

Behavior Therapy for the Treatment of Tourette's Disorder in India: A Patient Series from an Indian General Hospital Psychiatric Unit

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ABSTRACT

Reports on behavioral interventions for the treatment of Tourette's disorder (TD) from India are limited. This patient series describes the usefulness and feasibility of conducting behavioral interventions for patients with TD from an Indian general hospital psychiatric unit. Behavioral treatments in these seven consecutively treated adult/adolescent patients with TD included all components of habit reversal treatment, comprehensive behavioral intervention for tics, and exposure with response prevention in some patients. Patients were predominantly male, with adolescent-onset severe TD, typical features and psychiatric comorbidities, and poor response to multiple medications prior to the institution of behavior therapy. In addition to long delays in diagnosis, none of the patients or their caregivers had been informed by the doctors they had consulted earlier about TD or the need for behavioral treatments before attending our center. Institution of behavioral

treatments along with medications led to a 75% reduction in the severity of tics and reduction in comorbid symptoms. Patients and caregivers also reported similar rates of improvement as well as reductions in subjective distress and caregiver burden. Five patients have been followed up for seven months to seven years; apart from one patient, all others have had only minor exacerbations of tics during this period. This limited experience suggests that behavior therapies for TD can be successfully implemented in low-resource, non-specialized Indian settings. They are effective, and gains from such treatment are usually enduring.

Keywords: Tourette's disorder, behavior therapy, India

Tourette's disorder (TD) is a childhood-onset complex neurodevelopmental disorder characterized by multiple motor and vocal tics, frequently accompanied by comorbid psychiatric disorders.¹ The overall international prevalence of TD has

been estimated to be around 1% (range 0.4%–3.8%).² Although data from Asian countries is scarce, prevalence rates of 0.4%–0.56% have been reported.^{2,3} This suggests that TD is not rare, even among Asian populations.

Several behavioral treatments have been tried for the management of TD. These include habit reversal treatment (HRT), comprehensive behavioral intervention for tics (CBIT), exposure with response prevention (ERP), massed negative practice, self-monitoring, contingency management, relaxation training, cognitive behavioral treatment, biofeedback, and assertiveness training. However, the only ones with proven efficacy are HRT, CBIT, and ERP.^{4–8} Therefore, the current consensus on management of TD recommends psychoeducation when tics are transient or mild, behavioral interventions for tics of moderate severity, and combined treatment with medications (alpha-2 agonists or antipsychotics)

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and HRT, CBIT, or ERP when the tics are severe.⁴⁻⁶

The principal components of HRT are awareness training (self-detection of premonitory urges and tics), competing response training (learning voluntary behaviors physically incompatible with tics), and caregiver support (enhancing the patients' awareness and encouragement by caregivers). CBIT utilizes these core HRT components along with relaxation training, psychoeducation, and functional intervention (identifying events that worsen tic severity and developing strategies to manage them in daily life). ERP involves exposure to premonitory urges while refraining from indulging in tics.

Existing Indian literature on TD consists of one case series⁹ and about a dozen case reports. Most patients appear to have been treated with medications, with the administration of repetitive transcranial magnetic stimulation (rTMS) or deep brain stimulation in patients resistant to medications.^{10,11} Although HRT is mentioned in some instances,^{12,13} there is virtually no information on the behavioral management of TD. Therefore, there is a need to examine whether behavioral treatments are equally effective for TD in routine clinical settings in India.

This patient series describes seven consecutive patients from our center who were treated with a combination of behavioral techniques and medications. This report, representing our experience of behavior therapy for TD, is presented to demonstrate that behavioral interventions are feasible and effective even in low-resource, non-specialized settings such as general hospital psychiatric units in India.

Methods

This patient series describes behavior therapy carried out in seven consecutively treated adult/adolescent patients with TD attending the general hospital psychiatric unit of a multispecialty hospital in North India. The CARE guidelines for consensus-based clinical case reporting have been followed for this report.

Behavioral treatments were conducted by different trainee psychiatrists under the supervision of senior residents well versed in the behavioral management

of TD and the consultant psychiatrist. Interventions were based on principles of HRT, CBIT, and ERP. A standardized treatment protocol was followed for each patient. To begin with, TD was diagnosed according to the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV-TR or DSM-5) criteria. All possible alternative conditions were ruled out by careful history-taking, physical examination, and carrying out the necessary investigations. After establishing the diagnosis, a comprehensive clinical, behavioral, and psychosocial assessment of TD and comorbidities was carried out. Behavioral assessments followed the standard "A-B-C" approach, in which the types of tics, their antecedents, and consequences, including the associated subjective distress, environmental and other contextual factors influencing the occurrence of tics, and the motivation for treatment, were evaluated. Psychosocial assessments included (unstructured) evaluation of both patients and their caregivers about their knowledge regarding TD, emotional problems, disability caused by tics, social impact of tics, and caregiver burden. The Yale Global Tic Severity Scale (YGTSS) was used to rate tic severity. The Yale-Brown Obsessive Compulsive Scale (Y-BOCS) was used to rate the severity of obsessive compulsive disorder (OCD), and the Revised Connor's Parent Rating Scale was used to rate the severity of attention-deficit hyperactivity disorder (ADHD). Both patients and caregivers were educated about rating the progress of the treatment themselves.

Assessments were followed by the education of patients and caregivers about both TD and the behavioral treatments to be implemented. Relaxation training was undertaken next, as it forms a part of CBIT. Either autogenic training or Jacobson's progressive muscular techniques for relaxation were taught. Awareness training, consisting of self-monitoring by the patients with the caregivers' help, to enable early detection of premonitory urges and tics, was instituted after that. Competing responses were decided upon after discussions with patients and caregivers and then practiced in the presence of the trainees and caregivers. Individual support consisted of conventional supportive and expressive techniques to

enhance the patient's motivation for treatment and coping with the emotional consequences of tics. A typical behavioral treatment session lasted about 45 minutes. Response to behavioral interventions was monitored regularly by the trainees, the patients, and the caregivers; changes to the treatment plan were made based on the patients' needs. Both during the inpatient and follow-up phases, the generalization of the response was ensured by teaching the patients to manage tics triggered by events in the daily life. Caregivers were involved at each stage of the treatment. They were trained to gently point out the occurrence of tics to the patients, thereby enhancing their awareness of the tics. They assisted patients in practicing competing responses and provided encouragement and support. Caregivers were particularly encouraged to conduct sessions of their own so that they could become effective cotherapists once the patients went home. Additionally, their emotional and practical difficulties in caring for the patient were addressed by providing ongoing support. In one patient who had obvious triggers for tics, ERP was carried out in addition to HRT and CBIT. ERP for OC symptoms was also conducted in all four patients with OCD.

Two patients responded well to clonidine and did not require antipsychotics, which are usually second-line options for the pharmacological treatment of TD. Other patients received either clonidine and/or antipsychotics for tics. All patients received medications for the treatment of comorbid conditions. Three patients required changes in the medications while undertaking behavior therapy. Patients' written informed consent was obtained. Patient anonymity has been preserved.

Results

Table 1 depicts the details of clinical features and behavioral interventions.

Six of the seven patients were hospitalized for behavioral treatment after inadequate response to initial pharmacotherapy. All but one were male. They were all educated and mostly students. The mean age at presentation (\pm SD) was 24.42 (\pm 6.35) years. The mean age of onset of tics was 13.86 (\pm 4.88) years, but a proper diagnosis of TD was made only

TABLE 1.

Details of Behavioral Interventions for Tourette's Disorder

	Age (years)/sex	Age of onset (years)	Psychiatric comorbidity	Symptom profile*	Daily doses of medications	YGTSS Global severity score (maximum 100)		No. of BI sessions	Patients' reports of reduction in distress due to tics	Caregivers' overall estimate of the response to behavior therapy	Post-intervention follow-up
						Pre-treatment	Post-treatment				
1.	30 male	16	Schizophrenia OCD	Motor tics [†] Phonic tics (throat clearing, unintelligible sounds), self-injury	Clonidine 0.5 mg Clozapine 125 mg Sertraline 150 mg	90	24	120	60%	55%	Duration 3.5 years—major relapse once; responded to rTMS and change in medications
2.	17 male	12	ADHD OCD	Motor tics [†] Phonic tics (throat clearing, unintelligible sounds)	Clonidine 0.4 mg Methylphenidate 15 mg	73	24	40	70%	75%	Dropped out
3.	20 female	12	Dissociative disorder	Motor tics [†] Phonic tics (throat clearing)	Clonidine 0.3 mg	66	8	40	80%	80%	Dropped out
4.	22 male	13	OCD	Motor tics [†] Phonic tics (unintelligible sounds)	Haloperidol 0.75 mg Cloimipramine 75 mg	77	23	55	70%	75%	Duration 7 years—no relapses; improved functioning
5.	32 male	14	Generalized anxiety disorder and social phobia	Motor tics [†] Phonic tics (throat clearing)	Clonidine 0.3 mg Fluoxetine 20 mg	80	10	40	90%	90%	Duration 7 years—minor exacerbations when stressed; anxiety symptoms persist
6.	31 male	23	Major depression	Motor tics [†] Phonic tics (throat clearing), self-injury	Risperidone 2mg Escitalopram 20 mg	73	10	60	90%	80%	Duration 2 years—minor exacerbations when non-adherent
7.	19 male	7	OCD	Motor tics [†] Phonic tics (throat clearing)	Haloperidol 1.25 mg Escitalopram 20 mg	80	36	40	70%	80%	Duration 7 months—minor stress-related exacerbations

ADHD = attention-deficit hyperactivity disorder, BI = behavioral intervention, ERP = exposure and response prevention, OCD = obsessive compulsive disorder, rTMS = repetitive transcranial magnetic stimulation, YGTSS = Yale Global Tic Severity Score. *Other features included waxing and waning course, stress-related exacerbation of symptoms, ability to partially suppress tics voluntarily, premonitory urges, "just right" phenomenon. [†]Motor tics included shoulder, movements of limbs, head/neck, mouth, face and abdomen, eye blinking, repeated tapping, bowing, and touching.

by a mean age of 22.15 (\pm 5.25) years. In addition to the long delay in diagnosis, none of the patients and their caregivers had been informed about TD or the need for behavioral treatments by the doctors they had consulted earlier. All patients had at least one comorbid psychiatric diagnosis; the most common one was OCD, in four patients. The clinical profile

of TD was quite characteristic, with typical and common motor and phonic tics, premonitory urges, "just right" phenomenon, and the ability to partially suppress tics voluntarily. Other features included worsening with stress and associated problems such as self-injury.

Before starting the behavioral treatments, the response to multiple trials

of medication treatment among the patients had been poor. Accordingly, the tics were of moderate to severe intensity before starting behavior therapy. Mean pre-treatment YGTSS scores was 77 ± 7.53 , and all patients had a severe psychosocial impairment. According to their subjective estimates, there was considerable distress and burden among the caregivers.

Only one patient required 120 sessions of behavioral treatment. Managing his TD proved particularly difficult, because not only did he suffer from comorbid schizophrenia and OCD, but his pretreatment YGTSS score was also the highest (90). The rest of the patients required an average of about 46 sessions (45.83±9.17; range: 40–60) to achieve a reasonable response.

Treatment response was defined as a global severity score of 40 or less on the YGTSS, which corresponded with the presence of minimal/mild severity of tics and minimal/mild psychosocial impairment.¹⁴ According to this cutoff, a response was achieved in all the patients. Average YGTSS scores fell to 19.28±10.31 following treatment. The change in YGTSS scores (58/77) represented a 75% decrement in the symptom severity. In the four patients with OCD, Y-BOCS scores also decreased by about 64% from pretreatment scores of 28.0±7.0 to the posttreatment scores of 10.0±4.1. Symptoms of ADHD reduced by about 30% in the single patient with this co-morbidity. Patients reported an average reduction of 76% (range 60%–90%) in subjective distress due to the tics. Caregivers also reported an overall reduction of 76% in symptom severity. Additionally, the caregivers felt that the educative and supportive sessions had helped in reducing their psychological distress and burden of care. Thus, it was evident that all these patients who had responded poorly to initial medication treatment showed much better responses after the addition of behavioral treatments to their medication regimes.

Prospective follow-up was carried out for all the patients. However, two patients dropped out in the first few months after the completion of treatment. Five patients have remained on active follow-up for durations ranging from about seven months to seven years. Only one has suffered a major relapse, that is, a complete return of symptoms to the pretreatment levels. However, he responded well to rTMS and a change of medications. The rest have had minor exacerbations (i.e., the reappearance of 1–2 tics of milder intensity), which have responded to booster sessions of behavior therapy during outpatient follow-ups.

The booster sessions included all components of HRT and CBIT.

Discussion

All the seven patients of this study had a fairly typical picture of TD. The slightly later age of onset was in keeping with Indian reports^{9,12} and within the range of 4–15 years reported in cross-cultural studies among Asian populations.³ However, it appeared that minor tics such as eye blinking at younger ages had been missed in some patients because of the lack of awareness among the parents. The overwhelming male preponderance found in this report has been noted in cross-cultural studies but could also reflect the gender differences in health-seeking behavior in the Asian countries.^{2,3} The symptom profile, including the types of tics, the waxing and waning course, the high rates of associated comorbidities, and problems such as self-injury, was very similar to the existing literature on TD. This also endorsed the findings of cross-cultural studies that have concluded that the profile of TD is largely similar across cultures.^{2,3,15} Despite this typical presentation of TD, there was a delay in the diagnosis of about eight years. Other studies have also found that the mean lag between the onset of the symptoms and the diagnosis ranges 6–8 years.^{15–18} As in these studies, the principal cause of the delayed diagnosis among our patients was unawareness about TD among families as well as medical professionals. Later onset, prominence of comorbid OCD, and the ability to partially suppress the tics are some of the other factors, noted in the earlier reports^{16–18} that could have delayed the diagnosis in our patients.

The findings regarding behavioral interventions in this report are of greater significance because of the scarcity of data on this aspect from India. Behavioral interventions were carried out using a structured protocol, which included all the usual components of HRT and CBIT.^{4,19} Additional ERP was used in one patient for control of tics and in four patients for the treatment of OCD. Supportive sessions were carried out with both the patients and the caregivers for improving their understanding of the

treatment, helping them deal with the emotional and social consequences of the illnesses, and enhancing their motivation for treatment. Based on the YGTSS scores, about a 75% decrement in the tics was observed. Patients and caregivers also reported about 76% improvement in tics. There was a considerable improvement in the comorbidities such as OCD as well. Additionally, improvements in functioning and satisfaction with behavioral interventions, as well as reductions in associated distress and burden, were observed among patients and caregivers. Long-term effectiveness of the behavioral treatments was also evident from the fact that during follow-up ranging from several months to years, only one patient suffered a major relapse of the symptoms. Thus, the combination of medications and behavioral treatments proved to be more effective than medications when used alone.

HRT and CBIT are considered highly effective treatments for TD, with response rates of 33%–50%, while ERP is rated as “probably effective” because of fewer ERP trials.^{4,6,19,20} However, ERP is recommended as a first-line treatment for comorbid OCD.^{1,5} The symptoms of comorbid conditions such as OCD often cause greater distress and functional impairment in TD than the tics themselves. They can hinder the response to behavior therapy. Therefore, they require additional treatment.⁵ Though medications and behavioral management are found to be equally effective, our patients had severe TD where the combination of two treatments is more useful and is usually recommended.⁴ Available data also suggests that gains obtained with behavioral interventions are maintained over several months.^{4,6,19} Therefore, the results of behavioral interventions among our patients were remarkably similar to prior Western reports^{4–6} in terms of effectiveness, response rates, long-term outcome, and other benefits for both the patients and the caregivers.

HRT may help suppress tics through several different mechanisms.^{19,20} Since environmental factors influence the occurrence of tics, individualized HRT treatment mitigating their adverse impacts may reduce the frequency and severity of the tics. Premonitory urges are often

experienced as aversive events, generating anxiety that is only relieved by indulging in tics. During HRT, the patients learn to refrain from indulging in tics upon experiencing premonitory urges. This breaks the negative reinforcement cycle that maintains tics. The mechanisms of change in ERP may involve habituation or inhibitory learning, which reduces the need to resort to tics in response to the premonitory urges.²⁰ Improvement with behavioral treatments may also be mediated by biological mechanisms such as enhancement of frontal lobe activity that suppresses the basal-ganglia-mediated activation of tics.

Conclusions

Given that our findings represent uncontrolled data from a very small number of patients, only a few tentative conclusions can be made. To begin with, our experience has shown that it is feasible to conduct behavior therapies for TD in general hospital psychiatric settings in India, despite the comparative lack of resources and specialized skills for treatment. When behavioral treatments are carried out, results appear to be promising, particularly among those with severe TD with inadequate response to medications. Delays in diagnosis and initiation of adequate treatment, lack of awareness about TD, and limited access to behavioral treatment are common even in the West.^{4,19,20} That these may be far bigger problems in India is evident from the lack of Indian studies on this aspect. Moreover, our patient series also demonstrated that medical practitioners lack awareness about TD and its treatment. Consequently, they often fail to inform patients and caregivers about these aspects. Therefore, firstly there is an urgent need to conduct proper efficacy trials of behavioral interventions for TD in India. Skills for delivering these treatments from general hospital settings also have to be developed to increase the access to these effective forms

of treatment for TD. Finally, public and especially professional awareness regarding TD and its treatment, including the role of behavioral interventions, needs to be improved.

Declaration of Conflicting Interests

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