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Research Paper

Efficacy of Parent-Delivered, Home-Based Therapy for Tics

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ABSTRACT

Background: Although behavioral therapy is an effective approach to reduce tics in children and adults, there is an insufficient availability and accessibility of behavioral therapy in the community.**Objective:** The goal of the study was to test the clinical efficacy of home-based, parent-provided behavioral therapy in children with Tourette syndrome aged seven to 13 years.**Method:** An instructional habit reversal training-based video and guide was developed for use by parents. Eligible families, in this 10-week study, were enrolled in either a home-based therapy (DVD) group (received disk and written instructions) or an in-person therapist group (had scheduled visits with the therapist). Outcome scales included the Yale Global Tic Severity Scale, both the total Tic Severity Score and total Global Severity Score, and the parent report of Clinical Global Impressions of Improvement.**Results:** Forty-four children (mean age = 10.21 ± 1.69 years) were enrolled into either the DVD (n = 33) or in-person therapist (n = 11) groups. Eighteen completed the study—eight in the DVD and 10 in the in-person therapist group. Outcome measures showed significant reductions in Yale Global Tic Severity Scale change ratios: mean improvement on the Tic Severity Score was DVD 32.4% (P < 0.001) and in-person therapist 26.6% (P = 0.01); and for the Global Severity Score, DVD 33.7% (P < 0.001) and in-person therapist 26.7% (P < 0.001).**Conclusions:** Home-based, parent-administered habit reversal training behavioral therapy is efficacious for reducing tics in children. Telephone contacts early in the DVD treatment course might reduce the number of dropouts.

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Introduction

Tourette syndrome (TS) is a complex heterogeneous disorder characterized by the presence of chronic motor and vocal (phonic) tics and coexisting neuropsychiatric problems.¹⁻³ Tics are quick, rapid, sudden, recurrent, nonrhythmic motor movements or vocalizations that wax and wane; are exacerbated by stress, anxiety, and fatigue; and are often preceded by a premonitory urge. Tics may be mild and unobtrusive or very frequent, complex, forceful, intrusive, and self-injurious. The presence of tics can lead to psychosocial issues, bullying, physical discomfort, disruptions in the academic/workplace, and a poor quality of life. TS occurs worldwide with an estimated prevalence of about 1%.⁴ There is no cure

for tics, and therapy, either behavioral or pharmacologic, is indicated for functionally disabling tics.

There are three major behavioral interventions for tics including habit reversal training (HRT), comprehensive behavioral intervention for tics (CBIT), and exposure and response prevention. HRT contains awareness training (recognizing the urge or tic occurrence), competing response training (performing a socially discrete behavior that is physically incompatible with the tic and can be maintained for up to one minute), and social support (a home-based provider to deliver developmentally appropriate praise for correct implementation).⁵⁻¹¹ CBIT contains the three core components of HRT, plus relaxation and psychoeducational training, and function-based interventions to address internal and external factors that influence tic expression.^{10,12-15} Exposure and response prevention is a methodology that attempts to extend an individual's natural ability to suppress a tic and focuses on premonitory urges.^{16,17} Each of these methodologies minimize tic

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expression based on randomized, blinded, controlled trials. Meta-analyses have documented their efficacy, and expert opinion practice guidelines have recommended behavioral intervention as the initial therapeutic treatment for tics.^{15,18-20}

Despite the proven efficacy of behavioral therapy for TS, there is a longstanding dearth of trained therapists to assist affected individuals. For example, a survey of urban-based mental/behavioral health care providers found that less than 10% reported knowing how to implement HRT.²¹ Another study reported that treatment was unavailable to most families because of a deficiency of trained clinicians.²² When behavior therapy is accessible, there are long wait lists to see practitioners, families have to travel long distances, and costs may be prohibitive. To address some of these issues, recent experimental approaches have included group therapy,^{11,23} an intensive individual treatment format,²⁴ and the use of telemedicine over the internet.^{21,25-28}

The goal of the present study was (1) to develop a parent-administered, HRT-informed, instructional video and guide and (2) to test its efficacy compared with therapist-administered training. A similar approach has been beneficial in another childhood movement disorder, primary complex motor stereotypies.^{29,30} It was hypothesized that the HRT-based, parent-led, home-administered intervention would be an efficacious method to treat tics. We further speculated that the success of treatment would relate to the extent of involvement and enthusiasm of parents and the presence of co-occurring problems in the participants.

Methods

The Johns Hopkins Hospital Institutional Review Board approved this 10-week protocol. Individuals with TS, aged seven to 13 years, were recruited from either the Johns Hopkins Outpatient Pediatric Neurology Movement Disorder Clinic (HSS, Director) or a linked webpage. Written informed consent was obtained before beginning any study procedure.

The study coordinator (L.F.) screened all families over the telephone. Information gathered at initial screening included (1) a pediatric medical and developmental history questionnaire, (2) a comprehensive history of tics and prior therapies, (3) Yale Global Tic Severity Scale (YGTSS),³¹ (4) and screens for commonly occurring conditions such as anxiety/depression (Revised Child Anxiety and Depression Scale [RCADS])³² and attention-deficit/hyperactivity disorder (ADHD) (ADHD Rating Scale-5).³³ For individuals not previously evaluated at Johns Hopkins, a video documenting the patient's movements was required. A neurologist (H.S.S.) and neuropsychologist (E.M.M.) independently reviewed all information and qualified participants (based on inclusion/exclusion criteria—described below) were scheduled for a baseline evaluation.

Participants

Patients were included if they (1) met Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition, criteria for Tourette disorder; (2) were between ages seven and 13 years; (3) had observable moderate or greater tics, achieving a minimum score of greater than 20³⁴ for total tic severity on the YGTSS, and (4) had tic symptoms that were severe enough to warrant therapy. Concurrent use of medications for tic suppression, ADHD, or obsessive-compulsive disorder (OCD) was permitted, if the subject had been on a stable dose for more than four weeks and agreed to maintain a constant dosage throughout the study.

Exclusion criteria included (1) secondary tics, (2) significant medical illness or a chronic neurological condition (i.e., seizure disorder, developmental neurological conditions, acquired brain

injuries), and (3) current diagnosis of major depression, generalized anxiety disorder, separation anxiety disorder, autism spectrum disorder, intellectual disability, anorexia nervosa, bulimia, substance abuse, or psychotic symptoms (based on clinical evaluation). Individuals with significant OCD, not controlled by medication, and those with a prior history of behavioral treatment for tics were also excluded.

Educational HRT-based video and instructional guide, for parent home use

An educational HRT-informed training video was produced with the assistance of the Audiovisual Department at the Johns Hopkins Hospital. This instructional DVD (42 minutes) contains a 10-minute educational session on tics (HSS) and instructions provided by a licensed psychologist (M.S.) and informative vignettes for the remaining time. This parent-directed HRT video parallels the CBIT manual³⁵ with several exceptions. Specifically, the video incorporates psychoeducation, awareness training, and competing response (CR) training for tics. The video also emphasizes the differential reinforcement of incompatible behaviors (i.e., CRs) and other adaptive behaviors occurring in the absence of tics (i.e., reading a book without ticcing).

However, in contrast to the CBIT manual, this DVD did not specifically address or target several components including contingency management, inconvenience review, self-monitoring, and relaxation training. Second, whereas CBIT emphasizes the therapist-patient relationship, this parent-directed HRT video emphasizes the parent-child relationship. Parents received instructions to implement awareness training during the first week and to add CR training and differential reinforcement of incompatible/other in week two and beyond, adapting the reinforcement to the individual progress of the child.

Awareness training

Parent and child (1) choose the most bothersome tic, (2) develop an agreed operational definition of the target tic, and, (3) practice awareness training.

Competing response training

This component provides the child with an alternate motor behavior (i.e., CR) that he or she can enact in lieu of the tic. The CR is (1) physically incompatible with the tic, (2) discrete, (3) not painful, (4) generally compatible with functional behavior, (5) sustainable, and (6) transportable. The parent then instructs the child to practice the developed CR on command with adjustments and alterations made as necessary (e.g., controlled blinking for eye blinking tics). Following selection of the CR, the parent is taught to test the child's ability to reliably demonstrate the CR immediately and correctly when prompted.

Differential reinforcement of incompatible/other

This section includes parent education on (1) reinforcing the patient (verbal praise and a token to be exchanged for a reward) for independently using the CR in lieu of the tic; (2) encouraging the child to use the CR if he or she misses an opportunity to implement the CR; (3) rewarding the child when a functional task has been completed without tics or when the child starts to enact the tic, but without parental interruption, catches himself or herself, discontinuing the tic, and either performing the CR or engaging in an alternate adaptive activity; and (4) giving fewer rewards when the child is found to be performing the tic by a support person, but upon parental interruption, either performs the CR or engages in an alternate functional activity.

Study flow

Baseline evaluation

A member of the study group (H.S.S.) evaluated participants meeting screening criteria in person, confirming the diagnosis by clinical observation and interview and obtaining a videotaped YGTSS. Following confirmation of eligibility, each family was introduced to the therapist (S.M.) who provided a 30-minute overview of behavioral treatment for tics.

Randomization

After the baseline evaluation, families were computer-randomized by the study coordinator (L.F.) into either a home-based therapy (DVD) or an in-person therapist (IPT) trained study group (described below). The formal tic evaluator (H.S.S.) was unaware of the treatment category assignment. The study coordinator formally contacted all families, in both groups, after five weeks, and a final in-person assessment was scheduled at 10 weeks. Parents completed web-based online questionnaires (ADHD Rating Scale-5, Clinical Global Impressions of Improvement (CGI-I), and information about ongoing treatments—pharmacologic and behavioral) after five and 10 weeks.

Final visit

The final assessment included a YGTSS, conducted in person (H.S.S.) and videotaped. At the end of the evaluation, the treatment code was broken, and subsequent therapy was reviewed. Parent post-treatment questionnaires were completed online.

Treatment groups

Home-based therapy group

Each family received a copy of the training DVD and written instructions. The family was also provided contact information for the therapist and informed that they were permitted (and encouraged) to call with questions.

In-person therapist (IPT) groups

Participants in this group were to meet with the therapist eight times over the course of 10 weeks, weekly for the first six weeks and biweekly at weeks 8 and 10. Sessions followed a similar format for all participants, and approaches were similar to those on the DVD provided to the home-based treatment group. The therapist could individualize sessions based on the needs of each participant. For example, in some cases awareness training could occur across multiple sessions, whereas in other cases awareness training took place during only one session. Participants were instructed to practice at home between sessions. Session 1 focused on a review of the different categories of tics (e.g., eye/ocular, head and neck, vocal), and the participant selected the tic that would be targeted. Session 2 focused on awareness training. Session 3 focused on CR training. Each participant was taught to use deep diaphragmatic breathing as part of the CR training in addition to a CR specific to his or her tic. Session 4 focused on reinforcement training. During reinforcement training, the participant and a parent completed at least one 10-minute session to practice engaging in the CR behavior. The participant was instructed to engage in the CR behavior each time he or she felt the tic coming on (i.e., when the premonitory urge was detected, the participant engaged in the CR). The participant also had the opportunity to earn a predetermined reward, such as points exchangeable for goods, contingent on the absence of tics during the 10-minute reinforcement training session. Session 5 varied depending on the participant's needs. For example, some participants were ready to select a different tic and begin the treatment steps again, whereas others continued to receive

reinforcement training for the first tic. Session 6 focused on a review of HRT, and participants selected a new tic for application to apply HRT techniques. Sessions 7 and 8 focused on troubleshooting problems that arose as participants worked through the steps of therapy independently over the final weeks. Throughout the protocol, participants were encouraged to contact the therapist with any questions regarding implementation of HRT.

Assessment measures

Health screen

At screening, before the baseline visit, health and developmental information on the participant was obtained and included a parent-completed pediatric medical and developmental history questionnaire (a comprehensive review of the child's birth, medical history, and developmental milestones) and the following scales.

Revised Child Anxiety and Depression Scales (RCADS)³²

The RCADS is a 47-item rating measure that assesses anxiety, depression, and OCD symptoms. Parallel parent and child self-report measures are available. Symptoms are rated on a four-point Likert scale (0 = never, 3 = always). The six scales include Generalized Anxiety, Depression, Obsessive-Compulsive Disorder, Panic Disorder, Social Phobia, and Separation Anxiety. Grade-based T-scores for parent and self-report measures were obtained at baseline visit.

ADHD Rating Scale-5: Home Version³³

The ADHD Rating Scale-5 includes the diagnostic criteria for ADHD in the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders. There are two symptom subscales, Inattention (nine items) and Hyperactivity-Impulsivity (nine items). These 18 items are rated on a 4-point (0 to 3) Likert scale, with 0 = never or rarely, 1 = sometimes, 2 = often, and 3 = very often. In addition, the ADHD Rating Scale-5 assesses six domains of impairment: relationships with significant others, peer relationships, academic functioning, behavioral functioning, homework performance, and self-esteem. Norms for ages five to 17 years include a nationally represented standardization sample, and reliability and validity of scores has been demonstrated. Results were collected at baseline, five-week, and final (10-week) assessments. The Inattention and Hyperactivity subscales (raw scores) were used for analyses.

Yale Global Tic Severity Scale (YGTSS)³¹

The YGTSS is a clinician-completed measure consisting of a tic symptom checklist, motor and vocal tic severity ratings, and a global tic impairment rating. To ascertain tic severity ratings, the examiner rates five different dimensions of tic severity each on a 0 to 5 scale: tic number, frequency, duration, intensity, and complexity. Each of the dimensions is scored separately for motor and vocal tics to produce motor and vocal tic subscale scores (range 0 to 25). These subscales are then combined to produce a total Tic Severity Score (TSS) (range 0 to 50), with higher numbers indicating more severe tics. The YGTSS has demonstrated acceptable internal consistency and acceptable convergent and divergent validity.³¹ The TSS collected at baseline and final visit was the primary functional outcome measure for the study. The total Global Severity Score (GSS) includes the total tic severity score plus an associated impairment scale (range 0 to 50) that assesses tic-related disability during the past week.

Clinical Global Impressions of Improvement³⁶

The CGI-I is a parent-reported clinical impression of improvement. This seven-item scale asks the parent to rate the relative improvement of tics experienced by the patient since the beginning

of the study. Ratings include: 1 = very much better, 2 = much better, 3 = a little better, 4 = no change, 5 = a little worse, 6 = much worse, and 7 = very much worse. By convention, responders were defined as those who received a score of 1 or 2 on the CGI-I.¹⁰

Results

Forty-four children participated. Mean age (\pm S.D.) was 10.21 ± 1.69 (range 7 to 13 years) and included 37 boys and seven girls. The sample was 80% Caucasian, 2% African American, 4% Asian, and 14% multiracial; 11% reported Hispanic ethnicity. Six participants (14%) reported a prior diagnosis of ADHD. Mean age of onset for tics was six years (range: 24 to 72 months). At baseline, a total of seven participants (four in the DVD group, three in the IPT group) were taking tic-suppressing medications; six were taking medication for ADHD (four in the DVD group, two in the IPT group); and one was taking medication to treat OCD (DVD group).

Baseline characteristics for DVD ($n = 33$) and IPT ($n = 11$) groups are listed in Table 1. At study entry, there were no significant group differences in age, sex distribution, race, tic severity, hyperactivity/impulsivity, or parent- or child-rated symptoms of anxiety and depression. In contrast, the IPT group had significantly higher parent ratings of inattention at baseline, compared with the DVD group ($P = 0.021$, $\eta_p^2 = 0.124$). Despite the higher ratings of inattention at baseline in the IPT group, ADHD severity was not significantly associated with total tic severity at baseline (inattention: $r = 0.133$, $P = 0.395$; hyperactivity/impulsivity: $r = 0.171$, $P = 0.274$).

Forty-four participants were initially enrolled in the study. At study completion, 18 participants had completed the 10-week study in either the DVD group ($n = 8$) or the IPT group ($n = 10$). Based on baseline assessment, there were no significant group differences between study completers ($n = 18$) and those lost to follow-up ($n = 26$) in age, sex distribution, race, tic severity, ADHD symptoms, or child-rated symptoms of anxiety and depression (Table 2). In contrast, at baseline, the study completer group had

TABLE 1.
Baseline Differences Between DVD and In-Person Groups

Measure	DVD		P	η_p^2
	In-Person			
	n = 33	n = 11		
	Mean (S.D.)	Mean (S.D.)		
Age at study (years)	10.07 (1.89)	10.64 (0.77)	0.343	0.021
ADHD RS Inattention	8.59 (7.41)	14.45 (5.36)	0.021	0.124
ADHD RS H-I	6.87 (5.95)	8.27 (4.65)	0.484	0.012
YGTS Total Tic Severity	28.09 (5.63)	27.91 (4.44)	0.923	0.001
YGTS Total Score	49.15 (13.79)	50.18 (10.27)	0.822	0.001
RCADS-P GAD	42.03 (9.07)	46.73 (9.61)	0.152	0.049
RCADS-P Depression	40.44 (7.89)	45.09 (11.18)	0.138	0.053
RCADS-P OCD	35.88 (4.82)	36.27 (5.06)	0.817	0.001
RCADS-P Panic Disorder	42.41 (5.85)	41.91 (3.42)	0.792	0.002
RCADS-P Social Phobia	44.53 (10.93)	45.00 (9.37)	0.900	0.001
RCADS-P Separation Anxiety	47.28 (8.67)	50.00 (10.61)	0.417	0.017
RCADS-C GAD	41.12 (8.85)	45.73 (8.73)	0.141	0.051
RCADS-C Depression	46.36 (8.10)	48.64 (7.50)	0.417	0.016
RCADS-C OCD	42.18 (9.36)	44.82 (10.74)	0.440	0.014
RCADS-C Panic Disorder	46.88 (8.13)	49.36 (6.56)	0.365	0.020
RCADS-C Social Phobia	45.45 (11.52)	52.00 (12.05)	0.114	0.058
RCADS-C Separation Anxiety	50.18 (10.23)	48.64 (7.22)	0.646	0.005

Bold indicates clinical significance ($P < 0.05$).

Abbreviations:

ADHD RS = ADHD Rating Scale 5, Home Version

GAD = Generalized Anxiety Disorder

H-I = Hyperactivity/impulsivity

OCD = Obsessive-compulsive disorder

RCADS = Revised Child Anxiety and Depression Scale

YGTS = Yale Global Tic Severity Scale

TABLE 2.
Baseline Differences Between Study Completers and Lost to Follow-Up

Measure	Lost to F/U		P	η_p^2
	Completer			
	n = 26	n = 18		
	Mean (S.D.)	Mean (S.D.)		
Age at study (years)	10.10 (1.92)	10.38 (1.34)	0.588	0.007
ADHD RS Inattention	10.24 (7.49)	9.89 (7.36)	0.879	0.001
ADHD RS H-I	7.28 (6.20)	7.17 (4.90)	0.949	0.001
YGTS Total Tic Severity	28.08 (6.10)	28.00 (4.06)	0.963	0.001
YGTS Total Score	49.23 (14.44)	49.67 (10.67)	0.914	0.001
RCADS-P GAD	40.72 (8.99)	46.72 (8.89)	0.036	0.103
RCADS-P Depression	39.56 (8.04)	44.50 (9.56)	0.074	0.076
RCADS-P OCD	34.72 (4.66)	37.72 (4.63)	0.043	0.096
RCADS-P Panic Disorder	41.56 (4.47)	43.28 (6.28)	0.300	0.026
RCADS-P Social Phobia	43.08 (9.02)	46.83 (12.09)	0.250	0.032
RCADS-P Separation Anxiety	46.88 (9.33)	49.47 (8.81)	0.372	0.020
RCADS-C GAD	41.00 (9.10)	44.11 (8.64)	0.262	0.030
RCADS-C Depression	46.65 (8.30)	47.33 (7.59)	0.784	0.002
RCADS-C OCD	41.23 (8.66)	45.17 (10.79)	0.187	0.041
RCADS-C Panic Disorder	46.35 (7.98)	49.17 (7.37)	0.241	0.033
RCADS-C Social Phobia	45.35 (11.39)	49.61 (12.41)	0.246	0.032
RCADS-C Separation Anxiety	50.85 (10.97)	48.28 (6.94)	0.385	0.018

Bold indicates clinical significance ($P < 0.05$).

Abbreviations:

ADHD RS = ADHD Rating Scale 5, Home Version

GAD = Generalized Anxiety Disorder

H-I = Hyperactivity/impulsivity

OCD = Obsessive-compulsive disorder

RCADS = Revised Child Anxiety and Depression Scale

YGTS = Yale Global Tic Severity Scale

significantly higher parent-rated symptoms of participant OCD ($P = 0.043$, $\eta_p^2 = 0.096$) and generalized anxiety ($P = 0.036$, $\eta_p^2 = 0.124$), compared with the lost to follow-up group. Of those individuals originally randomized to the DVD group, 25 of 33 (76%) were lost to follow-up, compared with only one of 11 (9%) in the IPT group ($\chi^2 = 15.17$, $P < 0.001$). A listing of parent reasons for study dropout is presented in Table 3.

Primary outcomes for study completers were assessed by comparing baseline YGTS scores (TSS and GTS) with the last recorded assessment values for the YGTS using an analysis of only those participants who completed both baseline and 10-week follow-up ($n = 18$). Given baseline group differences in parent ratings of inattention, the ADHD Rating Scale-5 Inattention Scale total score was a covariate in this analysis.

Change ratios for baseline to 10-week scores are listed in Table 4. For YGTS total TSS was DVD 32.4% and IPT 26.6% and total GSS was DVD 33.7% and IPT 26.7%—in all instances with large effect size. Analyzing only individuals who completed the entire study, using repeated measures ANCOVA (analysis of covariance), with treatment group as the between group variable and time as the repeated variable, and covarying for baseline parent ratings of inattention, there was a significant effect of time, with scores across both groups decreasing over the 10-week treatment period [$F(1,15) = 4.93$, $P = 0.042$, $\eta_p^2 = 0.247$]. In contrast, the effects for group ($P = 0.826$) and the time-by-group interaction ($P = 0.918$) were not significant. These findings suggest that among the 18 individuals who completed the 10-week course of treatment, the improvement in the DVD group ($n = 8$) was not statistically different from the improvement for those who completed the IPT group ($n = 10$).

Secondary outcomes were examined using caregiver CGI-I scores for individuals completing the study at five- and 10-week follow-up (Table 5). For those individuals who completed the study, there were no significant overall differences in CGI-I between IPT and DVD group ratings at five weeks [$\chi^2(2) = 3.150$, $P = 0.533$] or at 10 weeks [$\chi^2(2) = 0.257$, $P = 0.879$]. Using a CGI-I score of “very much better” or “much better” to indicate a responder, values show an increase in proportion of responders between five and

TABLE 3.
Parent Reasons for Study Dropout

Reason Stated for Drop out	Number
Tics decreased dramatically or disappeared before starting treatment	4
Stopped participating; no reason given	8
Participant didn't like/wouldn't cooperate with home treatment	4
Difficulty with sessions	1
Family didn't have enough time/family schedule conflicts	2
Family sought alternative intervention	1
Tics became worse; too stressful for family	1
Participant had concussion and didn't want to continue	1
Became ineligible—started tic-suppressing medication during study	1
Started treatment for anxiety instead	1
Participated in treatment, but did not come for final evaluation visit	1
Reported "DVD didn't work"; sent new DVD, but still dropped out	1
TOTAL	26

10 weeks in both the DVD (12.5% to 50%) and IPT groups (20% to 40%).

A total of eight parents from the DVD group and 10 from the IPT group completed a post-treatment questionnaire. In the DVD group, all eight indicated they watched the DVD a total of one to five times over the 10 weeks, 87% said they felt the video was useful, and 100% said they would recommend the treatment to others. In the IPT group, the number of actual in-person training sessions were 6 ($n = 1$); 7 ($n = 2$); and 8 ($n = 7$), 100% said they felt the treatment was helpful, and all said they would recommend it to others.

Discussion

Habit reversal therapy and its more inclusive successor CBIT have both been shown to be beneficial for reducing tic severity in short- and long-term follow-up studies.³⁷ Furthermore, studies have negated concerns about behavioral therapy increasing tics, leading to the substitution of new tics, straining attentional resources, or causing increased stigmatization.^{38,39} Hence, based on their proven success, several treatment guidelines list behavioral therapy as first-line treatment for tics.^{1,17-20,40-42} Unfortunately, despite these recommendations, the limited availability of trained therapists, duration of treatment, financial costs, and lack of insurance coverage restrict accessibility to these therapies.^{43,44} Although steps are being taken to overcome barriers to access and availability, these approaches still require significant therapist time, effort, and cost. To address these restrictive factors,

investigators are currently evaluating the feasibility of internet-based and telehealth approaches.^{27,28,45} The current study examined an innovative approach—one that provides access/availability of behavior therapy but does not place additional burden on clinicians.

The results of this study show that both home-based DVD and in-person treatment groups had meaningful reductions in primary outcome measures. More specifically, in children with moderate or greater tics, there was a significant decline in baseline to 10-week scores for the YGTSS TSS (32.4%, $P < 0.001$, for DVD, 26.6%, $P = 0.01$, for IPT) and YGTSS GSS (33.7%, $P < 0.001$, for DVD and 26.7%, $P < 0.001$, for IPT). A clinically meaningful change in tic severity is a 25% decrease on the YGTSS-TSS score.⁴⁶ A therapeutic beneficial response was also confirmed in the parent-completed secondary measure (CGI-I); scores at 10 weeks indicate that tic symptoms were "much better" in 50% of the DVD and 40% of the IPT groups. In addition, a post-treatment questionnaire identified a parent recommendation of 87% in the DVD and 100% in the IPT group. Overall, these data confirm beneficial results demonstrated in prior randomized control trials that compared HRT/CBIT to either waitlist conditions (no treatment provided)^{47,48} or active comparison conditions.^{15,49-54} Data from the current study also support prior reports showing that the presence of co-occurring ADHD, OCD, and anxiety do not moderate responses to HRT.⁵⁵ As only four participants completing the protocol were receiving tic-suppressing pharmacotherapy, earlier suggestions that behavioral treatment outcome is reduced in individuals receiving medications could not be addressed.⁵⁵

Although confirmation of behavioral therapy for tics is important, the primary goal of this study was to determine the efficacy of a home-based DVD therapy. Despite the relatively small number of completers, those who completed the trial had improvement equal to that in the in-person treatment group. Nevertheless, these results occur in the context of a large dropout rate in the DVD group (25 of 33) when compared with the IPT group (one of 11). A comparison of data between dropouts and study completers showed no identifiable differences in baseline data regarding age, gender, race, baseline tic severity, or presence of co-morbid conditions. In contrast, the study completer group did have higher parent-rated symptom scores for OCD and general anxiety. As presented (Table 3), parent-provided reasons for dropout were variable, ranging from "no reason" to "alterations in tics" and "family issues." Involvement in therapy (whether DVD or IPT) clearly places an added burden on the family system. Thus, it is possible that some families may not have the capacity to fully pursue an independent treatment program.

Whether some of the dropouts would have failed to respond after completing the full 10-week trial is unknown. CGI-I data, showing greater improvement at 10, compared with five weeks of

TABLE 4.
Primary Outcome Measures at Baseline and Final (Week 10) Assessment for Participants Completing Study

	Baseline Mean (S.D.)	10 Week Mean (S.D.)	<i>P</i> (Difference)	η_p^2	Change Ratio (%)
YGTSS-Total Tic					
DVD ($n = 8$)	27.75 (3.62)	18.75 (5.47)	<0.001	0.866	32.4
In-person ($n = 10$)	28.20 (4.56)	20.70 (6.34)	0.010	0.537	26.6
YGTSS-GSS					
DVD ($n = 8$)	47.13 (12.19)	31.25 (7.56)	<0.001	0.835	33.7
In-person ($n = 10$)	51.70 (9.44)	37.90 (13.04)	<0.001	0.842	26.7

Abbreviations:

GSS = Global Severity Score

YGTSS = Yale Global Tic Severity Scale

Change ratio = (Baseline score – 10-week score)/Baseline score; *P* values in table represent significance levels for repeated measures ANCOVA (covarying for baseline ADHD Rating Scale 5 inattention score), comparing 10-week score to baseline score within each group.

TABLE 5.
CGI-I Parent Ratings at 5 and 10 Weeks; Number of DVD and IPT Study Completers Within Each Category CGI-I Ratings Subjects

	Very Much Better	Much Better	A Little Better	No Change	A Little Worse	Much Worse	Very Much Worse	Number
5-week								
DVD	1	0	5	1	1	0	0	8
In-person	0	2	5	2	1	0	0	10
10-week								
DVD	0	4	3	1	0	0	0	8
In-person	0	4	4	2	0	0	0	10

CGI-I is parent rating of clinical global impression of improvement.

treatment, in both groups, suggest a need for a more prolonged behavioral treatment period. Clearly, the direct and active involvement of parents is an essential factor, especially for the success of home-based therapy. Ongoing investigations are evaluating whether brief required telephone or email contacts between the parent and therapist, especially early in the treatment course, might be a beneficial addition to home-based therapy. Unfortunately, the small number of parents who pursued this assistance in the current study (two total phone calls, 16 total emails) limits the ability to directly test this hypothesis. In studies investigating behavioral therapy in children with complex motor stereotypies, the combination of home-based therapy and therapist contact (via telephone) was more beneficial in reducing movements than either solely home-based or IPT treatment.³⁰ Additional elements suggested to be predictors of behavioral benefit include the enrollment of individuals with a positive expectancy and families willing to commit sustained efforts.⁵⁵

This study has several limitations including a small number of subjects and the lack of comprehensive control comparison groups including a DVD treatment arm that provides educational material but suggests ignoring the tics and an IPT group that provides only encouragement. Furthermore, recognizing that the study therapist was required to teach and instruct families using information contained on the DVD, our results are not directly comparable to those using in-person training provided by an experienced, flexible behavioral therapist. Nevertheless, recognizing that many patients/families receive behavioral therapy from nonexperts, due to limited accessibility/availability of expert resources, the current data do support an important role for therapists with less experience. Finally, additional studies are required to determine the treatment response for specific (motor and vocal) tics, effects on comorbid psychosocial issues, alterations of premonitory urges, and long-term benefits.

This is the first study in TS comparing a parent-administered behavioral therapy with in-person, therapist-directed care program. Despite the current large dropout rate, recognizing the potential beneficial effect of home-based therapy, its ready availability, the reduced need for therapist involvement, and inexpensive cost, we suggest a stepwise approach to the treatment of tics, the first step being home-based HRT, especially when there is no acute psychosocial or physical requirement to reduce tics and/or there is a waitlist for behavioral therapy appointments. If parent-administered therapy is unsuccessful, formal CBIT/HRT, administered by trained therapists, should be initiated. Finally, if both behavioral approaches are unsuccessful, the treating physician should consider the initiation of pharmacotherapy.

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References

- Singer HS. Tics and Tourette syndrome. *Continuum*. 2019;25:936–958.
- Dale RC. Tics and Tourette: a clinical, pathophysiological and etiological review. *Curr Opin Pediatr*. 2017;29:665–673.
- Robertson MM, Eapen V, Singer HS, et al. Gilles de la Tourette syndrome. *Nat Rev Dis Primers*. 2017;3:16097.
- Scharf JM, Miller LL, Gauvin CA, Alabiso J, Mathews CA, Ben-Shlomo Y. Population prevalence of Tourette syndrome: a systematic review and meta-analysis: meta-analysis of TS prevalence. *Mov Disord*. 2015;30:221–228.
- Azrin NH, Nunn RG. Habit-reversal: a method of eliminating nervous habits and tics. *Behav Res Ther*. 1973;11:619–628.
- Azrin NH, Nunn RG. Habit Control in a Day: Simple, Effective, and Professionally Tested Techniques for Eliminating the Physical Habits that May Be Keeping You from Leading a Normal Life. 1st ed. New York, NY: Simon & Schuster; 1977.
- Woods DW, Miltenberger RG, Lumley VA. Sequential application of major habit-reversal components to treat motor tics in children. *J Appl Behav Anal*. 1996;29:483–493.
- Cook CR, Blacher J. Evidence-based psychosocial treatments for tic disorders. *Clin Psychol Sci Pract*. 2007;14:252–267.
- Himle MB, Woods DW, Piacentini JC, Walkup JT. Brief review of habit reversal training for Tourette syndrome. *J Child Neurol*. 2006;21:719–725.
- Piacentini J, Woods DW, Scahill L, et al. Behavior therapy for children with Tourette disorder: a randomized controlled trial. *JAMA*. 2010;303:1929–1937.
- Woods DW, Conelea CA, Himle MB. Behavior therapy for Tourette's disorder: utilization in a community sample and an emerging area of practice for psychologists. *Prof Psychol Res Pr*. 2010;41:518–525.
- Wile DJ, Pringsheim TM. Behavior therapy for Tourette syndrome: a systematic review and meta-analysis. *Curr Treat Options Neurol*. 2013;15:385–395.
- Wilhelm S, Peterson AL, Piacentini J, et al. Randomized trial of behavior therapy for adults with Tourette syndrome. *Arch Gen Psychiatry*. 2012;69:795–803.
- Scahill L, Woods DW, Himle MB, et al. Current controversies on the role of behavior therapy in Tourette syndrome: controversies on behavior therapy in TS. *Mov Disord*. 2013;28:1179–1183.
- McGuire JF, Piacentini J, Brennan EA, et al. A meta-analysis of behavior therapy for Tourette syndrome. *J Psychiatr Res*. 2014;50:106–112.
- Verdellen CWJ, Hoogduin CAL, Kato BS, Keijsers GPJ, Cath DC, Hoijtink HB. Habituation of premonitory sensations during exposure and response prevention treatment in Tourette's syndrome. *Behav Modif*. 2008;32:215–227.
- Verdellen C, Hartmann A, Murphy T, ESSTS Guidelines Group. European clinical guidelines for Tourette syndrome and other tic disorders. Part III: behavioural and psychosocial interventions. *Eur Child Adolesc Psychiatry*. 2011;20:197–207.
- Pringsheim T, Holler-Managan Y, Okun MS, et al. Comprehensive systematic review summary: treatment of tics in people with Tourette syndrome and chronic tic disorders. *Neurology*. 2019;92:907–915.
- Pringsheim T, Okun MS, Müller-Vahl K, et al. Practice guideline recommendations summary: treatment of tics in people with Tourette syndrome and chronic tic disorders. *Neurol*. 2019;92:896–906.
- Murphy TK, Lewin AB, Storch EA, Stock S. Practice parameter for the assessment and treatment of children and adolescents with tic disorders. *J Am Acad Child Adolesc Psychiatry*. 2013;52:1341–1359.
- Himle MB, Olufs E, Himle J, Tucker BTP, Woods DW. Behavior therapy for tics via videoconference delivery: an initial pilot test in children. *Cogn Behav Pract*. 2010;17:329–337.

22. Cuenca J, Glazebrook C, Kendall T, et al. Perceptions of treatment for tics among young people with Tourette syndrome and their parents: a mixed methods study. *BMC Psychiatry*. 2015;15:46.
23. Nissen JB, Kaergaard M, Laursen L, Parner E, Thomsen PH. Combined habit reversal training and exposure response prevention in a group setting compared to individual training: a randomized controlled clinical trial. *Eur Child Adolesc Psychiatry*. 2019;28:57–68.
24. Flanckbaum M, Rockmore L, Franklin ME. Intensive behavior therapy for tics: implications for clinical practice and overcoming barriers to treatment. *J Dev Phys Disabil*. 2011;23:61–69.
25. Andr n P, Aspvall K, Fern ndez de la Cruz L, et al. Therapist-guided and parent-guided internet-delivered behaviour therapy for paediatric Tourette's disorder: a pilot randomised controlled trial with long-term follow-up. *BMJ Open*. 2019;9:e024685.
26. Conelea CA, Wellen BCM. Tic treatment goes tech: a review of TicHelper.com. *Cogn Behav Pract*. 2017;24:374–381.
27. Himle MB, Freitag M, Walther M, Franklin SA, Ely L, Woods DW. A randomized pilot trial comparing videoconference versus face-to-face delivery of behavior therapy for childhood tic disorders. *Behav Res Ther*. 2012;50:565–570.
28. Jakubovski E, Reichert C, Karch A, Buddensiek N, Breuer D, M ller-Vahl K. The ONLINE-TICS study protocol: a randomized observer-blind clinical trial to demonstrate the efficacy and safety of internet-delivered behavioral treatment for adults with chronic tic disorders. *Front Psychiatry*. 2016;7:119.
29. Specht MW, Mahone EM, Kline T, et al. Efficacy of parent-delivered behavioral therapy for primary complex motor stereotypies. *Dev Med Child Neurol*. 2017;59:168–173.
30. Singer HS, Rajendran S, Waranch HR, Mahone EM. Home-based, therapist-assisted, therapy for young children with primary complex motor stereotypies. *Pediatr Neurol*. 2018;85:51–57.
31. Leckman J, Riddle MA, Hardin MT, et al. The Yale Global Tic Severity Scale: initial testing of a clinician-rated scale of tic severity. *J Am Acad Child Adolesc Psychiatry*. 1989;28:566–573.
32. Chorpita BF, Yim L, Moffitt C, Umemoto LA, Francis SE. Assessment of symptoms of DSM-IV anxiety and depression in children: a revised child anxiety and depression scale. *Behav Res Ther*. 2000;38:835–855.
33. DuPaul G, Power TJ, Anastopoulos AD, Reid R. *ADHD Rating Scale—5 for Children and Adolescents*. New York: Guilford Press; 2016.
34. Bloch MH, Leckman JF. Clinical course of Tourette syndrome. *J Psychosom Res*. 2009;67:497–501.
35. Woods DW, Piacentini J, Chang S, et al. *Managing Tourette Syndrome: a Behavioral Intervention for Children and Adults Therapist Guide*. New York, NY: Oxford University Press; 2008.
36. Guy W. Clinical global impressions (CGI) scale. In: Rush AJ, ed. *Handbook of Psychiatric Measures*. Washington, DC: American Psychiatric Association; 2000:100–102.
37. Essoe JK, Grados MA, Singer HS, Myers NS, McGuire JF. Evidence-based treatment of Tourette's disorder and chronic tic disorders. *Expert Rev Neurother*. 2019;19:1103–1115.
38. Capriotti MR, Himle MB, Woods DW. Behavioral treatments for Tourette syndrome. *J Obsessive Compuls Relat Disord*. 2014;3:415–420.
39. Peterson AL, McGuire JF, Wilhelm S, et al. An empirical examination of symptom substitution associated with behavior therapy for Tourette's disorder. *Behav Ther*. 2016;47:29–41.
40. Lewin AB, Mink JW, Bitsko RH, et al. Utility of the diagnostic interview schedule for children for assessing Tourette syndrome in children. *J Child Adolesc Psychopharmacol*. 2014;24:275–284.
41. Steeves T, McKinlay BD, Gorman D, et al. Canadian guidelines for evidence-based treatment of tic disorders: behavioural therapy, deep brain stimulation, and transcranial magnetic stimulation. *Can J Psychiatry*. 2012;57:144–151.
42. Martino D, Pringsheim TM. Tourette syndrome and other chronic tic disorders: an update on clinical management. *Expert Rev Neurother*. 2018;18:125–137.
43. Marcks BA, Woods DW, Teng EJ, Twohig MP. What do those who know, know? Investigating providers' knowledge about Tourette's syndrome and its treatment. *Cogn Behav Pract*. 2004;11:298–305.
44. Fr ndt O, Woods D, Ganos C. Behavioral therapy for Tourette syndrome and chronic tic disorders. *Neurol Clin Pract*. 2017;7:148–156.
45. Ricketts EJ, Gilbert DL, Zinner SH, et al. Pilot testing behavior therapy for chronic tic disorders in neurology and developmental pediatrics clinics. *J Child Neurol*. 2016;31:444–450.
46. Jeon S, Walkup JT, Woods DW, et al. Detecting a clinically meaningful change in tic severity in Tourette syndrome: a comparison of three methods. *Contemp Clin Trials*. 2013;36:414–420.
47. O'Connor K, Lavoie ME, Robert M. Preparation and motor potentials in chronic tic and Tourette syndromes. *Brain Cogn*. 2001;46:224–226.
48. Azrin NH, Peterson AL. Treatment of Tourette syndrome by habit reversal: a waiting-list control group comparison. *Behav Ther*. 1990;21:305–318.
49. Verdellen CWJ, Keijsers GPJ, Cath DC, Hoogduin CAL. Exposure with response prevention versus habit reversal in Tourette's syndrome: a controlled study. *Behav Res Ther*. 2004;42:501–511.
50. Rizzo R, Pellico A, Silvestri PR, Chiarotti F, Cardona F. A randomized controlled trial comparing behavioral, educational, and pharmacological treatments in youths with chronic tic disorder or Tourette syndrome. *Front Psychiatry*. 2018;9:100.
51. Azrin NH, Nunn RG, Frantz SE. Habit reversal vs. negative practice treatment of nervous tics. *Behav Ther*. 1980;11:169–178.
52. Wilhelm S, Deckersbach T, Coffey BJ, Bohne A, Peterson AL, Baer L. Habit reversal versus supportive psychotherapy for Tourette's disorder: a randomized controlled trial. *Am J Psychiatry*. 2003;160:1175–1177.
53. Deckersbach T, Rauch S, Buhlmann U, Wilhelm S. Habit reversal versus supportive psychotherapy in Tourette's disorder: a randomized controlled trial and predictors of treatment response. *Behav Res Ther*. 2006;44:1079–1090.
54. Yates R, Edwards K, King J, et al. Habit reversal training and educational group treatments for children with Tourette syndrome: a preliminary randomised controlled trial. *Behav Res Ther*. 2016;80:43–50.
55. Sukhodolsky DG, Woods DW, Piacentini J, et al. Moderators and predictors of response to behavior therapy for tics in Tourette syndrome. *Neurol*. 2017;88:1029–1036.