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Behavioral Therapy for Tourette Syndrome and Chronic Tic Disorders

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Abstract

Purpose of review: To summarize behavioral interventions for the treatment of primary tic disorders.

Recent findings: Although tics were attributed to a disordered weak volition, the shift towards neurobiological models of tic disorders also transformed nonpharmacologic treatment practices. Current international guidelines recommend habit reversal training, comprehensive behavioral intervention, and exposure and response prevention as first-line therapies for tics. Appropriate patient selection, including age and presence of comorbidities, are salient clinical features that merit consideration. Evidence for further behavioral interventions is also presented. **Summary:** Currently recommended behavioral interventions view tics as habitual responses that may be further strengthened through negative reinforcement. Although availability and costs related to these interventions may limit their effect, Internet-based and telehealth approaches may facilitate wide accessibility. Novel nonpharmacologic treatments that take different approaches, such as autonomic modulation or attention-based interventions, may also hold therapeutic promise. *Neurol Clin Pract* 2017;7:148-156



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Tics are sudden, rapid, recurrent, nonrhythmic motor movements or vocalizations¹ that are typically not context-related. Tics characteristically wax and wane in frequency and severity and are commonly preceded by an impending (interoceptive) sensation termed premonitory urge.² Although tics may present in many disorders, they are commonly encountered in primary tic disorders, such as Tourette syndrome (TS).

A fundamental characteristic of tics is that they can be voluntarily suppressed. This quality often aids the diagnostic distinction of tics from other hyperkinesias, such as chorea or myoclonus. Importantly, it is also the basis of most behavioral treatments in TS and other tic disorders, aiming to reinforce tic inhibitory capacity. On the other hand, this unusual phenomenon, the ability to exert voluntary control over involuntary movements, led to puzzling psychological

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Cognitive behavioral treatment focuses on changing the way a patient evaluates his or her actions and expectations in high-risk situations related to frequent “ticcing” behaviors.

hypotheses about the pathophysiology of tics and TS. This, coupled by commonly associated neuropsychiatric comorbidities, such as attention-deficit/hyperactivity disorder (ADHD), obsessive-compulsive disorders (OCD), and affective and personality disorders,³ further reinforced the moralizing view that tics originate in unusual characters and bodies that “lack willpower.”⁴

Clearly, research in tic disorders has come a long way. Aided by early developments in neuropharmacology and followed by thorough clinical characterization studies and findings from neuropathology, neurophysiology, and neuroimaging, we now know that tics and their associations are the result of aberrant brain development, particularly within the cortico-striato-thalamo-cortical circuits.⁵

Propelled by the change in the overall perception of tic disorders and most importantly by the systematic implementation of nonpharmacologic therapeutic strategies for tics, we provide a concise synthesis of behavioral treatments of the past, present, and future. Our aim is to offer a practical overview of the different therapeutic approaches and to reflect current guideline recommendations.^{6–8}

The past

Psychoanalytic and supportive psychotherapy Up to the beginning of the second half of the 20th century, psychoanalytic psychotherapy was the predominant treatment approach for tic disorders (the following information is based on Kushner⁴). Indeed, this was the result of early psychological conceptions including Jean Marc Gaspard Itard’s proposal that involuntary tics resulted from an underdeveloped will and could be strengthened by moral treatments (chapter 2), or Meige and Feindel’s position that tics were uncorrected infantile habits in people with inherited weakness of the will (chapters 4 and 12). The first psychoanalytic view of tics was presented by Sandor Ferenczi in 1921, who concluded that tics were “stereotyped equivalents of Onanism” and resulted from repressed masturbatory desires (chapter 4). This view later considered that tics only manifest in susceptible children who had experienced severe, repressed familial psychological conflicts and had difficulties with “ego synthesis” due to overprotective parents (chapter 7). Free association and dream interpretation were implemented as therapeutic strategies (chapter 7) and aimed at unmasking the presumed underlying unconscious mechanisms leading to tics. Other behavioral treatments included the separation of children from their parents, in order to eliminate the pathologic familial environment, and intensive exercise routines to channel motor urges in other more positive ways (chapter 7). Breathing exercises, hypnosis (chapters 3 and 12), and “mirror drills” in order to aid patients in becoming conscious about their tics and learning to control them were also applied.

Most aforementioned psychoanalytic-based therapies failed to lead to sustainable tic reduction (chapter 12), as initial misattributions of treatment success or relapse were later found to be no different from the naturally occurring waxing and waning of tics. Also, such therapeutic approaches were shown to be inferior to other nonpharmacologic treatments such as habit reversal therapy (HRT).^{9,10} Nevertheless, conversation-based supportive psychotherapy is often considered nowadays, as it focuses on problem solving and reflecting about current life issues.¹¹ Indeed, such an approach could improve the overall well-being, including life

Supplemental Data

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satisfaction and psychosocial functioning, of people with tics.¹⁰ However, supportive psychotherapy has yielded negative results with regard to tics themselves, compared to other treatment strategies such as HRT.^{9,12} Indeed, based on clinical practice rather than on empirical evidence, the European guideline and current reviews on behavioral therapies in tic disorders include recommendations on supportive psychotherapy or other psychosocial interventions (e.g., psychoeducation^{6,11,13}) as a component of overall management.

Massed (negative) practice (MP) MP is based on the idea that tics are learned maladaptive habits that can be replaced by more flexible or adaptive habits.¹⁴ It involves the accurate, effortful, repetitive, rapid, voluntary production of the individual's tics over a certain amount of time (e.g., 15–30 minutes) until tic frequency decreases or the tic disappears. This is considered to be the result of conditioned inhibition or fatigue, in turn leading to suppression/abandoning the tic.^{6,15} Numerous case reports (referenced in table e-1 at Neurology.org/cp) revealed mixed results, and there is only one randomized control study to date that examined the effects of MP compared to HRT in 22 participants with tics (16–62 years). MP led to a significant reduction of tics (30%), but was overall less effective than HRT (97%).¹⁶ Based on these data, MP appears to have only limited therapeutic value and is therefore not recommended in existing guidelines.^{6–8}

Other approaches In la cure de sommeil (sleep treatment), patients were suddenly awakened from their sleep, rendering them weak to “conscious resistance,” in order to be immediately subjected to behavioral interventions (chapter 9).⁴ In aversive therapy, patients had to repeat the word “shock” following every single tic (chapter 8). Assertiveness training considered “internal and external aspects” leading to tic manifestation and taught patients to respond more assertively to their external environment (table e-1).

The present

HRT, comprehensive behavioral intervention (CBIT), and exposure and response prevention (ERP) HRT is currently the most extensively researched and widely applied behavioral treatment for tic disorders.^{11,17,18} The original multicomponent HRT was based on the idea that tics resemble habits and was introduced by Azrin and Nunn¹⁹ in 1973. HRT comprises 9 different techniques within 4 discrete domains: (1) awareness training techniques in order to aid patients in detecting situations and early warning signs related to impeding tic behaviors; (2) competing response technique²⁰ instructing individuals to perform a movement that is incompatible with the tic using antagonistic muscles; (3) motivation techniques focusing on social and environmental consequences of tics; and (4) generalization, which represents a complete rehearsal of the HRT protocol and its transference to everyday life. Since Azrin and Nunn's¹⁹ original article, HRT has been further simplified, paring down the number of domains to a combined awareness and competing response training without losing its efficacy.²¹ HRT is also the main component of CBIT,^{12,22} which also involves psychoeducation, relaxation training, and functional intervention (identifying events and situations that worsen tic severity as well as developing strategies to manage them). To date, 14 randomized controlled trials (RCT) on HRT/CBIT in children and adults with tic disorders have been conducted (complete studies referenced in table e-2; for reviews, see references 6, 11, and 23), showing significant improvements in tic severity with treatment.

ERP is a different approach that was originally developed by Victor Meyer²⁴ in 1966 to treat obsessive-compulsive symptoms. It is based on the view that tics are conditioned responses to premonitory urges,^{25,26} and that weakening the association between impeding tics and premonitory urges will lead not only to meaningful tic reduction but also to reduction of urges.²⁵ During ERP, patients are asked to tolerate premonitory urges for longer periods of time (exposure; usually up to 2-hour sessions), while resisting tic execution (response prevention), resulting in an overall reduction of tic frequency outside therapeutic sessions.²⁶ To date, only 1 RCT has been published showing comparable efficacy between ERP and HRT.²⁷

Importantly, ERP targets many different tics at once, whereas HRT focuses on a limited number of tics at a time.²⁷ However, a common criticism of this study has been that the time allotted and number of treatment sessions for ERP in this trial was longer than for HRT, perhaps resulting in a greater training effect.

Habit reversal therapy/CBIT and ERP are currently recommended as first-line behavioral treatments in the existing guidelines and practice parameter,^{6–8} as well as in pragmatic treatment recommendations for children with chronic tic disorders and TS.¹³

Other approaches *Cognitive-behavioral treatment (CBT)* One RCT compared the outcome of a behavioral treatment group performing HRT based on a 13-step program introduced by Azrin and Peterson²⁸ in 1988 with that of a cognitive treatment group in which maladaptive expectations were replaced by reconstructed, more realistic anticipations, and found comparable results for both treatments.²⁹ In 2 further studies, the CBT program included components of HRT and was shown to be more effective than a waitlist control condition³⁰ and equally effective in patients with and without anti-tic medication.³¹ As CBT includes components of HRT and in the absence of well-controlled large RCTs, its added therapeutic value remains unclear.⁶

Contingency management (CM) and function-based interventions (FBI) CM considers the instances related to tic emergence and their influence on consequent tics, e.g., tic reduction when tics are ignored or tic reinforcement if tics become the focus of attention.^{6,11} Several case studies used CM (table e-1) and showed positive effects on tics. However, CM was mainly used in combination with other therapeutic approaches, largely as part of HRT,⁹ and, therefore, assessing its therapeutic efficacy on its own is difficult. FBI are a specialized form of CM that aim to identify and modify specific factors in the individual's environment that enhance or reduce tics. To our knowledge, there are only 2 case reports successfully using FBI for tic reduction so far (table e-1). The European guidelines of behavioral treatments recommend CM/FBI as second-line therapy or add-on behavioral treatment.⁶

Relaxation training (RT) The observation that stress, anger, anxiety, and excitement lead to increased tic burden led to the hypothesis that stress reduction might also decrease tics.³² RT is a hypernym for a number of strategies/techniques to reduce levels of stress, anger, and anxiety, thereby also leading to decreased muscular tension. It includes deep breathing techniques, progressive muscle relaxation training, and motor imagery, and is mostly applied as part of a multicomponent treatment (table e-1). In mono-component, case-control studies (n = 6), RT has been shown effective on its own (32% tic reduction), but less effective compared to other behavioral approaches such as HRT (55%) or self-monitoring (SM; 44%, also see next paragraph³³). Moreover, the effects of RT have been reported to be of limited duration.³⁴ Currently, RT is recommended by the European guidelines as second-line or add-on behavioral treatment.⁶

SM, awareness training, and mindfulness-based stress reduction SM attempts to facilitate tic awareness by teaching patients to recognize and systematically record their tics. There is a limited number of reported cases on SM (table e-1), and it is noteworthy that SM is often used as a component of HRT.

Mindfulness-based stress reduction is a method designed to promote the awareness of one's bodily sensations, emotions, and thoughts while increasing attentional control and decreasing physiologic arousal.³⁵ A single feasibility study on mindfulness-based tic treatment in 18 individuals with TS (aged 16–67 years) over a period of 8 weeks revealed that 58.8% of participants were treatment responders and showed significant improvement in tic severity and tic-related impairment that was maintained at 1-month follow-up. There was no control group in this pilot study.

Tension reduction is another method to teach patients to identify moments of inner tension leading to tic performance, and to transform the tic into more socially acceptable motor responses.³⁶

Nonpharmacologic treatments rely heavily on the experience and practice of individual therapists and hence they may not be available in certain parts of the world.

Also promising are studies based on self-report assessments and observational studies that demonstrate a beneficial effect of music and physical exercise in patients with tics.^{37–39} Finally, participation in a 10-week program of a novel cognitive psychophysiological model for the management of tics has also shown positive results in children and adults with tic disorders and TS.⁴⁰

There are currently no clear recommendations for the practice of these therapeutic methods in any of the existing guidelines for the treatment of tics.

The future

Internet-based training and telehealth approaches Current technologies allow delivering CBIT via videoconferences. A study evaluating the efficacy of this approach compared to face-to-face sessions showed comparable effect sizes on tic reduction between the 2 studied groups (n = 18, 10 videoconference⁴¹). Internet-delivered CBIT is also the basis of an ongoing study protocol,⁴² comparing the efficacy of online CBIT (n = 72) vs psychoeducation alone (n = 72) vs face-to-face CBIT (n = 16) over a 2-year period. Further, a waitlist-controlled voiceover Internet protocol for delivering CBIT pilot study was performed in a small group (n = 20) of children and adolescents with chronic tic disorders, showing promising results.⁴³ Importantly, the implementation and combination of aforementioned methods may facilitate wide accessibility to behavioral treatments and reduce related costs, thereby potentially increasing treatment frequency, factors that are often limiting for current treatment practices (also see below).

Autonomic modulation and neurofeedback Tic frequency is often linked to a patient's emotional state and higher baseline heart rate and blood pressure have been reported in patients with TS compared with healthy controls (for a review, see reference 44). Autonomic modulation might therefore have an influence on CNS function and tic occurrence.⁴⁴ Neurofeedback is a noninvasive method of autonomic modulation that is based on operant conditioning. It displays real-time physiologic processes such as brain activity, heart rate, or electrodermal activity and by using relevant feedback directs patients how to modulate their physiologic responses.⁴⁴ One study⁴⁵ reported significantly lower tics during relaxation biofeedback compared to arousal biofeedback using electrodermal activity biofeedback in 15 adults with TS, and more recently the combination of biofeedback with motor imagery (the participant imagines an action without performing it) showed promising results in adolescents with medication-refractory TS.⁴⁶ However, results from other studies yielded inconclusive or negative results (table e-1) and hence more research is required on this topic. Other autonomic modulation methods—for example, vagus nerve stimulation (pulsatile electrical vagus nerve stimulation to directly modulate afferent interoceptive signals)—may also hold therapeutic promise,⁴⁴ but the clinical relevance of such methods remains unclear.

Scaling between voluntary and involuntary behaviors A final comment on future treatment approaches is related to whether young patients should be focusing on their tics and urges, as in current therapeutic practice, or whether their attention should be shifted away from involuntary movements, i.e., reinforcing perception of voluntary motor signals. This idea hinges on similarities between tic suppression and thought suppression studies⁴⁷ and thereby on putative mechanisms of negative reinforcement: increasing awareness of tics during early stages of tic manifestation (e.g., parents and teachers asking children to stop ticcing) could

lead to a permanent increase in later stages in life. In the absence of any controlled longitudinal data, this hypothesis remains speculative but is often discussed by TS experts. A single study in 23 adolescents with TS examined factors related to awareness of voluntary actions and revealed that the capacity to inhibit tics was associated with the capacity to perceive voluntary motor signals.⁴⁸ This highlights that a possible by-product of improved action awareness could be an increase in voluntary control over tics. Whether data demonstrating the attenuating effects of physical exercise on tics^{37,38} are also in line with this suggestion remains unclear.

A practical approach to behavioral treatments of the present for tics

Evidence in the current practice of nonpharmacologic treatments for tics in primary tic disorders and TS supports the use of HRT/CBIT and ERP as first-line interventions, with preference to HRT/CBIT if available. Importantly, elements of HRT and ERP can be combined to manage different tics in the same patient. However, there are relevant limitations for both HRT/CBIT and ERP that include age of patients, tic severity, profile of comorbidities, treatment availability, cost, and insurance coverage. Indeed, younger children might have difficulties understanding the content of behavioral treatments, particularly if they have not developed a clear awareness of premonitory urges.¹³ Also, patients with multiple and severe tics may have difficulties in focusing on single urge/tic associations required in HRT/CBIT or may not be able to suppress their tics for more than the few seconds required in ERP. Although the effect of various psychiatric comorbidities on treatment response to HRT/CBIT/ERP remains unclear, it is likely that any symptoms that influence the ability of the patient to comply with required treatment procedures are likely to negatively affect the potential for treatment gains. Moreover, motivation can also often be a limiting factor, as many young patients and contrary to their environment might not find their tics distressing enough.

Nevertheless, published treatment manuals describing the step-by-step implementation of CBIT²² or ERP⁴⁷ can help therapists learn to conduct evidence-based interventions. Likewise, Tourette organizations, such as the Tourette Association of America (TAA) in the United States, have developed a formal training course that allows potential CBIT therapists to be trained and receive follow-up consultation as they begin to implement the intervention. Furthermore, a host of online educational resources about CBIT have been made available on the TAA Web site through their partnership with the US Centers for Disease Control and Prevention (tourette.org/research-medical/cbit-overview/).

In addition, therapy costs may not be covered by standard health care and hence access to behavioral treatments may be further limited. Of note, the treatment of tics alone may not suffice for many patients, and priority should be also given to the treatment of comorbidities such as ADHD or OCD, which in themselves may further complicate treatment of tics.¹³ In such cases, a combination of both behavioral and pharmacologic treatments may be required.¹³

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Take-home points

- The management of tic disorders involves both behavioral and pharmacologic treatment practices.
- Current international guidelines recommend habit reversal training, comprehensive behavioral intervention, and exposure and response prevention as first-line behavioral treatments for tics.
- Treatment practices should be tailored to the individual needs of patients, taking into consideration age, tic severity, and presence of neuropsychiatric comorbidities such as attention-deficit/hyperactivity disorder and obsessive-compulsive disorder.
- Internet-based and telehealth approaches may facilitate wide accessibility to behavioral treatments.
- Novel nonpharmacologic treatments that focus on modulation of autonomic symptoms or attention-based interventions may also hold therapeutic promise.

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