquisition and fidelity of implementation).

Overall Findings

This study provides initial evidence that parents can provide HRT to their children while receiving coaching from a behavioral therapist. These findings answer the primary question: Will parents of children with CTDs participating in a parent-mediated version of HRT learn to implement the strategies with adequate fidelity? Parent participants and one of the child participants found the treatment to be acceptable, while the second child's responses were invalid and uninterpretable. In addition, motivation to participate remained constant over time for the parent participants and one child participant. Impact on tic severity is difficult to determine; however, future studies may help to clarify the relationship between a parent-mediated version of HRT and its impact on tic severity with additional child participants.

Limitations of the Current Study

The most salient limitation to this study was the small number of participants. While results were replicated for both parent participants for fidelity of implementation, it was difficult to determine the impact of the intervention on tic severity for the child participants. In addition, further replication of results would have strengthened the conclusion regarding the utility of a parent-mediated version of HRT. Perhaps this study provides the preliminary support for a large-scale study that would provide definitive evidence that parents can implement this treatment and show positive effects on child tic frequency and severity.

The research design of the current study was a nonconcurrent multiple baseline across participants design. This model was used due to the study being conducted in an applied setting,

where it would be potentially unethical to withhold treatment. In effect, the decision to use this design accelerated initiation of the study for participants and allowed for participants to be added while other participants were already enrolled and participating in the study. However, this type of design potentially introduces history as a threat to internal validity (Christ, 2007; Kazdin, 2011). To more completely control for history, a multiple baseline design or a multiple probe design could have been utilized. However, in an analysis of concurrent versus nonconcurrent designs, Christ (2007) discusses how concurrent and nonconcurrent multiple baseline designs are sufficiently robust to determine experimental control, provided that a priori hypotheses and that duration of baseline phases are specified ahead of time in a nonconcurrent study. The current study does not completely meet criteria for a nonconcurrent study due to some overlapping data points during baseline and intervention; however, both criteria discussed by Christ were met in that there were assumptions made about parent fidelity, the primary variable (i.e., parents would demonstrate 0% of steps completed independently during baseline), and that baseline length was specified prior to the start of the study (i.e., three sessions for all participants). Given these factors, it is unlikely that history was a threat to internal validity.

Another limitation to the current study was attrition of one of the participants. This parent-child dyad chose to drop out of the study after completing the first intervention session. While personal reasons were cited, it is important to note that there may have been significant differences between this parent-child dyad and the two other parent-child dyads who remained in and completed the study and follow up.

Neither of the parent participants in the current study reached 100% on the fidelity checklist. This lack of mastery may be due to multiple reasons. First, the checklist may need additional evaluation as a tool to better determine if all of the steps included are necessary. Next,

parents may need additional time in intervention with coaching to reach the 100% mastery level. Ratings of fidelity of therapists delivering HRT or CBIT are currently unavailable for comparison, so it is currently not possible to compare parental fidelity in the current study to "typical" fidelity.

Similar to parent fidelity, fidelity of coaching was not evaluated in the current study. The aim of the study was not to collect information on therapists' fidelity; however, given that coaching was implemented by different researchers trained in HRT, the study is limited by this lack of fidelity data. Future research could be strengthened by taking data on therapists implementing coaching while delivering a parent-mediated version of HRT.

Implications for Future Research

This study is the first known attempt to employ a parent-mediated version of habit reversal. Results suggest potential utility of a parent coaching model with parents as therapists as a modality of treatment. Given difficulties with access to therapists trained in HRT (McGuire et al., 2015), a parent-mediated version of HRT may be useful with dissemination of treatment strategies. As discussed previously, parent-mediated interventions have resulted in positive outcomes for both parents (Baker, Landen, & Kashima, 1991; Brookman-Frazee, 2004; Eisenstadt, Eyberg, McNeil, Newcomb, & Funderburk, 1993; Koegel, Bimbela, & Schreibman, 1996; Ingersoll & Dvortcsak, 2010; McConachie & Diggle, 2007; McNeil & Hembree-Kigin, 2010) and children (Eisenstadt, Eyberg, McNeil, Newcomb, & Funderburk, 1993; McNeil & Hembree-Kigin, 2010; Strauss, Mancini, the SPC Group, & Fava, 2013; Strauss et al., 2012). Replication of these results with a larger sample will be important to determine more long-term utility for this type of treatment modality. In addition, a multiple baseline or a multiple probe

design could be used to address potential threats to internal validity. It may also be of benefit to replicate this study with older adolescents.

In addition to replication, future studies could investigate use of a parent-mediated version of HRT with more severe tics and a more varied topography of tics. While each parent-child dyad in the current study identified multiple tics, treatment often focused on the same tic from week to week. Similarly, tics were rated as low to moderately impairing. It would be helpful to replicate this study with children with more topographies of tics and more severity of tics.

With replication, it may be beneficial to investigate use of a parent-mediated model of HRT using a telehealth model. HRT as an intervention has been investigated using videoconferencing with positive results (Himle et al., 2010; Himle et al., 2012). By teaching parents to provide HRT, it could be possible that HRT is delivered more frequently and with higher fidelity in the home setting than if HRT is provided to children with the therapist as the primary change agent.

In addition, previous studies have not reported fidelity of implementation statistics for therapists delivering HRT. In a parent-mediated model, it will likely be important to include these statistics as the skills are likely to be new. The measure designed to assess fidelity of implementation was designed for the purpose of the current study. Future research could examine the psychometric properties of this measure.

One factor that was not measured during the current study was use of habit reversal strategies by the children. It may be beneficial to measure whether a child signals during awareness training to reach the suggested 80 percent awareness as suggested by Woods and colleagues (2008b) and whether s/he effectively uses the competing response. It may be difficult

to detect all instances of a competing response, such as in the case of diaphragmatic breathing. However, this information may be of value in future research to draw conclusions about use of HRT strategies and impact on tic severity.

Future research may also investigate child motivation as a primary variable. Due to patterned responding by one of the participants, it was difficult to determine whether or not child motivation to participate had an impact on treatment. It might be useful to implement parent-mediated HRT with children with different levels of initial treatment motivation (e.g., low, moderate, or high) to further investigate this variable.

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APPENDIX A

Clinical Interview

Age of child:

Race/ethnicity of child:

Race/ethnicity of parent(s):

Age of parent(s):

Parental occupation(s):

Parent(s) highest level of education obtained:

Parental marital status:

If divorced, custody arrangements:

Who lives at home with child?

Medications taken by child:

Psychiatric diagnoses of child:

Tic symptom history:

Family history of tics:

Current tics (topography, frequency, settings):

Favorite activities of child:

APPENDIX B

Fidelity Checklist for Parent-Mediated HRT

ID #: _____ Collected by: _____

Session #: _____ PR:____/REL:_____

	Observed		Coached?
Skill	**Yes**	**No**	**Coached (C) or Independent (I)**
Reviews significant events of past week, both positive* and negative, and their impact on tics with the child. *Must identify at least one positive event			
Reviews homework, problem solving any difficulties with noncompliance.			
Assigns points for behavioral reward system appropriately (i.e., for coming to session and whether homework completed).			
Creates or reviews inconvenience review (updating as necessary).			
Obtain SUDS rating on scale of 0 to 10 from child. *Must do for each tic.			
Create or review/update tic hierarchy with child and discusses which tic to focus on for treatment based on hierarchy.			
Identifies or reviews antecedents/consequences for tics (i.e., function-based assessment).			
Discusses or reviews potential strategies for the antecedents/consequences identified (i.e, function-based intervention). *Must discuss/review at least one strategy.			
Awareness training – introduction Parent introduces awareness training and describes purpose to child.			
Awareness training – response description Parent has child demonstrate tic and describe the tic in detail (i.e.,			

what it looks/sounds like, which muscles are moving) or reviews response description from previous session if already created.			
Awareness training – response description Parent points out to child if s/he fails to describe a key feature of the tic or agrees that child has adequately explained all aspects of the target tic.			
Awareness training – response description Parent works with child to establish any warning signs (premonitory urges/sensations) or reviews the premonitory urges identified in the previous session.			
Awareness training – acknowledge self-tic/premonitory urge Provides appropriate instructions for child identifying own tics (e.g., "If you feel the urge to tic or notice a tic occurs, put your pointer finger up"), including modeling the expected signal			
Awareness training – acknowledge self-tic/premonitory urge Provides appropriate praise for signaling or immediate constructive feedback (i.e., gentle reminder) for forgetting to signal *Must occur 80% of the time for "Yes" *If no tics occur, parent should provide reminder prompts to signal approximately every 1-2 minutes (at least 1 reminder has to occur for "Yes")			
Competing response – explanation Parents explain rules of a competing response to child. 1. Physically incompatible 2. Maintained for 30 seconds or until urge goes away 3. Socially inconspicuous 4. Less effortful than tic 5. Easily integrated into normal activity *Must include all rules for "Yes"			
Competing response – choice of behavior Parent and child work together to develop a competing response or parent reviews the competing response from the previous session.			
Competing response – model Provides appropriate instructions for using competing response (e.g., "If you feel the urge to tic or notice a tic occurs, use your exercise"), including modeling the competing response			
Competing response – teaching to child Parent has child demonstrate the competing response			

Competing response – reaction from parent Parent praises child for appropriate use of correct response or provides immediate corrective feedback (i.e., gentle reminder) if child misses tic *Must occur 80% of the time for "Yes" *If no tics occur, parent should provide reminder prompts to signal approximately every 1-2 minutes (at least 1 reminder has to occur for "Yes").			
Assigns appropriate homework based on session (e.g., self-monitoring, practice awareness training, practice competing response).			
Assigns points for the behavioral reward system appropriately at the end of the session for compliance with strategies during session.			
Total:			

Calculate fidelity: _____ "Yes" without "C"/ 21 total steps X 100 = _____%

APPENDIX C

Parent-Mediated HRT Session Timeline

Baseline session 1

- Explain study to parent and child
- Obtain informed consent/assent
- Therapist conducts clinical interview and YGTSS
- Parent fills out PTQ, MYTS, PMI, TEI-SF, TAQ
- Child fills out MYTS, TEI-SF, TAQ, and PUTS
- Therapist collects SUDS ratings from child for endorsed tics during YGTSS
- Baseline data collection – free play scenario with parent and child (30 minutes)
- Parents able to ask questions

Baseline session 2

- Parent fills out PTQ
- Therapist collects SUDS ratings from child for endorsed tics during YGTSS and PTQ
- Parent, therapist, and child discuss goals for intervention
- Therapist provides psychoeducation about tic disorders with handout
- Baseline data collection – free play scenario with parent and child (30 minutes)
- Parents able to ask questions

Baseline session 3

- Parent fills out PTQ
- Therapist collects SUDS ratings from child for endorsed tics during YGTSS and PTQ
- Therapist provides psychoeducation about tic disorders with handout
- Baseline data collection – free play scenario with parent and child (30 minutes)
- Therapist provides handout about function-based assessment and intervention
- Parents able to ask questions

Intervention session 1

- Parent fills out PTQ
- Therapist provides didactic training in function-based assessment, creating a tic hierarchy, and collection of SUDS rating
- Observation of intervention between parent and child with coaching
    - Fidelity rated through direct observation
    - Child tic frequency collected through direct observation (30 minutes)
- Feedback given to parents/questions answered by therapist
- Therapist gives handout about awareness training

Intervention session 2

- Parent fills out PTQ

- Therapist provides didactic training in awareness training (i.e., response description, response detection, early warning signal detection)
- Observation of intervention between parent and child with coaching
    - Fidelity rated through direct observation
    - Child tic frequency collected through direct observation (30 minutes)
- Feedback given to parents/questions answered by therapist
- Therapist gives handout about competing response training

Intervention session 3

- Parent fills out PTQ
- Therapist provides didactic training in competing response training (i.e., rules for selection, developing incompatible behavior w/ child)
- Observation of intervention between parent and child with coaching
    - Fidelity rated through direct observation
    - Child tic frequency collected through direct observation (30 minutes)
- Feedback given to parents/questions answered by therapist
- Therapist provides handout about combining all strategies
- Parent fills out PMI and MYTS
- Child fills out MYTS and PUTS

Intervention session 4

- Parent fills out PTQ
- Therapist provides didactic training about using all strategies together
- Observation of intervention between parent and child with coaching
    - Fidelity rated through direct observation
    - Child tic frequency collected through direct observation (30 minutes)
- Feedback given to parents/questions answered by therapist

Intervention 5

- Parent fills out PTQ
- Observation of intervention between parent and child with minimal coaching
    - Fidelity rated through direct observation
    - Child tic frequency collected through direct observation (30 minutes)
- Feedback given to parents/questions answered by therapist

Intervention 6

- Parent fills out PTQ
- Observation of intervention between parent and child without coaching
    - Fidelity rated through direct observation
    - Child tic frequency collected through direct observation (30 minutes)
- Feedback given to parents/questions answered by therapist
- Therapist discusses maintenance without coaching and relapse prevention

- Parent fills out TEI-SF, TAQ, PMI, MYTS
- Child fills out MYTS, TEI-SF, TAQ, and PUTS

One Month Follow-up

- Parent fills out PTQ
- Observation of intervention between parent and child without coaching
    - Fidelity rated through direct observation
    - Child tic frequency collected through direct observation (30 minutes)
- Feedback given to parents/questions answered by therapist
- Therapist discusses maintenance without coaching and relapse prevention
- Parent fills out TEI-SF, TAQ, PMI, MYTS
- Child fills out MYTS, TEI-SF, TAQ, and PUTS

APPENDIX D

Tables and Figures

Table D1

Results from the Treatment Evaluation Inventory, Short Form (TEI-SF) and the Treatment Acceptability Questionnaire (TAQ)

Scales - Tom	Pre-Intervention Session 1		Mid-Intervention Session 6		Post-Intervention Session 9		Post-Intervention One-Month Follow-Up	
	Parent	Child	Parent	Child	Parent	Child	Parent	Child
TEI-SF	32	26*	33	24*	38	19*	35	22*
TAQ	39	21*	42	37*	40	26*	41	27*

Scales - Nate	Pre-Intervention Session 1		Mid-Intervention Session 6		Post-Intervention Session 9		Post-Intervention One-Month Follow-Up	
	Parent	Child	Parent	Child	Parent	Child	Parent	Child
TEI-SF	38	41	38	42	38	40	38	37
TAQ	39	35	42	41	40	39	39	41

Combined Scales – Parent Scores Only*	Pre-Intervention Session 1	Mid-Intervention Session 6	Post-Intervention Session 9	Post-Intervention One-Month Follow-Up
	Means	Means	Means	Means
TEI-SF	35	35.5	38	36.5
TAQ	39	42	40	40

*Scores were not calculated for mean child participant on these measures due to concerns regarding Tom's method for answering questions and potential misunderstanding of questions.

Note: The TEI-SF has a minimum score of 9 and a maximum score of 45. Scores around 27 indicate "moderate" treatment acceptability. The TAQ has a minimum score of 6 and a maximum score of 42. Higher scores indicate higher treatment acceptability.

Table D2

Results from the Parent Motivation Inventory (PMI)

Scales - Mary	Pre-Intervention Session 1	Mid-Intervention Session 6	Post-Intervention Session 9	Post-Intervention One-Month Follow-Up
	Score	Score	Score	Score
Desire for Change	22	23	23	22
Readiness to Change	60	58	59	55
Perceived Ability to Change	15	15	15	14
Total	97	96	97	91

Scales - David	Pre-Intervention Session 1	Mid-Intervention Session 6	Post-Intervention Session 9	Post-Intervention One-Month Follow-Up
	Score	Score	Score	Score
Desire for Change	26	25	24	23
Readiness to Change	70	70	69	70
Perceived Ability to Change	16	18	18	19
Total	112	113	111	112

Combined Scales	Pre-Intervention Session 1	Mid-Intervention Session 6	Post-Intervention Session 9	Post-Intervention One-Month Follow-Up
	Mean	Mean	Mean	Mean
Desire for Change	24	24	23.5	22.5
Readiness to Change	65	64	64	62.5
Perceived Ability to Change	15.5	16.5	16.5	16.5
Total	104.5	104.5	104	101.5

Note: The scales for the PMI have the following minimum values: Desire for Change minimum value = 7; Readiness to Change minimum value = 14; Perceived Ability to Change minimum value = 4; and Total minimum value = 25. The scales for the PMI have the following maximum values: Desire for Change maximum value = 35; Readiness to Change maximum value = 70; Perceived Ability to Change maximum value = 20; and Total maximum value = 125. Higher scores indicated higher parental motivation.

Table D3

Results from the Motivation for Youth Treatment Scale (MYTS)

Scales – Mary/Tom	Pre-Intervention Session 1		Mid-Intervention Session 6		Post-Intervention Session 9		Post-Intervention One-Month Follow-Up	
	Parent	Child	Parent	Child	Parent	Child	Parent	Child
Problem Recognition	3	5	3	2	2.5	2	2.8	2.3
Treatment Readiness	4	3.3	3.8	3	4.5	2.3	3.8	2.3
Total	3.5	4.1	3.4	2.5	3.5	2.1	3.3	2.3

Scales – David/Nate	Pre-Intervention Session 1		Mid-Intervention Session 6		Post-Intervention Session 9		Post-Intervention One-Month Follow-Up	
	Parent	Child	Parent	Child	Parent	Child	Parent	Child
Problem Recognition	2.5	1.3	2	1.5	2	1.3	2.3	1.8
Treatment Readiness	5	4	5	5	5	5	5	4.3
Total	3.8	2.6	3.5	3.3	3.5	3.1	3.6	3

Combined Scales	Pre-Intervention Session 1 Means		Mid-Intervention Session 6 Means		Post-Intervention Session 9 Means		Post-Intervention One-Month Follow-Up Means	
	Parent	Child	Parent	Child	Parent	Child	Parent	Child
Problem Recognition	2.8	3.1	2.5	1.8	2.3	1.6	2.5	2
Treatment Readiness	4.5	3.6	4.4	4	4.8	3.6	4.4	3.3
Total	3.4	3.4	3.4	2.9	3.5	2.6	3.4	1.8

Note: The scales for the MYTS have a minimum score of 1 and a maximum score of 5. Scores below 2.9 are considered "Low," scores of 2.9 to 3.8 are considered "Medium," and scores of 3.9 or above are considered "High." All scores have been rounded to the nearest tenth when necessary to appropriately match these scales.

Table D4

Results from the Parent Tic Questionnaire (PTQ)

Session # / Child	1	2	3	4	5	6	7	8	9	10	Overall	
Tom											M	SD
Motor Tic Severity	0	0	0	0	0	0	0	0	0	0	0	0
Vocal Tic Severity	7	8	11	5	7	2	2	6	9	17	7.4	4.4
Total PTQ Score	7	8	11	5	7	2	2	6	9	17	7.4	0.4
Nate											M	SD
Motor Tic Severity	3	21	9	8	9	9	0	4	0	2	6.5	6.28
Vocal Tic Severity	0	2	0	0	0	0	0	0	0	2	0.4	0.84
Total PTQ Score	3	23	9	8	9	9	0	4	0	4	6.9	6.67
Combined	M	M	M	M	M	M	M	M	M	M	M	SD
Total PTQ Score	5	15.5	10	6.5	8	5.5	1	5	4.5	10.5	7.15	5.51

Note: All scales of the PTQ have a minimum value of 0. The scales for Motor and Vocal Tic Severity each have a maximum value of 112. The Total Tic Severity scale has a maximum value of 224. Higher scores on the Motor Tic Severity, Vocal Tic Severity, and Total scales indicate higher frequency and intensity of tics.

Table D5

Results from the Yale Global Tic Severity Scale (YGTSS)

Child	Pre-Intervention Session 1	Post-Intervention Session 9	Post-Intervention One-Month Follow-Up
Tom			
Motor Tic Severity	0	0	0
Vocal Tic Severity	11	11	11
Total Tic Severity	11	11	11
Impairment	10	10	10
Total YGTSS Score	21	21	21
Nate			
Motor Tic Severity	12	6	6
Vocal Tic Severity	1	1	1
Total Tic Severity	13	7	7
Impairment	10	0	0
Total YGTSS Score	23	7	7
Combined	M	M	M
Total YGTSS Score	22	14	14

Note: All scales of the YGTSS have a minimum value of 0. The scales for Motor and Vocal Tic Severity each have a maximum value of 25. The Total Tic Severity scale has a maximum value of 50. The Total YGTSS scores has a maximum value of 100. Higher scores on the Motor, Vocal, and Total Tic Severity scales indicate higher number, frequency, intensity, complexity, and interference from tics. The Total YGTSS score represents these same factors; however, it also indicates level of impairment from tics.

Table D6

Results from the Subjective Units of Distress Scale (SUDS) Ratings

Session #	1	2	3	4	5	6	7	8	9	10
Child										
Tom	Score	Score	Score	Score	Score	Score	Score	Score	Score	Score
/m/ sound	10	10	0	0	10	0	0	0	0	1
Throat clearing	N/A	N/A	N/A	N/A	N/A	N/A	N/A	8	0	0
Sniffing	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	N/A	10
Nate	Score	Score	Score	Score	Score	Score	Score	Score	Score	Score
Arm tense	0	0	2	4	6	2	0	1	0	0
Finger flex	0	0	0	0	2	1	0	0	0	0
Facial grimace	0	0	0	0	0	1	0	0	0	0
Eye blink	0	1	0	3	2	0	0	0	0	0

Note: The minimum possible score for this measure is 0 (i.e., tics are not a problem to me) and the maximum score possible is 10 (i.e., tics are the worst problem for me). Higher scores indicate higher perceived distress from tics, as reported by the child participants.

Table D7

Results from the Premonitory Urge for Tics Scale (PUTS)

Child Participants	Pre-Intervention Session 1	Mid-Intervention Session 6	Post-Intervention Session 9	Post-Intervention One-Month Follow-Up
	Score	Score	Score	Score
Tom	20	16	16	13
Nate	27	31	30	28
Mean Score	23.5	23.5	23	20.5

Note: The minimum possible score for this measure is 9 and the maximum score possible is 36. Higher scores indicate more awareness of a premonitory urge and potentially more distress and impairment.

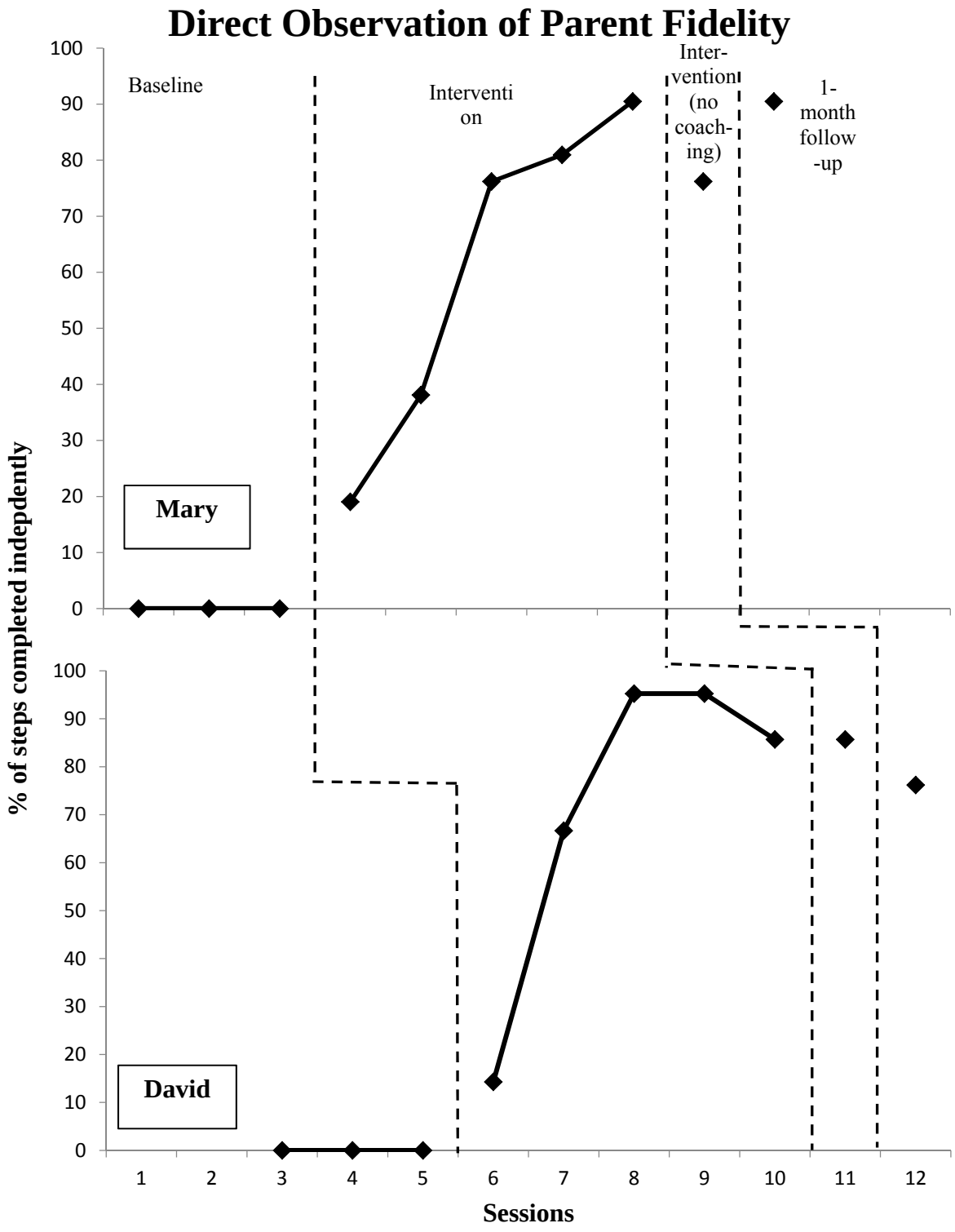

Figure D1. Parent fidelity across sessions for both parent participants.

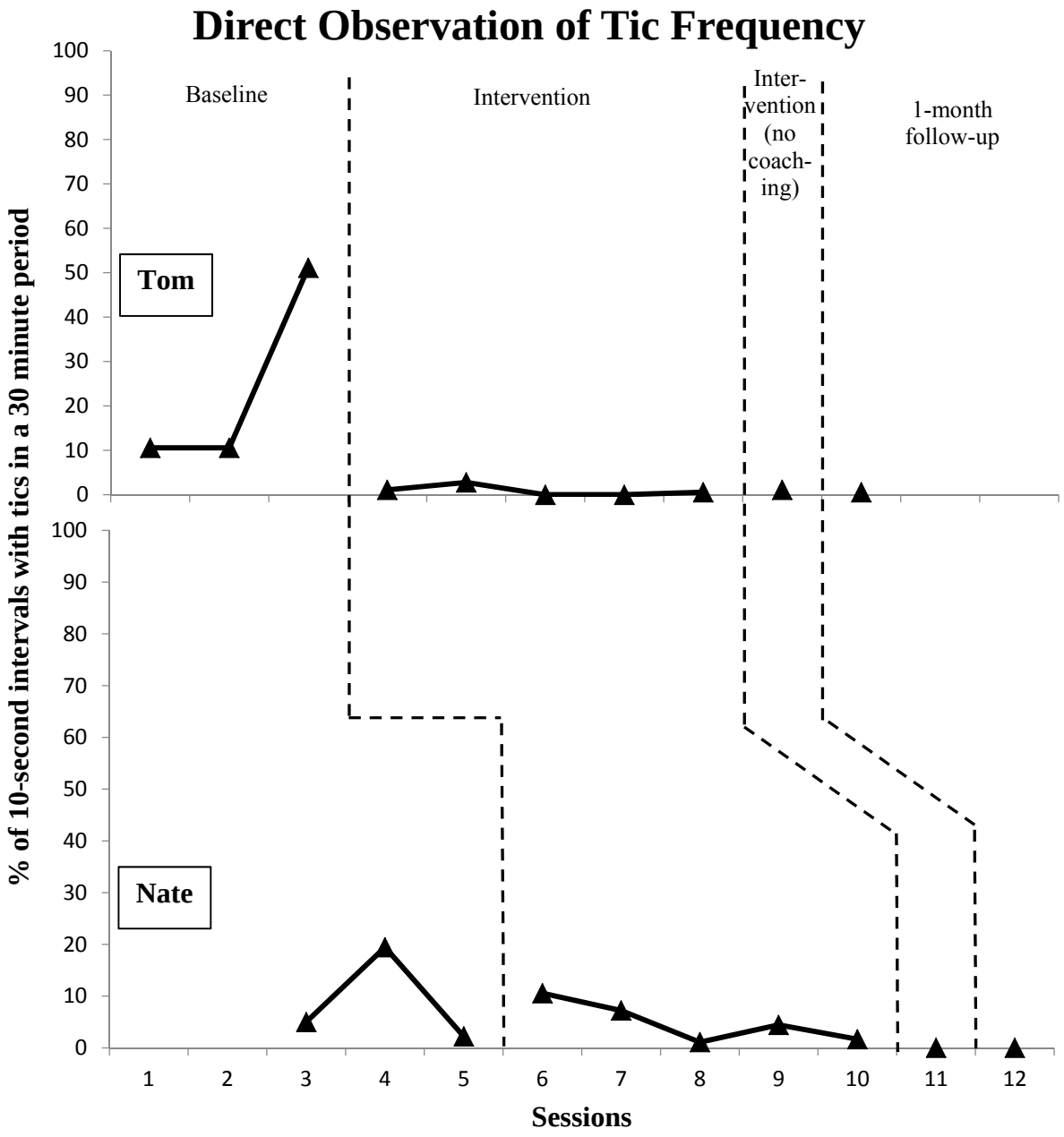

Figure D2. Tic frequency data of both child participants.

Note: Data were collected using 10-second partial interval schedule over a 30 minute period.

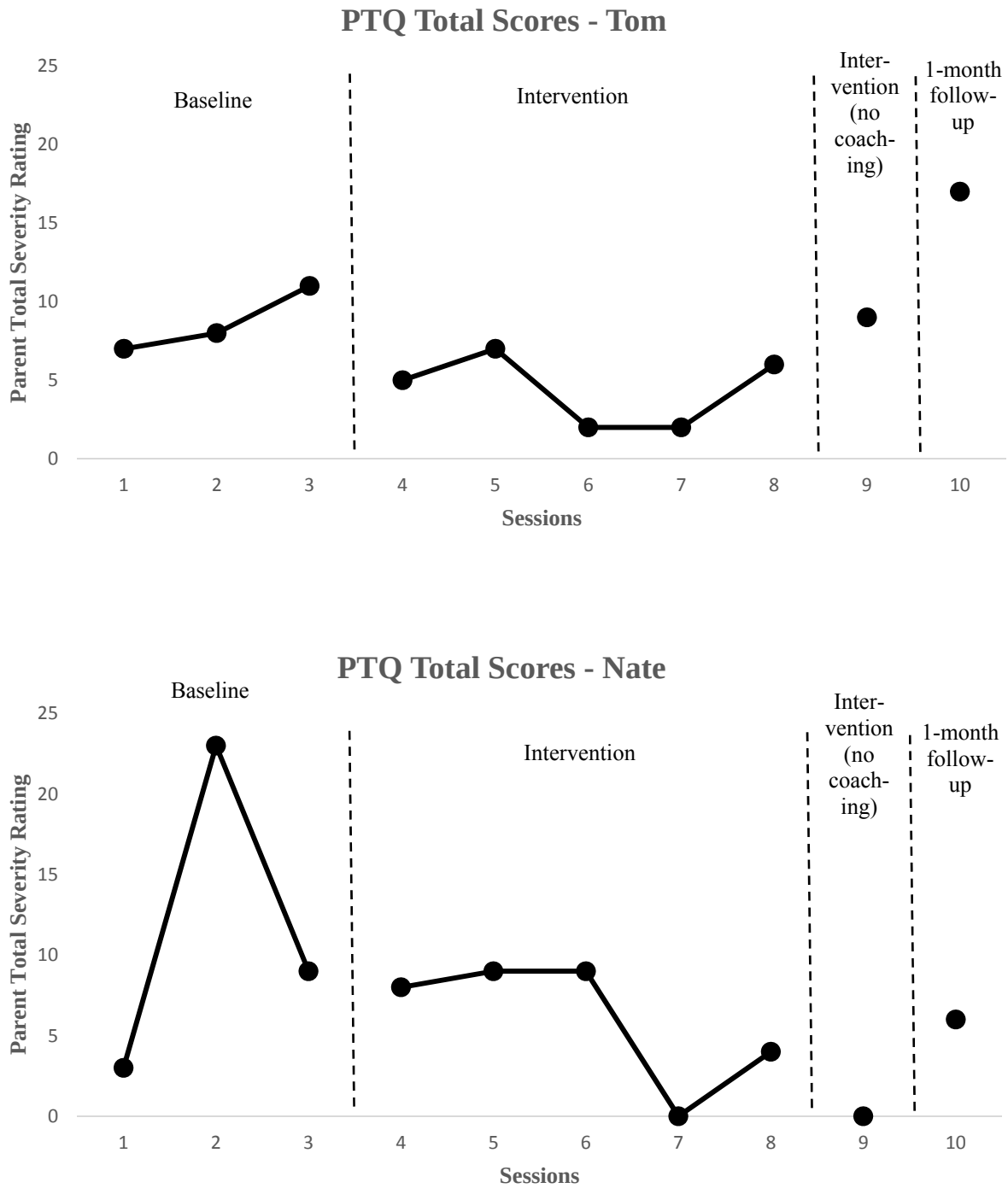

Figure D3. Total Severity scores from the Parent Tic Questionnaire (PTQ) for each child participant.

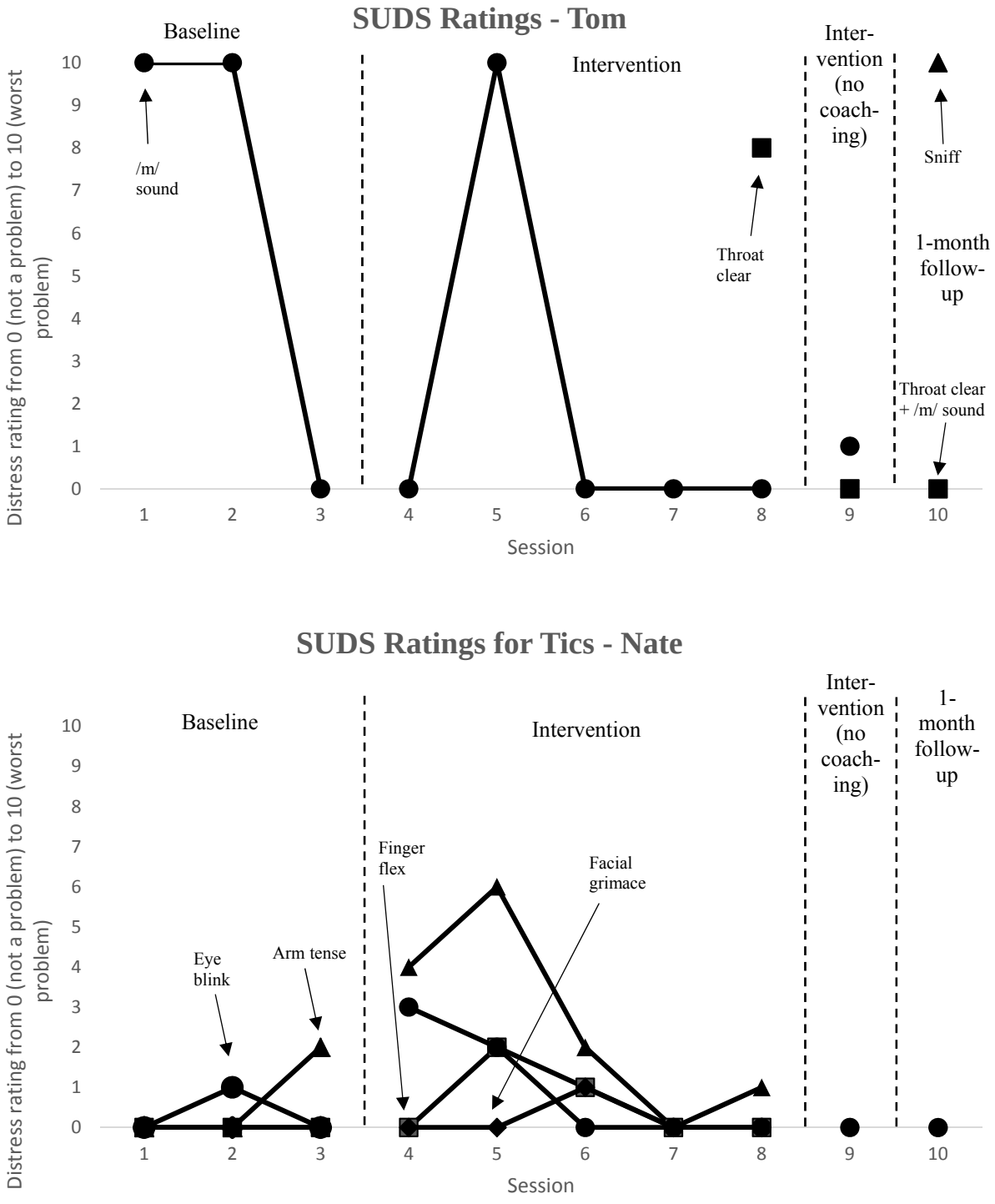

Figure D4. Ratings of subjective units of distress scale (SUDS) for each child participant.