Pragmatic Case Studies in Psychotherapy, http://pcsp.libraries.rutgers.edu Volume 13, Module 1, Article 1, pp. 1-37, 04-22-17 [copyright by author]

The Case of "Hiro": Treating Tourette Syndrome by Comprehensive Behavioral Intervention for Tics (CBIT)

JEREMY D. LICHTMAN a,b,c

^a Graduate School of Applied & Professional Psychology, Rutgers University-New Brunswick

^b Correspondence regarding this article should be sent to Jeremy Lichtman, Tourette Syndrome Clinic, Graduate School of Applied & Professional Psychology, 152 Frelinghuysen Road, Piscataway, NJ 08854 Email: jdlichtman@gmail.com

ABSTRACT

Cognitive-behavioral therapy (CBT) with Comprehensive Behavioral Intervention for Tics (CBIT) is an efficacious treatment for Tourette Syndrome (TS), with a manualized version available for guiding practitioners through treatment. However, CBIT is still a fairly new treatment for TS and rigorous training in its application is sparse. Additionally, potential treatment considerations are not thoroughly described in the manual. This dissertation presents a systematic case study that specifically analyzes the use of the manual with 10-year-old "Hiro," a South Asian Indian boy with TS who also had attention deficit hyperactivity disorder (ADHD), which is a confounding factor in the treatment of TS. To do this meant applying flexibility and an "hypothesizing-then-testing" mentality. The creation of an individualized case formulation and an associated treatment plan for Hiro were very valuable components in his treatment. The case study chronicles Hiro's successful treatment and explores how and when adherence to the manual was effective as well as how and when greater flexibility was required. Furthermore, the case study explores factors in the clinician/client relationship and how that relationship can prove a powerful tool in treatment. Options for how to maintain treatment goals after termination are also examined.

Key words: Tourette Syndrome; Tics Disorders; Comprehensive Behavioral Intervention for Tics (CBIT); Habit Reversal Training (HRT); cognitive-behavioral training (CBT); Attention Deficit Hyperactivity Disorder (ADHD); manualized therapy; individualized case formulation; case studies; clinical case studies

1. CASE CONTEXT AND METHOD

The Rationale for Selecting This Particular Client for Study

The goal of this case study was threefold: To analyze the clinical application of a manualized treatment for Tourette Syndrome (TS); to record and analyze how this application led to a successful treatment of TS—specifically, in what ways adherence to the manual was effective and what contravention from the manual was necessary—and to record the growth of a novice practitioner and analyze what I needed to learn to successfully treat my client.

^c Note: This article is a reformatted and edited version of my dissertation (Lichtman, 2015).

The treatment provided for this client was Comprehensive Behavioral Intervention for Tics (CBIT), which is a manualized, cognitive behavioral therapy (CBT) treatment for children and adults with TS (Woods et al., 2008). This client, who will be called "Hiro," was selected for this dissertation because he represents a fairly typical case of TS and was therefore a good candidate for a typical CBIT intervention. However, his case is interesting because early on in treatment it was discovered that exact replication of the manualized treatment was not working. Therefore, this paper will document my journey wading through a treatment that was at first unsuccessful, and how I discovered the need to adopt a more pragmatic approach when treating Hiro.

When treating Hiro, two significant complicating factors arose: one, his parents lost confidence in the treatment very quickly, when it was not immediately successful; and two, his Attention Deficit Hyper Activity Disorder (ADHD) symptoms confounded the treatment process. A third factor that arose later in treatment was that his family was moving out of the United States and a plan had to be developed to maintain treatment success and continue treatment for new tics even after treatment termination. These challenges forced me to disembark from the direct path provided by the treatment manual and to develop an individualized case formulation and related individualized treatment plan. This paper will describe that process as well as highlight how I mitigated the negative effects of the confounds as well as utilized Hiro's own strengths to generate an ultimately successful treatment plan.

In that vein, through a systematic single-case study design, this case study hopes to illuminate the following questions with regards to clients with TS:

- 1. Are all of the elements in the CBIT treatment plan always successful?
- 2. How does ADHD impact CBIT treatment success and what can be done about it?
- 3. How can a clinician respond to challenging elements in a TS case?
 - a. Create an individualized case formulation that includes flexibility, ingenuity, and highlights the client's strengths.
 - b. Implement an individualized treatment plan that ignores unhelpful elements from the manual and incorporates the client's strengths and interests.
 - c. Monitor the effective and ineffective elements of the intervention to address and overcome challenges that arise during treatment.

The Clinical Setting in Which This Case Took Place

This case took place in the Tourette Syndrome program at The Graduate School of Applied and Professional Psychology (GSAPP) at Rutgers University (RU). At the time of the case I was a student in my second year in the doctoral school psychology program and a part of the TS program treatment team. The director of the TS program, who is a licensed psychologist, and who is knowledgeable about TS and comorbid disorders and CBT in general, supervised me throughout the case. Hiro was assigned to me after his initial phone intake. After three intake sessions with me, it was decided that TS would be the focus of treatment. After a discussion of what treatment would involve, treatment was begun, and lasted for 19 sessions.

The Methodological Strategies Employed for Enhancing the Rigor of the Study

Detailed clinical notes were written up after each session and reviewed by my supervisor. DVD recordings were also taken for most of the sessions. Both my notes and DVD recordings were utilized to detect both effective and ineffective elements of treatment by my supervisor and myself. Additionally, five quantitative measures were provided: two at the outset of treatment to measure behavioral issues and ADHD; another two measures were used for initial assessment purposes and to provide continuous monitoring of the client's symptoms; and one was used when treatment started and was used at the beginning of each session. The measures and results are discussed below in the chapters on assessment (IV) and outcome (VIII).

2. THE CLIENT

Hiro is a 10 year-old Indian male who lives with his biological mother, father and 7 year-old brother. He is in the fifth grade and mainstreamed in public school. He has a primary diagnosis of TS and ADHD, and his parents noticed his first vocal tics when Hiro was 4 years old and his first motor tics when he was 8. Hiro's first vocal tic was an "mmm" sound; his first motor tic, an unusual eye-blinking movement. The symptoms of both motor and vocal tics were at their worst when Hiro was 8 years old. The onset of tics was gradual and, according to his parents, followed the typical pattern of TS, with waxing and waning periods of tic intensity.

Hiro was brought in to the TS program for CBIT by his parents as a means for reducing the severity of his tics. At the time of intake, Hiro presented with multiple different motor and vocal tics, including simple motor tics—brief, darting movements such as head jerking and eyeblinking; complex motor tics—longer, more purposeful-seeming movements, such as touching objects or bending his entire body over; and simple vocal tics, such as sniffing and grunting. Additionally, Hiro's ADHD symptoms were obvious at intake as they were expressed through significant fidgeting in his seat, standing up, and occasionally walking around the room. He was referred to the TS program by the New Jersey Center for Tourette Syndrome (NJCTS), a statewide advocacy organization, for treatment of his tics.

3. GUIDING CONCEPTION WITH RESEARCH AND CLINICAL EXPERIENCE

Defining Tourette Syndrome

According to the DSM-V, the following criteria have to be met for a diagnosis of Tourette Syndrome:

- Presence of multiple motor and at least one vocal tic; and
- The tics are present for more than a year since the first tic was present; and
- The onset of tics is before the age of 18; and
- The tics cannot be the result of substances or a different medical condition.

A tic is an involuntary motor movement or vocalization. The severity and heterogeneity of tics are vast (Leckman, Bloch, Scahill, & King, 2006). A motor tic can range from a simple, easily camouflaged movement, such as a rapid hand squeeze, to a more noticeable and exaggerated movement, such as a patterned way of jumping. Vocal tics also have a broad range of form and severity, such that a simple vocal tic may be a quiet cough that is barely noticeable or seemingly normative, while a more severe tic could be a loud or animal-like cry. Tic presentation follows a waxing and waning pattern over weeks and months, where tics can be very severe at one point and essentially nonexistent at another (Leckman, Bloch, Sukhodolsky, Scahill, & King, 2013).

While prevalence studies over the past decade have demonstrated a wide range of estimates of Tourette Syndrome (TS) in the population (Scahill, Dalsgaard, & Bradbury, 2013), it can be said that roughly 3 to 5 per 10,000 adolescents and 10 to 13 per 10,000 children are affected by the disorder (Piacentini, Pearlman, & Peris, 2007), with boys affected at roughly three to five times the rate as girls (Bitsko et al., 2014). The worst age for tics tends to be between 10 and 12 years old; children's tics also tend to become more complex as they grow older (Bloch, 2013). Despite this, without treatment between one-third and one-half of children with TS experience a significant decline in tics when they reach adolescence (Bloch et al., 2006).

Tics often arise from a heightened sensitivity to stimuli (Conelea & Woods, 2008). When the feeling preceding the tic is internally motivated, that feeling is called a premonitory urge, of which children tend to become more aware as they grow older (Woods, Piacentini, Himle, & Chang, 2005). Most individuals with Tourette Syndrome claim that their tics are preceded by a premonitory urge, which is often described as an itch or tickle-like feeling that increases in tenor and can only be satiated with the performance of the tic (Leckman, Walker, & Cohen, 1993). While individuals with TS can suppress their tics for a time, this is difficult and requires attention that then cannot be placed on other social or occupational activities (Jankovic, 1997). Additional internal stimuli that can affect tics include emotional states, such as fatigue and anxiety, which often cause an increase in frequency and severity of tics; even positive emotional states, such as excitement, can cause an escalation of tics (Bornstein, Steffl, & Hammond, 1990).

External contingencies, such as social gatherings, playing video games, or time prior to public speaking, can also have an impact on tic symptomology in both positive and negative directions (Silva, Munoz, Barickman, & Friedhoff, 1995). Specifically, activities that will increase negative emotional arousal will likely increase tics while activities that require either concentration or goal-directed motor control (e.g., sports) could decrease the presence of tics (Leckman et al., 2013; Silva et al., 1995).

Behavioral Model of Tourette Syndrome

While this section will discuss a behavioral model for TS, it is not meant to imply that tics are purely learned behaviors with no biological components. In fact, a later section will discuss some of the biological factors in TS. However, elements of TS can certainly be understood based on principles of behavioral psychology. To begin, I will give a basic and brief overview of behavioral principles. I will then show how TS can be understood via those principles.

Behaviorists, such as Cooper, Heron, and Heward, (2007) would say that all human beings perform behaviors, which are the activities of living organisms. Behaviors, however, do not happen in a vacuum, but are influenced by the environment around them. According to this view, behaviors are actually responses to the environment, which can be described physically, temporally, or by its effects on behaviors (Cooper et al., 2007). Environmental factors can be broadly divvied up into two event categories: antecedents and consequences. Antecedents are the environmental factors that precede behaviors while consequences are the ones that follow behaviors (Cooper et al., 2007). A key component of behavior theory is that the environment can be shaped to affect the behaviors that are exhibited (Cooper et al., 2007).

When viewed through this lens, tics are behaviors that are affected by the environment but can also be shaped by changes in the environment. Antecedent factors will therefore be events that immediately precede the tics and can either increase or decrease their frequency and severity (Capriotti & Woods, 2013). These antecedents can be external, such as playing video games or reading a book (Conelea & Woods, 2008) or internal, such as boredom (Bornstein et al., 1990). How antecedents affect tics can vary broadly (Silva et al., 1995), but some constants include stress, which seems to exacerbate tics for almost everyone (Eapen, Fox-Hiley, Banerjee, & Robertson, 2004) and relaxation, which seems to decrease tics for almost everyone (Silva et al., 1995).

Consequences, according to this model, are the responses to tics immediately or soon after they occur. According to a Capriotti and Woods (2013) survey of the literature, attention from adults or parents can increase the frequency of tics while rewards for stopping a tic can decrease the occurrence of that tic. The idea is that consequences following a tic can reinforce the tics, while rewards for not performing a tic can positively reinforce the inhibition of a tic.

A special kind of antecedent in relation to TS is the premonitory urge that was discussed earlier. According to a behavioral model, the premonitory urge is viewed as an aversive stimulus. When the tic-behavior is performed, it alleviates the aversive feelings of the premonitory urge, therefore negatively reinforcing the performance of the tic (Capriotti & Woods, 2013). Considering that tics are consistently repeated behaviors, this relationship between the performance of tics and the alleviation of the premonitory urge is particularly insidious and reinforcing. This model for understanding TS is particularly important for cognitive behavioral therapy of TS, as will be explained in greater detail below.

Biological Components of Tourette Syndrome

There are multiple lenses through which a biological understanding of TS can be viewed, including, but not limited to, cellular pathology, electrophysiology (Orth, 2013), neurochemistry (Singer, 2013), and neurobiology (Greene, Black, & Schlaggar, 2013; Mink, 2001; Mink, 2003). This section will focus on the neurobiology of TS, and specifically the area of the brain known as the basal ganglia.

The basal ganglia form an area of the brain that sits at the base of the forebrain and is strongly connected to the cerebral cortex, thalamus, and brainstem (Stocco, Lebiere, & Anderson, 2010). According to Stocco et al. (2010), some of the main functions of the basal

ganglia include control and regulation of the motor and premotor areas of the brain, which have an impact on voluntary movements. The basal ganglia do this by either performing or not performing an inhibitory action on motor-control areas in the brain. According to these researchers, the basal ganglia are also involved in nondeclarative (i.e., implicit) learning in humans and stimulus-response association learning in animals.

The basal ganglia affect motor control through direct and indirect pathways (Greene et al., 2013). The process begins with the area of the basal ganglia known as the striatum receiving excitatory input from the cortex, causing a neuron to "fire" and send a message to the next brain area (Carlson, 2013). When the direct pathway is activated, the striatum then inhibits pathways leading to the thalamus, restricting the neuron from "firing," and sending a message to the next area in the brain (Carlson, 2013); at this point, the thalamus is then released from its resting state (Greene et al., 2013; Stocco et al., 2010). According to Greene et al. (2013), once released from the resting state, the thalamus excites the cortex, leading to motor performance. When the indirect pathway is engaged, the striatum causes the thalamus to remain in its resting state, thus not exciting the cortex, thereby restricting motor movement. In this sense, the basal ganglia is involved in both exciting and inhibiting the thalamus, which in turn affects motor movement in general.

The pediatric neurologist Jonathan Mink (2001) suggests that there can be aberrant activity in the striatum, leading to tics. According to Mink, there is inappropriate activity in different areas of the striatum that liberates the thalamus from its resting state and causes activity in the motor areas of the cortex. In this theory, activity in different areas of the striatum is directly connected to different tic expression (Greene et al., 2013; Mink, 2001). In other words, there is an excess of activity in the striatum of an individual with TS as compared to someone without TS. This extra activity engages the direct pathway from the striatum to the thalamus, exciting the thalamus, which in turn excites the motor cortex and leads to tics (Mink, 2001). This theory is supported by research on the postmortem TS brain, as well as on micro-stimulation and microinjection of the striatum (Greene et al., 2013).

Treatment of Tourette Syndrome

History of Behavioral Interventions for Tourette Syndrome

As mentioned previously, the treatment used in this case was CBIT. However, there is a history of behavioral interventions developed and used before the advent of CBIT (Capriotti & Woods, 2013), four of which were important in the eventual development of CBIT and which had varying ranges of effectiveness in my own treatment of Hiro.

One treatment was self-monitoring (Ollendick, 1981), which required an individual to become considerably aware of their tics. This treatment was found to have little effect on its own, but it is thought to be a useful auxiliary in CBT for tics (Woods, Miltenberger, & Lumley, 1996). The second was contingency management, a method that rewarded individuals for attempting to stop their tics (Wagaman, Miltenberger, & Williams, 1995). Relaxation training for suppressing tics (Bergin, Waranch, Brown, Carson, & Singer, 1998), the third treatment, was thought to be helpful because a person's tics worsen when stressed (Eapen et al., 2004). Like

self-monitoring, it was discovered that contingency management and relaxation training are better utilized as adjuncts to CBT for tics than on their own (Woods et al., 2008). Habit Reversal Training (HRT), the fourth intervention, has been found to be the most effective behavioral treatment for tics (Bate, Malouff, Thorsteinsson, & Bhullar 2011) and will be covered in greater detail in the next section.

Comprehensive Behavioral Intervention for Tics (CBIT)

CBIT is, strictly speaking, an eight-session empirically validated treatment for TS (Woods et al., 2008), although the actual number of sessions necessary can vary widely. At its core, CBIT uses HRT to help people who suffer from TS. HRT itself incorporates three different parts. The first is awareness training (Woods et al., 2008). This part of HRT requires the client to become extensively aware of their tics. The first step in this is to develop a thorough definition of the target tic. For example, a simple head-shaking tic might be defined as "a slight inclination of the head backwards, followed by a rapid, straight and vertical jerk of the head forward at a 30 to 60 degree angle." Next, the client is instructed to raise their finger every time the tic occurs, while simultaneously holding a conversation with the clinician about something unrelated to tics (e.g., hobbies). The point of this is to create a strong awareness of the tic even without explicit focus on it, as would be the case outside of the therapist's office (Capriotti & Woods, 2013). While this conversation is being held, the clinician praises the client for each successful recognition of their tics, but also informs the client when the client fails to acknowledge the tic. When the client reaches 80% efficiency in recognizing tic occurrence, the client and clinician develop a thorough definition of the premonitory urge (Leckman et al., 1993) that precedes the target tic. For the head jerking tic mentioned above, the definition might be "a pressure-like feeling at the crown of the head down through the forehead." The client and clinician then repeat the previously mentioned sequence, although this time the client raises their finger whenever they feel the premonitory urge as opposed to the tic itself.

Creation of a competing response (CR) is the next piece in HRT. The CR is a behavior that is incompatible with the tic; can be performed in any environment and for extended periods of time; and, ideally, is socially inconspicuous and less impairing than the tic (Woods et al., 2008). For example, the CR for the head-jerking tic might include a slight tensing of the neck muscles while holding the chin against the chest and/or looking straight ahead. Whatever competing response is created will have to be effective as well as amenable to the client. For this reason, creation of a CR is often a trial-and-error process that requires creativity and patience until the right CR is found.

Once the CR is created, the client will perform the CR for the target tic in treatment sessions whenever they feel the premonitory urge for the tic and, will sustain the CR for either a full minute or until the urge to tic dissipates. If the client forgets to use their CR and tics instead, the clinician will remind the client to use their CR, which they will then perform for a full minute. During this part of the treatment session, the client and clinician converse about something or engage in an activity unrelated to tics. The idea, again, is to simulate the use of the CR in the real world as much as possible.

The final component to HRT is social support (Woods et al., 2008). The identified social support system will usually be a parent, spouse, or some other family member. The clinician's job is to train the social support person to recognize when the tic occurs and praise the client when they use their CR or gently remind them when they do not. It is critical that the social support person realize that they are not to praise or admonish the non-occurrence of tics, but *only to support* the use of the CR skills outside of therapy.

CBIT incorporates many other components of behavioral treatments, such as functional-analytic-based treatments, relaxation training, and reward systems (Woods et al., 2008). In theory, the function-based elements of CBIT are among the most important, alongside HRT. Function-based treatment for TS is broken up into two parts in CBIT: First, an assessment of what variables contribute to both increased and decreased severity of tics; second, shaping the antecedents and consequences of the client's tics to help reduce tics. Antecedent functional interventions can include informing relevant people (e.g. teachers, classmates) of what TS is; reducing time spent in tic-inducing environments or activities (e.g. playing video games); or coaching the client to speak about having TS. A change in consequence can include not comforting a child after the performance of a tic. Also, if tics are used as an escape mechanism (e.g. child gets out of taking tests because of tics), discontinuing the escape procedures is another form of a consequence-based functional-intervention.

The following is a summary outline of CBIT based on the Woods et al. (2008) manual for treating children and adults with TS. The first two sessions include providing a rationale for treatment, psychoeducation about TS, creation of a tic hierarchy (a list of the present tics and the levels of inconvenience attributed to each), functional assessment of tics, and creation of a reward system. The skills taught in these first two sessions are monitoring and awareness of the tics (for both the client and the family), HRT for the first tic on the hierarchy, and functional intervention strategies for the first tic on the hierarchy. The rest of the sessions all include review of treatment procedures for the tics already covered, and HRT and functional-based assessment and intervention strategies for a new tic on the hierarchy. Relaxation training, incorporating diaphragmatic breathing and progressive muscle relaxation (PMR), is introduced in sessions four and five and is reviewed through the rest of treatment. Every session concludes with homework to review the skills developed in treatment. CBIT also incorporates other important elements for treatment, such as establishing rapport with the client, educating the client about TS, practicing the CR outside of treatment, and identifying a social support person to help the client with practice.

Cognitive Behavioral Therapy Mechanisms of Change

In an effort to pinpoint the exact mechanism of change in HRT, Woods et al. (1996) sequentially measured the effects of awareness training, self-monitoring outside of the session, social support, and use of CR on a small group of children with motor tics. They found that different elements of the treatment had effects for different children. For half the children it took awareness training, social support, and use of a CR to decrease tic symptomology. Awareness training and self-monitoring together were effective for one fourth of the group, while awareness training alone was effective for the last quarter.

Awareness training may be effective at tic reduction because of the enhanced awareness itself, either in the training or in the self-monitoring (Ladouceur, 1979). In this sense, awareness training itself might serve as an active treatment. Alternatively, awareness of the tic itself might serve as an aversive stimulus that punishes engagement of the tic (Nelson & Hayes, 1981), or as motivating the individual to avoid the tic by suppressing it (Woods et al., 1996).

HRT may be effective because it habituates the individual with tics to the premonitory urge preceding the tic. This perspective implies two possibilities. One is that the premonitory urge is the actual aversive stimulus for people with tics. This theory is supported by research showing that performance of the tic provides relief from the premonitory urge (Scahill, Leckman, & Marek, 1995). The other possibility implied is that tics are voluntary actions that are performed to relieve the unpleasant feelings preceding the tic, which is how one physician with TS described his tics. According to him, the only "difference between a tic and a normal action is that a tic happens because of an irrepressible physical urge" (Turtle & Robertson, 2008). He concludes that the only way to satiate the sensation is to perform the tic. It has also been suggested that the premonitory urge is in fact a neutral event that becomes negatively associated with the aversive physical and social consequences of the tic (Best, 2009).

If the tics are negatively reinforced by relief from the premonitory urge, then performance of the CR may be blocking the tic and allowing for habituation to the aversive premonitory urge to occur. The ideas here are akin to how exposures in anxiety treatments habituate anxious individuals to the feared content, teaching them that they will be okay even when exposed to the source of their anxiety (Kendall, Furr, & Podell, 2010). With HRT for tics, awareness of the premonitory urge, coupled with performance of the CR for at least a minute or until the urge dissipates, teaches the individual that they can experience the premonitory urge, abstain from ticcing, and eventually the urge will go away.

CBT Versus Psychopharmacology

The question of whether or not to use medication in the treatment of any psychological pathology is often a tricky one. In the case of TS, it is further compounded by the lack of consistent and clear literature on what drugs work best, which is further affected by the waxing and waning cycles of TS as part of its normative developmental patterns (Roessner et al., 2011). Two important concepts to keep in mind while determining what treatment to provide for TS are equifinality—the idea that a single disorder can be caused via different pathways—and multifinality—the idea that the same internal or external event can lead to different outcomes in different individuals, some adaptive and others maladaptive (Hinshaw, 2013; Holmbeck, Devine, & Bruno, 2010). These ideas are useful because if TS can have different etiologies in different individuals, it is easier to understand why different pharmacological and behavioral treatments are effective for different people.

One family of drugs used for TS treatment is antihypertensive, also known as alpha 2 agonists. These drugs were originally used to lower blood pressure and for ADHD treatment in youth (Roessner et al., 2011), but have also been found to be effective for treating mild to moderate TS. The two drugs in this family used for TS treatment are guanfacine and clonidine.

Common side effects of these drugs include headaches, fatigue, somnolence, and nausea, although some of these side effects, such as somnolence and fatigue, are known to remit after 2 weeks. Due to the mild nature of the adverse effects of these medications, these drugs are often used as the first line of defense for TS pharmacological treatment.

A second line of pharmacological treatment for TS is dopamine agonists. Dopamine agonists are also known as antipsychotic drugs. These drugs fall into two categories: typical, which are older, and atypical, which are newer. An effective, typical antipsychotic for tics is haloperidol, which has a fairly large evidence base behind it (Roessner & Rothenberger, 2013) but also a number of serious side effects that range from drowsiness to weight gain and sexual dysfunction, though the side effects can be as severe as extrapyramidal symptoms, such as dystonias and Parkinson-like symtpoms. (Sallee, Nesbitt, Jackson, Sine, & Sethuraman, 1997). An effective atypical antipsychotic for tics is risperidone, which is similarly as effective as haloperidol, but with similar yet less severe side effects (Scahill, Leckman, Schultz, Katsovich, & Peterson, 2003).

While different medications have been studied for the treatment of TS, the seemingly most effective are those that target dopaminergic hyperfunction (Bloch, State, & Pittenger, 2011). In a review of the literature by Singer (2013), studies have found increased number of striatal and cortical dopamine receptors in clients with TS as well as increased dopamine dispersal in their brains. For this reason, dopamine agonists, which work by blocking dopamine reception in the brain, are among the most effective pharmacological treatments for TS (Robertson, 2000). A common critique of the medication trials with TS has been the lack of long-term studies evaluating the long-term effects of medication on tics for individuals with TS (Roessner et al., 2011)

Having already described the form of CBT known as the CBIT/HRT treatment for TS, I will now provide a brief summary of some of the findings that show CBIT/HRT for TS to be an effective treatment and reasonable alternative to medications. Reviews of both large, randomized controlled trials and smaller studies, by Himle, Woods, Piacentini, & Walkup (2006) and Cook and Blacher (2007), as well as a meta-analysis by Bate et al. (2011), have shown that HRT for TS has a consistent and clinically significant impact on reducing tics. One large, randomized, and methodologically rigorous study by Piacentini et al. (2010) showed CBIT to be significantly more effective than Supportive Psychotherapy (SP) for youth with TS, while a study by Wilhelm et al. (2012) showed similar results for CBIT with adults. Both studies showed over 80% of patients in the CBIT group maintaining gains at 6 months post treatment. HRT-based treatment also meets the APA Division 12 criteria for being a well-established intervention for tics (Cook & Blacher, 2007).

When directly comparing CBT to psychopharmacology, the advantages of CBT treatments rest mainly in the lack of side effects. Additionally, they may provide a sense of control that medical treatments could lack, as well as lasting effects that would otherwise dissipate with the discontinuation of medication. That being said, from my own clinical experience, medications are sometimes needed for more severe cases of TS before behavioral treatments can have any effect; it may be that the tics are so severe with some clients that medications are needed to minimize their extreme frequencies and duration before attempting

behavioral treatment. At the same time, it is important to note that a study of CBT for TS in both medicated and unmedicated participants by O'Connor et al. (2009) showed no difference in tic reduction between the two groups. However, in cases where TS comorbidities, such as ADHD, are prominent, medication may be a necessary measure for the comorbid pathology before CBT for TS can be effective (Roessner et al., 2011; Scahill et al., 2006).

Behavior, Biology, and TS: An Interactive Model

As mentioned above, from a behavioral perspective, tic behaviors are performed to relieve the negative stimulus that is the premonitory urge, and is reinforced through the principles of negative reinforcement (Scahill et al., 1995). From a biological perspective, there is inappropriate activity in the basal ganglia that causes the tic and is reinforced by the neurochemical dopamine (Mink, 2001). Dopamine therefore appears to be a key component of understanding how a behavioral perspective of TS can be integrated with a biological understanding of TS, for those who are genetically susceptible to TS.

The basal ganglia form an area of the brain that is replete with dopamine receptors (Mink, 2001). In studies where mice are connected to electrodes that stimulate dopamine production, the mice have pressed levers to activate the electrodes to the point of starvation and even death (Ridley, 1999). This indicates that activities that increase dopamine can motivate repeated behaviors because dopamine production causes feelings of pleasure, or at least enhances the motivational salience of the behaviors (Berridge & Kringelbach, 2008). Given that antipsychotic drugs that reduce dopamine production have been effective in reducing tics, it is reasonable to believe that production of tic behavior is related to dopamine.

Given that the basal ganglia and dopamine have also been implicated in nondeclarative learning (Stocco et al., 2010), a picture of how biology and behavior regarding tics becomes plausible. In this picture, a tic is a nondeclarative learned response where performance of the tic reduces the premonitory urge. This reduction is reinforced because of an increase in dopamine, which either creates feelings of relief (i.e., dissipation of the premonitory urge) or enhances the motivation to produce the tic behavior in response to the premonitory urge. This is similar to the mouse hitting the lever. Just as the mouse learns that hitting the lever creates a feeling which is mediated through a dopamine related mechanism, an individual with TS learns that performing the tic creates a positive feeling, or at least decreases a negative feeling, which is also mediated through a dopamine related mechanism. This is obviously a very tentative hypothesis, but research on TS related to dopamine and the basal ganglia support it as a possibility.

Psychosocial Implications and Common Comorbidities of TS and the Importance of TS Treatment

TS rarely presents alone. An individual with TS is significantly more likely to be diagnosed with ADHD and obsessive-compulsive disorder (OCD) than their peers (Gaze, Kepley, & Walkup, 2006; Kadesjö & Gillberg, 2000). Typical ADHD symptoms include difficulty with focusing on tasks or people, forgetfulness, and hyperactivity. An interesting perspective provided by Denckla (2006) proposes that there are actually two separate conditions: TS with ADHD and pure TS. She presents data indicating that individuals with TS alone have

enhanced motor speed, higher IQ, and no learning disabilities compared to the TS with ADHD population. Gaze et al (2006) posits a number of theories as to the high comorbidity rate between ADHD and TS, two of which suggest that there is a underlying genetic abnormalities in neurotransmitter functioning that leads to both TS and ADHD. Regardless, comorbid TS and ADHD lead to greater social difficulties for children than TS alone (Carter et al., 2000). Additionally, Eddy et al. (2011) reported that children with both diagnoses have lower perceived quality of life.

Typical OCD symptoms include obsessions, such as extreme and irrational fear of germs and contamination, and compulsions, such as repeated and excessive hand washing as a means of relieving the anxious obsession. Youth with TS have more obsessive-compulsive symptoms (OCS) than do their peers (Grad, Pelcovitz, Olson, Mathews, & Grad, 1987). One theory as to the higher rates of OCD and OCS in the TS population posits that it is a dopaminergic dysfunction that may serve as a common pathophysiologic mechanism in both TS and OCD (Denys et al., 2013).

Children with TS are also commonly viewed as less popular and often report teasing because of their tics (Bawden, Stokes, Camfield, Camfield, & Salisbury, 1998; Stokes, Bawden, Backman, Dooley, & Camfield, 1991). Other psychiatric conditions such as anxiety, depression, and learning disabilities may also coincide with TS, further complicating the lives of those with TS (Gaze, Kepley, & Walkup, 2006).

4. ASSESSMENT OF THE CLIENT'S PRESENTING PROBLEMS, GOALS, STRENGTHS, AND HISTORY

Presenting Problems

Hiro presented for treatment with moderate TS symptoms. He entered treatment at this time because his parents were worried that his tics would continue to worsen and impede his future ability to succeed. At the time of intake, Hiro presented with multiple motor and vocal tics, including simple motor tics—brief, darting movements such as head jerking and eyeblinking; complex motor tics—longer, more purposeful-seeming movements such as touching objects or bending his entire body over; and simple vocal tics, such as sniffing and grunting. In addition, Hiro's ADHD symptoms were obvious at intake as they were expressed through significant fidgeting in his seat, getting out of his seat, and occasionally walking around the room.

Relevant Background Information

Hiro was born full-term with no major complications. Hiro met all developmental milestones, with complications only in overactivity, impulsivity, unusual fearfulness, and sleeping. However, Hiro grew out of sleeping issues and unusual fearfulness. Hiro had an adenoidectomy at the age of two due to frequent colds and allergies. Hiro continued to have frequent cold and allergy symptoms until the age of 8. At the time of treatment, Hiro had minor

dust and milk allergies. Hiro and his parents denied current or past depression, suicidality,

Quantitative Assessment

Yale Global Tic Severity Scale (YGTSS)

trauma, abuse, or major medical concerns.

The YGTSS is a clinician-rated evaluation tool that is given in a semi-structured interview format and measures tic severity and overall impairment due to tic symptoms (Leckman et al., 1989; Scahill & Leckman, 1995). Its psychometric properties are considered among the best for TS scales (Kompoliti & Goetz, 1997). The YGTSS assesses the current tic repertoire, both as endorsed by the client and observed by the clinician, and the severity on multiple dimensions, including number of tics, frequency, intensity, complexity, and interference.

These dimensions are rated on a 0 to 5 scale, with 0 representing lowest level of severity and 5 representing highest level of severity. There is also a Tic Impairment Scale that ranges from 0 to 50 and is anchored at 10-point benchmarks. These scales are then combined to equal a Total Motor Tic Score, which ranges from 0 to 25; a Total Phonic Tic Score, which also ranges from 0 to 25; and a Total Tic Score, which ranges from 0 to 50. A Total Tic Score of 9 or lower is indicative of minimal tic symptoms; a score between 10 and 19 is considered mild; a score between 20 and 30 is rated as moderate; and a score between 31 and 50 is considered a marked level of tic symptoms. Storch et al. (2011) proposes that a 35% change, or 6 or 7 point change, in the Total Tic Score should be considered a clinically significant treatment response.

The YGTSS was completed at the beginning of treatment and again at the end. The YGTSS was then completed at a follow up evaluation three months after treatment ended, and again six months post treatment (Table 1). At the beginning of treatment, Hiro received a Total Tic Impairment score of 26 (Total Motor Tic Score=14; Total Phonic Tic Score=12), which placed him in the moderate level of tic severity before treatment. He received a 10 on the impairment scale, indicating that his tics cause him minimal difficulties with family life, self-esteem, social acceptance, or school functioning.

Children's Yale-Brown Obsessive Compulsive Scale (CY-BOCS)

The CY-BOCS is a clinician-rated evaluation tool that is given in a semi-structured interview format and measures obsessive and compulsive symptoms that are related to OCD and has been noted to have high reliability and validity (Goodman et al., 1989). The CY-BOCS assesses current and past obsessive and compulsive repertoires, both as endorsed by the client and observed by the clinician, as well as the severity of the obsessive thoughts and the compulsive behaviors. The severity of the obsessions and compulsions are measured on multiple dimensions, including Time Occupied by Thoughts/Compulsions, Interference, Distress, Resistance Against Obsessions/Compulsions, and Degree of Control over Obsessions. Scores on the CY-BOCS range from 0 to 40, with a score of 0 to 7 implying subclinical, 8 to 15 implying mild, 16 to 23 implying moderate, 24-31 implying severe, and 32-40 representing extreme.

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The CY-BOCS was completed by Hiro at the beginning and again at the end of treatment. At the beginning of treatment, Hiro received a total score of 18 (Obsessions = 8; Compulsions =10), indicating moderate level of OCD severity (Table 2). At the time, Hiro reported obsessions that included the intrusive word "toodles." Hiro's compulsions involved touching his pencil as well as using his pencil to, in his words, "breathe." In other words, he would imagine his pencil was like a straw and would breathe in and out of it. Hiro reported a daily occurrence of symptoms, with symptom-free periods of multiple hours. Clinical interpretation of the CY-BOCS suggested that Hiro may have OCD; however, Hiro may have confused normal thoughts for obsessions and tic behaviors as compulsions. Therefore, it was my clinical judgment that Hiro better met the criteria for TS alone, as opposed to TS and OCD.

Achenbach: Child Behavior Checklist (CBCL; Achenbach, 2001)

The CBCL is a self-report questionnaire that describes a parent's perception of their child across a spectrum of common child problem areas. Hiro scored in the borderline range for the Attention Problem scale and in the normal range for all other scales of the CBCL. The scores indicate that Hiro's parents perceive that their child has some difficulties with attention that warrant action.

Achenbach: Teacher Report Form (TRF; Achenbach, 2001)

The TRF is a self-report questionnaire that describes a teacher's perception of a student across a spectrum of common child problem areas. This was scored by one of Hiro's teachers. Hiro scored in the clinically significant range for the Hyperactivity-Impulsivity subscale. Hiro scored in the normal range on all other scales of the TRF. The scores indicate that Hiro's teacher perceived that he has difficulties with hyperactivity and impulsivity.

Conner 3 – Teacher Report (http://www.mhs.com/product.aspx?gr=cli&id=overview&prod=conners3)

The teacher version of the Conners 3 is a self-report that describes a teacher's perception of a student in terms of attention, hyperactivity, impulsivity, conduct, and oppositionality. The same teacher who filled out the Achenbach TRF scored this. Hiro was in the clinically significant range on the following scales: Inattention, Hyperactivity/Impulsivity, DSM-IV-TR Inattentive, and DSM-IV-TR ADHD Hyperactive-Impulsive. He was in the borderline range for Executive Functioning. He was in the normal range for all other scales. The scores suggest a high likelihood that Hiro met the diagnosis for ADHD—Combined Type.

Conners 3 – Parent Report (http://www.mhs.com/product.aspx?gr=cli&id=overview&prod=conners3)

The parent version of the Conners 3 is a self-report that describes a parent's perception of their child in terms of attention, hyperactivity, impulsivity, conduct, and oppositionality. Hiro was in the clinically significant range on the following scales: Inattention, Hyperactivity/Impulsivity, and Executive Functioning. He was in the normal range for all other scales. The scores suggest a high likelihood that Hiro met the diagnosis for ADHD-Inattentive Type and possibly for ADHD-Combined Type.

Diagnosis (Table 3)

At the onset of treatment, Hiro met the DSM-5 (American Psychiatric Association, 2013) criteria for Tourette Syndrome and Attention-Deficit Hyperactivity Disorder—Combined Type. Hiro's diagnosis of Tourette Syndrome was based upon past neurologists' diagnoses along with his current description of his tics, scores on the YGTSS, observed motor and vocal tics, and reported tic history. His diagnosis of ADHD – Combined Type was based upon both Hiro's and his parents' current description of his attentional difficulties and impulsivity, in addition to the Conners 3 and Achenbach report forms by the parents and Hiro's teacher. Hiro's CY-BOCS score suggested a diagnosis of OCD. However, based on a clinical interview with Hiro, the fact that individuals with TS often have OCS but not full-blown OCD (Grad et al., 1987), and clinical interpretation of Hiro's descriptions of his OCD symptoms as being better understood as his using vivid descriptions of what were actually normative behaviors, Hiro's OCD symptoms seem better understood as a manifestation of his Tourette Syndrome alone.

Table 3 presents a DSM-IV diagnosis of Hiro to include the Global Assessment of Functioning (GAF) Scale that was removed from the DSM-5.

Strengths

Hiro has a good relationship with his parents and younger brother. He is bright, as noted by his parents, his teachers, and myself. He loves to learn new things and his parents reported that he is constantly surprising them with his ever-growing knowledge base. He also has a positive relationship with his teachers—something that is often, unfortunately, not the case for students with ADHD (Sukhodolsky et al., 2003). He also has a group of friends and does well socially; and he even related to me during treatment that he was comfortable talking about both his tics and treatment with his friends and teachers.

Hiro's interests include reading, running around, and video games—knowledge of the last proved critical to successful treatment. Hiro also has an incredibly active and powerful imagination, which served to be a key element to successful treatment. In addition, he also has a fun-loving, silly personality. Further, Hiro displayed impressive insight into his tics. As treatment progressed, Hiro was able to describe his tics and premonitory urges in dramatic detail and was also able to recognize their occurrence with high accuracy.

5. CASE FORMULATION AND TREATMENT PLAN

Formulation

Before treatment began, the following informed my conception of Hiro's case: Tourette Syndrome (TS) is a neurological disorder with childhood onset and presents with varying motor and vocal tics and, given that, accurately described Hiro's pathological traits. Since TS is neurological in origin, it is a condition that Hiro was born with. Environmental factors that might affect it are unknown. Additionally, Hiro did manifest behaviors associated with a diagnosis of ADHD Combined Type, a common comorbidity with TS (Denckla, 2006). However, this did not seem an important factor in his case formulation because his ADHD did not seem to negatively

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impact him in other areas of his life, such as grades, relationships with his peers, or his relationship with his parents. Given the lack of negative impact his ADHD had had on his life thus far, I assumed it also wouldn't impact treatment designed to mitigate his tics.

Treatment Plan and List of Treatment Goals

While there are 30 years' worth of discussion in the literature on behavioral interventions for TS, very few clinicians seem familiar with this literature (Woods et al, 2008). Fortunately, at the GSAPP TS program, team members are specifically trained to utilize behavioral treatment for tics, and therefore that was my clear first choice when deciding on what treatment plan to create. Because TS so often presents with comorbidities (Gaze et al., 2006) and kids with TS often are impacted in multiple ways (Stokes et al., 1991), these factors are generally important to take into consideration when developing treatment. However, with Hiro, as the tics themselves appeared to be the only significant stressor—and even then, mostly just for his parents—they were the main target of treatment.

The manual most recognized for children with TS is Woods et al.'s (2008) *Managing Tourette Syndrome: A Behavioral Intervention for Children and Adults*. This manual is composed of multiple elements for treatment of tics, including relaxation training, a functional assessment of the environment to reduce exposure to tic-inducing settings, and Habit Reversal Training to specifically target tic management and reduction. The manual also includes the use of a behavioral reward system for performance of homework. However, at the suggestion of my supervisor, I did not incorporate that element because we wanted to develop in Hiro a sense of internal motivation.

Given Hiro's presentation, his initial treatment goals were the following:

- -- Develop buy-in for treatment and belief from parents and from Hiro in the efficacy of treatment.
- -- Discover and functionally deal with environmental conditions that may increase tic symptoms.
- -- Develop relaxation skills to help manage tics.
- -- Create awareness of tics when they occur.
- -- Create awareness of premonitory urges before tics occur.
- -- Develop Competing Responses (CR) to negate tics.
- -- Decrease number and frequency of tics as recorded on the YGTSS and SUDS (Subjective Units of Discomfort Scale, a measure of how much a given tic bothers and causes subjective discomfort for an individual).
- -- Teach skills so that Hiro can continue creating CRs for new tics post treatment.

These goals were presented, more or less, in the order that the manual laid out treatment. The first goal was critical to the success of the other goals, and thus needed to be met before attempting any further goals. The last two treatment goals, on the other hand, were envisioned as natural outcomes of successful treatment, as opposed to actionable in and of themselves.

The Case of "Hiro": Treating Tourette Syndrome by
Comprehensive Behavioral Intervention for Tics (CBIT)

J.D. Lichtman

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6. COURSE OF TREATMENT

Phase I: By the Book (Sessions 1-5)

Sessions 1-2: Testing the Client's Feet in the Water

For this point in treatment, the manual suggested a few steps, beginning with psychoeducation about TS. The creation of a Tic Hierarchy, a list of current tics in order of which are the most impeding, was stressed as a very important component of treatment, as well as establishing a "Subjective Units of Distress Scale" (SUDS) for all tics on the Hierarchy. We were also to create a Tic Inconvenience list, involving a functional assessment of the environmental factors that contribute to increased ticcing and that provided a practical means of establishing ways that the tics impeded Hiro's functioning. Lastly, we began HRT for the first tic on the Hierarchy. Before we began treatment, I knew Hiro's parents were anxious about how his tics would impact him in his life. For this reason, I disclosed that I myself had TS during the intake sessions as a means of relaying that TS was not a fatal diagnosis and that Hiro's tics did not destine him to a future of disappointment and lack of success. I believe this level of disclosure strengthened Hiro's parents trust in me and Hiro's belief that I understood the difficulties of managing tics.

In our first session, I explained to Hiro that the first tic we would work on would be the one that bothered him the most. The idea was to develop buy-in from him, so we wanted him to be motivated to change the tic. In this first session, Hiro was able to operationalize both his eyerolling tic and the premonitory urge that preceded it in a clear manner. He described the tic as a rolling of both eyes first up, and then immediately in a leftward direction, with the movement ending with his eyes facing downwards. The entire movement lasted about three to five seconds and usually appeared in bouts of three to four eye rolls. The premonitory urge consisted of tenseness in the tops of both eyes that then proceeded to the left and then the bottom of his eyes (see Table 4 for a list of operational definitions of tics, premonitory urges, and competing responses).

While performing the functional assessment, Hiro's parents suggested that his tics were at their worst when he first came home from school each day. Given that, we decided that Hiro and his mother would play catch when he first came home. We hypothesized that if he were able to have a more fun arrival home from school, he would be more relaxed for the rest of the evening and his tics would not be as bad. For homework for this first session and the next, we implemented this time to play ball, and had both Hiro and his parents keep track of his tics during the week.

After consulting with my supervisor, we decided to switch our first target tic to a jaw clenching, since eye-related tics tend to be very trenchant and we wanted to start with an easier tic to help build up Hiro's sense of self efficacy. In the second session, Hiro was again able to operationalize both his jaw tic and the premonitory urge that preceded it in a clear manner. He described the tic as a clenching of the back jaw muscles together, without the teeth ever touching. The premonitory urge consisted of tenseness in the back of both his jaw muscles at the same time.

I carried out all of the components of the treatment manual at this point and the tentative beginning of a positive relationship was forming between myself and Hiro, and between myself and his parents. I used these first few sessions to continue to build the rapport that I had initiated during the intake sessions. One of Hiro's favorite topics of conversations was a video game called Clash of Clans. This video game would remain one of our primary topics of conversation while using CBIT for his tics for the rest of treatment. As I will discuss later, I was actually able to incorporate Hiro's love of this game into treatment.

Sessions 3-4: Success is in the Eye of the Beholder

While the first sessions seemed to go well, by the third session Hiro's parents began losing faith in treatment, as revealed through their arrival times to treatment. Up to this point, for the intake sessions and the first two sessions, Hiro's parents had always arrived early to the clinic. However, his parents started arriving five to fifteen minutes late during this phase.

While Hiro's parents were starting to doubt the treatment at this point, it's important to note Hiro's own experience. In the third session, we developed our first CR together—in this case, for his jaw tic. This session is a great example of the experimental, trial-and-error nature of CBIT. The first CR we developed, holding his jaw open while breathing in a slow, controlled manner, had no effect on Hiro's resistance to tic performance. Thus, we developed a second CR that had Hiro holding his jaw closed instead. While performing this CR, Hiro described to me that his CR was the Barbarian King in his Clash of Clans game, while the tic was the evil Pikies. In this analogy, the Barbarian King (his CR) was protecting the village from the Pikies (his tic). This session would later serve as monumental for me, because it provided me with the idea of using Hiro's imagination as a clinical tool.

At this point I was still strictly adhering to the manual, adding new elements, such as relaxation techniques, as stipulated. However, by session four I dropped the functional assessment piece at the suggestion of my supervisor. At this point, I began to see success of the HRT component of treatment, as did Hiro. However, Hiro's mother told me that he was not practicing at home, thereby negatively affecting buy-in from Hiro's parents.

Session 5: On the Precipice

Knowing that Hiro's parents' buy-in was hanging in the balance, I spent the beginning of this session in conversation with them, trying to explain that treatment patience was needed and that success in the treatment would not be immediate. During this conversation, Hiro's mother said that she thought his impulsivity, part of his ADHD diagnosis, might be impeding his ability to succeed. At this point I acknowledged his mother's concern and replied that I would both think about what she said and discuss it with my supervisor. When meeting with Hiro during this session I maintained adherence to the manual—which in this context meant maintaining the relaxation components of treatment, despite any evidence that it was effective for Hiro—but I had a growing concern that something had to change for treatment to succeed.

Interlude: Reformulation of Case and Goals

At this point, I realized my original conceptualization of the case was inadequate. Hiro's ADHD symptoms seemed to be significantly detracting from his ability to use his CR when at home, thereby making it seem to his parents that he was not gaining at all from treatment. Accordingly, an updated conceptualization of this case required incorporating his ADHD and understanding how that impacted our treatment goals. As Barkley (1997) attests, ADHD causes deficits in self-regulation for motivation and motor control. In the case of Hiro, who also has TS, this appeared to have created a situation where, even though he knew how to use a CR, he would tic impulsively before even thinking about using his CR, which is what was happening at home and why he wasn't practicing his CR at home. My conclusion was that he did not lack the skills, only the attentional motivation and control to effectively use the skills he had been learning.

As part of this formulation, I recognized that success during treatment sessions, as opposed to homework, was the result of two factors: (1) During sessions I acted as Hiro's proxy executive functioning, as is often the case in ADHD treatment (Kazdin, 2008), generating the self-regulation piece that was missing at home; and (2) Coming to session was fun for him because we spoke about video games, a topic of great interest to him; therefore, he was motivated to perform his CR because that meant continued video game discussion. To replicate these missing features in his homework, I decided to incorporate a behavior chart to motivate Hiro to practice at home and keep him focused on his homework tasks. A copy of the table and instructions for its use are provided in Table 5.

This reformulation of Hiro's case also required an updated version of goals. Below is the list of the original goals. Each bold * represents a goal that I felt at this point was unsuccessful and detracting from the most successful element of treatment, the HRT component, while an unbolded × represents a goal that I had yet to meet but was retaining for treatment. The goals with no marks beside had been partially met or were in the process of being met at the time.

- -- Develop buy-in for treatment and belief from parents and from Hiro in the efficacy of treatment. X
- -- Discover and functionally deal with environmental conditions that may increase tic symptoms. **★**
- -- Develop relaxation skills to help manage tics. **≭**
- -- Create awareness of tics when they occur.
- -- Create awareness of premonitory urges before tics occur.
- -- Develop Competing Responses (CR) to negate tics.
- -- Decrease number and frequency of tics as recorded on the YGTSS and SUDS. X
- -- Teach skills so that Hiro can continue creating CRs for new tics post treatment.

At this point I decided to focus in on the HRT components of the goals, build stronger buy-in from Hiro's parents, and add successful use of the behavior chart to the goals.

Phase II: I Go My Own Way: Slight Deviations with Big Differences (Sessions 6-7)

In our sixth session I introduced the behavior plan outlined in Table 5 for homework assistance. First I introduced it to Hiro. I instructed him in its use, explaining that he could earn stickers in treatment and at home, and should put the stickers on himself and pick the amount of video game time he wanted as his reward. Initially, Hiro did not believe that his parents would let him play video games as a reward as he and his brother were only allowed to play on weekends. However, he humored me and we moved on to work on both a grunting tic and a squeaking tic, because the same CR, diaphragmatic breathing, was used for each. At the end of session, I had him explain the behavior plan to his parents and he was amazed and excited that they were on board.

There are at least two reasons why I think this reward system was effective. The first is that it was another way of bringing his favorite game, Clash of Clans, into the therapeutic setting. Hiro wanted to be a video game designer when he became an adult, so I believe incorporating video games as a treatment component was a way of showing him that adults took his video game ambitions seriously and not just as a childhood fancy. Secondly, Hiro's home was already very structured and organized, allowing the reward system to become a functional part of an already fairly systemic home schedule.

By our seventh session a few impressive things had occurred. First, Hiro came in with a fully completed homework form. He was very proud of himself, and while his parents still felt he was not using his CR's enough outside of the practice time per day, it was clear that they were satisfied as well. They began showing up to clinic on time again. I indicated to them that at this point, it would be better to focus just on the practice sessions and move on to generalizing his skills at a later point. Second, Hiro began taking more initiative during sessions. While he felt the CR we had developed in this session was working well, he was not as confident that it would continue to hold up. Therefore, he had us develop a backup CR, just in case he needed it.

At this point, Hiro's SUDS also started to drop significantly on the tics we were working on. As Hiro's tics were decreasing, he also had an epiphany: Up to this point, he did not feel that his tics bothered him much. However, as he was learning to manage them on his own, he confessed that they bothered him more than he recognized and it felt amazing that he was able to manage them so well.

Phase III: Success in the Eyes of Everyone (Sessions 8-12)

At this point, treatment really hit a really good groove. My relationship with Hiro was growing, success with CRs was increasing, SUDS were decreasing, and homework time was incrementally increasing from a half hour to an hour, then to 72 minutes, and finally to 90 minutes by Session 12. Hiro also reported that he was starting to generalize his tic management techniques to outside of therapy and homework time. This added to his growing sense of self-efficacy when it came to his tics. Hiro's parents also noted his dramatic progress at this point. During check-ins with them, they would comment that they did not realize that Hiro could manage such control of his tics, and that they were very proud of him and his progress.

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In our eighth session, Hiro excitedly reported that all of his friends and teachers knew about his CRs, and his teachers would even remind him to use them. In this session we also incorporated a new piece to treatment: quizzes. Up to this point, after we chose which next tic on the hierarchy to work on, I would lead Hiro through the different steps of HRT. In this session, however, after we picked the tic to focus on, I asked him what the next part of treatment entailed. Hiro looked puzzled at this change in procedure, then hesitantly offered that he would describe his tic. I praised him enthusiastically and then explained that from now on, he would be indicating which step went next for creating each new CR for a tic.

Hiro came into our ninth session having had a rough week with his tics, a fairly typical example of the natural waxing and waning cycle of tics. Not only had his grunting tic, which we had already developed a CR for, come back, but it proved even more resistant to his CR. Additionally, he had developed a new jaw tic (another example of a common phenomenon in TS: the development of new tics at arbitrary times) that was causing him pain in his ear. When given a choice as to which tic to work on, Hiro chose the new jaw tic. We had a successful session and by the end, when it was suggested that his practice time be increased to an hour, Hiro was happy with that change.

For our tenth session Hiro decided he wanted to work on an eye-blinking tic, even though his grunting tic persisted. However, the first few CR's we developed were not working. At this point, I started introducing visualization for particularly challenging tics such as this one. The idea behind visualization, which was inspired both by Hiro's previous use of analogy between his tics and CRs with Clash of Clans as well as advice from my supervisor, was that Hiro would picture various characters from the Clash of Clans game, as the premonitory urge, the tic, and the CR. The premonitory urges were enemy characters, the tic itself was the attack from the enemies on his home base, and the CR was his character's retaliation against the enemy. As Hiro practiced his CR, he simultaneously visualized it as his troops as they were protecting his city from the enemies, the premonitory urge and the tic. This use of his powerful imagination was enormously helpful for this tic, and would continue to be a potent tool for particularly challenging tics in later sessions.

In our eleventh session Hiro and I discussed treatment thus far. Hiro reported that he felt he had gained a great deal from treatment. He explained that when he first started treatment and the clinician asked him to explain the first tic he was working on in detail, he felt that the whole thing was silly. However, he said that after three treatment sessions he had already started to see improvement.

I learned that Hiro was moving back to India with his family soon. While I knew that we had made very important progress up to that point, I also knew he was not ready to embark on his own just yet. Therefore, I needed to set up a plan to prepare and empower Hiro to continue the great work we had begun.

Phase IV: Beginning of the End (Sessions 13-18)

To prepare Hiro for post-treatment success, I came up with the following plan:

- 1. Session 14: Hiro's parents would observe me going through a typical session with Hiro, with me pausing along the way to explain what we were doing and why.
- 2. Session 15: Hiro and I would meet alone to process how the experience of his parents watching the previous session was for him.
- 3. Session 16: Same as Session 14.
- 4. Session 17: In this session, Hiro's parents would run treatment while I observed and offered praise and critique.
- 5. Session 18: Same as Session 17.

The concept was that after this mini-training "seminar," Hiro's parents would be able to continue treatment with him after termination. This would mean weekly "sessions" for Hiro with his parents in which they would together develop new CRs for new tics and the momentum from treatment would continue. The plan was to empower both Hiro and his parents so that treatment was a success for them as a family, not just for myself as the clinician.

In our thirteenth session I laid out this plan, which Hiro's parents found highly encouraging, and they expressed relief that a plan was in place. In this session, we also returned to the snorting tic, because Hiro stated a need for a stronger CR. We again used visualization here to help bolster the CR process.

Hiro's parents sat in on the fourteenth session with Hiro and myself. Before we began, I confirmed with Hiro that he would feel comfortable with his parents present. Once he approved the change, the session commenced. I explained to his parents that the first step was to pick a tic to work on, after which we did just that. Next, I explained that Hiro needed to come up with a comprehensive description of his tic. We continued this way for the rest of the session. Throughout this process, Hiro's parents took notes while asking clarifying questions. I made certain to model using constructive praise for Hiro throughout the session and explained to the parents later that that was also an important element of treatment.

In our fifteenth session, when discussing how Hiro felt about having his parents in the room with us during treatment, he replied that it did not bother him and that he even thought it was good that they were learning so they could help him maintain and advance his tic management skills in India. We also used this session to discuss any anxieties Hiro might have been harboring about moving to India and specifically about explaining his tics to other people while there. Regarding the move to India itself, Hiro said he would prefer to stay in America, but understood that moving to India is what his family needed to do. However, he said he was determined to move back to the U.S. for college—he even had a specific college in California in mind where they have a video game design major. Regarding his tics, he said he was not worried about needing to explain what they were and that he was confident people would understand and not treat him differently because of it. This relaxed, confident, and determined attitude was emblematic of how Hiro approached most things in his life; this admirable strength was one that I tried to encourage and utilize throughout treatment.

The sixteenth session proceeded in much the same way as the fourteenth, as intended. In the seventeenth session_Hiro's parents ran the show for the first time in our therapy meeting. Before we began, I laid out a schedule for the session: Hiro's parents would run the session for 15 minutes, then we would break to discuss how it was going, and then they would continue with the session, after which we would end with another discussion of how that went. To help ease the process, I began with a brief overview of the whole HRT process. During the first break, I checked in with both Hiro and his parents to see how they felt the session was going. Hiro said his parents were "15% annoying," but that it was otherwise going well. Hiro's parents acknowledged that they moved from the awareness training piece to development of the CR too quickly and therefore did not create a powerful enough CR for the tic.

When Hiro said his parents were being annoying, I started thinking about what made our sessions together so successful. As I've noted before, a part of our success was the relationship we had developed and the fact that Hiro had the chance to talk about his favorite topic, video games, as much as he wanted in session while I participated. Therefore, I determined that a way to help Hiro's parents succeed in maintaining treatment while in India would be to make the time Hiro spent with his parents on HRT also a time where he was allowed to talk about whatever topic interested him while his parents would have to participate and show excitement for the topic. Throughout the session I interrupted only to remind Hiro's parents to praise Hiro for each step along the way. At the end of the session, I praised Hiro's parents' hard work in the session as well as their performance. Finally, I introduced the idea that HRT time with his parents could also be a time for Hiro to talk about video games with them. Hiro seemed excited by this prospect, while his parents were more hesitant. However, they acknowledged that it made sense and agreed to try.

Our final session together mimicked the previous session. However, a major difference was that Hiro's parents were more self-confident in their performance of HRT with Hiro. They did say that they did not "get the kid stuff" and had a hard time talking about video games with Hiro. Nonetheless, they did try, and when asked about it, Hiro said they had done a fairly good job. Also, due to the difficult nature of the tic we tried tackling in this session (a hiccup-like noise), no successful CR was developed in the session. I informed Hiro's parents that this was okay, that not every session would lead to the best possible CR, that this tic was particularly challenging (as vocal tics tend to be), that they did a great job in the session, and that they should not be discouraged.

In this last session together, we also reviewed the natural waxing and waning course of TS and how that could continue throughout the rest of his life; we also reviewed the fact that through treatment, Hiro now had the tools to help manage his tics, even if they increased in severity.

After having worked together for almost a year, saying goodbye was difficult for all of us. Hiro's parents expressed gratitude not just for the treatment, but for being a friend to Hiro and providing a safe place where he could test out ideas and grow as an individual. Hiro also expressed thanks for our time together and said he would miss me, but that he also hoped we could be friends in the future and we could together develop a computer app for CBIT for tics.

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I conveyed to Hiro and his family how much I had learnt from them, how wonderful it was to work with such a bright and creative boy as Hiro, and how impressed I was by Hiro's family and their dedication to helping Hiro, even when it meant stepping outside their own comfort zone. As our session came to a close, I also requested that we all be in touch in both three months and six months for follow-up booster sessions through Skype, to which Hiro and his parents responded with enthusiastic agreement. I expressed my gratitude that they would be willing to do that and said that I was looking forward to seeing how Hiro was doing in three months' time.

7. THERAPY MONITORING AND USE OF FEEDBACK INFORMATION

My treatment with Hiro was monitored on a weekly basis through notes, DVD recordings, and supervision. During supervision I would provide a recap of the previous session and my supervisor would have read my notes and often watched the DVD of the session. This led to great feedback from my supervisor, who has expertise in TS treatment, and helped me adapt the CBIT manual into a fluid and novel individualized treatment plan.

Two psychometric measures, the YGTSS and CY-BOCS, were also used to track Hiro's symptoms before and after treatment (see Tables 1 and 2); and another measure, Hiro's Subjective Units of Discomfort (SUDS) was used to monitor progress on a session-by-session basis (see Table 6). While the CY-BOCS and especially the YGTSS allowed me to measure treatment success overall, Hiro's SUDS ratings were instrumental for measuring our weekly progress and providing cues as to where to go next.

8. CONCLUDING EVALUATION OF THE THERAPY'S PROCESS AND OUTCOME

The Outcome of Hiro's Therapy

Hiro's treatment was rocky in the very beginning, but soon after I made the decision to alter treatment to more uniquely fit his presentation, success in treatment picked up significantly. Hiro's treatment success can be seen in both the quantitative and qualitative data that is described below. In general, Hiro learned to use HRT to manage all of the tics that we covered in our 18 sessions together. Additionally, Hiro learned to better recognize his tics and developed a more comfortable relationship with his symptoms. By the end, both Hiro and his parents were content with therapy, enjoyed coming, and walked away with the skills to maintain treatment gains and continue treatment without my direct involvement.

Quantitative Results

As shown in Table 1, when treatment first began, Hiro had a Total Tic Impairment Score of 26 on the *Yale Global Tic Severity Scale (YGTSS)*, which put him in the moderate level of tic severity, and an Impairment score of 10, indicating mild negative impact on general life satisfaction caused by tics. At the end of treatment, Hiro had a Total Tic Impairment Score of 14,

a 12-point drop, which put him in the mild range of impairment and an Impairment score of 0, indicating no negative impact of tics on Hiro's general life satisfaction.

At his three-month post-treatment session, Hiro's Total Tic Impairment score had decreased even more to an 11, maintaining his "mild" range of symptoms. At his six-month follow-up, Hiro's score was a 14, the same as it was when we completed treatment together. These scores demonstrate that Hiro and his family were able to continue utilizing his skills to manage his tics even after our treatment together was completed. This low score is even more impressive given the fact that Hiro had only recently endured significant changes (e.g., moving to India, starting a new school, etc.) and that changes often lead to an increase in tic symptomology. I believe this is a testament to how well Hiro learned tic management skills, to his own flexible and resilient personality, and to the informed support from his parents.

Qualitative Results

The following is a list of our original goals:

- -- Develop buy-in for treatment and belief from parents and from Hiro in the efficacy of treatment. ✓
- -- Discover and functionally deal with environmental conditions that may increase tic symptoms. x
- -- Develop relaxation skills to help manage tics. x
- -- Create awareness of tics when they occur. ✓
- -- Create awareness of premonitory urges before tics occur. ✓
- -- Develop Competing Responses (CR) to negate tics. ✓
- -- Decrease number and frequency of tics as recorded on the YGTSS and SUDS. ✓
- -- Teach skills so that Hiro can continue creating CRs for new tics post treatment. ✓

There are x's next to the treatment goals that we dropped and check marks next to the goals that were accomplished. As can be seen, the focuses of treatment after my reformulation of Hiro's case were on developing a relationship with Hiro and his parents, decreasing frequency and intensity of his tics, and teaching Hiro the skills to maintain treatment goals. By the end of treatment Hiro had made tremendous progress in terms of comfort with his tics, using his CR's, reducing his tics, and developing skills to maintain and continue his success.

It is instructive to look at Hiro's Tic Hierarchy (Table 6), and at his Subjective Units of Discomfort Scores (SUDS). For the tics that we had worked on, his SUDS were continuously decreasing, with only the occasional spike in specific or new tics. While this is not a direct indicator of fewer symptoms present, it does reflect how Hiro felt about each tic we worked on after we worked on it. Overall, and more specifically, Hiro's decreasing SUDS shows a level of comfort developed around his tics that were not there before. Additionally, this level of comfort can be gleaned in the three-and six-month follow-up sessions. Even though Hiro developed new tics, he was confident that he could keep developing new CR's that would eventually work. His

faith in his own abilities to manage his tics was something that was not present when we first began treatment together but grew over time and continued to grow even through follow-up sessions.

Discussion

Through my treatment with Hiro I learned a number of valuable lessons regarding both manualized treatment and myself as a practitioner. Regarding manualized treatment for TS, as applied to Hiro's case, I discovered the following: (a) Having only eight sessions was too brief for effective treatment; (b) there was nothing in the manual for garnering parental buy-in, which for Hiro's case was critical; (c) there were a very limited number of competing responses provided in the manual, which therefore required significantly greater creativity on my part to invent more; (d) there was no recourse in the manual for particularly challenging tics (e.g., Hiro's grunting tic); (e) the manual did not address Hiro's ADHD comorbidity problem; and, (f) for Hiro's case, there were treatment elements—like the Functional Based Assessment—that were unnecessary and proved unwieldy. This is not to say that the manual did not provide a very solid, basic outline for treatment of TS—it certainly did. But if I had not had the support, knowledge, and help of my supervisor, I would not have been able to provide the service that I was able to deliver.

As Fishman notes in his *The Case for Pragmatic Psychology*, "Past cases provide guidance for understanding and action in present cases because of those features of past cases which are similar to features of the present cases. In short, we 'learn from experience'" (1999, p. 192). Given the limited experience I had with clients with TS up to that point, my supervisor, with his significant amount of experience, essentially served as my database for dealing with the challenges of my case. Fishman's (1999; 2005) pragmatic case study model adequately captured the approach I took with my client, both because it represented the pragmatically inspired, individualized-case-formulation approach I took when deviating from the manual and because it provided a systematic means for evaluating the treatment from a "mixed methods" perspective, that is, from both a qualitative and quantitative perspective (Dattilio, Edwards, & Fishman, 2010).

Ultimately, between my supervisor's wide array of knowledge regarding treating TS and the tools of the pragmatic case study approach, I was able to treat my client successfully and to the best of my ability. In this sense, the pragmatic case study approach served as the solution for the challenges that the typical, manual-based approach could not. Thus, as mentioned above, if not for my supervisor, I would have been at a loss for how to best treat Hiro. For that reason, I support the notion put forth by Fishman (1999) for a database that could provide numerous case studies—such as this one—for whenever a clinician is faced with the challenges of treating someone for whom a manual, or the practitioner's general knowledge, is not enough.

Two final, but very important, lessons I learned from working with Hiro are the following. First, Hiro's own impressive imagination and ingenuity taught me the value of utilizing those same traits in therapy. From Hiro, I learned that the manual will not provide the answers we sometimes need and that, as therapists, we need to tap into our own reservoirs of ingenuity and skills to have the confidence to try new things. For me, this included using video-

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game-inspired visualizations to help Hiro with tough tics, and developing a parent-training model for HRT so that Hiro's family could continue treatment without me.

Finally, I think it can be argued that it was the therapeutic bond I developed with Hiro that contributed so much to our success together. I knew that every week, Hiro looked forward to coming to treatment. A part of that was his own budding sense of self-efficacy when it came to managing his tics. But a large part, I believe, was because I provided a space for him to talk enthusiastically and knowingly about his personal interests with an adult who attentively listened and respected these interests. This contributed so much to our collaborative work together, and I believe we could not have had the success we did without it.

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Table 1: Hiro's Scores on the Yale Global Tic Severity Scale (YGTSS)

Measure	Pre	Post	3-month follow up	6-month follow up
Total Tic Impairment Score*	26	14	11	14
Range of severity	Moderate	Mild	Mild	Mild
Impairment Score**	10	0	10	10
Range of severity	Minimal	None	Minimal	Minimal

^{*}Total YGTSS score ranges of severity groupings for patients are as follows: 0-9 (Minimal), 10-19 (Mild), 20-30 (Moderate), 31-50 (Marked)

<u>Table 2: Hiro's Scores on the Children's Yale-Brown Obsessive Compulsive Scale (CY-BOCS)</u>

Measure	Pre	Post	3-month follow up	6-month follow up	
Overall CY- BOCS Score*	18	0	0	0	
Range of severity	Moderate	Subclinical	Subclinical	Subclinical	

^{*}Total CY-BOCS score ranges of severity groupings for patients who have both obsessions and compulsions are as follows: 0-7 (subclinical), 8-15 (Mild), 16-23 (Moderate), 24-31 (Severe), 32-40 (Extreme)

^{**} Impairment Score ranges of severity groupings for patients are as follows: 0 (None), 10 (Minimal), 20 (Mild), 30 (Moderate), 40 (Marked), 50 (Severe)

Table 3: Hiro's Diagnosis at Beginning and End of Treatment

Axis		S		DSM-IV Diagnosis at End of Therapy		
Axis I				Tourette Syndrome Attention Deficit Hyperactivity Disorder— Combined Type		
Axis II	V71.09	No diagnosis	V71.09	No diagnosis		
Axis III		None		None		
Axis IV		None		None		
Axis V		GAF = 65		GAF = 80		

Table 4: Tic Hierarchy with Operational Definitions of Tics and Competing Responses, page 1

Tic	Operational Definition of Tic	Operational Definition of Premonitory Urge	Competing Response
Jaw clenching*	Clenching the back jaw muscles together, but without the teeth ever touching	Tenseness in the back of both of his jaw muscles at the same time	Holding his jaw closed.
Grunting	A deep, forceful exhalations of breathe from the mouth, coming rom deep in the chest. Occurs in multiple bouts	Tenseness in the center of chest	Diaphragmatic breathing
Head shaking side to side	A shaking of his head to the right first at about a 75-degree angle and then immediately to the left to a similar degree. This occurred at least 2 or 3 times each time	A tenseness that starts on the right side and then moves to the left side almost immediately afterwards.	Holding chin down to chest while tensing neck muscles
Head shaking up	Very quickly jerking head back at a 45 degree angle at least three or four times	Tenseness that begins in the back of his skull and moves down to the middle of the neck	Holding head back and tensing neck muscles
Jaw click	A sharp, forceful jerk of the jaw to the left and then the right. Occurs in bouts.	Tenseness in the jaw/cheek bone area, starting in the left jaw and then moving to the right as well	Holding jaw slightly open while tightening the jaw muscles.
Blinking	Sudden, fast eye blinking occurring in bouts of multiple blinks	Tense feeling in both the tops of the eye balls and in the bottom of the eye lids	Slow, controlled blinking with mental visualization for assistance
Snorting	A sharp, forceful exhale of breath from the nostrils that occurs in bouts.	Tenseness in the upper area of both nostrils	Diaphragmatic breathing with the intake happening through the nose and the exhale through the mouth

^{*} Tics are listed in order that they were worked on in therapy sessions

Table 4: Tic Hierarchy with Operational Definitions of Tics and Competing Responses, page 2

Tic	Operational Definition of Tic	Operational Definition of Premonitory Urge	Competing Response		
Head-shoulder shake*	Moving head down towards his left shoulder, while simultaneously moving his should up towards his ear; followed by same movement, but to the right. Occurred in bouts of at least 3 at a time.	Tenseness that starts in left shoulder, but immediately and quickly travels up to the left side of the head, around the ear. After beginning of tic is performed, tenseness then moves to the right shoulder and follows same patter as the left side. Tenseness moves back and forth between sides.	Pulling shoulders down and tensing neck muscles		
Eye rolling	Eyes move up in a forceful manner, then slowly rotate to the right and go around full circle	Tenseness that begins in the upper area of the eye ball and then travels around the eyeball and the eye socket from the right to the left	Squeezing the eyes shut tightly and holding, with visualization for assistance when needed		
"Mmm" noise	A "mmm" type noise that is caused by a forceful exhale of breathe through tightly held lips, with vocalizations from the throat at the same time	Tenseness in throat.	Tightening throat muscles while imaging a permeable wall, in the area where the premonitory urge is, that lets air in, but does not let the tic out		
Growling	A noise, similar to the sound of a dog growling, that comes from the throat and is an extended noise	Tenseness in the whole of the throat	Diaphragmatic breathing while tightening throat muscles and utilizing visualization when necessary to strengthen CR		
Hiccup	A short, sharp noise, that involved a very quick intake of breath through the nose and a tightening of the area where the throat and nose connect	Tenseness in the area where the nose and throat meet	Diaphragmatic breathing while simultaneously tightening the throat muscles		

^{*} Tics are listed in order that they were worked on in therapy sessions

Table 5: Homework Form: CBIT Practice Period Tracker*

Day	1	2	3	4	5	6

*Instructions: Practice your competing response 6x a day for X minutes at a time over a XX minute period of time. For every X minutes where competing response is used, Hiro will place a sticker on the chart to track progress. If 3 stickers are earned, XXX minutes of video game time is earned. If 6 stickers are earned, XXXX minutes of video game time is earned. Stickers earned that are below the 3 or 6 sticker mark can be added from the previous week to the next week.

Table 6: Hiro's Tic Hierarchy with SUD Scores

Tic	First time present	Second time present	Third time present	Session 16	Session 17	Session 18	3- month follow up	6- month follow up
Jaw Clenching*	9**	8	2	0	0	0	0	0
Grunting	10	5	10	0	0	0	0	0
Head shaking side to side	10	6	0	0	0	1	0	1
Head shaking up	6	4	0	1	0	1	0	0
Jaw click	10	0	1	0	0	0	0	0
Blinking	1	3	5	1	0	1	0	0
Snorting	5	0	0	1	0	0	0	0
Head- shoulder shake	5	1	0	0	0	1	0	0
Eye rolling	8	6	1	2	0	1	0	0
"Mmm" noise	10	0	0	1	2	0	0	0
Growling				4	0	2	0	0
Hiccup					7	2	2	4
Nose Wrinkling							3	0
Stomach Tensing							5	5
Wheezing								7

^{*} Tics are listed in order that they were worked on in therapy sessions

^{**} The numbers in the table cells indicate the SUDS score for a particular presentation of a particular tic.