

# TOURETTE’S SYNDROME MANAGED BY ONLINE COMPREHENSIVE BEHAVIOURAL INTERVENTION FOR TICS (CBiT): A CASE REPORT

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## INTRODUCTION:

Tourette’s syndrome is a neurodevelopmental disorder commonly presenting in young males below 18 years of age and persisting for more than one year with multiple motor and vocal tics [1]. Tics are sudden, rapid, recurrent, and non-rhythmic motor movements or vocalizations commonly preceded by an urge [2]. Most cases of Tourette syndrome show significant improvement or resolve entirely by adulthood [3]. Patients whose tics persist into adulthood suffer from higher anxiety, low self-esteem, socioeconomic status, and poor quality of life [4]. Tourette syndrome is frequently associated with other common childhood disorders such as attention deficit hyperactivity disorder (ADHD) (60-80%), obsessive compulsive disorder (OCD) (11-80%), anxiety, depression (13-76%), migraine (25%) and self-injurious behaviour [5,6].

According to the American Academy of Neurology’s practice guidelines, Tourette syndrome can be managed with behavioural, pharmacological, and surgical modalities [7]. Behavioural therapy includes exposure and response prevention therapy (ERP), habit reversal therapy (HRT), and comprehensive behavioural intervention for tics (CBiT), which is considered the safest and first line of treatment [4,7].

CBiT is typically a behavioural intervention that includes psycho-education, HRT, functional analysis, and relaxation training and improves the patient’s recognition of the initial urge by providing a competing response or motor movement incompatible with the corresponding tic [8,9].

## CASE PRESENTATION:



A 12-year-old female child, the eldest of two siblings and a student of class 7, belonging to an upper-middle class nuclear family, presented to a tertiary care facility in south India via online consultation in February 2020 with complaints of irresistible gestures and odd sounds that were produced involuntarily and persisted for a period of one to two minutes. With only one episode per day, these episodes happened three to four times every week.

Her peers initially noticed the sounds and gestures in class, and perceived it as verbally abusive behaviour and reported it to the class teacher, who took the child to the sick room, where she had specific peculiar involuntary motor movements involving her upper limb along with vocal tics such as clearing of the throat. All episodes occurred when the child was at school, as the parents were not initially observant of the tics.

The school administration suggested that the child require medical attention from the school's on-site health team involving a psychiatrist and psychologist, who met with the parents and the child. After ruling out organic causes with a thorough neurological examination, and medical and radiological investigations, the diagnosis of Tourette's disorder with co-morbid depression was made. However, the parents believed there were linguistic and cultural barriers because the school medical team was from a different country than the child and her family. In addition, they found it challenging to proceed with the same team for treatment, so they sought an online consultation from our team.

The parents and the child were psychoeducation about the illness. Comprehensive behavioural intervention therapy (CBiT) was chosen because of the ongoing COVID pandemic and the family's preference for a non-pharmacological mode of treatment. After taking informed consent for online CBIT, sessions with the

index child were scheduled weekly with the mother as the co-therapist. The initial sessions involved detailed history-taking. Apart from tics, history also indicated that the child had displayed reclusive behaviour and a deterioration in academic performance after her father’s haemorrhagic stroke in January 2019. The symptoms of depressed mood, decreased appetite, decreased psychomotor activity, and feelings of helplessness and worthlessness were suggestive of comorbid depression. No past or family history suggestive of any psychiatric, behavioural illness, or other medical conditions were present. No family history of substance was present.

During the mental status examination, a rapport was built with the child. The patient discussed her low academic performance, anxiety about her father’s health, and the absence of suitable social support from her mother and school. The patient scored 24 on the Yale Global Tic Severity Scale (YGTSS) at that time, indicating moderate tic disorder, and a score of 3 on the Clinical Global Impressions Scale (CGI), suggesting mild illness severity. The child demonstrated a need for parental attention and acceptance on the Children’s Apperception Test (CAT) and anxiety over rejection and abandonment. The primary evidence in favour of the thesis was reaction formation and regression. The child was average in her studies, as confirmed by Malin’s intelligence scale for children (MISC).

Therapy was first targeted to control general anxiety and depressive symptoms, and then vocal tics and upper extremity movement tics were treated. The patient complied well with the therapy and did as she was told when given directions.

Initially, the motor tics were targeted (SUD: 75), and once their SUD started decreasing, vocal tics were also targeted. The course of improvement in tics with SUD values is given in Table 1.

Tic	Session 1	Session 2	Session 3	Session 16	Session 17	Session 18	3-month follow up	6-mon
Head shaking up	5	3	0	0	0	0	0	0
Jaw click	10	0	1	0	0	0	0	0
Blinking	5	3	2	1	0	0	0	0
Snorting	5	0	0	1	0	0	0	0
Head-shoulder shake	5	1	0	0	0	1	0	0
Eye rolling	7	6	1	0	0	0	0	0
“Mmm” noise	10	0	0	1	0	0	0	0

Table 1: Tic Hierarchy with SUD Scores

The sessions were held weekly and the scale was also applied weekly. Only the sessions which show improvement have been reported in Table 1. Both the child and the parent-reported clinical improvement, including a decrease in the frequency of the tic episodes to once every 2-3 weeks, an improvement in the child’s quality of life, more significant social interaction at home and school, and an improvement in academic achievement. In April 2020, the YGTSS revealed a score of 12, suggesting improvement in both motor & vocal tics). A complete absent tic episode and a YGTSS score of 6, indicating a practically symptom-free condition, were seen after six months of weekly therapy sessions, which continued till July 2020.

The child’s academic performance had improved, as evidenced by her improved assessment exam grades, regular attendance in class, and compliance with school rules. CBIT was continued as a follow-up treatment for six months because the child showed good compliance and significant symptom improvement.

In August 2020, MSE revealed a reactive child with no worries, low mood, and improved social interaction, whose fundamental mental functions remained the same. In addition, she could effectively regulate stressors in terms of frequency, severity, and duration of a stress response.

The child is currently under follow-up monthly, planned for 12 months till February 2021. Before initiating the management, the therapist and the parents of the child during the therapeutic alliance entered into a contract to continue follow-ups till the child feels a complete resolution of her symptoms and improved

psychosocial functioning. Thus 12 months period was taken to achieve the treatment goals but was not prefixed as such.

## **DISCUSSION:**

The effectiveness and long-term prognosis of behavioural therapies in the index case were similar to those reported in earlier studies [10–12]. The most extensively studied behavioural treatments for tic disorders are HRT and CBIT [13,14]. Relapse prevention programs, relaxation training, function-based assessment and therapies, and psycho-educational sessions are components of CBIT. This elaborated CBIT treatment programme includes conventional HRT components with other methods recommended by understanding chronic tic disorder (CTD) [15,16]. CBIT is considered superior to pharmacotherapy in enhancing psychosocial functioning and appears to have fewer adverse effects, especially if the patient is motivated to undertake lengthy therapy. For those with CTD who want to improve their psychosocial functioning but are concerned about the side effects of pharmacotherapy, CBIT may be an alternative [17].

Good prognostic indicators include early diagnosis, the prompt intervention of parents, psychosocial support, average IQ of the child, regular follow-up, and negative family history. Females with CTD experience peak symptoms later, less age-related remission, and worse tic impairment, especially in adulthood [18]. The decision to initiate therapy is based on how disabling the child's symptoms are to their normal development and schooling. Children responded to CBIT in trials more frequently than adults (53% vs. 38%, respectively) [16,19]. Nevertheless, given that children responded to the control treatment more than adults (psychoeducation and supportive therapy; 19% versus 7%), this may reflect factors other than the particular efficacy of CBIT in these age groups. Children as young as nine years old have participated in studies showing the effectiveness of behaviour treatment [19].

The current case significantly contributes to the scientific literature by showing that CBIT utilized as a part of an intensive outpatient program carried out via an online platform might lower tic intensity. Additionally, recent studies have demonstrated that CBIT, when administered online via videoconferencing mode, can lessen the severity of tics [20]. In a study comparing the efficacy of this method to in-person meetings (n = 18, 10 videoconferences), comparable effect sizes on tic reduction were discovered [21]. The foundation of a study protocol that compares the effectiveness of online CBIT (n = 72), psychoeducation alone (n = 72), and face-to-face CBIT (n = 16) over two years primarily involves internet-delivered CBIT [22]. Also, a small (n = 20) group of kids and teenagers participated in a waitlist-controlled voiceover Internet protocol for delivering CBIT pilot study, with promising outcomes [23]. Three children receiving remotely delivered CBIT, demonstrated clinically substantial reductions in tic frequency in an initial pilot investigation [24]. By implementing this method of treatment delivery, the potential treatment catchment areas are expanded, making CBIT more accessible to a wider range of patients who are limited by location, transportation, or cost and enabling patients to concentrate on developing and implementing their competing responses without interference from work or school [25]. About 30% of TS patients experience anxiety or depression at some point [26]. Suicidal ideas and attempts occur in about 10% of young individuals with tic disorders [27,28].

A large epidemiological cohort study from the Swedish National Patient Register found that adults with TS had a roughly four-fold greater chance of both suicide attempts and fatalities [29]. Screening for depression and anxiety symptoms is especially important in individuals with a family history of these disorders.

## **CONCLUSION:**

According to current guidelines for children with persistent tic disorders and TS, habit reversal therapy, CBIT, and ERP are advised as first-line behavioural treatments. However, using these methods in conjunction with internet-based and telehealth approaches may improve general accessibility to behavioural treatments, reduce related expenses, and boost treatment frequency. This case shows how online-based CBIT can decrease tic severity and disability caused by the tics while also improving quality of life.

## **CONFLICTS OF INTEREST:**

None declared.

## **AUTHOR CONTRIBUTION:**

All the authors contributed equally in drafting, editing, revising and finalizing the case report.

## **ETHICAL APPROVAL:**

The ethical approval was not required for the case report as per the country's guidelines.

## **CONSENT:**

Written informed consent was obtained from the patient to publish this report.

## **DATA AVAILABILITY STATEMENT:**

The data that support the findings of this article are available from the corresponding author upon reasonable request.

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