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journal homepage: www.elsevier.com/locate/jocrdBehavioral treatments for Tourette syndrome [☆]Matthew R. Capriotti ^a, Michael B. Himle ^b, Douglas W. Woods ^{c,*}^a University of Wisconsin-Milwaukee, Milwaukee, WI, United States^b University of Utah, Salt Lake City, UT, United States^c Department of Psychology, Texas A&M University, College Station, TX 77845, United States

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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.

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1. Introduction

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, 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, Dat 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 cortico-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 (Shprecher & Kurlan, 2009; Waldon, Hill, Termine, Balottin, & Cavanna, 2013). 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 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, 2000; 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) show metabolic changes in the CSTC system following treatment (Schwartz, Stoessel, Baxter, Martin, & Phelps, 1996).

2. 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

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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., antecedent 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; Himle 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 tic-exacerbating 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, 1996; 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 tics in the presence of the urge, thereby breaking the negative reinforcement cycle.

3. 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.

3.1. 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 1 min 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 min. One tic is targeted each week and the patient is instructed to practice the HRT exercises between sessions.

3.2. 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., 2008a). 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.

3.3. 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

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