

Intensive outpatient comprehensive behavioral intervention for tics: A case series

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Abstract

Recent randomized clinical trials have established the efficacy of Comprehensive Behavioral Intervention for Tics (CBIT) in treating children and adults with Tourette syndrome and persistent tic disorders. However, the standard CBIT protocol uses a weekly outpatient treatment format (*i.e.*, 8 sessions over 10 wk), which may be inconvenient or impractical for some patients, particularly patients, who are required to travel long distances in order to receive care. In contrast, an intensive outpatient program may increase accessibility to evidence-based behavioral treatments for Tourette syndrome and other persistent tic disorders by eliminating the necessity of repeated travel. This case series evaluated the use of an intensive outpatient program CBIT (IOP CBIT) for the treatment of 2 preadolescent males (ages 10 and 14 years) with Tourette syndrome. The IOP CBIT treatment protocol included several hours of daily treatment over a 4-d period. Both children evi-

denced notable reductions in their tics and maintained treatment gains at follow-up. Moreover, both patients and their parents expressed treatment satisfaction with the IOP CBIT format. This case series addresses an important research gap in the behavioral treatment of tic disorders literature. The patients' treatment outcomes indicate that IOP CBIT is a promising treatment that warrants more systematic investigation.

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Key words: Tourette syndrome; Tics; Habit reversal; Intensive outpatient; Behavior therapy

Core tip: Comprehensive Behavioral Intervention for Tics (CBIT) is an empirically supported treatment for individuals with Tourette syndrome. However, the standard, weekly outpatient format of CBIT may preclude some from receiving care. This is the first case series to examine the treatment outcomes of intensive outpatient CBIT (Intensive Outpatient Program CBIT) in children. Despite marked differences between the two boy's presentations, outcomes for both cases were positive.

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INTRODUCTION

Tourette syndrome (TS) is a disorder characterized by multiple motor tics and at least one vocal tic that occur regularly and are present for at least 12 mo^[1]. On average, tics emerge between the ages of 3 and 8 years, peak between 10 and 12 years, and decrease in adulthood^[2,3]. An estimated 60% of children with Tourette syndrome also

meet diagnostic criteria for at least one psychological disorder, with attention deficit hyperactive disorder (ADHD) being the most common comorbid condition^[4], followed by obsessive compulsive disorder (OCD), social anxiety, depression, and externalizing behaviors^[4-6].

Standard treatments for tourette syndrome and persistent tics

Historically, pharmacologic interventions have been used as the first-line treatment for symptom management in Tourette syndrome patients^[7]. However, medications require long-term continuous use and are associated with negative side effects that frequently lead to discontinuation of treatment (for review, see^[7-9]). Alternatively, behavioral interventions reduce concerns regarding negative side effects and potential long-term consequences of prolonged medication use. A number of behavioral interventions have been examined (for review, see^[10]) with habit reversal therapy^[11] garnering the most support (for review, see^[12]). Habit reversal consists of awareness training, contingency management, relaxation training, competing response training, social support, and generalization training.

Comprehensive Behavioral Intervention for Tics (CBIT)^[13] is a multiple-component behavioral treatment for Tourette syndrome and persistent tic disorders that expands on the original habit reversal therapy protocol and includes additional emphasis on psychoeducation, functional interventions, and relapse prevention. Recently, two large randomized controlled trials examined the efficacy of CBIT compared to supportive therapy in adults and children diagnosed with Tourette syndrome and persistent tic disorders. The child study ($n = 126$; mean age 11.7 years) found that CBIT was superior to a psychoeducation and supportive therapy comparison condition in reducing tics (52.5% *vs* 18.5%, respectively)^[14]. The adult study ($n = 122$; 16-69 years) also found superior results for CBIT, with 38.1% of the participants who received CBIT *vs* 6.4% in the psychoeducation and supportive therapy condition experiencing a significant improvement in their tics symptoms at post-treatment^[15]. Importantly, both adults and children in the CBIT condition maintained treatment gains and reported decreased psychological symptoms at the six-month follow-up.

Taken together, these findings indicate that CBIT produces similar outcomes as medication without the side effects and that patients continue to experience benefits after treatment is completed^[14]. In response to mounting evidence, CBIT is now considered a first-line treatment for persistent tic disorders in Europe^[12] and Canada^[16].

Rationale for intensive outpatient CBIT

The standard outpatient CBIT protocol is comprised of eight sessions that are completed over 10 wk, followed by three monthly booster sessions. However, weekly sessions may be inconvenient or impractical for some patients depending on the complexity of their symptoms or their accessibility to care. Instead, these patients may benefit from an intensive outpatient program (IOP) that

compresses CBIT into a week-long protocol. An IOP can help extend treatment catchment areas and compensate for the current lack of CBIT providers. Importantly, IOP also allows for patients to practice CBIT without the distraction of school or work. This is particularly relevant to the use of the competing response procedure, which is to be implemented upon the detection of a premonitory urge to tic or the actual occurrence of a tic. The IOP CBIT allows patients to dedicate time specifically to detecting urges and tics and implementing the competing responses without the distractions of day-to-day life. To date, no studies have been published evaluating the effectiveness of an IOP CBIT. However, Flancbaum *et al*^[17] presented a case study detailing the outcome of a 25-year-old male diagnosed with TS who traveled to the United States in order to receive seven sessions of adapted CBIT over two weeks^[13]. The patient reported notable decreases in tic frequency and subjective distress and high treatment satisfaction at posttreatment, although he also reported a lapse in his tic symptoms when he returned home.

Little is known about the benefits of IOP CBIT, but there is precedence for treating children with an IOP behavioral program. For example, Whiteside and colleagues^[18] present a case series of three adolescents who received 10 sessions of exposure and response prevention for OCD over five days. Each of the three adolescents experienced a decrease in OCD symptoms at post-treatment, and two maintained gains after three months. Moreover, an IOP (one session) protocol has been used to treat specific phobia in children and has demonstrated efficacy in three randomized controlled trials (for review, see^[19]).

Goals of the case series

The current case series addresses an important limitation in the literature by examining whether IOP CBIT can help quickly reduce tic severity in two youth diagnosed with TS. Although the boys in the case series differed markedly by age, ethnicity, psychological symptoms, behavioral distress, and tic severity, and although they were treated by different treatment teams (see Table 1), both evidenced a notable reduction in tics and maintained their treatment gains. The patients and their parents provided written informed consent for this case series.

CASE REPORT

Patient A

Patient A (see Table 1) was a 10-year-old Asian-American male in the fourth grade. He was placed in the gifted-and-talented program and advanced mathematics. He maintained good grades but had occasional behavioral problems at school. He had several friends and was involved in piano, karate, and chess.

Patient A's tics were first noticed by his second grade teacher when he was 7 years old. He was evaluated by a neurologist and a psychologist a year prior to receiving IOP CBIT. Both diagnosed him with Tourette syndrome.

Table 1 Summary of patient information

	Patient A	Patient B
Age (yr)	10	14
Ethnicity	Asian-American	African-American
Academic history	4 th grade Gifted and talented	9 th grade Dysgraphia, low-intellectual functioning, and disorder of written expression
Psychological history	Tourette syndrome	Tourette syndrome, ADHD, specific phobia, anxiety, insomnia, and stuttering
Tic interference	Minimal	Significant
Type (number) of tics	Motor (5)	Motor (6) and vocal (4)
Treatment teams	Psychologist, psychology postdoctoral fellow, and psychology intern	Psychologist and two psychology postdoctoral fellows
Length of treatment	4 d, 8 treatment sessions	4 d, 3 treatment sessions

ADHD: Attention deficit hyperactive disorder.

Table 2 Patient A and B's outcome assessment scores

Measures	Patient A				Patient B								
	Baseline	1 wk posttreatment	1 mo follow-up	7 mo follow-up	Baseline		1 wk posttreatment		1 mo follow-up		6 mo follow-up		
	M	M	M	M	M	P	M	P	M	P	M	P	
YGSS													
Total	15	9	6	5	21	18	15	13	11	12	14	13	
Number	2	1	1	2	5	4	4	4	2	2	2	2	
Frequency	4	4	2	2	3	3	3	3	2	2	2	3	
Intensity	4	3	2	1	4	4	2	2	3	4	2	4	
Complexity	3	0	0	0	4	3	2	2	2	3	4	3	
Interference	2	1	1	0	5	4	4	2	2	1	4	1	
CGI-SI	4	3	2	2	5		3-4		3		4		
CGI-I		1	1	1	-		2		2		3		

M: Motor tic; P: Phonic tic; YGTSS: Yale Global Tic Severity Scale (Clinical Cut-off: 14), YGTSS subscales are out 5, with 0: None, 5: Severe; CGI-SI: Clinical Global Impressions-Severity of Illness (0: Not assessed; 1: Normal; 2: Borderline; 3: Mild; 4: Moderate; 5: Mark; 6: Severe; 7: Extreme); CGI-I: Clinical Global Impression-Improvement (0: Not assessed; 1: Very much improved; 2: Much improved; 3: Improved; 4: Minimal improvement; 5: No change; 6: Minimal worse; 7: Much worse; 8: Very much worse).

The neurologist recommended medication, which his parents decided against, and the psychologist recommended yoga and family therapy. They attended two sessions of family therapy but discontinued treatment after deciding that it was not helpful. After researching behavioral treatments on the Internet, Patient A's mother contacted one of the authors (ALP) to inquire about receiving CBIT for her son. Since the family would be required to travel to another city to receive CBIT, the staff and his mother agreed to use an IOP CBIT protocol. The patient presented for care in March 2013. At baseline, he and his mother reported that he experienced frequent facial tics that interfered with piano practice and chess competitions, but the tics did not interfere with his academic or social functioning. However, his mother was concerned that he would have tic-related social difficulties when he started middle school the following year.

Baseline assessment

A baseline assessment was conducted by a master's level independent evaluator (IE), who was not involved in the patient's treatment. The Yale Global Tic Severity Scale (YGSS)^[20] and the Clinical Global Impression Scale (CGI)^[21] were administered at baseline, posttreatment, and follow-up and were the main outcome measures for treat-

ment (see Table 2). The YGTSS, a semi-structured clinical interview, is routinely used in the TS literature and has well established psychometric properties (*e.g.*,^[14, 20, 22, 23]). It provides a Total Motor Tic Score (range: 0-25), Total Phonic Tic Score (range: 0-25), Total Tic Score (range: 0-50); past studies have used YGTSS Total Scores greater than 13 as a cut-off for clinically significant tics (> 9 if patient has only motor or vocal tics; *e.g.*,^[14]). A decrease of 4 points on the YGTSS is considered clinically meaningful in children^[14]. The YGTSS was conducted by the IE and was completed by the patient with the help of his mother. The CGI-S and CGI-I scales are well-established rating tools applicable to all psychiatric disorders^[21]. The CGI-S scale is used to assess treatment response in patients. The CGI-S requires the clinician to rate the severity of the patient's illness at the time of the assessment, relative to the clinician's past experience with patients who have the same diagnosis. The CGI-I requires the clinician to rate how much the patient's illness has improved or worsened relative to a baseline state.

The IE also administered the Kiddie Schedule for Affective Disorders and Schizophrenia-Present and Lifetime Version^[24], a semi-structured clinical interview designed to determine present episode and lifetime history of psychiatric illness based on the diagnostic criteria of the

Table 3 Overview of Patient A's treatment schedule

	Day 1	Day 2	Day 3	Day 4
Session Number	1:90 min	3:60 min	5:60 min	7:60 min
	Review history	Review OOSAs	Review OOSAs	Review OOSAs
	Treatment rationale	Inconvenience review	Inconvenience review	Inconvenience review
	Psychoeducation	Review treatment tic 1	Review treatment tic 1-3	Review treatment tic 1-3
	Tic hierarchy	Functional intervention	Functional intervention	Functional intervention
	Introduce function-based interventions	Competing response tic 2	Competing response tic 2	Competing response tic 2 Review relaxation
	Introduce reward program	Reward review	PMR	Relapse prevention
	Teach tic monitoring		Reward review	Reward review
Lunch (2 h)	Monitor tic 1	Monitor tics 1, 2	Monitor tics 1-3	Monitor tics 1-3
	Functional assessment	Practice CRs 1, 2	Practice CRs 1-3	Practice CRs 1-3
Session Number	2:90 min	4:60 min	6:60 min	8:60 min
	Review OOSAs	Review OOSAs	Review OOSAs	Review OOSAs
	Inconvenience review	Inconvenience review	Inconvenience review	Inconvenience review
	Functional assessment and treatment tic 1	Review treatment tic 1 and 2	Review treatment tic 1-3	Review treatment tic 1-3
	Competing response tic 1	Functional intervention tic 3	Functional intervention tic 2	Functional intervention
	Reward review	Competing response tic 3	Competing response tic 2	Competing response tic 2
		Introduce relaxation	Review relaxation	Review relaxation
		Diaphragmatic breathing	Reward review	Relapse prevention
		Reward review		Reward review
OOSAs	Practice CR for tic 1 Monitor 30 min	Practice CRs tics 1-3 Monitor 30 min Relaxed breathing	Practice CRs tics 1-3 Monitor 30 min Relaxation	Posttreatment assessment

CR: Competing response; OOSA: Out of session assignment; PMR: Progressive muscle relaxation.

Table 4 Patient's A tic symptom hierarchy tracker ratings

	S1	S3	S4	S5	S6	S7	S8	1 mo	6 mo
Eye Blink	6	6	4	4	3	1	2	0	1
Upper Lip	4	4	4	1	1	0	1	0	0
Facial Grimace	6	6	0	1	1	0	0	0	0
Neck Jerk	9	3	2	1	1	0	1	1	0
Nose Flair	4	4	0	0	0	0	0	0	0

Session 2 (S2) scores were not recorded; Subjective Unit of Distress Scale range from 0 to 10, with 0: No Distress; 10: Extreme distress.

Diagnostic and Statistical Manual of Mental Disorders, fourth edition, text revision^[25]. In addition, the patient completed the Premonitory Urge Scale^[26], the Child Yale-Brown Obsessive Compulsive Scale^[27,28], and the ADHD Interview^[29] at baseline. These commonly used measures were selected to provide a comprehensive evaluation of the patient's tic-related symptoms and psychiatric functioning.

The assessment confirmed a diagnosis of Tourette syndrome with evidence of clinically meaningful motor tics. He had a history of vocal tics but was not experiencing them at the time of the assessment. He did not endorse OCD symptoms and reported only minimal ADHD symptoms. Patient did not meet diagnostic criteria for other Axis I diagnoses.

Formulation, rationale, and treatment plan

Environmental and social factors are believed to play a significant role in tic manifestation^[22]. CBIT is an evidence-based behavioral treatment that recognizes and targets these factors. The patient completed eight ses-

sions (60 to 90 min each) of CBIT over four consecutive days (see Table 3). The protocol was administered by a treatment team, including two licensed psychologists and a pre-doctoral psychology intern.

Course of treatment

Psychosocial and tic history were gathered and treatment rationale was provided in Session 1. Consistent with the CBIT manual, Session 1 focused on information gathering and providing a treatment overview and rationale. Patient A's mother was already well versed on TS. Patient A and his mother identified five current motor tics (neck jerk, eye blink, upper lip tic, facial grimace, and nose flair) that occurred in isolation and as a single complex tic (see Table 4). Tic monitoring and the role of social support to encourage skill use were introduced and a reward program was established to reinforce treatment compliance.

The Tic Hassle worksheet^[30], the functional assessment procedure, and the competing response procedure were introduced in Session 2 and were conducted each session for the remainder of treatment. The Tic Hassle worksheet uses a Subjective Units of Distress Scale (SUDS), in which patients verbally rate their level of distress on an 11-point scale, with 0 representing minimal distress and 10 representing extreme distress. In Session 2, Patient A identified neck pain (SUDS = 9), people noticing (SUDS = 3), his grandfather staring at him (SUDS = 4), interruption of piano practice (SUDS = 6), and increasing the time it takes to complete school work (SUDS = 5) as tic hassles. By the end of the eighth session, he no longer experienced distress from these hassles. Patient A and his mother had difficulty completing the functional

assessment for the individual tics throughout treatment. They reported that the tics occurred with equal frequency across all settings and denied consequences following tic occurrence. Moreover, due to the format of treatment, they had little to no opportunity to implement relevant function-based interventions at home.

Competing response training focused on one tic at a time, starting with his most distressing tic (*i.e.*, neck jerk tic). This component of treatment requires that patients become more aware of their tics and premonitory urges. Consequently, Patient A was asked to describe his tic and its corresponding premonitory urge, identify each time he engaged in the tic during the training, and then identify each time he experienced the premonitory urge. Next, the patient and the provider collaboratively selected an appropriate competing response. Effective competing responses are physically incompatible with and less conspicuous than the tic, can be performed for at least 60 s, and do not disrupt normal activity^[13]. For example, the competing response for the patient's neck jerk tic involved having him gently move his chin forward and focusing on one spot each time he experience the tic or the premonitory urge. Self-monitoring indicated that his neck jerk tic occurred frequently (30 times in 5 min). By Session 4, he described decreased neck pain, and his mother reported a notable decrease in the neck jerk tic. The competing response training was implemented for his eye-blink and lip tics in Sessions 3 and 4, respectively. His competing responses included slow, rhythmic blinking for the eye tic and pursing his lips gently together for the lip tic. He demonstrated quick mastery over the lip tic but continued to have difficulty with his eye-blink tic. Consequently, the eye-blink tic remained the focus of CBIT for Sessions 5 to 8. By Session 8, both he and his mother reported improvement in his eye-blink tic, although his mother still occasionally had to prompt him to engage in his competing response.

Relaxation training was initiated in Session 4, and relapse prevention was discussed in Sessions 7 and 8. Since they had previously disagreed about what constituted a tic, both the patient and his mother were asked to discuss how they would handle new tics should they emerge. Ways to communicate about potential tics were explored, and a plan for this type of conversation was developed. At the end of treatment, both Patient A and his mother expressed high treatment satisfaction.

Patient B

Patient B (see Table 1) was a 14-year-old African-American male. He participated in a home school program in which he attended classes several times per week outside his home and was also involved in track and field. At the time of the intervention, Patient B had not shared information about his diagnosis with peers. As a result, he often suppressed his vocal and motor tics when around peers and then released his urges to tic at home. At intake, Patient B's vocal and motor tics had occurred for approximately six months. He had already been evaluated

by pediatric neurology and developmental pediatrics and was prescribed methylphenidate for ADHD, clonidine for tics, and melatonin for sleep. Patient B completed an magnetic resonance imaging and electroencephalography with pediatric neurology, and it was determined that he did not present with epilepsy or other neurological concerns. When the tics were unresponsive to medication intervention, Patient B and his family were referred for behavioral treatment. Patient B's psychological history was positive for developmental delays, ADHD, and learning difficulties, with no prior history of tics. He had previously undergone treatment for specific phobia and stuttering.

Patient B presented for behavioral treatment in Spring 2012. Given the severity of his behaviors at his initial appointment (*i.e.*, grabbing his mother's arm, punching the floor, and difficulty starting and stopping movements), consultation with ALP was sought regarding the appropriateness of outpatient services. It was determined that he might benefit from an intensive outpatient treatment protocol, which started in Summer 2012.

Baseline assessment

A baseline assessment of Patient B's current functioning was conducted by a master's level IE, who was not involved in the treatment delivery, and included the YGTSS^[20] and the clinician-rated CGI^[21] and the Hopkins Motor/Vocal Tic Scale (HMVTS)^[31]. Only the YGTSS and CGI were used as outcome assessments, and additional information about their psychometric properties can be found under Patient A's baseline assessment section. The assessment confirmed the diagnosis of TS and indicated that Patient B was experiencing clinically significant vocal and motor tics (as defined^[14]). Specifically, Patient B and his parents reported six motor and four vocal tics, which significantly interfered with family interactions and had begun to interfere with his peer relationships.

Formulation, rationale, and treatment plan

The treatment team met with Patient B and his family to discuss treatment options including IOP CBIT. The family had already exhausted many other options in the community with little to no success, and his parents were hopeful that this alternative approach would alleviate his symptoms. Given the severity of his symptoms, Patient B continued to take clonidine during his participation in IOP CBIT.

Patient B and his parents attended one baseline assessment session and three IOP CBIT sessions over the course of four consecutive days. Although only three IOP CBIT sessions were conducted, the total amount of time spent for the intervention was comparable to that of the standard eight session CBIT protocol. During the course of treatment, Patient B's parents observed through a one-way mirror. Treatment was administered by a team consisting of a board certified child and adolescent psychologist and two child and adolescent post-doctoral fellows (see Table 5 for a summary of the specific treatment schedule).

Table 5 Overview of Patient B's treatment schedule

	Day 1	Day 2	Day 3	Day 4
Session Number	S1: (1.5 h)	S2: (3.5 h)	S3: (3.5 h)	S4: (3.5 h)
	Baseline assessment	Psychoeducation	Review relaxation OOSAs	Competing response
	Meet therapists	Introduce relaxation	Tic hassles form	Inconvenience review
	Introductions	Stress <i>vs</i> relaxation	Competing responses	Relaxation practice
	Treatment overview	Relaxation postures	Inconvenience review	Review OOSAs
		PMR + 12	Review tic 1 and 2	Review CR for all tics
		Diaphragmatic breathing	Competing responses 1, 2	Summarize progress
		Visual imagery	Review treatment tic 1	Emphasize social support
		Awareness training	Practice relaxation	Reward review
		Psychoeducation about tics	Competing response 1 and 2	
		Rationale for treatment	Assign homework	
		Tic Sx hierarchy		
		Feedback about assessment		
OOSAs		Practice relaxation	CR tics 1-2	F(x) based interventions
		Practice PRM + 12	Monitor 15 min, 3-4x	Relaxation
		Practice visual imagery	F(x) based interventions	Family and social support
			Relaxed breathing	

PMR: Progressive muscle relaxation; Sx: Symptoms; CR: Competing response; OOSA: Out of session assignment; F(x) = Function.

Course of treatment

During the baseline assessment, Patient B and his parents identified six current motor tics (grabbing/touching, putting napkins in mouth, full-body twitches, open mouth with head nodding, "closing" self into small spaces, tapping surfaces) and four vocal tics (screaming, humming, repeating self, and "Aahh" sounds). The treatment agenda, rationale, tic monitoring, and family support were discussed with Patient B and his parents.

CBIT was initiated in Session 2. The treatment team provided psychoeducation about tic disorders, the rationale for competing response training, awareness training, and the stress and relaxation responses. The team also engaged the patient in several relaxation strategies and progressive muscle relaxation, which yielded a notable decrease in his tics. A hierarchy of the patient's current tics was developed. Patient B was assigned relaxation and tic monitoring homework, which was reviewed at the start of the next session (see Table 5).

The following day, Patient B reported some benefit with homework while his mother reported a reduction in the severity and intensity of the grabbing tic in public places. Session 3 focused on completing the tic hassles worksheet^[30], in which Patient B described the grabbing tic and vocal tic as most bothersome. He identified arm pain (SUDS = 9), parental dependence (SUDS = 11), and annoying others (SUDS = 7) as tic hassles. Patient B reported that his vocal tic was embarrassing (SUDS = 10). Competing response training was implemented with one tic at a time, starting with the most distressing tic (grabbing), followed by the vocal tic. For the grabbing tic, Patient B was asked to practice squeezing his hands together and pushing them down as a competing response. For the vocal tic, Patient B was instructed to clench his teeth and push his tongue against the roof of his mouth as his competing response.

On Day 4, the treatment team reviewed the homework, in which the patient's mother observed only one

vocal tic during several discrete tic observation periods. Patient B reported not having any tics or urges while at a friend's house, and he stated that he did not feel as though he was suppressing his tics. Because Patient B was still reporting some difficulty identifying premonitory urges, a token economy was also implemented during the session whereby Patient B earned points towards a desirable reward for detecting a premonitory urge by notifying the provider (*i.e.*, raising his finger) and engaging in the appropriate competing response. Patient B responded positively to the token economy and was motivated to identify premonitory urges. He was also able to resist the urge to tic or engaged in the tic for markedly less time compared to pretreatment. At the end of the session, Patient B and his mother reported improvement in the awareness of his tics and premonitory urges. The treatment team also practiced relaxation strategies, summarized treatment progress, and discussed relapse prevention during the last part of the session. Providers emphasized the importance of ongoing social and family support.

Over four days of assessment and treatment, behavioral improvement was observed and noted by all three providers, Patient B, and his parents. Patient B reported feeling skeptical at the beginning of the week about whether this treatment would be effective, but at the last session he stated, "I stand corrected." Patient B was able to control his tics by either stopping them from occurring, notifying providers when he was about to have one, and/or decreasing the length of time spent engaging in specific tic behaviors. Overall, Patient B and his parents verbally reported high treatment satisfaction.

Results

Patient A: By the end of treatment, Patient A and his mother reported a clinically meaningful decrease in his tic severity as assessed by the YGTSS and the CGI. Importantly, his tic severity scores had decreased further by the

one-month follow-up (see Table 2). Following their one-month follow-up assessment, Patient A and his mother attended a 60-min booster session, in which his mother reported that she only occasionally noticed a slight eye-blink tic. Patient A disagreed with his mother that this was a tic. He also reported that although he still experienced an urge to tic, the urge was less severe and occurred less frequently. Both Patient A and his mother reported continued treatment satisfaction with IOP CBIT at follow-up. Patient A and his mother reported continued treatment gains at the seven-month follow-up and high treatment satisfaction. More specifically, he reported that he continued to occasionally experience a slight eye tic. However, both he and his mother agreed that the tic was not noticeable to others and did not cause him interference. No new tics emerged from the time of treatment completion through the seven-month follow-up period.

Patient B: The YGTSS (see Table 5) and HMVTS were administered by the same IE and completed by Patient B with the help of his parents at one week, one month, and six months. Overall, the assessments revealed clinically meaningful improvement in Patient B's functioning (see Table 2). At the one-week follow up, there was an overall reduction in number of tics (10 *vs* 2). The parents reported that Patient B had not engaged in the grabbing tic in the previous week, that he was more aware of the urge to grab, and that he was able to apply a more appropriate competing response (*i.e.*, walking away, distraction, breathing). During follow-up interviews, Patient B was observed using several appropriate competing responses (*i.e.*, crossing his arms, sitting on his hands) in reaction to the urge to grab others. Patient B also reported feeling "a lot better," stating, "I don't really tic as much." He also reported that the duration of his tics had decreased, that he was less bothered by his tics, and that the tics were less noticeable to others in public. The parents confirmed his impressions. Patient B was also provided with additional suggestions on competing responses to use for the remaining tics. The treatment team reviewed the follow-up plan with parents, which included booster sessions.

At the one-month follow-up, the YGTSS revealed that treatment gains had been maintained, and Patient B demonstrated a reduction in tic number, frequency, and interference of both motor and vocal tics. By the six-month follow-up, Patient B was exhibiting a slight increase in the frequency of vocal tics and an increase in the complexity and interference of motor tics (see Table 2); however, Patient B admitted to not practicing the breathing and relaxation strategies. Therefore, booster sessions were scheduled to review IOP CBIT components.

Although Patient B's presentation at six months post-treatment revealed some regression (as seen in Table 2), the family expressed their appreciation for Patient B's progress and his ability to function better at home and at school. The family also stated that the tics had become "so subtle" that he was no longer concerned or upset by them.

DISCUSSION

The current case series describes the implementation of an intensive outpatient behavioral treatment with two preadolescents who presented with Tourette syndrome. Despite their different presentations, both patients demonstrated treatment gains following the IOP CBIT intervention. The generalizability of the current case series is unknown at this time. However, IOP CBIT may be appropriate for individuals who present with moderate to severe tics, those who are experiencing clinically significant impairment in daily academic and social functioning, and for individuals and their families who desire to experience a quick reduction in motor and/or vocal tics. On the other hand, individuals who might not be good candidates for CBIT include those with oppositional and/or defiant behaviors, since adherence to the treatment protocol would likely prove to be a challenge. In addition, because it is important for individuals with Tourette's to receive adequate psychosocial support in monitoring and reducing their tics, those with chaotic or limited family and social support systems may find this protocol challenging.

It should be noted that many individuals with Tourette syndrome experience a waxing and waning of symptoms over time and that many tics resolve on their own^[2,3]. Although CBIT is not a cure for Tourette syndrome, based on the current case series, individuals who follow the treatment protocol can expect to learn tools and skills to better manage their tics, understand their premonitory urges, reduce the negative impact of the tics on their lives, and experience improvement in their overall academic and social functioning^[14].

Limitations and future directions

Despite addressing an important gap in the literature pertaining to the use of an intensive outpatient CBIT approach with children and adolescents, there are several limitations to the current case series that should be noted. First, an intensive treatment approach requires a time commitment from parents and patients that would likely require a parent to request time away from work and/or a child to be absent from school. This might present a financial challenge for some parents and possibly create academic stressors for some children. It also raises the question about the most convenient time to deliver an intensive outpatient intervention for children and adolescents. That is, Patient A received his treatment during a planned school break, while Patient B was seen during the summer. Clinicians should consider and discuss the time commitment it takes for families to participate in this type of intensive treatment approach.

Second, both patients received treatment at academic medical centers without individual fee-for-service costs as a part of psychology internship and postdoctoral training programs. It is possible that many families might find paying out-of-pocket for an intensive outpatient treatment to be a financial burden. Moreover, with the increased limitations placed on behavioral health services by managed

care organizations, insurance companies might be unable or unwilling to pay for an intensive outpatient program. Future research should examine the generalizability of a comprehensive behavioral intervention for TS in the community at large. Third, receiving treatment as part of a research study or through a military treatment facility would also facilitate access to services for this population. While members of the current treatment teams received training, consultation, and/or supervision from one of the leading researchers (ALP) in the field of TS, accessibility of behavioral health providers who are trained in CBIT might be more limited in other geographical areas. Both families reported feeling grateful that they had access to the current treatment teams. Agencies and educational institutions would greatly benefit the community by offering more training opportunities for behavioral health providers in the treatment of TS. The Tourette Syndrome Association has sponsored many CBIT training programs.

Finally, the intensive outpatient program implemented in the current treatment protocol might compromise the external validity of the intervention. Both patients received the intensive treatment at a much faster pace compared to traditional therapy, creating an artificial environment in which to practice the skills learned. This limited both patients' ability to practice the functional based interventions in their everyday environments at a more natural pace. Future research should continue to examine the generalizability and long-term benefits of IOP CBIT. Future research should also consider a single subject research design or an experimental research design to include a control group receiving traditional weekly CBIT with the experimental group receiving IOP CBIT over 3-4 d.

The current case series adds an important piece to the scientific literature on the behavioral treatment of Tourette syndrome and persistent tic disorders by demonstrating that Cognitive Behavioral Intervention for Tics employed as part of an intensive outpatient program can reduce tic severity. The use of an intensive outpatient program incorporating Comprehensive Behavioral Intervention for Tics appears to offer several benefits. First, the patients in this case study were able to make notable progress over the span of 1 wk *vs* 10 wk. Additionally, IOP CBIT allows patients to focus almost exclusively on developing and practicing their competing responses without the interference of work or school. IOP CBIT also expands the potential treatment catchment areas, which would make CBIT more accessible to a wider range of patients who would otherwise be limited by geography or expense. Importantly, an IOP CBIT has the potential to help compensate for the current lack of CBIT providers.

COMMENTS

Case characteristics

Both Patients A (10-year-old male) and B (14-year-old male) experienced multiple tics that were consistent with Tourette syndrome.

Clinical diagnosis

Patients A and B both met diagnostic criteria for Tourette syndrome.

Differential diagnosis

Patients A and B were both assessed for attention deficit hyperactive disorder and obsessive compulsive disorder.

Treatment

Patients completed intensive outpatient Comprehensive Behavioral Interventions for Tics (CBIT).

Related reports

Although previous studies support the use of CBIT, when delivered in eight weekly sessions, more research is needed to determine whether an intensive outpatient format can improve tic management in children with a persistent tic disorders; however, the treatment outcome of these two cases are promising.

Experiences and lessons

This case series represents the first report of treatment outcomes following an intensive outpatient CBIT protocol for children. Although future research is required before more definitive conclusions can be reached, the findings of this case series suggest that Intensive Outpatient Program CBIT may reduce tic symptoms in children with Tourette syndrome.

Peer review

This is a template for a valuable modification of CBIT for those who desire thorough management in a short period of time. This represents a promising approach that merits confirmation by other investigators in other settings.

REFERENCES

- 1 **American Psychiatric Association.** Diagnostic and statistical manual of mental disorders, Fifth edition. Arlington, VA: American Psychiatric Association, 2013
- 2 **Jankovic J.** Tourette's syndrome. *N Engl J Med* 2001; **345**: 1184-1192 [PMID: 11642235 DOI: 10.1056/NEJMra010032]
- 3 **Leckman JF, Zhang H, Vitale A, Lahnin F, Lynch K, Bondi C, Kim YS, Peterson BS.** Course of tic severity in Tourette syndrome: the first two decades. *Pediatrics* 1998; **102**: 14-19 [PMID: 9651407 DOI: 10.1542/peds.102.1.14]
- 4 **Specht M, Woods D, Piacentini J, Scahill L, Wilhelm S, Peterson A, Walkup J.** Clinical characteristics of children and adolescents with a primary tic disorder. *J Dev Phys Disabil* 2011; **23**: 15-31 [DOI: 10.1007/s10882-010-9223-z]
- 5 **Bloch MH, Leckman JF.** Clinical course of Tourette syndrome. *J Psychosom Res* 2009; **67**: 497-501 [PMID: 19913654 DOI: 10.1016/j.jpsychores.2009.09.002]
- 6 **Rizzo R, Gulisano M, Cali PV, Curatolo P.** Long term clinical course of Tourette syndrome. *Brain Dev* 2012; **34**: 667-673 [PMID: 22178151 DOI: 10.1016/j.braindev.2011.11.006]
- 7 **Scahill L, Erenberg G, Berlin CM, Budman C, Coffey BJ, Jankovic J, Kiessling L, King RA, Kurlan R, Lang A, Mink J, Murphy T, Zinner S, Walkup J.** Contemporary assessment and pharmacotherapy of Tourette syndrome. *NeuroRx* 2006; **3**: 192-206 [PMID: 16554257 DOI: 10.1016/j.nurx.2006.01.009]
- 8 **Peterson AL, Azrin NH.** Behavioral and pharmacological treatments for Tourette syndrome: A review. *Appl Prev Psychol* 1993; **2**: 231-242 [DOI: 10.1016/S0962-1849(05)80093-9]
- 9 **Peterson AL, Campise RL, Azrin NH.** Behavioral and pharmacological treatments for tic and habit disorders: a review. *J Dev Behav Pediatr* 1994; **15**: 430-441 [PMID: 7884015 DOI: 10.1097/00004703-199412000-00007]
- 10 **Peterson AL.** Psychosocial management of tics and intentional repetitive behaviors associated with Tourette syndrome. In Woods DW, Piacentini JC, Walkup JT (Eds), *Treating Tourette syndrome and tic disorders: A guide for practitioners* (pp. 154-184). New York: Guildford Press, 2007
- 11 **Azrin NH, Nunn RG.** Habit reversal: A method of eliminating nervous habits and tics. *Behav Res Ther* 1973; **11**: 619-628 [DOI: 10.1016/0005-7967(73)90119-8]
- 12 **Verdellen C, van de Griendt J, Hartmann A, Murphy T.** European clinical guidelines for Tourette syndrome and other tic disorders. Part III: behavioural and psychosocial interven-

- tions. *Eur Child Adolesc Psychiatry* 2011; **20**: 197-207 [PMID: 21445725 DOI: 10.1007/s00787-011-0167-3]
- 13 **Woods DW**, Piacentini JC, Chang SW, Deckebach T, Ginsburg GS, Peterson AL, Wilhelm S. Managing Tourette syndrome: A behavioral intervention for children and adults. Therapist guide. New York: Oxford University Press, 2008
 - 14 **Piacentini J**, Woods DW, Scahill L, Wilhelm S, Peterson AL, Chang S, Ginsburg GS, Deckebach T, Dziura J, Levi-Pearl S, Walkup JT. Behavior therapy for children with Tourette disorder: a randomized controlled trial. *JAMA* 2010; **303**: 1929-1937 [PMID: 20483969 DOI: 10.1001/jama.2010.607]
 - 15 **Wilhelm S**, Peterson AL, Piacentini J, Woods DW, Deckebach T, Sukhodolsky DG, Chang S, Liu H, Dziura J, Walkup JT, Scahill L. Randomized trial of behavior therapy for adults with Tourette syndrome. *Arch Gen Psychiatry* 2012; **69**: 795-803 [PMID: 22868933 DOI: 10.1001/archgenpsychiatry.]
 - 16 **Steeves T**, McKinlay BD, Gorman D, Billingshurst L, Day L, Carroll A, Dion Y, Doja A, Luscombe S, Sandor P, Pringsheim T. Canadian guidelines for the evidence-based treatment of tic disorders: behavioural therapy, deep brain stimulation, and transcranial magnetic stimulation. *Can J Psychiatry* 2012; **57**: 144-151 [PMID: 22398000]
 - 17 **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 [DOI: 10.1007/s10882-010-9222-0]
 - 18 **Whiteside SP**, Brown AM, Abramowitz JS. Five-day intensive treatment for adolescent OCD: a case series. *J Anxiety Disord* 2008; **22**: 495-504 [PMID: 17543497 DOI: 10.1016/j.janxdis.2007.05.001]
 - 19 **Davis TE 3rd**, Ollendick TH, Ost LG. Intensive Treatment of Specific Phobias in Children and Adolescents. *Cogn Behav Pract* 2009; **16**: 294-303 [PMID: 20161063 DOI: 10.1016/j.cbpra.2008.12.008]
 - 20 **Leckman JF**, Riddle MA, Hardin MT, Ort SI, Swartz KL, Stevenson J, Cohen DJ. 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 [PMID: 2768151 DOI: 10.1097/00004583-198907000-00015]
 - 21 **Guy W**, Bonato R. Manual for the ECDEU Assessment Battery (2nd ed.). Chevy Chase, MD: U.S. Department of Health, Education, and Welfare, National Institute of Mental Health, 1970
 - 22 **Caurin B**, Serrano M, Fernández-Alvarez E, Campistol J, Pérez-Dueñas B. Environmental circumstances influencing tic expression in children. *Eur J Paediatr Neurol* 2014; **18**: 157-162 [PMID: 24210363 DOI: 10.1016/j.ejpn.2013.10.002]
 - 23 **Storch EA**, Murphy TK, Geffken GR, Sajid M, Allen P, Roberti JW, Goodman WK. Reliability and validity of the Yale Global Tic Severity Scale. *Psychol Assess* 2005; **17**: 486-491 [PMID: 16393016 DOI: 10.1037/1040-3590.17.4.486.]
 - 24 **Kaufman J**, Birmaher B, Brent D, Rao U, Flynn C, Moreci P, Williamson D, Ryan N. Schedule for Affective Disorders and Schizophrenia for School-Age Children-Present and Lifetime Version (K-SADS-PL): initial reliability and validity data. *J Am Acad Child Adolesc Psychiatry* 1997; **36**: 980-988 [PMID: 9204677 DOI: 10.1097/00004583-199707000-00021]
 - 25 **American Psychiatric Association**. Diagnostic and statistical manual of mental disorders (4th ed., text rev.). Washington, DC: Author, 2000
 - 26 **Woods DW**, Piacentini J, Himle MB, Chang S. Premonitory Urge for Tics Scale (PUTS): initial psychometric results and examination of the premonitory urge phenomenon in youths with Tic disorders. *J Dev Behav Pediatr* 2005; **26**: 397-403 [PMID: 16344654 DOI: 10.1097/00004703-200512000-00001]
 - 27 **Goodman WK**, Price LH, Rasmussen SA, Mazure C, Fleischmann RL, Hill CL, Heninger GR, Charney DS. The Yale-Brown Obsessive Compulsive Scale. I. Development, use, and reliability. *Arch Gen Psychiatry* 1989; **46**: 1006-1011 [PMID: 2684084 DOI: 10.1001/archpsyc.1989.01810110048007]
 - 28 **Goodman WK**, Price LH, Rasmussen SA, Mazure C, Delgado P, Heninger GR, Charney DS. The Yale-Brown Obsessive Compulsive Scale. II. Validity. *Arch Gen Psychiatry* 1989; **46**: 1012-1016 [PMID: 2510699 DOI: 10.1001/archpsyc]
 - 29 **Scahill L**, Chappell PB, Kim YS, Schultz RT, Katsovich L, Shepherd E, Arnsten AF, Cohen DJ, Leckman JF. A placebo-controlled study of guanfacine in the treatment of children with tic disorders and attention deficit hyperactivity disorder. *Am J Psychiatry* 2001; **158**: 1067-1074 [PMID: 11431228 DOI: 10.1176/appi.ajp.158.7.1067]
 - 30 **Woods D**, Piacentini J, Chang S, Deckebach T, Ginsburg GS, Peterson A, Wilhelm S. Managing Tourette Syndrome: A Behavioral Intervention. Parent Workbook. New York: Oxford University Press, 2008
 - 31 **Walkup JT**, Rosenberg LA, Brown J, Singer HS. The validity of instruments measuring tic severity in Tourette's syndrome. *J Am Acad Child Adolesc Psychiatry* 1992; **31**: 472-477 [PMID: 1592779 DOI: 10.1097/00004583-199205000-00013]

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