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Behavior Therapy for Tic Disorders: An Evidenced-based Review and New Directions for Treatment Research

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Abstract

Behavior therapy is an evidenced-based intervention with moderate-to-large treatment effects in reducing tic symptom severity among individuals with Persistent Tic Disorders (PTDs) and Tourette’s Disorder (TD). This review describes the behavioral treatment model for tics, delineates components of evidence-based behavior therapy for tics, and reviews the empirical support among randomized controlled trials for individuals with PTDs or TD. Additionally, this review discusses several challenges confronting the behavioral management of tics, highlights emerging solutions for these challenges, and outlines new directions for treatment research.

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Human and Animal Rights and Informed Consent

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Keywords

Tourette Disorder; Persistent Tic Disorder; PTDs; Treatment Outcome; Comprehensive Behavioral Intervention For Tics; Habit Reversal Training; Tourette's Syndrome; developmental disorders

INTRODUCTION

Tics are sudden rapid non-rhythmic motor movements or vocalizations that can be simple (rapid, meaningless) or complex (purposeful, orchestrated) in nature [1]. Tics are relatively common among school-aged youth for brief periods of time, but often do not continue beyond six months [2]. A chronic or Persistent Tic Disorder (PTD) is characterized by the presence of either a single or multiple motor or vocal tic(s), but not both, that persisted longer than a year; with a diagnosis of Tourette's Disorder being conferred when both motor and phonic tics are present (although not necessarily concurrently) for longer than a year [1]. Persistent Tic Disorders and Tourette's Disorder (collectively referred to as PTDs henceforth) affect approximately 0.4–1.6% of youth [3, 4]. For youth with PTD, symptoms typically onset around six years of age [5], and exhibit a fluctuating course with peaks in symptom severity that stabilize over a period of weeks [6]. For the majority of youth, tics reach their greatest severity in adolescence—increasing in number, type, and frequency—but subside in early adulthood in many cases [5, 7]. Tic symptoms show minimal difference between youth and adults with PTD [8], with the most common bothersome tics including eye blinking, head jerks, sniffing, throat clearing, and other complex motor tics [9]. In addition to tics, individuals with PTD typically present with co-occurring psychiatric disorders [e.g., anxiety disorders, attention deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD)] [10–13], mood and behavioral problems (e.g., disruptive behaviors, rage attacks, anger problems, suicidal thoughts and/or behaviors) [14–16], and social difficulties (e.g., peer victimization, social deficits, low self-concept) [17–22]. *Also see Hanks et al. [22] in this issue.* Tics and co-occurring problems can cause individuals with PTD to experience significant impairment [21, 23, 24], and a poor quality of life [25, 26]. Thus, effective treatments are needed for individuals with PTD to efficiently manage their tics and co-occurring symptoms.

Historically, tic symptom severity has been managed using psychotropic medications, such as antipsychotic agents and/or alpha-2 agonists [27]. A meta-analysis of five randomized controlled trials (RCTs) of antipsychotic medications identified a significant but moderate reduction in tic severity relative to placebo [effect size = 0.58], with no significant difference between medications types [28]. Additionally, a meta-analysis of six RCTs of alpha-2 agonist medications identified a statistically significant albeit small reduction in tic severity relative to placebo (effect size = 0.31) that was moderate when limited to RCTs in which individuals had both PTD and ADHD (effect size = 0.68) [28]. Despite their efficacy, these medications are often accompanied by side effects that can limit long term use [27]. Moreover, while medication management significantly reduces tic severity, some troublesome tics may remain. Thus, individuals with PTD have to confront and cope with tics even when receiving evidence-based pharmacotherapy.

In addition to pharmacotherapy, behavior therapy has demonstrated success in reducing tic severity for individuals with PTD in RCTs [29–36], and can serve as a stand-alone intervention or augmentation strategy to existing pharmacotherapy. Indeed, professional organizations have recommended behavior therapy as the first-line intervention for youth with PTD who have mild-to-moderate tic severity [37–39]. This paper provides an evidence-based review of behavior therapy to manage tic symptoms for individuals with PTDs. This paper also discusses the challenges confronting the behavioral management of tics, highlights emerging solutions for these challenges, and outlines new directions for treatment research.

BEHAVIOR THERAPY

Behavioral interventions for tics have existed for several decades, with early case reports having been published in the 1960's and 1970's [40, 41]. Although initial case reports suggested tics to be nervous habits [40], more recent conceptualizations of behavioral interventions acknowledge the neurobiological basis of the condition [42, 43]. Multiple types of behavioral interventions have been evaluated in RCTs for the treatment of individuals with PTD [e.g., habit reversal training (HRT), mass negative practice (MNP), awareness training (AT), exposure response prevention (ERP)] [38], but only HRT and its successor the Comprehensive Behavioral Intervention for Tics (CBIT) have consistently demonstrated efficacy in RCTs and meta-analyses [44].

Habit reversal training (HRT) is a multiple component intervention that can include psychoeducation, awareness training, competing response training, generalization training, self-monitoring, relaxation training, behavioral rewards, motivational procedures, and social support [45]. The therapeutic components of HRT are detailed in Table 1. Although it can include multiple components, the core therapeutic skills of HRT are awareness training, competing response training, and social support [46]. The CBIT utilizes these core HRT components along with relaxation training and behavioral rewards, and incorporates functional assessments and function-based interventions to mitigate daily life factors that can exacerbate tic symptoms [43]. Table 1 describes the functional assessment and interventions in greater detail.

Behavioral Treatment Model

Behavioral therapies like HRT and CBIT are based on the belief that tics have a neurobiological basis, but that both external and internal cues can serve as antecedents that influence tic symptoms [43]. External cues can include specific activities (e.g., playing sports, musical instruments, doing paperwork) and environmental/situational situations (e.g., returning home, specific family members), with internal cues commonly including premonitory urges and/or internal mood states (e.g., anxiety). Individuals with PTD experience corresponding consequences associated with external and internal factors that can subsequently reinforce tic symptoms. For example, a premonitory urge is an internal sensation that individuals describe as an aversive “urge”, “feeling”, “impulse” and/or “pressure” that often precedes a tic [47, 48]. Individuals with PTD report that the performance of the tic and/or multiple tics alleviates the distressing premonitory urge [47]. Consequently, the tic becomes negatively reinforced because it produces a reduction in the

aversive sensation of the premonitory urge. In HRT and CBIT, individuals learn to identify antecedents to tic symptoms and implement competing responses that break the negative reinforcement cycle. This allows for individuals to habituate to internal aversive triggers like premonitory urges and modification of consequences for external factors (e.g., avoidance of homework or social situations due to increased tics).

Empirical Evidence

Multiple case reports have highlighted the benefit of behavior therapy for individuals as young as five [49] and old as 75 years of age [50]. Eight published RCTs having evaluated the efficacy of behavior therapy relative to either waitlist conditions [30, 34] or an active comparison condition in youth and adults [e.g., MNP, ERP, psychoeducation and supportive therapy (PST)] [29, 31–33, 35, 36]. Azrin and colleagues (1980) compared HRT and MNP using a one-to-two session treatment protocol in 22 youth and adults [29]. Mass negative practice involves the voluntary rapid and repeated performance of tics for a defined period of time that is interspersed with brief periods of rest [38]. Azrin and colleagues found that participants receiving HRT exhibited a 92% reduction in tic frequency relative to a 33% reduction in tic frequency among the MNP condition four weeks post-treatment [29].

Azrin and Peterson (1990) compared HRT to a waitlist condition in 10 youth and adults with PTD, with participants in the HRT condition receiving an average of seven sessions over a four month period [30]. Using direct observation tic frequency counts conducted within the clinic, Azrin and Peterson found that tic frequency reduced by an average of 93% in the HRT condition compared to an average 14% reduction waitlist group [30]. O'Connor and colleagues (2001) compared a 13 session HRT treatment package to a waitlist control condition among 69 individuals with PTD [34]. O'Connor and colleagues observed that participants receiving HRT exhibited greater reductions in video observations of tic frequency and severity compared to the waitlist control condition [34].

In 2003, Wilhelm and colleagues compared the efficacy of HRT relative to PST in a 14 session treatment protocol for 32 adults with PTD [35]. Wilhelm and colleagues found a large within-group effect size for reduction of tic severity in the HRT group ($d = 1.50$), with no meaningful within-group effect observed in the PST group ($d = -0.03$) [35]. Verdellen and colleagues (2004) compared 10 sessions of HRT to 12 sessions of ERP in 43 youth and adults with PTD [33]. Exposure response prevention is a behavioral intervention similar to HRT and CBIT, but differs in its approach to managing tic symptoms. In ERP, participants are exposed to sensations (e.g., premonitory urges) and stimuli that elicit tics for a prolonged period of time and practice suppressing/resisting the tic (rather than engaging in a competing response) [33, 38]. Through repeated exposure and resistance, individuals are believed to habituate to the sensations and stimuli that elicit the tics. Similar to HRT and CBIT, this is believed to discontinue the negative reinforcement cycle wherein the individual experiences relief in aversive sensation (i.e., the premonitory urge) following the tic. The ERP group exhibited an averaged 8.6 point reduction on the YGTSS compared to an average 4.4 point reduction on the YGTSS in the HRT group. Although these group differences approached statistical significance ($p = 0.06$), the noted difference in therapeutic contact between treatment conditions confounds these findings (12 120-minute ERP sessions versus 10 60-

minute HRT sessions) [33]. In 2006, Deckersbach and colleagues compared a 12 session treatment protocol of HRT to PST in 30 adults with PTD [36]. Although no significant difference in tic severity was observed at pre-treatment, Deckersbach and colleagues found that participants in the HRT condition had lower tic severity at the mid-treatment and post-treatment assessments compared to the PST condition [36].

Piacentini and colleagues (2010) conducted a large-scale multi-center RCT of 8 sessions of CBIT compared to 8 sessions of PST over 10 weeks in 126 youth with PTD [31]. Piacentini and colleagues found that CBIT was associated with a 7.6 point decrease on the YGTSS compared to a 3.5 point decrease in the PST condition on the YGTSS ($p < 0.001$, effect size = 0.68)[31]. On the Clinical Global Impression of Improvement (CGI-I) [51], 52.5% of participants in the CBIT condition and 18.5% of participants in the PST condition exhibited a positive response to treatment [31]. Therapeutic gains observed during the acute treatment period were found to maintain after six months, with no adverse consequences [31, 52]. In 2012, Wilhelm and colleagues (2012) conducted a parallel large-scale multi-center RCT of 8 sessions of CBIT compared to 8 sessions of PST over 10 weeks in 122 older adolescents and adults with PTD [32]. Wilhelm and colleagues found that participants in the CBIT group had a 25.8% decrease on the YGTSS compared to an 11.5% decrease in the PST group ($p < 0.001$, effect size = 0.57) [32]. On the CGI-I, 38.1% of participants in the CBIT condition and 6.8% in the PST condition exhibited a positive response to treatment [32]. Treatment gains were observed up to six months after acute treatment [32]. Follow-up analyses across these two large CBIT trials identified that CBIT was efficacious across all identified tic symptoms clusters [8]. An individualized examination of treatment response of specific bothersome tics and tic characteristics in these CBIT trials found that specific tics were more likely to improve and remit with CBIT relative to PST, with broader tic characteristic analyses revealing that tics with a premonitory urge exhibited greater improvement and remission with CBIT compared to PST [9].

In 2014, McGuire and colleagues conducted a random effects meta-analysis of behavior therapy RCTs and examined moderators of treatment effects [44]. Behavior therapy had a moderate-to-large effect (effect size = 0.67–0.94) relative to comparison conditions [44]. Additionally, participants receiving behavior therapy were more likely to exhibit a treatment response relative to comparison conditions (odds ratio = 5.77), with a number needed-to-treat of three [44]. Moderator analysis identified that greater mean participant age and average number of therapy sessions was associated with larger treatment effects, whereas an increased percentage of participants with co-occurring ADHD was associated with smaller treatment effects [44].

CHALLENGES CONFRONTING BEHAVIOR THERAPY FOR TICS

Although behavior therapy has considerable empirical support for reducing tic symptom severity, several challenges exist to its wide spread use and implementation as a first-line intervention for individuals with PTD. This section outlines these challenges and highlights potential emerging solutions.

Clinician Concerns Regarding Possible Negative Consequences of Behavior Therapy

Some clinicians have expressed concerns that behavior therapy may have unintended negative consequences for individuals with PTSD. These include concerns that focusing attention on tics will make them worse, tic suppression produces “rebound” effects, and that behavior therapy may result in symptom substitution [53]. Although these concerns are well-intentioned, there exists empirical evidence that contradicts many of these misconceptions. First, there is a belief among healthcare providers that either talking about tics [54] or focusing attention on tics will make them worse [55]. While tic-related discussions and direct attention to tics can momentarily increase tic frequency [56, 57], awareness training that includes focused attention on individual tics and self-monitoring of tics has been shown to actually produce modest reductions in tics [46]. Second, a considerable percentage of healthcare providers believe that tic suppression strategies may inadvertently produce increased tic frequency above initial levels referred to as a “rebound effect” [54]. The best evidence to empirically evaluate this concern comes from Verdellen and colleagues (2007) who compared tic frequency in a 15 minute video observations before and after ERP therapy sessions in which participants practiced tic suppression [58]. After each one of the therapy sessions, the mean post-session tic counts were lower than baseline tic frequency demonstrating that tic suppression activities did not result in worsening of tic symptoms [58]. Third, some clinicians have raised concerns that behavior therapy can result in symptom substitution based on anecdotal case reports [54, 59, 60]. Symptom substitution is a long-standing concern that dichotomizes psychodynamic and behavioral therapies [61]. Symptom substitution refers to the belief that the behavioral treatment of one tic may result in the appearance of a new tic and/or an increase in severity of other non-targeted tics. For example, if an arm tic was treated with behavior therapy, the concern may be that the patient may develop a new tic (e.g., leg kick) in response to suppressing their original tic and/or experience a worsening in severity of non-targeted tics. Despite possible evidence in among case reports [59, 60], systematic empirical evaluations have found no evidence of symptom substitution in response to behavior therapy for tics for individuals with PTSD [62].

Limited Accessibility and Availability

While behavior therapy has demonstrated its efficacy for managing tic severity in structured RCTs, there are a limited number of clinicians trained in evidence-based behavioral practice [63]. This poses a considerable challenge for managing tics with behavior therapy in community settings as there is likely availability and access to trained treatment providers [63, 64]. As a result, many individuals with PTSD may be prohibited from either receiving behavior therapy and/or experience long-wait periods prior to treatment initiation. Several innovative solutions have been explored to address this challenge. First, the Tourette Association of America (previously the Tourette Syndrome Association) and the U.S. Centers for Disease Control and Prevention have collaborated to develop training opportunities to increase the number of clinician’s trained in evidence-based behavior therapy [64]. Second, several studies have examined novel treatment delivery modalities to increase access and availability of behavior therapy. These innovations include intensive behavior therapy treatment protocols [65, 66], and telemedicine approaches to delivering behavior therapy [67–70]. Third, given the limited number of mental health professionals trained in behavior therapy, other professionals such as physical therapists have been

exploring the possibility of conducting behavior therapy for PTD. Rowe, Yuen, and Due (2013) conducted an open-label trial of 8 sessions of CBIT for PTD conducted by physical therapists in 30 youth with PTD [71]. Rowe and colleagues found that youth exhibited a reduced number of tics and had improved scores on the Parent Tic Questionnaire [71, 72]. Although these collective approaches have shown considerable promise, future research is needed to continue to explore approaches to increasing the availability and accessibility of behavior therapy for individuals with PTD.

Limited Therapeutic Response to Behavior Therapy

Although many individuals receiving behavior therapy experience significant reductions in tic severity, these behavioral interventions do not often result in complete tic remission. Indeed, a positive response to treatment among individuals with PTD corresponds with a 25–35% reduction in tic severity on the YGTSS [73, 74]. Thus, there is a clear need to enhance therapeutic outcomes with behavioral interventions. Several possible approaches to enhance therapeutic outcomes may prove useful for individuals with PTD based on the limited available evidence. First, the addition of adjunctive therapeutic components to existing evidence-based behavioral interventions may prove beneficial. Franklin and colleagues (2011) [75] found that adding acceptance and commitment therapy (ACT) to evidence-based behavior therapy yielded additional benefit relative to evidence-based behavior therapy alone in a small pilot study of youth with PTD [75]. Second, the augmentation of behavioral interventions with pharmacotherapy may result in enhanced therapeutic outcomes. Lyon and colleagues (2010) examined whether methylphenidate (MPH) enhanced youth's ability to suppress tics relative to placebo in a RCT [76]. Although Lyon found minimal difference in youth's ability to enhance tic suppression between groups [76], it is important to note that tic suppression is distinctly different from evidence-based behavior therapies. Thus, the combination of MPH and behavior therapy may still prove useful as co-occurring ADHD was found to attenuate treatment effects in a recent meta-analysis [44]. Finally, the use of cognitive enhancers may serve as another possibility to augment therapeutic outcomes with behavior therapy. Broadly, cognitive enhancers are compounds that are believed to augment psychosocial interventions to produce expedited and/or enhanced therapeutic benefit (e.g., d-cycloserine). Cognitive enhancers have demonstrated some promise among related disorders like OCD in enhancing therapeutic outcome and expediting treatment gains [77]. Despite the possible potential of all three augmentation approaches, considerable future research is needed to investigate evidence-based augmentation strategies for behavior therapy.

Limited treatments targeting other problems encountered by individuals with PTD

The predominant focus of pharmacological and behavioral interventions for individuals with PTD has been reduction in tic symptom severity. This treatment approach is predicated on the assumption that tic severity is predominantly responsible for the distress, impairment, and poor quality of life experienced by individuals with PTD. While many individuals experience significant improvement with pharmacological or behavioral interventions, a considerable percentage of individuals continue to experience distress and impairment from tics and associated problems (e.g., social problems, social deficits, peer victimization, poor self-perception). Moreover, as complete symptom remission is infrequent with either

intervention, individuals have to learn effective coping skills to manage daily challenges associated with tics. Taken together, there is a clear need for evidence-based interventions to reduce impairment, improve the quality of life, and develop effective coping strategies among individuals with PTD. Although recognized as an important aspect of treatment in evidence-based practice parameters [37], there have only been two studies that have evaluated treatments to address these concerns. First, Storch and colleagues (2012) developed a modular cognitive behavioral therapy (CBT) called “Living with Tics” (LWT) that aimed to promote resiliency and coping skills to manage the psychosocial consequences of tics among youth with PTD [78]. The LWT intervention was evaluated in an open-label case series of 8 youth with PTD. Youth exhibited significant reductions in tic-related impairment and tic severity on the YGTSS, as well as experienced improved self-concept and quality of life [78]. On the CGI-I, 6 of the 8 youth (75%) were considered treatment responders. Second, McGuire and colleagues (2014) extended the initial findings by Storch and colleagues (2012) by incorporating additional modules into the LWT intervention and evaluated its efficacy compared to a waitlist condition in a RCT of 24 youth with PTD [78, 79]. Youth in the LWT ($n = 12$) group exhibited significantly reduced tic-related impairment on the YGTSS and improved quality of life relative to the waitlist condition [79]. Ten youth (83%) in the LWT group were considered treatment responders on the CGI-I compared to four youth in the waitlist condition (33%) [79]. Treatment gains in the LWT group were maintained at a one-month follow-up assessment [79]. While the modular LWT intervention has demonstrated benefit as a stand-alone treatment, it can also serve as important therapeutic complements to either behavior therapy or pharmacological interventions. Although demonstrating considerable promise, additional research is needed to develop and evaluate further interventions focused on reducing impairment and promoting quality of life among individuals with PTD--especially among adults.

CONCLUSION

In summary, behavior therapy is an established evidence-based intervention for the management of tics characteristic of PTD. Behavior therapy has demonstrated its efficacy across multiple RCTs, and produces treatment effects comparable to those observed with antipsychotic medications. Indeed, professional organizations recommend behavior therapy as a first-line intervention for individuals with mild-to-moderate tic severity [37], with recent research highlighting the counterintuitive benefit of behavior therapy for tics with premonitory urges that tend to have greater severity [9]. While behavior therapy offers the promise of significant tic reduction in the majority of cases, some challenges remain that confront the regular utilization of behavior therapy as a first-line intervention for individuals with PTD. Additionally, further efforts are needed to enhance therapeutic outcomes for individuals who fail to exhibit a clinically meaningful response to behavior therapy, and refine related interventions to promote coping skills to manage the adverse psychosocial consequences of tics. Taken together, these challenges highlight the importance of ongoing behavioral treatment research to improve the therapeutic outcomes and quality of life for individuals with PTD.

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Table 1

Components of Habit Reversal Training (HRT) and Comprehensive Behavioral Interventions for Tics (CBIT)

Therapeutic Component	Description
Psychoeducation	Provides an overview of tic disorders. Information typically includes: diagnostic criteria; clinical features; developmental course; co-occurring conditions and difficulties; etiological factors; prevalence to reduce stigma; and correct any misconceptions.
Awareness Training	Clinician collaborates with the patient to develop operational definition of tic and identify early tic signals (e.g., premonitory urge, early tic movements). Once defined, patient practices recognizing tic occurrence in session and/or engages in simulations of tic to enhance awareness.
Competing Response Training	After awareness training, the clinician assists the patient in developing an alternative behavior (called a competing response) that is physically incompatible with the tic. The behavior should be able to be used for 1 minute or until the premonitory urge subsides, and must be socially discrete. The patient practices using the identified competing response in session contingent upon the early tic signals, the actual tic, or a simulation of the tic.
Generalization Training	As the patient demonstrates appropriate awareness and implementation of competing response for a tic, the clinician asks the patient to generalize these skills into daily activities. In session, this may include having the patient practice using the competing response in a normal conversation. Outside of session, the patient is asked to practice using the competing response at home and/or in other relevant situations/settings.
Self-Monitoring	A clinician instructs the patient and/or social support person (e.g., parent, significant other) to monitor targeted tics during a period of time when the tics are most likely to occur. Monitoring includes identification of the activity, the specific tic, the number of times the tic was observed for each monitoring period. Typical weekly monitoring can include 3 to 4 periods for up to 30 minutes. When possible, patients and social support person should monitor tics concurrently.
Relaxation Training	The clinician instructs the patient in the performance of diaphragmatic breathing, and progressive muscle relaxation (PMR: tensing and relaxing individual muscle groups) in session. These activities can relieve stress and reduce muscle tension, which may serve as antecedents or consequences of tics. After the patient develops competency in these skills, the patient is asked to identify times and places to practice outside of session.
Function-based Assessment	The clinician interviews the patient and/or parent to identify any <i>antecedents</i> (settings, situations, or mood states occurring before tics) and <i>consequences</i> (situations, or reactions following tics) associated with tic worsening for each individual tic. A self-report form can also be used to gather data regarding antecedents and consequences to tics to facilitate the interview.
Function-based Intervention	After completion of the function-based assessment, the clinician uses the information to collaborate with the patient and/or parent to develop interventions that reduce the impact of antecedents and consequences on tics. The goal of the interventions is to reduce tics and tic-related functional impairment.
Behavioral Rewards	In collaboration with the patient and/or parent, the clinician develops a developmentally-appropriate behavioral reward system to provide motivation for treatment engagement. Afterward, the clinician administered behavioral rewards at each session (e.g., points) contingent upon the performance of specified behaviors (e.g., session attendance, homework completion). The patient can then exchange these session-based rewards for rewards outside of session provided by the parent.
Review of Tic Difficulties	The clinician and patient collaboratively develop a list of bothersome aspects of having tics. The patient rates the degree of distress for each identified difficulty on a 0 to 10 scale. The patient revises and re-evaluates the difficulties identified on this list throughout treatment.
Relapse Prevention	The clinician reviews the patient's progress in treatment and interventions for bothersome tics targeted in treatment (e.g., function-based interventions, competing responses). The clinician discusses with the patient strategies for managing new tics that may develop using behavioral strategies in case of worsening tic symptoms. The clinician also discusses ongoing use of social support and ongoing implementation of skills learned in treatment outside of therapy.