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Behavioral Treatments for Tourette Syndrome

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Abstract

Tourette syndrome (TS) is a neuropsychiatric condition associated with substantial distress and functional impairment. Pharmacotherapy has traditionally been considered the first-line intervention for this condition, but there is strong evidence that behavior therapy is a comparably effective treatment option. Here we review empirically supported behavior therapy protocols for treating TS and the evidence associated with each. Potential mechanisms through which behavior therapy operates and concerns surrounding the utilization of behavioral interventions are also discussed.

Tourette Syndrome (TS) is a childhood-onset neurobiological disorder characterized by multiple motor and vocal tics that have persisted for at least one year since initial onset (American Psychiatric Association, 2013). In addition to the tics, most adults (Leckman, Walker, & Cohen, 1993) and many children (Banaschewski, Woerner, & Rothenberger, 2003) report that their tics are preceded by unpleasant somatic sensations (i.e., premonitory urges) that build up upon attempts to suppress tics and are temporarily alleviated by tic performance (Kwak, Vuong, & Jankovic, 2003).

Although the exact cause of TS is unknown, there is considerable evidence that tics result from structural and/or functional dysfunction within cortio-striato-thalamo-cortical (CSTC) circuitry (Singer & Minzer, 2003). Not surprisingly, pharmacotherapy has historically been considered the first-line intervention and has shown moderate degrees of effectiveness for reducing tics (Waldon, Hill, Termine, Balottin, & Cavanna, 2013; Shprecher & Kurlan, 2009). However, pharmacotherapy is not effective for all patients and can yield aversive short and long-term side effects that limit its use (Waldon et al., 2013). In light of these limitations, there has been a growing interest in non-drug (including behavioral) treatments for TS. Behavioral interventions are based on the assumption that tics have a biological origin, but their expression is influenced by contextual variables (Conelea & Woods, 2008a; Woods & Himle, 2004). Behavior therapy (BT) focuses on modifying environmental factors

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that influence tic severity and teaches patients specific skills they can use to better manage their tics (Himle, Woods, Piacentini, & Walkup, 2006).

The use of non-pharmacological treatment for TS may represent a shift in thinking for practitioners who typically treat with more conventional pharmacotherapies. However, the use of non-drug strategies to manage or treat the effects of a biological abnormality is not uncommon. For example, attention-deficit hyperactivity disorder and obsessive-compulsive disorder (OCD) have both been associated with functional and structural abnormalities within the CSTC system (Hale, Hariri, & McCracken, 2006; Saxena & Rauch, 2000) and are commonly comorbid with TS (Freeman et al., 2000). Both of these disorders are commonly treated with pharmacotherapy (Goldman, Genel, Bezman, & Slanetz, 1998; Greist & Jefferson, 1998). However, there is considerable evidence that BT or a combination of BT and medication is effective for managing these disorders (Abramowitz, 1997; Pelham, Wheeler, & Chronis, 1998). In fact, in OCD, there is compelling evidence that those who respond to BT (in the form of exposure and response prevention, EXRP) show metabolic changes in the CSTC system following treatment (Schwartz, Stoessl, Baxter, Martin, & Phelps, 1996).

Rationale for Use of Behavior Therapy to Manage TS

The rationale for using BT to treat TS is based on the idea that tics are *the product of* a specific neurobiological dysfunction, but are also *influenced* by environmental factors. Within the behavioral model, the terms "environmental" and "contextual" variables are often used interchangeably to describe events that occur before or after the tic that make the tic more or less likely to happen or otherwise influence the frequency and intensity of tics. These events can be internal (i.e., occurring within the body and experienced only by the person) or external (i.e., occurring outside of the patient). A behavioral model posits that tics become predictably influenced by these variables. This conceptualization allows for the fact that tics can essentially be "pushed" and "pulled" by environmental factors without insinuating they are volitional. The goal of behavior therapy is to modify these environmental factors in order to reduce tic severity.

Several variables occurring before tics (i.e., contextual variables) have been found to influence tic expression. Anxiety and social situations are frequently reported to exacerbate tics (Himle et al., in press; O'Connor, Brisebois, Brault, Robillard, & Loiselle, 2003; Silva, Munoz, Barickman, & Friedhoff, 1995). Other variables that have been reported to influence tics include talking about tics, engaging in sedentary activities, experiencing boredom, transitioning between activities, and being observed (Dufrene, Watson, Echevarria, & Weaver, 2013; Piacentini et al., 2006; Silva et al., 1995; Woods, Watson, Wolfe, Twohig, & Friman, 2001). However, the effects of contextual variables are idiosyncratic. For example, one study (O'Connor et al., 2003) found that 50% of an adult sample reported increased tics in social situations but 30% reported decreases in tics and 20% reported no change. In light of this, one goal of behavior therapy is to first conduct an individualized assessment of ticexacerbating factors and then eliminate or change tic-exacerbating variables, when possible, in order to decrease tic severity. Multiple studies have shown that environmental reactions to tics (e.g., escape from demanding tasks when tics occur, receiving attention when tics occur, receiving rewards for suppressing tics) can influence tic expression (Carr, Taylor, Wallander, & Reiss, 1994; Himle et al., in press; Watson & Sterling, 1998) and suppression (Himle & Woods, 2005; Woods & Himle, 2004). Another important reaction to tics that occurs within the patient's own body is the aforementioned reduction of the premonitory urge immediately after a tic is performed. From a behavioral perspective, the facts that premonitory urges are experienced as aversive, dissipate after the tic has been completed, and this dissipation is experienced as relief suggests that tics can be strengthened via negative reinforcement (i.e., due to tic-contingent reduction of the urge; Capriotti, Brandt, Turkel, Lee, & Woods, in press; Woods, Piacentini, Himle, & Chang, 2005). In other words, the tic is strengthened because it reduces the aversive urge. In line with this urge-reduction model, a second goal of BT for TS is to teach the patient strategies to refrain from ticcing in the presence of the urge, thereby breaking the negative reinforcement cycle.

History and Overview of Behavior Therapy for TS

Various behavioral therapies have been developed for TS over the past 40 years. Two approaches, habit reversal training (HRT; and its expanded version, Comprehensive Behavioral Intervention for Tics; CBIT) and exposure with response prevention (EXRP), have garnered the strongest empirical support and, therefore, will be reviewed here.

Habit Reversal Training.

Evidence for HRT as a promising treatment for tics first emerged in 1973 (Azrin & Nunn, 1973). The goal of HRT is to teach the patient to recognize the occurrence of each tic (or pre-tic warning sign) and engage in a response that directly interrupts performance of the tic. HRT has three primary components: awareness training (AT), competing response training (CRT), and social support.

AT teaches the patient to detect and discriminate the occurrence of each tic and/or pre-tic warning sign (e.g., a premonitory urge). This is done by having the patient signal to the therapist each time a target tic occurs during a session along with the therapist providing feedback to the patient when the patient misses a tic. AT continues until the patient can detect all, or nearly all, of his/her tics during the session, at which point the patient is taught to predict tic occurrence by detecting the earliest parts of the tic movement along with premonitory urges.

After AT is mastered, the patient is taught to interrupt the tic by engaging in a specific competing response (CR). A CR is any behavior that is (a) physically incompatible with the tic, (b) socially inconspicuous, and (c) easily performed by the patient across a variety of contexts and situations. The patient is instructed to use the CR whenever he/she detects a premonitory urge, notices the beginning of a tic, or completes a tic. The patient is asked to use this CR for one minute or until the premonitory urge dissipates, whichever is longer. For example, with an arm-jerking tic, the CR might be to gently press the elbow to the side of the body. A different CR is devised for each tic targeted in treatment.

Social support involves teaching a parent, spouse, or other support person to provide gentle prompts to use the CR when tics occur without the patient's noticing. The social support person is also taught to praise the patient for practicing the competing response exercises. It is important to note that the occurrence or nonoccurrence of tics is never praised or prompted, but rather the patient is reinforced for using his/her therapy skills.

Typically, HRT is taught over the course of 8–11 individual therapy sessions, each lasting 60–90 minutes. One tic is targeted each week and the patient is instructed to practice the HRT exercises between sessions.

Comprehensive Behavior Therapy for Tics.

More recently, researchers have developed an enhanced behavioral treatment package called Comprehensive Behavioral Intervention for Tics (CBIT). CBIT is a manualized treatment that adds several therapeutic tools to the standard HRT protocol (Woods et al., 2008). Most importantly, an individualized, function-based assessment is conducted to systematically evaluate idiosyncratic contextual factors that may be exacerbating the patient's tics. Based on this assessment, modifications are introduced to eliminate or modify tic-exacerbating factors in order to create a more "tic-neutral environment." Traditional HRT skills are then taught, along with relaxation skills such as progressive muscle relaxation and diaphragmatic breathing. For pediatric patients, CBIT also employs a behavioral reward system to facilitate participation in in-session therapeutic activities and between-session practice of CBIT skills.

Exposure and Response Prevention.

Based on the similarity between the urge-reduction model of TS (O'Connor, Gareau, Borgeat, 1997; Woods et al., 2005) and the anxiety-reduction model of OCD (Mataix-Cols, Rosario-Campos, & Leckman, 2005), specific BT procedures shown to be effective for treating OCD have been modified for application to TS. In particular, exposure and response prevention (EXRP), the gold standard BT for OCD, exposes patients to anxiety-evoking stimuli (exposure) while simultaneously refraining from performing compulsions to reduce anxiety (response prevention). Although the exact mechanism by which EXRP is effective is unclear, it appears that repeated exposure to anxiety-provoking stimuli, coupled with response prevention, reduces anxiety through the processes of anxiety habituation and/or inhibitory learning (Craske et al., 2008). Applied to TS, the goal of EXRP is to expose the patient to aversive premonitory urges while he/she refrains from ticcing for an extended period of time (e.g., 90 min). It should be noted that this approach differs substantially from simply suppressing tics for very brief periods (e.g., 30 s), as patients may attempt to do outside of a behavioral intervention. The rationale is that, through repeated practice with prolonged tic suppression, the patient will habituate to the urge and tic severity will reduce over time. EXRP sessions involve instructing individuals to suppress their tics (but without providing specific strategies for doing so) while focusing on, and rating, any bodily sensations and premonitory urges that they experience (Verdellen, Keijsers, Cath, & Hoogduin, 2006). When a tic occurs, the therapist acts as a "coach" by encouraging the patient to improve his/her suppression. Patients are also encouraged to bring personal "ticexacerbating" objects into the session in order to elicit premonitory urges during the suppression sessions.

Review of Treatment Outcomes for Behavior Therapy for TS

As a broad category of treatments, BT for TS has been shown to be effective in multiple studies using both randomized controlled trial and small-N controlled experimental designs. As many recent reviews have evaluated the published work on BT for TS (Bate, Malouff, Thorsteinsson, & Bhullar, 2011; Capriotti & Woods, 2013; Cook & Blacher, 2007; Himle et al., 2006), only the general findings are discussed here.

Across numerous studies conducted since 1973, research clearly demonstrates the efficacy of HRT over wait-list, massed practice, relaxation training alone and supportive therapy (Cook & Blacher, 2007; Himle et al., 2006; Piacentini et al., 2010; Woods, Conelea, & Himle, 2006). Recently, the largest studies of BT for TS to date were completed (Piacentini et al., 2010; Wilhelm et al., 2012). In the pediatric Comprehensive Behavioral Treatment for Tics (CBIT) trial, 126 children with TS and chronic tic disorders were randomly assigned to receive either 10 sessions of CBIT or 10 sessions of psychoeducation and supportive psychotherapy. Results showed that CBIT was more effective than psychoeducation and supportive therapy at reducing tic severity, with individuals assigned to the CBIT group demonstrating tic reduction comparable to what has been reported in trials of atypical antipsychotics (Scahill et al., 2013). In addition, the vast majority (i.e., 87%) of those who responded to CBIT at the end of 10 weeks (53% of CBIT recipients were acute phase responders), maintained their gains at 6-months post-treatment. Furthermore, CBIT responders demonstrated improvements in social functioning and decreases in anxiety and disruptive behavior at six-month follow-up, indicating that BT focused on tics may have beneficial effect in other important, but untargeted, clinical domains (Woods et al., 2011).

In a parallel adult trial (N=122), CBIT also demonstrated outcomes superior to a psychoeducation and supportive therapy intervention. 38% of adult patients showed a clinically significant response to CBIT (compared to 6% in the psychoeducation and supportive therapy condition), and 80% of these responders maintained their gains at 6-month follow up. Across both the pediatric and adult studies, attrition and withdrawal rates were low, and treatment-related adverse events were rare and benign. Taken together, the results of these trials suggest that CBIT is a safe, efficacious, and long-lasting treatment for TS in both children and adults.

Although HRT/CBIT has the strongest empirical support (among the behavior therapies) for reducing tics, EXRP has also shown to be effective in preliminary studies. In the only clinical trial of EXRP conducted to date, Verdellen and colleagues (2004) compared EXRP to HRT and found the two treatments were equally effective at reducing tics. However, in this study the EXRP treatment group received double the in-session treatment time as the HRT group (24 hrs vs. 10 hrs), thereby confounding the conclusion that EXRP is as effective as HRT/CBIT. Although EXRP appears to be a promising treatment for TS, more research is needed to further evaluate its efficacy (Cook & Blacher, 2007).

Overall, a number of well-controlled, randomized allocation trials support the safety, efficacy, and durability of behavior therapy for treating TS in both children and adults. In fact, two of these trials (i.e., the CBIT studies) enrolled more participants than almost every

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other TS treatment study to date, including both pharmacological and behavioral intervention trials. Based on this evidence, recently published European (Verdellen et al., 2011) and Canadian (Steeves et al., 2012) guidelines for treating TS have suggested that behavior therapy be used as a "first line" intervention with patients seeking treatment for the first time. Additionally, HRT/CBIT qualifies as a "well-established" treatment for TS under the guidelines described by the American Psychological Association's Division 12 Task Force on the Promotion and Dissemination of Psychological Procedures (Chambless et al., 1998; Cook & Blacher, 2007).

Potential Mechanisms of Change in Behavior Therapy for TS

Although the efficacy of BT for reducing tics is well established, the mechanism by which it is effective remains unclear. In their original treatment development work, Azrin and Nunn (1973) proposed that because tics are performed at such a high frequency, the tic becomes habitual and automatic and, over time, leads to over-strengthening of the muscles involved in tic performance. In line with this conceptualization, they proposed that HRT works because performance of the CR strengthens muscles antagonistic to those involved in the tic (Azrin & Nunn, 1973). However, the explanation has been challenged by at least one study showing that contingent application of a CR that is topographically dissimilar to the tic (i.e., does not involve the contraction of antagonistic muscles) can effectively reduce tics in some cases (Sharenow, Fuqua, & Miltenberger, 1989, Woods et al., 1996).

Another hypothesized behavioral mechanism is based on the assumption that tics are maintained, in part, because they function to alleviate premonitory urges (Capriotti et al., in press; Woods et al., 2005). If this is correct, HRT may work in one of two ways. First, it is possible that HRT simply trains a new behavioral tendency to replace the tic in the presence of the urge. If the competing behavior is reinforced more frequently or more strongly than the tic, then the patient's response tendency will be allocated to the competing response. Another possibility is that HRT may interrupt or delay the tic thereby allowing a patient to habituate to the aversive premonitory urge. Habituation is said to occur when intense or unusual sensory stimulation is first experienced as aversive, but becomes less noticeable and less bothersome with the passage of time. With repeated practice, habituation occurs more rapidly and the initial exposure to the stimulus produces a less aversive reaction in the patient. By performing a competing response, patients learn to engage in behaviors other than the tic when the urge is present, thereby allowing habituation to occur. With continued practice, the urge becomes less frequent and intense, and the tic occurs less frequently and/or forcefully. This view draws empirical support from a study in which three of four persons receiving HRT reported a reduction in premonitory urge as well as a reduction in tic frequency across EXRP sessions (Hoogduin, Verdellen, & Cath, 1997) and from a replication of these findings for patients receiving ERP in a larger scale trial (Verdellen et al., 2004).

Naturally, behavioral researchers tend to focus on behavioral rather than biological levels of analysis. As such, the bulk of research on the mechanisms of change has focused on behavioral processes. However, it is also important to consider the neurocognitive processes that underlie successful nonpharmacological tic management. There is evidence to suggest

that children and adolescents with TS have increased cognitive control of bodily movements compared to non-TS matched controls (Mueller, Jackson, Dhalla, Datsopoulos, & Hollis, 2006). This finding presents an apparent paradox, as tics are involved *impaired* control of movement. However, it is possible that, over time, some individuals with TS develop enhanced top-down control of movements through repeated efforts to suppress their tics. This may account for the fact that most individuals with tics experience significant remission of symptoms in adulthood (Leckman et al., 1998).

Similarly, neuroimaging research that has examined the role of the basal ganglia in tic suppression suggests that the regulation of movement occurring within this region is impaired in persons with TS. The neural mechanisms underlying tic suppression are unclear and to date only one imaging study has been conducted. Peterson and colleagues (1998) collected fMRI data from 22 adults with TS during brief (40-second) epochs of rest and tic suppression and found that, compared to when subjects were at rest (no suppression), neural activity during suppression decreased in the basal ganglia and increased in the somatosensory and attention-related cortical regions. The study also found a strong inverse correlation between the general severity of TS and reductions in basal ganglia activity during suppression. The authors concluded that suppression was an attention-related task and that failure to suppress was related to an inability to down-regulate subcortical (i.e., basal ganglia) neural activity. Accordingly, BT may serve to increase activation in these regions during voluntary tic suppression, thereby augmenting the brain's ability to control unwanted movements.

Factors Influencing the Utilization of Behavior Therapy for TS

It is often difficult to promote utilization of effective nonpharmacological treatments by clinicians (Jankovic & Kurlan, 2011; Scahill et al., 2013; Woods, Conelea, & Himle, 2010). The development of evidence-based behavioral therapies has grown considerably in the past quarter century, and as a results a noticeable gap has emerged between what is considered "best practice" by the research literature, and those treatments that are utilized in the clinical community (Weissman, et al., 2006). Since many, if not most, people with TS seek initial consultation and treatment from neurologists and other physicians, research psychologists concerned with the dissemination of BT for TS must not only promote the uptake of BT amongst professionals in their own discipline (i.e., psychology, psychiatry, social work), but also devise strategies to disseminate knowledge about treatments to professionals outside of the mental health field. With the growing amount of evidence on the efficacy of BT for TS, the medical community has increasingly begun to consider behavior therapy as a viable treatment option. However, surveys of practicing psychologists and physicians who treat TS have found that relatively few reported knowing how to implement BT for TS (Marcks, Woods, Teng, & Twohig, 2004).

In addition to the relatively low availability of professionals trained in BT for TS, research also suggests that there may be hesitancy among some professionals to recommend BT for TS due to beliefs about the negative effects of such treatments. One concern is the belief that treating some tics may cause an increase in non-targeted tics or cause new tics to emerge (i.e., symptom substitution; Nurnberger & Hingtgen, 1973). To date, symptom substitution

of this sort has not been seen in empirical studies. At least one study designed to directly test this hypothesis failed to reliably demonstrate symptom substitution (Woods, Twohig, Flessner, & Roloff, 2003). Additionally, the decreases in overal tic severity documented in many trials of BT for TS argue against this phenomenon, as symptom substitution would have resulted in compensatory increases in non-treated or novel tics, resulting in equivalent pre- and post-treatment levels of total tic severity. A second persisting concern is that BT may lead to a "tic rebound effect," such that tics increase after periods of voluntary suppression (Marcks et al., 2004). This implies that BT might teach patients to temporarily stop tics, but that this suppression leads to high-frequency bouts after efforts to suppress tics cease. While a rebound effect has been observed in one study following medication withdrawal (Leckman et al., 1986), several studies have failed to show immediate rebound following prolonged periods of behavioral tic suppression (Specht et al., 2013; Verdellen, Hoogduin, & Keijsers, 2007; Woods et al., 2008). In addition, only 1 child (1.6%) in the pediatric CBIT trial reported tic worsening during active behavior therapy compared to 4 children (6.2%) in the control condition (Piacentini et al., 2010). Collectively, these studies suggest that a suppression-related tic exacerbation is not of sufficient concern to preclude the use of BT.

A third concern regarding the use of BT for TS is that practicing a competing response may strain a patient's attentional resources, thereby diminishing the patient's ability to participate in other cognitively demanding tasks like schoolwork. To date, one study tested this hypothesis by examining performance on an attention-demanding task during periods of ticsuppression compared to non-suppression baseline periods (Conelea & Woods, 2008b). The study found that tic suppression had a mild detrimental effect on task performance, thus providing limited support for concerns about the attentional demands of competing response use. However, participants in this study had not received suggestions for specific competing responses or tic suppression strategies taught in BT for TS, nor did they practice reinforced suppression repeatedly, as patients do in BT. As the practice of any new skill is likely to detract from cognitive performance on concurrent tasks until the newly-acquired task is practiced to proficiency, it is quite possible that any learning or attentional deficits seen during suppression for these participants (who had received no prior BT) would have waned throughout BT. Future research is needed to examine these potential detrimental effects of tic suppression on attention as patients throughout the course of BT. Such a study would provide a more thorough and valid examination of attention-detracting effects of tic suppression in the context of BT for TS. Additionally, it should be noted that tics and associated premonitory urges distract from focused attention in and of themselves (for a first-person account, see Kane, 1994). Therefore, any distracting effect of suppression associated with BT must be weighed against the reduced distraction from tics with successful treatment.

A final tic-related concern is that BT requires patients to pay increased attention to his/her tics, which might have a tic-worsening effect. Indeed, research suggests that both anxiety and talking about tics can increase tic frequency (O'Connor et al., 2003; Silva et al., 1995; Storch et al., 2007). However, research has demonstrated that clinician-recommended exercises that involve paying increased attention to tics (either via self-monitoring or awareness training) have either no effect, or a somewhat beneficial effect on symptom

severity (Cook & Blacher, 2007; Ollendick, 1981, Woods, Miltenberger, & Lumley, 1996; Wiskow & Klatt, 2013). Additionally, studies showing that increases in tic frequency can be caused by talking about tics also indicate that these effects quickly disappear once the topic of conversation shifts away from tics (Dufrene et al., 2013; Woods et al., 2001). This argues against the concern that talking about tics in therapy would lead to global or enduring increases in tic severity.

Finally, there is some concern that the BT will inadvertently lead to "psychologization" of TS which may in turn increase in stigma surrounding TS (Jankovic & Kurlan, 2011). This is an understandable concern given how some in the professional psychological community conceptualized TS in the past (e.g., the psychoanalytic movement of the early 1900s; see Germiniani, Miranda, Ferenczy, Munhoz, & Teive, 2012; Kushner, 1999). Medical professionals and advocacy groups have made great strides in educating the general population about TS and decreasing stigma surrounding the disorder by emphasizing its place as a medical condition. Some are concerned that behavioral interventions may create potentially harmful misconceptions in the general public. To some, the efficacy of HRT could be misconstrued as evidence that tics are voluntary or that persons with TS can stop their tics simply by "trying hard enough." Although such a view is inconsistent with a behavioral understanding of TS, it is the responsibility of behavior therapy practitioners to accurately present this treatment to consumer to the public. Done effectively, behavior therapy appears to be a useful and desirable treatment option for persons with TS.

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