Comprehensive Behavioral Intervention to Improve Occupational Performance in Children With Tourette Disorder

Jan Rowe, Hon K. Yuen, Leon S. Dure

OBJECTIVE. We evaluated the efficacy of a comprehensive behavioral intervention for tics (CBIT) program to reduce tic severity and improve occupational performance in children with tic disorder using a one-group pretest–posttest design.

METHOD. Thirty children with tic disorder completed an eight-session CBIT program. The program focused on habit reversal, relaxation training, and function-based approaches to address how the environment and social situations (antecedents and consequences) sustain or influence tic severity.

RESULTS. We observed significant reduction in the number of tics and improvement in scores on the Parent Tic Questionnaire, Subjective Units of Distress Scale, and Child Occupational Self Assessment after CBIT compared with scores at baseline.

CONCLUSION. Findings provided support that CBIT reduced the number of tic expressions, tic severity, and level of distress associated with tic and improved these children's self-perception of their competence in and importance of performing everyday activities (i.e., occupational performance).


Tourette disorder (TD; also called Tourette syndrome or chronic tic disorder) is a childhood-onset neurobehavioral disorder characterized by multiple motor and vocal tics that fluctuate in severity and last for ≥1 yr (Du et al., 2010; Leckman, Bloch, Scahill, & King, 2006). Tics are rapid, recurrent, and stereotypical motor movements or vocalizations that are involuntary and nonrhythmic; tics typically occur many times per day and often in bouts or in various combinations (Leckman et al., 2006). The more common motor tics include eye blinking, head jerking, shoulder shrugging, and facial grimacing; common vocal tics include snorting, sniffing, and grunting (Chang, Himle, Tucker, Woods, & Piacentini, 2009). Tics typically emerge at age 5 yr, peak in severity during early adolescence, then decline during young adulthood (Leckman et al., 2006). Premonitory sensory urges before tic occurrence are often identified in children >8 yr old (Du et al., 2010). Common comorbidities of TD include attention deficit hyperactivity disorder, obsessive–compulsive disorder, and symptoms of anxiety and depression (Leckman et al., 2006).

Tic expressions can create problems with body function (e.g., pain; Riley & Lang, 1989), limitations in activities (e.g., writing), and restrictions in participation (e.g., social, sports, recreational activities) that may lead to social isolation (Meucci, Leonardi, Zibordi, & Nardocci, 2009). Tic disorders can also cause marked distress and significant impairment in the child’s interpersonal interactions and relationships, such as problems in making and keeping friends (Champion, Fulton, & Shady, 1988), emotional well-being (e.g., being made...
fun of or bullied; Zinner, Conelea, Glew, Woods, & Budman, 2012), and self-esteem. The cumulative effect of moderate to severe tic expression over the school years can have a deleterious effect on behavioral health and academic and occupational performance (Packer, 2005).

A variety of behavioral treatments have shown some success in tic management (Cook & Blacher, 2007). The most compelling evidence for managing tics is that for habit reversal training (Azrin & Peterson, 1988), with tic awareness training and competing response training as the most crucial elements of the program (Miltenberger, Fuqua, & Woods, 1998). Comprehensive behavioral intervention for tics (CBIT; Piacentini et al., 2010), recently shown to be effective to reduce tics and tic-related impairment in children with tics, consists of habit reversal and relaxation training and function-based approaches that address how the environment and social situations sustain or influence tic severity.

Most behavioral treatments are focused primarily on the reduction of the number, frequency, and intensity of tics without investigating their impact on the performance of the child’s everyday and school activities. This area is especially relevant to occupational therapy practitioners because these treatments are not a cure for tics but reduce the severity of tic expression. Therefore, a better understanding of the impact of the behavioral treatment on the child’s occupational performance may assist the therapist to better design the intervention and improve long-term outcomes.

The aim of this study was to investigate the efficacy of a CBIT program, a replication of Piacentini et al.’s (2010) study, for reduction in the number of tic expressions, tic severity, level of distress associated with tics, and improvement in perceptions of the competence in and importance of performing everyday activities (i.e., occupational performance) among children with tic disorder.

Method

Research Design

We used a one-group pretest–posttest research design. The study was approved by the institutional review board of the University of Alabama at Birmingham (UAB), where the study was conducted. Signed informed consent was obtained from the parent with verbal consent from the participant.

Participants

A convenience sample of 30 children with TD was obtained through the movement disorder clinic at UAB Children’s Hospital and from pediatric neurologists in the Birmingham, Alabama, area. Participants eligible for the study were children or youth <20 yr old who were diagnosed with TD and had at least one motor or vocal tic occurring daily. In addition, the children had to be able to understand and follow verbal commands in English. The exclusion criteria were unwillingness to address tic problems, unstable medical conditions, diagnosis of autism spectrum disorder, traumatic head injury, psychosis, or moderate to severe intellectual disabilities.

Instruments

Child Occupational Self Assessment. The Child Occupational Self Assessment, Version 2.1 (COSA; Keller, Kafkes, Basu, Federico, & Kielhofner, 2006) was designed to measure self-perception of the occupational performance profile in children and youth (Kramer, Kielhofner, & Smith, 2010). The COSA comprises two subscales: Occupational Competence and Values (importance). Each subscale has 25 items representing a range of everyday activities that most children encounter at home, in school, and in their communities. Each item is rated on two separate 4-point rating scales on the basis of how competent the children perceive themselves to be in doing these occupations and how important these same occupations are to them. The COSA was administered according to the guidelines provided in the assessment manual (Keller et al., 2006). The responses on each rating scale were recoded as follows: for Competence, 1 = I have a big problem doing this; 2 = I have a little problem doing this; 3 = I do this OK; and 4 = I am really good at this; for Values, 1 = not really important to me; 2 = important to me; 3 = really important to me; and 4 = most important of all to me. A composite score for each subscale is formed by summing scores on the 25 items (range = 25–100), with a higher score indicating more competence in performing these activities or placement of more value (importance) on these activities.

Results from the Rasch analysis indicated that the 25 items fit the unidimensional construct that each subscale was intended to measure (Kramer et al., 2010). The COSA has demonstrated acceptable content (as evident by only one item on the Values subscale not meeting the Rasch model fit requirement of having mean square fit statistics <1.4) and substantive validity (as evident by >85% of children meeting the fit statistics requirement on both subscales) in diverse children and youth groups ages 6–18 yr (Kramer et al., 2010).
Subjective Units of Distress Scale. The Subjective Units of Distress Scale (SUDS; Woods et al., 2008) was originally developed by Wolpe (1958). It is a scale ranging from 0 to 100 that measures the subjective intensity of distress experienced by an individual. Woods et al. (2008) modified the number of rating points to 11 (i.e., 0–10), with 0 indicating no distress and 10 indicating maximum distress, for measuring children’s subjective level of distress associated with each identified tic. The SUDS was significantly and negatively correlated with clinicians’ rating of general functioning (r = −.44; Tanner, 2012).

Parent Tic Questionnaire. The Parent Tic Questionnaire (PTQ; Chang et al., 2009) was designed to assess the presence, frequency, and intensity of the child’s motor and vocal tics during the previous week as reported by the parent. The PTQ includes questions on 14 motor and 14 vocal tics. Parents indicate whether their child exhibits any of the 28 tics and rate the frequency and intensity of each tic separately on a rating scale of 1 to 4, with higher scores indicating greater frequency and stronger intensity. Details on the scale anchors have been documented elsewhere (Chang et al., 2009). A severity score for each tic is formed by summing the ratings on the frequency and intensity components, taking into consideration the presence or absence of the tic. Separate total motor and vocal tic severity scores are formed by summing the rating of all motor and vocal tics, respectively. A grand total severity score is formed by combining the scores from both motor and verbal tics (range = 0–224). The PTQ demonstrated acceptable internal consistency (α = .86–.90), temporal stability (intraclass correlation coefficient = .84), and convergent validity (significantly correlated with clinicians’ rating of tic severity, r = .72) in a clinic sample (Chang et al., 2009).

Tic Symptom Hierarchy Tracker. In addition to the COSA, SUDS, and PTQ, we used the number of tics identified in the Tic Symptom Hierarchy Tracker (TSHT; Woods et al., 2008), in which the participant lists his or her current tics in a hierarchical order of bothersomeness, as an additional outcome measure.

Intervention

Parents of children with TD who agreed to let their children participate scheduled a baseline evaluation at the UAB Pediatric Tourette Syndrome Clinic. The CBIT program was delivered through a manualized protocol consisting of eight sessions. The first session and part of the second session were for information gathering, baseline assessment, and rapport building. The first six sessions were held weekly, and the subsequent two sessions were conducted in alternating weeks (see Table 1 for the specific evaluation and treatment across the eight sessions). At the end of each session, homework was assigned to the participant to maximize self-management and develop positive habits.

At the end of Session 1, the therapist requested that the participant and parent each monitor the first tic identified on the TSHT using separate monitoring sheets (Woods et al., 2008). The monitoring sheet documents the nature of the selected tic for intervention, day and time of tic occurrence, activity or event in which the participant was engaging when the tic occurred, and a tally count of occurrences (Woods et al., 2008). In addition, the participant was asked to complete a daily functional assessment form (self-report) about the frequency of other tics, the context, the feelings (physical and emotional) experienced, and the reactions from others.

During Session 2, the therapist reviewed the participant’s completion of the assigned homework. The participant identified his or her current tics in hierarchical order from the most to least bothersome using the TSHT. As understood by the participant, bothersome could indicate physical pain, embarrassment, annoyance, inconvenience, or distraction from meaningful occupations.

<table>
<thead>
<tr>
<th>Week</th>
<th>Session</th>
<th>Intervention</th>
<th>Evaluation</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>1</td>
<td>—</td>
<td>COSA, PTQ, TSHT</td>
</tr>
<tr>
<td>2</td>
<td>2</td>
<td>CRT</td>
<td>TSHT, SUDS</td>
</tr>
<tr>
<td>3</td>
<td>3</td>
<td>CRT</td>
<td>TSHT</td>
</tr>
<tr>
<td>4</td>
<td>4</td>
<td>CRT, relaxation (e.g., diaphragmatic breathing) training</td>
<td>TSHT, SUDS</td>
</tr>
<tr>
<td>5</td>
<td>5</td>
<td>CRT, progressive muscle relaxation exercise training</td>
<td>TSHT</td>
</tr>
<tr>
<td>6</td>
<td>6</td>
<td>CRT</td>
<td>TSHT, SUDS</td>
</tr>
<tr>
<td>7</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>8</td>
<td>7</td>
<td>CRT</td>
<td>TSHT</td>
</tr>
<tr>
<td>9</td>
<td>—</td>
<td>—</td>
<td>—</td>
</tr>
<tr>
<td>10</td>
<td>8 (post-CBIT)</td>
<td>CRT, relapse prevention strategies</td>
<td>COSA, PTQ, TSHT, SUDS</td>
</tr>
</tbody>
</table>

Note. — = For Sessions 7 and 8, treatment was provided in alternating weeks; therefore, the participants did not come for therapy on Weeks 7 and 9. CBIT = comprehensive behavioral intervention for tics; COSA = Child Occupational Self Assessment; CRT = competing response training; PTQ = Parent Tic Questionnaire; SUDS = Subjective Units of Distress Scale; TSHT = Tic Symptom Hierarchy Tracker.
The participant then defined the tic features to make sure the parent and the therapist understood all aspects of the tic expression. The participant used the SUDS to rate the level of distress of each identified tic. Initially, the participant chose to work on the most bothersome tic (i.e., Tic 1).

The therapist then conducted a function-based assessment of the tic selected for intervention through which the therapist identified its antecedents and consequences. Most of the antecedents were related to school environment (e.g., types of classes) or people around the participant, because anxiety, stress, or fatigue can intensify the severity of tics. Subsequently, strategies (e.g., eliminating negative attention) were developed to modify antecedent situations or consequences, thereby decreasing the likelihood of the situation that intensified the tic severity.

Habit reversal training was introduced in Session 2, in which the participant was taught to increase the awareness of sensations before the occurrence of the selected tic (i.e., premonitory sensory urges) as well as the sequence of the tic as it occurred. Strategies that helped the participant increase the awareness of the tic were to slow the tic down or simulate the tic. Awareness training might also include having the participant hold up a finger each time he or she first became aware of signs that a tic was about to occur. The therapist and participant then developed a competing behavioral response to replace the tic as soon as the participant was aware of the emergence of the premonitory sensation. A competing behavioral response is a voluntary behavior that is physically incompatible with the execution of the tic but compatible with maintaining participation in ongoing activities (Piacentini et al., 2010). The participant practiced the competing response for 1 min while engaging in a daily activity that was normally affected or during which the tic normally occurred. As part of the program, parents prompted and reinforced the participant for performing the competing response at home whenever the premonitory sensation was emerging as well as during and after the tic’s occurrence.

If the competing response was not effective in managing the tic, an alternate competing response was developed in the next session. Within each session, one tic from the TSHT was addressed. Moving to management of another tic in subsequent sessions happened only if progress had occurred on the prior one. The TSHT was revisited in each subsequent session to note any changes in the occurrence of tics or level of distress and to alter the order of tics being addressed. As part of the participant’s weekly homework assignment, he or she was requested to complete a monitoring form for each new tic, which included the tic signal and competing response that helped manage it (Woods et al., 2008).

In addition, relaxation techniques such as diaphragmatic breathing and progressive muscle relaxation exercises were taught and practiced in Sessions 4 and 5 (or earlier to serve as a competing response for vocal tics) and incorporated into homework assignments. In Sessions 6 and 7, the participant learned to develop competing responses for new tics, and Session 8 focused on relapse prevention strategies. In the last three sessions (i.e., Sessions 6–8), the therapist reviewed the rules for coming up with the competing response. The focus in this phase of therapy was to ensure the participant and parent understood the strategies to identify competing responses for future tics. In addition, the participant was asked to rehearse all former successful competing responses for clarity and completeness. To provide visual feedback on progression, the therapist presented a form with a list of all the tics, signals that the participant had encountered (was still experiencing or had resolved), and competing responses with which he or she was successful or unsuccessful in replacing the tics. Last, if a need was identified for environmental cueing or for the participant to have picture or word menus to remind him or her to practice the competing response, these last three sessions would allow the participant (as well as the parent) to try out the cues or modify them before discharge.

Each CBIT session lasted about 60 min except for the first two sessions, which included the evaluation component and lasted for 90 min. The individualized, manual-based CBIT program was carried out by a licensed occupational therapist (Jan Rowe) with >2 yr of experience in conducting CBIT for children with tic disorder. The therapist had completed a 2-day training workshop on delivering the CBIT program, which was conducted by Douglas Woods, one of the founders of the CBIT program. To establish intervention fidelity, the therapist pilot-tested the protocol with five children with tics for procedure refinement and to ensure consistency of delivery across participants.

Data Collection

The occupational therapist (Rowe) collected the participant’s sociodemographic information, medical history, and medication information from the parent and administered the following assessments: COSA (Kramer et al., 2010), PTQ (Chang et al., 2009), TSHT, and SUDS (Woods et al., 2008) at baseline (the first two sessions) and the eighth session (see Table 1).

Data Analysis

Because scores on several outcome measures (i.e., COSA, SUDS, PTQ) did not meet the assumptions of normality,
we used nonparametric statistical methods to analyze the data. A nonparametric Wilcoxon signed-ranks test was performed to test the hypotheses that participants would have a fewer number of identified tics and that scores on the SUDS, PTQ (severity), and COSA (Competence and Values) would significantly improve at the completion of the CBIT program compared with scores at baseline. We assessed associations between number of tics reported by the participant and the parent and the COSA scores using the Spearman rank-order correlation. Statistical significance was set at \( p < .05 \).

Results

Thirty participants (21 boys, 9 girls; 27 White, 3 Black) completed the CBIT program. Four were diagnosed with chronic tic disorders, and 26 had Tourette syndrome. Participants ranged in age from 7 to 19 yr (mean \( M \) ± standard deviation \( SD \) = 12.3 ± 3.2 yr), with 1 participant who was 19. Most participants \((n = 27)\) lived with both parents. Reported comorbid diagnoses included 7 diagnosed with attention deficit hyperactivity disorder, 10 with obsessive–compulsive disorder, and 14 with general or social anxiety. Five participants took tic-suppressing medications (\( \alpha \)-2-adrenergic agonists and atypical neuroleptics) at baseline. All but 1 participant completed the entire eight-session CBIT program in 10 wk.

A significant reduction in the number of identified tics and improvement in scores on the SUDS, PTQ, and COSA (Occupational Competence and Values) was observed post-CBIT compared with baseline \((all ps < .0001; see Figure 1 and Table 2)\).

Compared with the three to four tics exhibited at baseline by the majority of the participants, one-third of the participants post-CBIT were tic free and another one-third reported exhibiting only one tic. On completion of the CBIT program, 2 participants reported that they no longer required tic-suppressing medications because they either did not have any tics or were able to manage occasional tics with the strategies learned in the CBIT program.

Significant associations between the Occupational Competence and Values scores at both baseline \((r_s = .48, p = .007)\) and post-CBIT \((r_s = .53, p = .003)\) were observed. The Values scores on the COSA were significantly correlated with number of tics reported on the TSHT \((r_s = -.51, p = .004)\) and PTQ \((r_s = -.48, p = .008)\) post-CBIT. The Competence scores on the COSA were significantly correlated with number of tics reported on the PTQ \((r_s = -.42, p = .022)\), and the association between Competence scores on the COSA and number of tics reported on the TSHT bordered on significance \((r_s = -.36, p = .05)\) post-CBIT. No significant correlation was observed between scores on the COSA (Competence or Values) and number of tics reported with these two instruments (TSHT and PTQ) pre-CBIT. The number of tics reported on the TSHT and PTQ was significantly correlated at both baseline \((r_s = .55, p = .001)\) and post-CBIT \((r_s = .91, p < .0001)\). Finally, motor tics were reported to occur more often \((M = 4.3 vs. M = 2.0)\) and be more severe than vocal tics on the PTQ \((total motor severity score, M \pm SD = 25.2 \pm 14.9; total vocal severity score, M \pm SD = 11.8 \pm 14.7)\).

Discussion

The results of this pilot study are consistent with findings from a previous large randomized controlled trial indicating that CBIT is effective in reducing tic severity (Piacentini et al., 2010). In addition, this study revealed that CBIT helps enhance participants’ self-perceived occupational performance as indicated by their improvement in scores on the COSA (Competence and Values). Schoolwork, socializing with friends and classmates, having time to do the things they wanted to do, and several basic activities of daily living (including sleep, hygiene, and eating) were the occupations that participants reported most often interrupted by the tics. Improvement in the ability to perform various everyday occupations related to participants’ roles is essential for their success in academic, home, and community engagement. Because Occupational Competence and Values scores were significantly correlated, participants who reported problems with (i.e., were less competent at) completing homework or schoolwork in a timely manner...
were likely to value these occupations as less important. Once participants were able to manage their tics, enabling them to focus on schoolwork and homework, their perceived ability to complete this work in a timely manner increased, as did the value or importance they assigned to these occupations. Finally, consistent with the generalization treatment effect for CBIT reported in the literature (Woods & Twohig, 2002), some of the untreated tics experienced by the participants in the current study also decreased after about five CBIT treatment sessions (see Figure 1).

Limitations

Because of the limitations of the one-group pretest-posttest design used in this study (such as maturation as a major threat to internal validity), we cannot provide a definite conclusion regarding the efficacy of CBIT in tic reduction and improvement in participants’ occupational performance. In addition, the COSA may not be an ideal tool to measure the impact of tics on a child’s occupational performance because it was not originally designed for that purpose; it may also not be ideal because of its self-report nature. However, our data indicate that the COSA was sensitive to changes in intervention for tics. The score changes on the COSA were also consistent with the anecdotal information that we received from parents at the end of the program. For example, one parent reported, “My child is trying things now they would have never tried before going through the program”; another parent commented, “[My son] still has tics but he can manage them now and they don’t affect his willingness to engage like we saw before the CBIT program.” Other potential assessments such as the Short Child Occupational Performance Evaluation (SCOPE; Bowyer, Kramer, Kielhofner, Maziero-Barbosa, & Girolami, 2007), which determines how a child’s volition, habituation, skills, and the environment facilitate or restrict participation, may serve as a supplementary outcome measure. However, the SCOPE requires gathering information in a variety of ways (i.e., observation in various natural settings, interviews with teachers, chart review), which was not feasible for this pilot study.

Future Research

More objective evaluations of the number, frequency, and intensity of tics and children’s occupational performance, including school performance, are essential to support the value of CBIT. To reduce recall bias, these evaluations may include frequency count of tics and tic intensity and disruption in the classroom, community settings, and home environment using momentary time sampling or interval recording strategies. In addition, further study should investigate the sustainability of CBIT as it relates to relapse and the benefit of booster sessions.

Implications for Occupational Therapy Practice

The purpose of this pilot study was to evaluate the efficacy of an 8-wk CBIT program to reduce tic severity and improve occupational performance in children with tic disorder. The results have the following implications for occupational therapy practice:

- Occupational therapists have a long history of using the cognitive–behavioral frame of reference to treat children with various behavioral disorders. Through the implementation of CBIT, occupational therapy practitioners can help support successful occupational performance and participation among children with TD.

- The unique aspect of occupational therapy in the treatment of children with TD is that the therapeutic goal of the intervention is not only to assist the children in managing tic expression but also to improve their occupational performance in school, home, and community settings.

- The CBIT treatment protocol (one 30–60 min session/wk for six to seven sessions) makes it possible for school-based occupational therapists to implement this therapy for the treatment of children with TD.

### Table 2. Improvement in the Outcome Measures After Completion of the Eight-Session CBIT

<table>
<thead>
<tr>
<th>Measure</th>
<th>Baseline Mean ± SD</th>
<th>Median (IQR)</th>
<th>Post-CBIT Mean ± SD</th>
<th>Median (IQR)</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>No. of tics (TSHT)</td>
<td>4.63 ± 2.3</td>
<td>4.0 (3)</td>
<td>1.1 ± 1.1</td>
<td>1.0 (2)</td>
<td>&lt;.0001</td>
</tr>
<tr>
<td>SUDS</td>
<td>6.7 ± 1.7</td>
<td>7.0 (3)</td>
<td>1.5 ± 1.9</td>
<td>1.0 (2)</td>
<td>&lt;.0001</td>
</tr>
<tr>
<td>PTQ—Total Severity COSA</td>
<td>36.9 ± 25.8</td>
<td>30.5 (39)</td>
<td>3.3 ± 3.7</td>
<td>2.0 (6)</td>
<td>&lt;.0001</td>
</tr>
<tr>
<td>Values</td>
<td>71.0 ± 19.8</td>
<td>69.0 (30)</td>
<td>86.5 ± 15.7</td>
<td>87.0 (24)</td>
<td>.0001</td>
</tr>
</tbody>
</table>

Note. CBIT = comprehensive behavioral intervention for tics; COSA = Child Occupational Self Assessment; IQR = interquartile range; PTQ = Parent Tic Questionnaire; SD = standard deviation; SUDS = Subjective Units of Distress Scale; TSHT = Tic Symptom Hierarchy Tracker.
Conclusion

Results support that CBIT reduced the number of tic expressions, tic severity, and level of distress associated with tics and improved these children’s self-perception of their competence in and the importance of performing everyday activities (i.e., occupational performance). Both child-perceived distress level related to tic expression (i.e., SUDS score) and parent perception of the child’s tic symptom severity (i.e., PTQ score) were reduced to minimal. ▲

Acknowledgments

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References


