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Highlights

- Tics have an average age of onset of 5–7 years of age.
- Comprehensive Behavior Therapy for Tics showed good outcomes for youth ages 5–8.
- Some comorbid symptoms and parent behavior toward tics also showed positive change.

Abstract

The Comprehensive Behavioral Intervention for Tic Disorders (CBIT) has demonstrated efficacy in large randomized controlled trials for children (≥9 yrs), adolescents and adults with Tourette Syndrome and Chronic Tic Disorders. Given the early age of onset for tic disorders, a large portion of affected individuals with chronic tic disorders are less than 9 years of age and appropriate developmental adaptations of behavioral treatment have not yet been tested. The goal of this study was to adapt and evaluate the acceptability and utility of a family-based adaptation of CBIT for children under 9 years of age. Children 5–8 years of age (*N* = 15) with chronic tics were recruited from three study sites. CBIT was adapted for use with young children and included habit reversal strategies introduced in a developmentally appropriate game format and function-based interventions to reduce family accommodation of and attention to tic symptoms.

Children and parents described high level of treatment satisfaction and study retention rate was 100%. Treatment response rate was 54% (CGI-I = 1 or 2) with a significant decrease in the YGTSS total score (Cohen's *d* = 0.73) that was largely maintained at 3-month and 1-year follow-up assessments. Treatment was associated with reduction of some symptoms of tic-related comorbid syndromes and with changes in parental accommodation and attention to tics.

Future research should determine if parental attention to tics and symptom accommodation are important mediators of treatment outcome, or if participating in this intervention at a younger age may prevent the chronic course of tic symptoms.

Keywords

Tourette Syndrome, behavior therapy, young children

Persistent Tic Disorders (PTD), including Tourette's Disorder (TD), are childhood-onset conditions, present in around 1% (TD) to 4% (PTD) of children, and are often associated with marked impairment in day-to-day functioning (Kurlan et al., 2001, Scahill et al., 2014, Scharf et al., 2012). Tics have an average age of onset of 5–7 years old with 20% of preschoolers and 8% of early-school-age children evidencing symptoms (Gadow, Nolan, Sprafkin, & Schwarz, 2002). Of those with chronic tic symptoms, up to 60% reported initial tic onset prior to age 7 (Burd, Freeman, Klug, & Kerbeshian, 2006) and 35% of clinically ascertained children with tics experienced their worst-ever tics at age 8 or younger (Leckman, Bloch, King, & Scahill, 2006).

Although the Comprehensive Behavioral Intervention for Tics (CBIT) has demonstrated efficacy in adults (Wilhelm et al., 2012) and youth as young as 9 years of age (Piacentini et al., 2010), there is a gap in the scientific literature supporting this approach for youth below age 9. Given the lack of empirically supported behavioral treatments for younger children, despite the early age of onset and frequency of functionally impairing tics prior to age 9 noted above, treatment professionals can find themselves in the difficult position of either providing psychoeducation and monitoring (i.e., "watchful waiting") or considering a pharmacological approach, despite realistic concerns about medication efficacy and side effects (Dure and DeWolfe, 2006, Kompoliti et al., 2010, Pringsheim and Pearce, 2010, Stevens et al., 2009). Clinical experience and select case reports (e.g., Feldman, Storch, & Murphy, 2011) suggest that elements of CBIT may be applicable and useful in the treatment of tics in young children, but a systematic adaptation and scientific evaluation has not yet been published. Studies support that age is a predictor of tic suppression (Conelea et al., 2018), such that young children may lack the self-awareness and self-control required for successful execution of CBIT strategies, providing rationale for a developmentally appropriate adaptation. Moreover, the functional and environmental factors that reinforce tic expression or tic suppression are persistent and present early in the course of the disorder (Greene et al., 2015, Himle et al., 2014), suggesting that enhanced functional interventions for young children may impact the frequency, severity, or course of tics. The current study describes the developmental adaptation of CBIT for use with children aged 5 to 8 years as well as initial data regarding the feasibility and utility of this adapted version (CBIT-JR).

CBIT is based on a biobehavioral model which posits that tics stem from a neurobiological substrate, but that tic severity and course are determined by an interaction between the individual's biological vulnerability and both internal and external environmental factors. For example, tic severity is diminished during calm, focused activities and made worse by stressful or exciting situations and fatigue. Over time, specific situations can lead to persistent tic worsening through positive and negative behavioral reinforcement. Interpersonal responses to the person ticcing, including positive attention to tics via reassurance or support, as well as negative attention such as reprimands or being told to "stop ticcing," likely maintain or increase tic frequency through external positive reinforcement cycles. Avoidance of activities associated with increased tics (e.g., class work, homework, chores, or stressful social events) may be experienced as helpful in the short term, but from a behavioral point of view such avoidance of challenging activities may serve to maintain or worsen tics in the long term via negative reinforcement (Conelea and Woods, 2008, Himle et al., 2014).

The CBIT protocol includes a multicomponent and individualized intervention that addresses each of the potential pathways that reinforce tic severity (Woods et al., 2008a). The first component, habit reversal training (HRT), is comprised of awareness training to enhance cognizance of the premonitory urges that typically trigger tic expression, and competing response training to effectively block tic expression, thus disrupting the link between urge and tic and eliminating the internal negative reinforcement cycle thought to maintain tics. Functional intervention, the second component, addresses those interpersonal situations that inadvertently result in tic worsening— positive and preferential attention to tics when they are at peak severity, criticism or scrutiny of the person when ticcing, and/or avoidance (escape) of activities associated with tic worsening. Antecedent factors associated with tic exacerbations (e.g., stress/anxiety, boredom, intense arousal) are also addressed by this component of treatment. Given the central role that social reactions often play in tic exacerbation

and maintenance, psychoeducation about tics is systematically provided to parents, siblings, and other prominent individuals in the child's life.

The original child CBIT trial, conducted with 126 youth ages 9–17, used a randomized controlled design to compare the efficacy of CBIT to manualized psychoeducation about tics plus supportive therapy (PST) (Piacentini et al., 2010). Overall, 53% of CBIT participants were rated as much or very much improved by independent clinical evaluators blind to treatment status compared to only 19% of youth receiving PST. CBIT was also associated with a 31% decrease in YGTSS Total Score compared to 18% for PST ($p < .01$), yielding a moderate effect size ($d = .68$). Benchmarking CBIT outcomes against those from medication trials suggests reasonable similarity for these two treatment modalities (Rizzo et al., 2018, Scahill et al., 2013).

From a functional perspective, when tics are met with attention from others, they may be maintained or strengthened as a result (Capriotti et al., 2014, Watson and Sterling, 1998). Although clinical experience suggests that young children with tics may initially be less aware than older youth of the environmental reactions their tics elicit, growing awareness of these reactions may not only worsen tics, but also contribute to the development and maintenance of premonitory urges (Capriotti, Espil, Conelea, & Woods, 2013). Given their emotional salience, parental reactions to their children's tics may be especially powerful in this regard (Watson & Sterling, 1998). Thus, early intervention, with specific emphasis on eliminating tic-triggered reactions from parents and others in the child's environment, may not only serve to reduce acute tic severity, but also prevent progression to more severe and chronic tic disorder and associated morbidity.

Study Aims

The aim of this study was to adapt the current CBIT manual (Woods et al., 2008a, Woods et al., 2008b) for use with the families of 5- to 8-year-old children with chronic tic disorder and to document the feasibility, acceptability, and initial utility of the adapted intervention in a small open trial of 15 youngsters.

Methods

Study Design

This was an open trial pilot study to assess the acceptability of a treatment program with developmentally appropriate modifications for young children and the feasibility of study procedures to test the utility of this treatment adaptation. Approval for human subjects research was obtained from the Institutional Review Boards at each participating study site prior to initiation of study procedures.

Study participants

Fifteen children were enrolled and completed study procedures across three geographically diverse university or academic medical center study sites (5 at Weill Cornell Medicine, 4 at University of Wisconsin–Milwaukee, and 6 at UCLA Semel Institute for Neuroscience and Human Behavior). Each study site had a specialty clinic and/or research program for youth with tic disorders, through which study participants were recruited. Careful phone screens were completed with parents to ensure likelihood of study entry at the initial baseline assessment. This was done to decrease the potential

burden to parents and young children of coming in for a lengthy assessment, which includes potentially reinforcing attention paid to tics, without high likelihood of meeting study criteria.

Eligible participants were children between 5 and 8 years of age presenting with clinically significant tics for at least 6 months duration. A minimum baseline score on the Yale Global Tic Severity Scale of 16 (10 for motor tics only) and a minimum Clinical Global Impressions Severity score of 4 or higher was required for study inclusion. Participants who presented on medication were required to be stable on the current dose of the medication with no planned changes. Participants were excluded from the study if they presented with a pervasive developmental disorder diagnosis, an IQ estimate below 70, or if a comorbid disorder (e.g. Attention Deficit Hyperactivity Disorder [ADHD], Obsessive Compulsive Disorder [OCD], Oppositional Defiant Disorder [ODD]) or an anxiety disorder was the primary concern requiring immediate treatment. Children presenting with these comorbidities were included if the tics were deemed to be the primary concern and the other symptoms were either already under sufficient control or were not assessed to need immediate intervention.

Study treatment

The study team completed a line-by-line review of the existing CBIT protocol to identify those elements requiring modification for younger children and to draft age-appropriate exercises and procedures for this group. The basic structure and components of the treatment remained similar to the original CBIT, however some developmental adaptations were made. The study intervention included 6 intervention sessions delivered over 8 weeks (four consecutive weeks for Sessions 1–4, with Sessions 5 and 6 delivered with an off week in between (see Figure 1 for a session-by-session breakdown of interventions). Session 1 was designed to be 90 minutes, and the remaining sessions were 45–50 minutes, depending on the behavior, tolerance, and participation of the child. The first session of the study treatment was conducted with parents alone because of the importance of psychoeducation and to underscore the crucial role of functional assessment and intervention with parents. The goal of psychoeducation was to normalize the experience of tics in young children, to correct parents' misbeliefs or misattributions about their child's tics, and to ensure parent understanding of and commitment to the treatment model, and the important role of parents in the intervention. The importance and rationale for functional assessment and intervention were reviewed with parents in this first session. The remaining sessions were completed jointly with the child and parent(s).

FIGURE 1. Session Breakdown of Interventions from the CBIT JR Study Manual.

Session 1: Meet with Parents only. Information Gathering to supplement initial assessment; Provide rationale for CBIT; Parent psychoeducation and support; Complete Tic Functional Assessment and review of child's daily routine. Create a Tic Hierarchy

Session 2: Meet with Child alone to build rapport and introduce The Opposite Game. Meet with child and parent together to continue functional assessment and environmental modification strategies for first tic. Practice general Competing Response Training through The Opposite Game for first tic.

Session 3: Meet with child and parent together unless extenuating circumstances do not permit. Practice with parents and child together at end of session if parents are not involved for the entire session. Practice competing response training via The Opposite Game and review environmental modification for next tic.

Session 4: Continue competing response training via The Opposite Game and environmental modification for next tic as described above.
Session 5: Continue competing response training via The Opposite Game and environmental modification for next tic as described above.
Session 6: Review, discuss plan for continued practice. Discuss relapse prevention.

The concepts underlying HRT were taught to the children through a novel modality called "The Opposite Game" (TOG). TOG was a developmentally sensitive approach designed to enhance child awareness and control of bodily sensations and movements, to teach the concept of opposite body actions, and to assess the child's readiness for HRT. TOG was similar to the childhood game "Simon Says" in a call-and-response style, with the emphasis, however, on having the child respond with the opposite body action. TOG utilized a set of "playing cards," including "command cards" that had a phrase and image of common tic body movements (e.g., "shrug your shoulders" or "sniff in through your nose") and "response cards" with a phrase and image of opposite body movements similar to competing responses (e.g., "pull shoulders down" or "gently blow air out of your nose"). The therapist showed a "command card" or demonstrated a body movement after which the child would pick out a corresponding "response card" and/or demonstrate the opposite body action that could be consistent with a competing response. Blank cards were also available to make new cards specific to the child's tics as needed. Children who were able to successfully complete TOG moved on to applying and practicing opposite behaviors (e.g., competing responses) contingent on tic urges or expression with their therapist, maintaining the developmentally sensitive play approach. Parents were actively involved in all sessions and gradually took greater responsibility for delivering TOG and/or HRT under the guidance of the study therapist and continuing the practice at home. Children earned tokens or stickers for participation and effort throughout each session, which could be turned in for predetermined prizes or rewards at designated intervals.

The rationale for a 6-session intervention was based on the assumption that young children would have fewer or less severe tics than older youth, and thus would require less time to complete the intervention. Significant consideration was also given to avoid paying excessive attention to the child's tics due to the understanding that more attention (positive or negative) could exacerbate tics. Anticipating that some young children participating in the study may not be aware that they have abnormal movements, we originally designed the intervention to minimize the child's awareness that the therapy was related to tics. However, all children who participated in this study were aware, to some degree, of their tics, so this feature did not prove necessary and was subsequently relaxed.

Treatment was delivered at each site by a licensed psychologist or a master's-level trainee supervised by an expert in the treatment of tic disorders. All therapists had prior experience providing behavioral treatment for tic disorders. All therapy sessions were videotaped, and study therapists participated in a weekly phone call to discuss any issues with treatment and to ensure maximal fidelity with the treatment protocol.

Measures

A full assessment battery, including the measures outlined below, was completed at baseline, prior to the start of the study treatment (Week 0). Following the 8-week intervention period, the assessment battery was repeated to assess posttreatment changes (Week 8). Participants were asked to return for a follow-up assessment 3 months following the completion of the study treatment (Week 21). Finally, the participants completed another assessment 1 year following their enrollment in the study (Week 52). Assessments were completed by a trained psychologist or doctoral student with supervision from an expert in clinical assessment of tic disorders. Posttreatment and follow-up evaluations were completed by a clinician who was not the treating clinician. Due to the open-trial nature of the study, evaluators were aware that children were receiving an active treatment, but were unaware of any details about treatment progress. Prior to posttreatment and follow-up evaluations, families were encouraged to minimize discussion of specific treatment activities with the evaluator.

All assessments were videotaped and a subset were co-rated to ensure interrater reliability.

Clinician Measures

The primary outcome measures were the Yale Global Tic Severity Scale (YGTSS; Leckman et al., 1989) and the Clinical Global Impressions-Improvement Scale (CGI-I; Guy, 1976).

The YGTSS is a clinician-rated scale that includes an inventory of current tics present in the past week and ratings of the number, frequency, intensity, complexity, and interference of tics, each rated on a 0–5 scale. These ratings are summed to achieve separate scores for motor and vocal tics (each ranging from 0–25) and a combined total tic score (range: 0–50). An associated impairment rating assesses tic-related disability during the last week (range: 0–50). The YGTSS has demonstrated good stability (Storch et al., 2005) and good convergent and discriminant validity (Leckman et al., 1989, Storch et al., 2005).

The CGI-I is a clinician-rated score used to assess overall treatment response with ratings from 1–7, with lower scores indicating improvement. Scores of 1 (*very much improved*) or 2 (*much improved*) indicate a positive treatment response. This scale has been used to assess global tic-related impairment and improvement, and to signify treatment response as a primary outcome measure in gold-standard trials involving patients with TS (e.g., Piacentini et al., 2010, Wilhelm et al., 2012).

Overall psychosocial functioning was also assessed using the Children's Global Assessment Scale (CGAS; Shaffer et al., 1983), a validated, clinician-administered rating of overall functioning (range: 0–100, with higher scores reflecting better functioning).

The Anxiety Disorders Interview Schedule-IV (ADIS-IV; Albano & Silverman, 2004) was administered at baseline assessment to assess for the presence of non-tic psychiatric conditions. The ADIS-IV is a gold-standard, semistructured clinical interview of DSM-IV internalizing and externalizing diagnoses.

Parent Questionnaires

The Parent Tic Questionnaire (PTQ; Chang, Himle, Tucker, Woods, & Piacentini, 2009) is parent-report inventory that assesses the number, frequency, and intensity of a child's motor and vocal tics. Total motor and vocal tic scores are summed to provide a total tic severity score. The PTQ has good internal consistency ($\alpha = 0.80$ to 0.86), excellent test-retest reliability (ICC = .84 to .89), good convergent

validity with the YGTSS, and good discriminant validity from hyperactive, obsessive-compulsive, and externalizing symptoms (Ricketts et al., 2018).

The Tic Accommodation and Reactions Scale (TARS; Capriotti et al., 2015) is a psychometrically sound measure of environmental accommodation and social reactions to a child's tics; the parent-report version was used in this study. The TARS demonstrates good internal consistency for the parent-report version (α = .89) and good convergent and discriminant validity.

The Child Behavior Checklist (CBCL; Achenbach, 1991) was used as the primary dimensional measure of comorbid internalizing and externalizing symptoms. The CBCL is a 118-item parent-report measure of child psychopathology that yields subscale scores corresponding to psychiatric diagnoses (i.e., DSM-oriented subscales), which are normed by child age with mean *t* = 50 and standard deviation of 10. The CBCL has strong psychometric properties, including high test-retest reliability (ICC = .95), high internal consistency (α = .78 to .97), and good convergent and discriminant validity (Achenbach & Rescorla, 2001).

Treatment Acceptability

To assess acceptability and feasibility of the modified treatment approach, all families completed an 8-item treatment acceptability questionnaire posttreatment assessing what they liked about treatment, disliked about treatment, how helpful they thought treatment was, and whether they would recommend this treatment to a friend or other family member. Items were rated on a 4-point Likert scale from 0–3, with 3 indicating maximal satisfaction with and helpfulness of the study intervention. Scores from the 8 items were then summed to create a range of possible scores from 0–24. Session attendance and treatment completion were also monitored as markers of feasibility and acceptability.

Results

Missing Data

All subjects completed baseline and posttreatment assessments. Twelve participants (80%) provided data at the 3-month follow-up assessment, and 10 participants (67%) provided data at the 1-year follow-up assessment. Little's MCAR test on total scores for YGTSS Severity, YGTSS Impairment, DSM subscales of CBCL, PTQ indicated that data were missing at random (chi square = 115.42, *df* = 239, *p* = 1.0).

At baseline, one participant had an invalid response pattern on the PTQ and therefore had to be excluded from all analyses of this measure. Missing data were carried forward from the posttreatment assessment (i.e., last observation available) for analyses of shorter-term maintenance (Week 0 vs Week 21). For analyses related to long-term maintenance (Week 0 vs Week 52), treatment mechanism, and changes in global and non-tic aspects of psychosocial functioning, cases with missing data were excluded on a pairwise basis.

Sample Characteristics

The average age at study entry was 6.7 years (range: 5–8 years). The average age of tic onset in these children was 3.9 years and the mean duration of the tics at study entry was 2.8 years. Two-thirds of the sample (67%) was male, which is consistent with the gender distribution for early-onset tic disorders. All children were of Caucasian ethnicity and 80% lived with both biological parents. The majority of the

sample (87%) met criteria for Tourette Syndrome, and the remaining participants (13%) met criteria for Chronic Motor Tic Disorder. One-fifth of the sample reported psychiatric comorbidity, either OCD (13%) or ADHD (7%), which is less than the typically high rates of comorbidity that are common in individuals with TS, but likely due to the young age of our sample. There was a site difference in baseline mean YGTSS severity that approached statistical significance (baseline YGTSS means: UCLA: 25.8 [7.4]; UWM = 18.0 [6.5]; WCM = 25.4 [6.0]; $p = .06$). Study site differences were nonsignificant ($p > .24$) for all other variables tested (age, gender, duration of tics, age at onset, and parental education).

Treatment Feasibility and Acceptability

Session attendance was very good, with 80% of the sample attending all treatment sessions, and 20% of the sample missing just one session. Subject satisfaction with the intervention was high. Although data were missing for one family, the mean score on the treatment acceptability questionnaire was 21.68. An analysis of specific individual items from the treatment acceptability measure indicated high "overall satisfaction with the program," strong likelihood that the parents "would recommend the program to a friend," and that the parents "would return to the program if they needed additional help for their child." Each of these items received a mean score of 2.79/3.0. All participants indicated that the intervention helped their child deal more effectively with their tics ($M = 2.71$).

Changes in tic Severity

Significant decreases in tic severity were seen throughout the study period, as evidenced by scores on YGTSS and Parent Tic Questionnaire (Table 1). The average total YGTSS score dropped significantly from baseline to the Week 8 posttreatment assessment ($t_{14} = 3.51$, $p < .01$, Cohen's $d = 0.73$), as did the YGTSS motor tic severity scores ($t_{14} = 3.38$, $p < .01$, $d = 0.95$). Vocal tic severity scores decreased marginally ($t_{14} = 1.93$, $p = .08$, $d = 0.50$). Posttreatment improvements in YGTSS scores were maintained in full at the 3-month follow-up assessment, at which time tics were comparable to posttreatment levels for total tic severity, motor tic severity, and vocal tic severity. The effect sizes from baseline to 3-month follow-up were largely maintained or showed continued improvement for change in total tic severity ($d = 1.0$), motor tic severity ($d = 1.0$), and vocal tic severity ($d = 0.95$). At posttreatment, 50% of subjects with available data (7/14) were classified as treatment responders (as indicated by a CGI–Improvement rating of "very much improved" or "much improved").

Table 1. Study Outcomes

Outcome	**BL M (*SD*)**	**Post M (*SD*)**	**3 mo FU M (*SD*)**	**BL-Post Effect Size (*d*)**	**BL-3mo FU Effect Size (*d*)**
YGTSS[a]					
Total Tic Severity	22.7 (9.1)	14.5 (10.1)	13.7 (6.5)	0.73*	1.00**
Motor Tic Severity	14.1 (3.6)	8.3 (6.9)	9.6 (4.3)	0.95**	1.22**
Vocal Tic Severity	9.5 (5.0)	6.3 (5.3)	4.1 (3.8)	0.50^	1.17**
Impairment	19.0 (9.9)	7.0 (8.8)	9.2 (10.0)	1.03**	1.25**
PTQ[b]					
Total Tic Severity	48.2 (24.7)	26.1 (25.0)	23.4 (15.6)	0.74*	1.23**
Motor Tic Severity	35.2 (23.6)	15.3 (18.6)	13.4 (12.5)	0.85**	1.20**
Vocal Tic Severity	13.0 (9.7)	10.9 (10.4)	6.6 (6.8)	0.19	0.91**

TARS	8.25 (8.2)	2.83 (3.1)	2.0 (1.9)	0.68*	1.00**

Note. FU = follow-up. BL = Baseline assessment. Post = Post treatment assessment. 3 mo FU = 3 month follow up assessment. BL-Post = change from baseline assessment to post treatment assessment. BL-3moFU = change from baseline to three month follow-up assessment. YGTSS = Yale Global Tic Severity Scale. PTQ = Parent Tic Questionnaire. TARS = Tic Accommodations and Reactions Scale.
^ $p < .10$. * $p < .05$. ** $p < .01$.
[a]For YGTSS, $n = 15$ for BL vs. Post analyses and $n = 12$ for BL vs. FU analyses.
[b]For PTQ, $n = 14$ for BL vs. Post analyses and $n = 9$ for BL vs. FU analyses.

Parents reported a significant decrease in total tic severity ($t_{13} = 2.76$, $p = .02$, $d = 1.25$) on the PTQ from baseline to posttreatment that was maintained at 3-month follow-up (see Table 1). Significant and durable decreases in tic-related impairment were also observed throughout the treatment and follow-up periods (see Table 1). YGTSS Impairment scores decreased significantly from baseline to posttreatment ($t_{14} = 4.0$, $p < .01$, $d = 1.03$), and these gains were maintained at 3-month follow-up.

Changes in Non-Tic Psychiatric Symptoms and Global Functioning

Positive outcomes were observed for children's functioning, both globally and in specific non-tic areas. Global psychosocial functioning, as measured by the CGAS, increased from baseline ($M = 65.0$, $SD = 11.4$) to posttreatment ($M = 75.8$; $SD = 8.7$; $t_{14} = 4.7$, $p < .01$), and persisted to 3-month follow-up ($M = 75.6$, $SD = 8.1$). Table 2 summarizes changes in non-tic psychiatric symptom domains, as measured by CBCL DSM-oriented subscales. Scores for Somatic and ADHD CBCL subscales decreased significantly from pre- to posttreatment (Table 2). Some decreases were also noted on the Affective, Oppositional Defiant, and Conduct subscales; however, these changes were not statistically significant.

Table 2. Changes in Comorbid Psychiatric Symptoms

Symptom Domain	**BL M (*SD*)**	**Post M (*SD*)**	**BL-Post Effect Size (*d*)**
Affective	56.6 (7.2)	54.2 (6.7)	0.24
Anxiety	59.2 (10.5)	59.8 (10.3)	−0.04
Somatic	64.4 (14.6)	57.7 (12.3)	1.23**
ADHD	54.3 (5.4)	52.4 (4.3)	0.90*
ODD	55.9 (5.4)	54.4 (5.5)	0.18
Conduct	53.2 (4.9)	52.8 (2.5)	0.76

Note. BL = Baseline assessment. Post = post treatment assessment. BL-Post = change from baseline to post treatment assessment. ADHD = Attention Deficit Hyperactivity Disorder. ODD = Oppositional Defiant Disorder. ns = non-significant.
Baseline and posttreatment scores on DSM subscales of the CBCL. $n = 9$ for all analyses.
* $p < .05$. ** $p < .05$.

1-Year Follow-Up

Evaluation of 1-year follow-up data showed strong maintenance of acute treatment gains, though participants were not limited from pursuing additional treatment if desired during the 1-year follow up period, so these data should be interpreted with that in mind. The mean YGTSS total tic score was 13.0 at 1-year follow-up ($SD = 7.2$), which was not significantly changed from posttreatment ($M = 13.5$;

SD = 11.2; t_9 = .15, *p* = .88). Tic-related impairment scores on the YGTSS were generally low (*M* = 8.0, *SD* = 7.5) and did not differ from posttreatment (*M* = 8.5, *SD* = 10.0; t_9 = .16, *p* = .88). Seventy percent of subjects assessed would not have met requirement for study entry based on their global severity at 1-year follow-up (i.e., CGI: Severity <3). Global psychosocial functioning remained high at one-year follow-up (CGAS: *M* = 76.2, *SD* = 14.0) and did not differ significantly from posttreatment levels (*M* = 75.7, *SD* = 8,1; t_8 = 0.49, *p* = .64). Small sample size and missing data did not allow for a close examination of change trajectories, but positive treatment outcomes demonstrated at posttreatment were largely maintained at 1-year follow-up.

Change in Parent Behavior

Given the increased focus on parent psychoeducation and training in this developmental adaptation of CBIT, we measured tic-related parental accommodations and reactions via the TARS over the course of treatment. TARS scores decreased significantly from pretreatment to posttreatment (baseline: *M* = 8.25, *SD* = 8.2; posttreatment: *M* = 2.83, *SD* = 3.1; t_{11} = 2.35, *p* = .038, *d* = 0.68), potentially demonstrating the efficacy of the parent focused interventions and the treatment sensitivity of the TARS.

Discussion

CBIT administered in a developmentally sensitive manner for younger children appears to be both acceptable and feasible, and shows acute and durable benefits for reduction of tics. Treatment benefit extended beyond improvement in tics, as a decrease in symptom and impairment ratings was reported for both tics and other comorbid symptoms. The treatment was shown to be acceptable to young children and their parents, as exhibited by the 100% retention rate for participants completing treatment and the high satisfaction ratings provided by parents. Moreover, our assessment, training, and treatment strategies were all feasible, suggesting these procedures would work well for a larger, adequately powered trial. Although the sample size was too small to identify potential moderators, the repeated measures for identifying potential mechanisms of change were feasible to collect.

The degree of tic symptom change and clinical response found in this study (*d* = .73 for change in YGTSS Total Tic Score) are comparable to those observed in the original child CBIT trial (*d* = 0.68; Piacentini et al., 2010), suggesting the benefit of the CBIT approach extends to this younger demographic. The treatment also resulted in reported change in somatic and ADHD symptoms, which may have a variety of potential explanations. The Somatic Scale of the CBCL may have been picking up somatic sequelae of tics, including the pain, soreness, or sensations associated with repeated ticcing and premonitory urges. Tics can also contribute to distractibility and difficulty staying on task. It is also possible that parental psychoeducation and functional interventions may have led parents to be more effective in ignoring minor somatic complaints from the youth and/or to overall positive behavioral changes (e.g., decrease in externalizing behavior as a result of increased parental consistency and reinforcement of non-tic behaviors). In general, the rate of comorbidity in this sample was less than in other studies of youth with TS, and while it is possible that the low rate of comorbidity influenced study outcomes, the rate of comorbidity in this sample is likely related to the younger age of this sample, which is below the modal age of onset and clinically relevant functional impairments for conditions such as OCD and ADHD.

Some of our initial assumptions were challenged by this study. We assumed that a younger sample would present with fewer or less severe tics, but the number, frequency, and severity of tics in this younger sample were similar to those of the larger randomized controlled trial with older patients. We also assumed young children would be unable to engage in HRT due to limited awareness of tics and urges, which also proved to be somewhat inaccurate. The majority of these youth showed awareness of tics and tic urges in a manner that was facilitative of treatment with HRT. Clinical consensus from study clinicians suggested that TOG was a helpful tool to orient the young participants, particularly those aged 5 to 6 years, to concepts of body awareness and opposite body actions pertinent to HRT. After practicing TOG in early sessions, these subjects were then ready for traditional HRT. The 7- and 8-year-old participants were often able to understand the HRT concepts early on; however; TOG remained a fun, developmentally appropriate and clinically meaningful tool for maintaining motivation and attention in session.

Inherent in our intervention adaptations was the assumption that increasing time spent with parents on psychoeducation and functional interventions would add additional benefit to the overall treatment. Unfortunately, the nature of our data collection does not permit a clear dismantling of intervention components to assess relative efficacy of functional interventions versus habit reversal strategies, but both appear to have been helpful, feasible, and important from clinician report and parent feedback. Future research should examine the temporal relationship between change in parental accommodation and attention to tics and change in the frequency and severity of tics. The degree to which change in parent behavior and other functional variables predicts the change in tic symptoms will determine the degree to which these processes should be targeted in clinic-based treatments, potentially prior to involving the youth in treatment at all, and could inform broad-reaching guidance to parents seeking consultation for tics from a variety of health professionals in different settings (pediatrics, schools, occupational therapy, etc). In general, we believe there is significant clinical and empirical evidence across many diagnostic categories to suggest that involving parents in short-term behavioral interventions for youth improves the acceptability, efficacy, and durability of the treatment overall (Bennett et al., 2013, Piacentini et al., 2011). For example, the increased focus on parent involvement and behavioral management training to modify functionally related antecedents and consequences may be one pathway through which the intervention impacted change in severity ratings for comorbid syndromes, such as ADHD.

Clinical Recommendations

Clinical experience and these pilot data suggest that CBIT can be a useful treatment modality for young children with chronic tics and should include developmentally modified habit reversal strategies for youth coupled with psychoeducation and functional interventions for parents. This offers an alternative pathway from the “wait and see” approach that has been recommended to parents in the past. When a young child first presents with new-onset tics, we recommend to parents a period of “educated observation.” Parents should be advised not to react to tics with a great deal of positive or negative attention, nor overly burden the child with several questions about the tics. Instead, parents should observe and become more aware of the potential environmental, situational, and/or interpersonal situations that correlate with increases in tic frequency or severity. Tics in young children may be transient and resolve on their own; thus, treatment may not be necessary when a young child first exhibits tics. The decision to bring a child in for treatment should be made once tics become

chronic and/or impairing, and the benefits of treatment outweigh the additional time and attention paid to tics by participating in a 6- to 10-week treatment program.

Parents of young children who are concerned about tics may be advised to see a care provider who is expert in the behavioral management of tics for an initial parent-only consultation. Psychoeducation for parents early on will assist with their assessment of tic severity and chronicity, inform appropriate decision making about the timing and type of treatment, and may potentially impact the course or severity of tics (Capriotti et al., 2013, Nussey et al., 2013). Parents should be advised that reacting to tics with a lot of positive or negative attention, changing the child's regular routine or behavioral expectations, and/or overly accommodating tics risks increasing the frequency and severity of tics. Some children do not experience distress or impairment from tics and thus may not be motivated for treatment. These children should not be forced to participate in treatment until they are ready or unless the tics are interfering in other domains (e.g., the teacher has recommended intervention because tics are disruptive in class, or the child is experiencing bullying or social rejection). At the same time, tics can be a marker for later comorbidities, so clinicians and parents can be aware of and watchful for the increased rate of ADHD, OCD, and/or anxiety in youth with tics. When tics are persistent, cause the child distress and/or impairment, or are associated with significant comorbidity, then individual behavioral intervention with a trained clinician may be appropriate and helpful for young children.

Limitations

The present study was subject to a number of limitations, including those inherent in an open trial with small sample size. First, although evaluators who completed clinician-rated assessment measures were blind to treatment progress, they were aware that the child was receiving treatment. Thus, their responses may have been globally biased in favor of identifying improvement over time. Second, the lack of a no-treatment control group leaves open the possibility that symptom change over the study period was due to nontreatment factors. Third, due to the small sample size, we used complete-case analysis to evaluate outcomes, which may have resulted in inaccurate estimates of long-term treatment durability, as 5/15 subjects did not provide data at 1-year follow-up. Fourth, we evaluated possible secondary treatment effects on a relatively large number of psychiatric symptom domains, and, due to low sample size, we were unable to correct for these multiple comparisons without compromising already limited statistical power. Fifth, we were unable to obtain a valid measure of premonitory urge severity, as no measure has been validated for youth in this age range. Sixth, our sample consisted of Caucasian children predominantly from two-parent households; thus, future research with more heterogeneous samples is needed to establish the generality of the effects observed in this study. Finally, 50% of the sample did not meet a priori criteria for treatment response at posttreatment, and we do not have adequate data due to study design and small sample to surmise what factors contributed to this pattern of response. The field at large has not yet systematically identified whether other clinical factors, such as longer treatment duration or the addition of medication, would significantly augment treatment response. Future study of the CBIT-JR intervention specifically, and behavioral approaches to CTD treatment broadly, would aid in addressing these limitations.

Despite the aforementioned limitations, the present results support continued clinical and research applications of CBIT in young children with tics. The study procedures were feasible and acceptable, and clinical benefits were similar to those in the larger randomized controlled study of CBIT. A large trial of CBIT in younger children, such as those who participated in this study, would more clearly define the effects of early behavioral intervention for tics. At the same time, continued investigation of the behavioral and biological mechanisms involved in tic expression is crucial to future efforts to understand the natural course of tic disorders, as well as to develop more potent interventions for them. At present, CBIT offers a nonpharmacological intervention approach for children, adolescents, and adults with treatment effect sizes consistent with many medication trials (Scahill et al., 2013), which is particularly appealing for many families of young children, such as those in this investigation.

Conflict of Interest Statement

The authors declare that there are no conflicts of interest.

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