Scandinavian Journal of Psychology

Empirical Article

Open Access

Group-based comprehensive behavioral intervention for tics (CBIT) for adults with Tourette syndrome or chronic tic disorders: A pilot study

Morten Bekk, Karete J. Meland, Erna Moen, Liv I. Nøstvik, Anne-Line Gausdal, Benjamin Hummelen

First published: 20 June 2023

https://doi.org/10.1111/sjop.12942

Due to safety procedures at the Oslo University Hospital, data are not publicly available. Access to data may be obtained by sending a request to the Data Protection Officer at the Oslo University Hospital: **personvern@ous-hf.no**, with a copy to the corresponding author.

Before inclusion in the study, patients gave written consent to participate. The Data

Abstract

Comprehensive behavioral intervention for tics (CBIT) administered individually is an effective treatment for tics. However, the effectiveness of CBIT administered in groups for adults with Tourette syndrome and chronic tic disorders has not been investigated yet. This pilot study examined the effectiveness of group-based CBIT with respect to reduction of tic severity and tic-related impairment, as well as improvement of tic-related quality of life. Data from 26 patients were included in the intention-to-treat analyses. The Yale Global Tic Severity Scale was used to assess total tic severity and tic-related impairment. The Gilles de la Tourette – Quality of Life Scale was used to assess tic-related quality of life. These measures were administered at three points in time: at pretreatment, posttreatment, and 1-year follow-up. The results showed a significant reduction of total tic severity from pretreatment to 1-year follow-up, with larges effect sizes. Tic-related impairment and tic-related quality of life also improved significantly, although the effect sizes were smaller. Motor tics showed a stronger reduction than vocal tics. Additional analysis revealed that all change was achieved during treatment and that this effect was maintained from posttreatment to 1-year follow-up. The results of this study indicate that group-based CBIT is a promising treatment for tics.

INTRODUCTION

Tourette syndrome (TS) is a neurodevelopmental disorder, typically with onset in childhood, and characterized by motor and vocal tics. The prevalence of TS is estimated to be close to 1% among children between 5 and 18 years of age (Robertson *et al*., **2017**). Tic severity declines with age, and it has been suggested that two-thirds improve significantly in tic symptoms by adulthood (Bloch & Leckman, **2009**). Community and clinical studies have shown that approximately 90% of individuals with TS have other comorbid diagnoses such as attention deficit hyperactivity disorder (ADHD), obsessive-compulsive disorder (OCD), anxiety disorders, and depression (Freeman *et al*., **2000**; Hirschtritt *et al*., **2015**; Khalifa & AL, **2006**). Both tics and comorbid psychiatric conditions are associated with psychosocial and functional impairment and undermine quality of life (Cavanna *et al*., **2013**; Conelea *et al*., **2013**).

Tics are defined as sudden, rapid, repetitive, and non-rhythmic motor movements or vocalizations. They are classified into simple motor and vocal tics that involve a single muscle group or simple meaningless sounds, on the one hand, and complex motor and vocal tics that involve multiple muscle groups, either simultaneously or in rapid succession, or complete words or short phrases, on the other. Unpleasant internal sensory sensations, named premonitory urges, commonly precede tics and are temporarily relieved by performance of the tic. Many individuals with tics are able to suppress their tics for a limited period of time, but this takes a lot of effort and may negatively impact concentration and interfere with functioning, for instance, at work or in social situations (Himle *et al*., **2007**; Matsuda, Kono, Nonaka, Fujio & Kano, **2016**). The onset of motor tics, usually observable in the face or head area, precedes vocal tics, and simple tics usually precede complex tics.

Tics exhibit fluctuations over time in type, frequency, intensity, and complexity and are influenced by psychological and contextual factors (Conelea & Woods, **2008**; Eapen, Fox-Hiley, Banerjee & Robertson, **2004**; Himle *et al*., **2014**; Misirlisoy *et al*., **2015**; Piacentini *et al*., **2006**). It has been suggested that that tic fluctuations predominately occur in relation to four types of partially overlapping scenarios: high or low sensory stimulation; anxiogenic situations; frustrating and anger-inducing contingencies; and fatigue and sleep loss (Godar & Bortolato, **2017**). On the other hand, activities or tasks that require focused concentration appear to relieve tics. Thus, psychological factors appear to play an important role in the exacerbation of tics. If it is possible to modify these psychological mechanisms, tics and tic-related difficulties may be reduced considerably.

Behavioral interventions such as habit reversal training (HRT), comprehensive behavioral intervention for tics (CBIT), and exposure with response prevention (ERP) were found in a meta-analysis by McGuire *et al*. (**2014**) to be effective treatments for tics. This meta-analysis included eight randomized controlled trial (RCT) studies of individual treatment with behavioral interventions for tics and found moderate to high treatment effect sizes (effect sizes = 0.67–0.94) compared with all control conditions, such as wait-list or an active comparison condition (e.g., awareness training, supportive therapy, and psychoeducation). A large RCT included in this review found that CBIT was superior to both psychoeducation and supportive therapy. Of interest, this study found that motor tics decreased more during the 10-week treatment than vocal tics (Wilhelm *et al*., **2012**). Similar results were found by Yates *et al*. (**2016**) in a sample of children with TS who were offered HRT group treatment.

Whether this effect also will occur in adult patients treated with HRT or CBIT in groups has not been investigated yet.

CBIT administered in groups for adults can provide additional benefits to individual treatment. Group-based CBIT for adults opens group members up for mutual support, sharing experiences in a safe context, and enhancing motivation to accomplish homework assignments. However, effects of behavioral interventions given in groups have been examined only in children and young people (Dabrowski *et al*., **2018**; Gur, Zimmerman-Brenner, Fattal-Valevski, Rotstein & Pilowsky Peleg, **2022**; Heijerman-Holtgrefe *et al*., **2021**; Nissen, Kaergaard, Laursen, Parner & Thomsen, **2019**; Yates *et al*., **2016**; Zimmerman-Brenner *et al*., **2022**). Overall, the results of these studies support the efficacy of behavioral interventions for tics delivered in groups as compared with control conditions. However, some inconsistences in findings have been reported. For instance, Yates *et al*. (**2016**) found that group-based HRT was more effective in reducing motor tics, but not vocal tics, compared with group-based educational intervention. Dabrowski *et al*. (**2018**) found significant improvement in tic severity and quality of life for both group-based HRT and group-based educational intervention at 12 months follow-up. Nissen, Kaergaard, Laursen, Parner, and Thomsen (**2019**) compared the effectiveness of a combination of HRT and ERP delivered in groups and delivered individually. Findings showed a greater reduction in tic-related disability in the individual setting, but both groups showed improvement in tic severity. A recent study by Gur, Zimmerman-Brenner, Fattal-Valevski, Rotstein, and Pilowsky Peleg (**2022**) found that CBIT delivered in groups had better effect on cognitive and emotional regulation abilities as compared with group-based educational intervention for tics, showing efficacy of CBIT beyond the well-documented efficacy of CBIT on tic severity. In sum, findings of group-based behavioral interventions for tics for children and youth are promising, but further studies are needed. For adults, empirical support for the effectiveness of group-based behavioral interventions for tic severity is lacking.

Adverse experiences associated with tics, such as attention from others contingent on tics, negative judgement, teasing, and exclusion, may cause exacerbation of tics and psychological distress such as embarrassment, shame, and fear of negative evaluation. Such negative and distressing experiences can be difficult to anticipate and respond to, and they may lead to frantic efforts to camouflage or suppress tics in social settings. This may be exhausting and compromise the quality of social interactions. Avoidance of social situations may also be a protective strategy for some patients (Conelea *et al*., **2013**; Malli, Forrester-Jones & Triantafyllopoulou, **2019**; Smith, Fox & Trayner, **2015**; Wadman, Tischler & Jackson, **2013**; Weingarden *et al*., **2018**). Psychosocial aspects of living with tics, such as adverse and unpredictable reactions to tics coupled with emotional reactions, have been described as a dominant feature of the experiences of people with TS. Accordingly, how to manage these social challenges can be considered an important aspect in the treatment of TS (Buckser, **2008**).

CBIT administered in groups for adults with TS or chronic tic disorders (CTD) may promote management of such social and emotional challenges of living with tics. Several therapeutic factors have been proposed with group treatment, such as vicarious and interpersonal learning, sense of belonging and relatedness, experiencing universality, altruism, and instillation of hope (Yalom & Leszcz, **2005**). Thus, employing CBIT in groups may contribute to enhanced therapeutic effects on other co-occurring problems related to living with tics and to enhanced quality of life.

To our knowledge, this pilot study is the first that examines the effectiveness of CBIT delivered in groups for adults with TS. The aim of the present pilot study was to explore the effectiveness of group-based CBIT for adults with TS or CTD by examining short-term and long-term outcomes of tic severity, tic-related impairment, and tic-specific quality of life. We expected a significant reduction of tic severity and tic-related impairment during the course of the project, as well as significant improvement of tic-specific quality of life. We also investigated differences in outcomes for vocal tics and motor tics separately and hypothesized that motor tics would show a stronger decrease than vocal tics. We further explored which aspects of quality of life improved most substantially (psychological, physical, obsessional, or cognitive). Finally, we examined differences in change rates for two different time periods, i.e., from pretreatment to posttreatment and from posttreatment to 1-year follow-up.

METHOD

Before inclusion in the study, patients gave written consent to participate. The Data Protection Officer at the Oslo University Hospital has approved the study.

Sample

Inclusion criteria were individuals aged over 18 years with an established diagnosis of TS or CTD and with no planned changes in medication during the treatment period. Exclusion criteria included the following: presence of schizophrenia spectrum disorders, current substance addiction, intellectual disability, brain injuries, current severe depression, and current suicidal ideation.

Patients were referred by general practitioners, neurologists, and mental health practitioners at outpatient clinics. Referring clinicians were informed about both inclusion and exclusion criteria. Altogether, 41 patients were referred to the treatment program. Three patients did not meet up for pretreatment assessment for unknown reasons. Seven patients were evaluated as qualified for CBIT but withdrew after pretreatment assessment due to interference with work or studies, time constraints, or great travel distance between home and clinic. Three patients received CBIT individually according to their own wishes. One patient experienced obsessive-compulsive symptoms as the main problem and was referred to OCD treatment. Finally, one patient was excluded because their tic-like symptoms could better be explained as stereotypies. There were 26 patients, 13 females and 13 males, who began group treatment, of whom one dropped out of treatment by session 4. Thus, data from 26 patients were included in the intention-to-treat analyses (see Fig. 1).

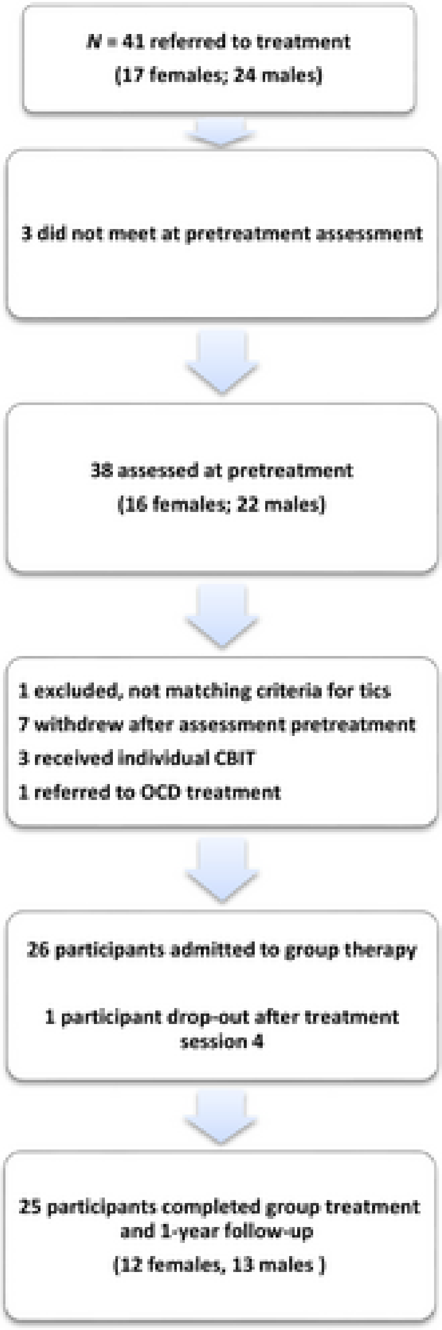

Fig. 1

Open in figure viewerPowerPoint

Diagram of participant flow through the study.

The ages of the patients included in the intention-to-treat group ranged from 20 to 70 years, with a mean age of 36 years. Eight patients were diagnosed with TS or CTD before age 18, whereas 18 patients were diagnosed after age 18. Seven patients were on medication for tics at pretreatment, i.e., pimozide ($n = 4$), aripiprazole ($n = 2$), and risperidone ($n = 1$). One patient received injections with botulinum toxin because of neck pain due to tics. Patients were encouraged not to change medication during the project period. The Yale-Brown Obsessive-Compulsive Scale (Y-BOCS) was used to assess obsessive-compulsive symptoms. It consists of 10 items, 5 items covering the severity of obsessions and 5 items covering the severity of compulsions. Total scores ranges between 0 and 40. It is a widely used clinician-administered interview for assessing the presence and the severity of OCD symptoms in adults (Goodman *et al*., 1989a; Goodman *et al*., 1989b). Y-BOCS scores were available for 21 patients. Seven patients (33.3%) had a total score beyond 16 points, a moderate clinical score that is considered the clinical cutoff score for OCD. Moreover, the Adult ADHD Self-Report Scale, version 1.1 (ASRS v1.1) (Kessler *et al*., **2005**; Kessler *et al*., **2007**) was used for the assessment of ADHD symptoms. These scores were available for 18 patients. Four or more confirming answers to the first six questions is indicative of ADHD. Seven patients (39%) had a score of 4 or higher, indicating presence of ADHD.

Measures

Yale Global Tic Severity Scale (YGTSS); YGTSS Total Tic Severity Score (TTSS)

The YGTSS is a multidimensional clinician-rated scale that measures tic severity over two time periods, i.e., the last 7 to 10 days and the worst period ever. In line with previous studies, the YGTSS was divided into the YGTSS Total Tic Severity Score (TTSS) and the YGTSS Impairment Score (McGuire *et al*., **2015**; McGuire *et al*., **2018**). The TTSS is rated over five domains: number of tics; frequency of tics; intensity of tics; complexity of tics; and interference. Motor and vocal tics are rated separately in each of the five domains, with item scores ranging between 0 and 5 points. TTSS is the sum of each of the five domains with a maximum score of 50 points. Higher scores indicate greater tic severity. The YGTSS has demonstrated good reliability and validity (McGuire *et al*., **2018**).

Yale Global Tic Severity Scale; YGTSS Impairment Score

The YGTSS Impairment Score rates the overall impact tics have on self-esteem, family life, social acceptance, or school or work functioning from none (= 0), minimal (= 10), mild (= 20), moderate (= 30), marked (= 40), to severe (= 50).

Clinical Global Impression of Improvement (CGI-I)

The CGI-I requires clinicians to rate improvement compared with baseline on a seven-point scale (i.e., “very much improved,” “much improved,” “minimally improved,” “no change,” “minimally worse,” “much worse,” to “very much worse”). The CGI-I is well-validated in treatment studies of tic disorders (Jeon *et al*., **2013**; McGuire *et al*., **2020**). However, in this study, CGI-I scores were set in collaboration with the patients, which deviates somewhat from the original procedures in which clinicians set a score without patients' input. In more detail, CGI-I was administered by clinicians at 1-year follow-up by asking patients how they would rate improvement compared with baseline on the seven-point scale, followed by a discussion of the participants' experience of treatment response in order to validate the original rating. Because of the long follow-up period of this study, we assume that this administration method is more reliable than the traditional method. However, we did not test the reliability of this adapted method.

The Gilles de la Tourette Syndrome – Quality of Life Scale (GTS-QoL)

The GTS-QoL is a 27-item self-report questionnaire that measures health-related quality of life in patients with TS (Cavanna, **2020**; Cavanna *et al*., **2008**). Higher scores indicate greater negative impact on health-related quality of life. The questionnaire comprises four subscales. The psychological subscale includes aspects such as depressed mood, isolation, lack of self-confidence, and anxiety. The physical subscale focuses on aspects that pertain to daily functioning in relation to tics such as embarrassing gestures, difficulties in daily activities, involuntary swearing, and pain or injuries. The obsessional subscale is mostly related to the repetitive aspects of OCD and includes symptoms such as repeating words and copying the behavior of people. Finally, the cognitive subscale includes characteristics such as memory problems, difficulties concentrating, and losing important things.

Other measures

Patients were assessed at pretreatment using the Schema for Registration of Tics (SRT). This is a Norwegian questionnaire, based on a model by the Yale Tourette Clinic, and developed by the National Resource Center for ADHD and Tourette Syndrome, Oslo University Hospital. The questionnaire comprises items mapping both current and previous presence of simple and complex motor tics and simple and complex vocal tics. In the course of the project, the SRT was discontinued as an outcome measure since it had become clear that it was sufficient to include the YGTSS only. The SRT was therefore not included in the follow-up evaluations. However, the SRT was still useful for imputation purposes of missing data at pretreatment. Information about the SRT can be obtained on request.

Procedures

The patients were assessed at three points in time: at pretreatment; at posttreatment (i.e., at the third booster session); and at 1-year follow-up (i.e., at the final treatment session 1 year after the third booster session). The interviews were conducted by two therapists participating in the study, the first and second author. In order to achieve consensus in scoring of the YGTSS, the therapists independently assessed six patients at pretreatment. One therapist administered the interview and scored the YGTSS while the other therapist observed the interview and made an independent scoring. Roles as interviewer and observer were alternated. After each assessment the scores were discussed.

A total of eight groups were conducted in this pilot project. The groups consisted of three or four patients, except the last group, which included only two patients. We intended to include three patients in this group, but one patient canceled treatment a few weeks before starting up.

Since we had no previous experience with group-based CBIT, we decided to conduct all group meetings with two therapists. The first author participated in all eight groups, the third author participated in the first four groups, and the second author in the last four groups.

Ahead of the treatment project, all three therapists had completed courses in HRT, provided by the Regional Resource Center for Autism, ADHD and Tourette Syndrome, Health Region South-East, Oslo University Hospital. The second and third authors had previous experience with group-based HRT for adult patients with hair pulling disorder and skin picking disorder.

Treatment

The CBIT treatment was carried out at an outpatient department specializing in the treatment of OCD-related disorders in accordance with the treatment manual for adults, i.e., *Managing Tourette Syndrome: A Behavioral Intervention for Children and Adults: Therapist Guide* (Woods *et al*., 2008a). The manual comprises thirteen chapters covering the content of the first eight weekly structured sessions and three monthly follow-up booster sessions. See Table 1 for a detailed overview of the sessions. The treatment program evaluated in this study also included a follow-up session 1 year after completion of the last booster session. Booster sessions and the 1-year follow-up session included reviews of HRT procedures, along with progress updates for each participant. Each group met weekly, for 3 h, over 8 weeks. This was a modification of the original intervention with six weekly sessions and then 2-week intervals between sessions 6, 7, and 8. This modification was implemented for practical reasons. During the project period, patients were offered three psychoeducational meetings in which they could invite family members and other significant persons. These meetings were delivered in addition to the eight treatment sessions and the three booster sessions. Each group was offered one meeting after treatment session 4 as a part of the treatment, held by the therapists who informed about TS and CBIT. During the project period, two psychoeducational meetings were held by the Norwegian Tourette Association to inform participants about TS and the Norwegian Tourette Association. The three therapists participated in these two meetings.

Table 1. Overview of the sessions

Session 1	Round of presentations: All participants and therapists in the group present themselves. Psychoeducation: About Tourette syndrome/tics, and the treatment rationale. All group members create their own tic hierarchy.
Session 2	Review of habit reversal training principles, awareness training, and competing response training for their chosen tic (i.e., first tic; conducted individually). Functional assessment and treatment strategies for their chosen tic (same as the tic above).
Session 3	HRT for their next chosen tic. Functional assessment and treatment strategies for their next chosen tic (same as the tic above).
Session 4	HRT for their next chosen tic. Functional assessment and treatment strategies for their next chosen tic (same as the tic above). Relaxation exercises.
Meeting with significant others	Plenary information about tics, TS, and the treatment rationale. Support to patients and their next of kin and significant others.
Session 5	HRT for their next chosen tic. Functional assessment and treatment strategies for their next chosen tic (same as the tic above). Relaxation exercises.
Session 6	HRT for their next chosen tic. Functional assessment and treatment strategies for their next chosen tic (same as the tic above).

	Relaxation exercises.
Session 7	HRT for their next chosen tic. Functional assessment and treatment strategies for their next chosen tic (same as the tic above). Relaxation exercises.
Session 8	HRT for the patients' remaining tics. Functional assessment and treatment strategies for the patients' remaining tics. Relaxation exercises. Ending the therapy: discussion of what has been meaningful throughout therapy, and relapse prevention strategies.
Booster meetings 1, 2, and 3	Review of treatment techniques for all treated tics, relaxation techniques, and relapse prevention strategies, along with progress updates for each patient.
One-year follow-up meeting	Review of treatment techniques for all treated tics, relaxation techniques, and relapse prevention strategies, along with progress updates for each patient.

- *Notes*: Each session lasted 180 min. All sessions were conducted in a group setting, except session 2. Each session after session 2 included review of events of the previous week, treatment procedures, home assignments, and an update of tic hierarchy and inconvenience since the last session. Due to individual progress in habit reversal training (HRT) for tics, some patients needed more than one session to get tic control for a tic. During the project period, two additional meetings were held by the Norwegian Tourette Association to inform participants about TS and the Norwegian Tourette Association. These meetings were open to all patients participating in the project, their significant others, and other interested parties.

CBIT is a multicomponent intervention and comprises (a) psychoeducation, (b) HRT, (c) functional interventions, and (d) relaxation training (Woods *et al*., **2008a**). HRT is a core component in CBIT and a method for management of tics. HRT consists of several procedures, such as tic-awareness training, which aims at becoming aware of the premonitory somatic sensations that precede tics to facilitate early tic detection. When aware, the patients practice blocking the tic with another alternative movement incompatible with the tic, named a competing response. The competing response is intended to prevent the expression of the tic and block the link between the relief of the premonitory somatic sensation and expression of the tic. Functional interventions target identifying external and psychological factors associated with tic exacerbation, to minimize these factors by modifying tic-prone situations or working out strategies to manage such situations. Psychoeducation includes information about tics, their course, causes, and comorbidities, and CBIT in order to increase knowledge about the condition and treatment. Relaxation training aims at reducing tensions and stress and may help to lessen tics and help manage anxiety problems (McGuire *et al*., **2020**; Woods *et al*., **2008a**).

Practicing the competing response, including awareness training, for the first target tic was carried out individually in order to ensure understanding of the principles for HRT and working out the patient's first competing response. Awareness and competing response training was introduced in treatment session 2, in accordance with the manual. The two therapists worked individually with each patient in the group setting, while the other patients witnessed the interventions. After the first two patients had completed these training procedures, the roles were switched. Further training on awareness, competing responses, functional interventions, psychoeducation, and relaxation training were carried out in the group setting.

Functional interventions were based on each patient's assessments of situations in their everyday lives to identify situations in which tics usually occurred frequently, using the Functional Assessment Form – Adult version in the workbook, *Managing Tourette Syndrome: A Behavioral Intervention: Adult Workbook* (Woods *et al*., **2008b**). Functional interventions can encompass a range of different interventions tailored to the patient's unique situation. The patients were helpful to each other both by working out strategies to manage situations and proposing ideas on how to modify situations associated with intensified tics by exchanging experiences, reflections, and suggestions. The therapists facilitated these mutual exchanges by asking explorative questions.

Statistical analysis

The mixed models procedure in SPSS (version 26) was used to assess clinical change for YGTSS Total Tic Severity Score (TTSS), YGTSS Impairment Score, and GTS-QoL. The time component was entered as the independent variable in the model and coded as 0, 1, or 2, corresponding to the three assessment occasions (pretreatment, posttreatment, and 1-year follow-up). Model parameters were estimated by restricted maximum likelihood estimation. The Akaike Information Criterion (AIC) was used to evaluate model fit. By including a random intercept at the individual level, model fit improved slightly for TTSS (AIC reduced from 443.5 to 441.1) and GTS-QoL (AIC reduced from 549.3 to 527.8). For YGTSS Impairment Score, the final Hessian matrix was not positive definite, and therefore, the random intercept was not included in the model. Mathematically, the random intercept model used in our study is identical to a repeated measurement ANOVA (Fitzmaurice, Laird & Ware, **2004**), facilitating comparisons with other studies in the field, e.g., of McGuire *et al*. (**2020**).

In order to compare change rates for the two different time periods, i.e., from pretreatment to posttreatment and from posttreatment to 1-year follow-up, a spline model with the knot at the second measurement occasion was evaluated. Compared with the linear model, the model fit of the spline model improved considerably. For TTSS, AIC reduced from 441.1 to 428.6; for YGTSS–Impairment Score, AIC reduced from 487.3 to 479.2; and for GTS-QoL, AIC reduced from 527.8 to 513.3.

In addition to common statistical effect sizes for mixed models (t and F), we also reported Cohens'd for longitudinal analyses based on the t test, i.e., $t/\sqrt{n}$.

Missing data

Errors in administration routines and participants' absence at the third booster session (posttreatment) or 1-year follow-up session are the main reasons for missing data. We assumed that the data are “missing completely at random” because errors in administration routines are not related to patient or treatment factors. In addition, patients used to give plausible reasons for their absence, lending further support to the missing totally at random hypothesis. Since maximum likelihood estimation was used, it should therefore not be necessary to apply corrective measures (Enders, 2010). However, the number of patients with missing data was too small to examine the missing completely at random assumption statistically. Therefore, we imputed missing values at those measurement occasions where it was possible to apply other measures. At pretreatment, this was realized using the Schema for Registration of Tics (SRT). Among the four patients who had missing TTSS at pretreatment, three had filled in SRT. The correlation between TTSS and SRT was 0.52, which was considered large enough to impute the missing data by regressing SRT on TTSS (simple linear regression; predicted values were used for the imputation). Since the SRT was discontinued during the project, we could not use the same procedures for the follow-up evaluations. In order to examine whether missing data at the second and third measurement occasions had an impact on effect sizes and significance levels, we supplied the main analyses by a set of mixed models analyses on data that were imputed using "last observation carried forward."

After imputation of the pretreatment variables, one patient had missing TTSS at pretreatment, seven patients had missing TTSS at posttreatment, and seven patients had missing TTSS at 1-year follow-up. Overall, 14 patients had TTSS for all three measurement occasions, nine patients had TTSS for two occasions, and three patients had TTSS for one occasion. CGI-I values were available for 20 patients at 1-year follow-up, with six missing CGI-I assessments at 1-year follow-up. Among the six patients with missing CGI-I, two patients had CGI-I at posttreatment, which were included in the results. Thus after imputation of these values, four patients had missing CGI-I in the 1-year follow-up analyses.

RESULTS

Linear change

Table 2 gives the results of the linear mixed models analyses. There was significant change for all three measures. For TTSS, the slope estimate was −6.7, which implies that TTSS reduced by 6.7 points on average for each time period, i.e., from pretreatment to posttreatment and from posttreatment to 1-year follow-up. Thus, given an intercept of 26.1, there was an overall reduction of TTSS from 26.1 at pretreatment to 12.7 at 1-year follow-up. This effect was highly significant ($p < 0.001$) and had a substantial effect size ($F = 37.0$; Cohen's $d = 1.20$). These results are illustrated in Fig. 2, showing a reduction of tic symptoms across the three points of measurement.

Table 2. Results from the mixed models analyses

	B_0	B_1	*SE*	95% CI	*df*	*F*	*t*	*p*	*d*
YGTSS-TTSS	26.1	−6.7	1.1	−8.9 to −4.5	42.5	37.0	−6.1	<0.001	1.20
TTSS motoric tic severity	16.0	−3.6	0.5	−4.7 to −2.5	38.5	47.1	−6.9	<0.001	1.35
TTSS vocal tic severity	10.2	−3.1	0.8	−4.7 to −1.6	40.5	16.9	−4.1	<0.001	0.80

	B_0	B_1	*SE*	95% CI	*df*	*F*	*t*	*p*	*d*
YGTSS Impairment Score	29.8	−8.2	2.0	−12.2 to −4.2	60.0	17.1	−4.1	<0.001	0.80
GTS-QoL	38.1	−6.6	1.9	−10.5 to −2.8	39.3	12.0	−3.5	<0.001	0.69

- *Note*: TTSS = Total Tic Severity Score; GTS-QoL = Gilles de la Tourette Quality of Life Scale; B_0 = intercept; B_1 = slope; *d* = Cohen's *d*.

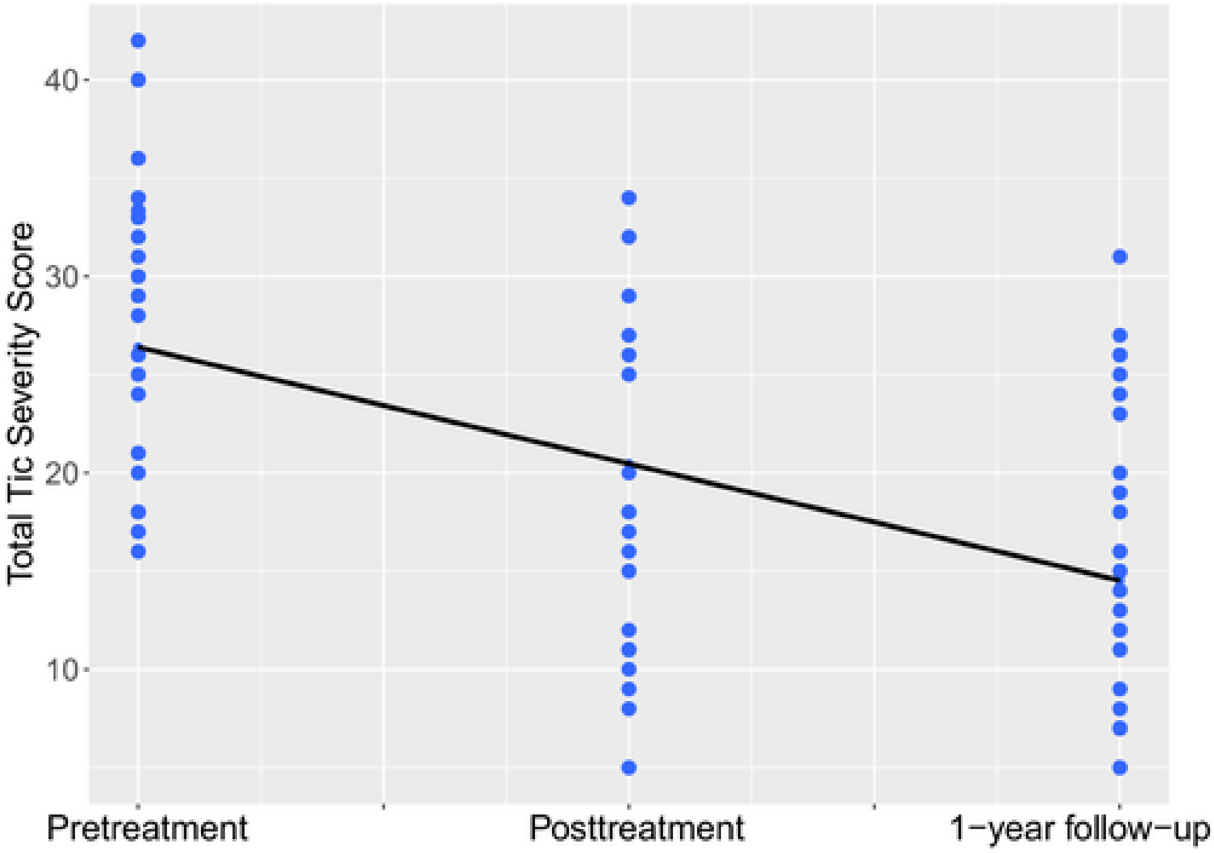

Fig. 2

Open in figure viewerPowerPoint

Linear change of Total Tic Severity Score.

YGTSS Impairment Score also diminished significantly, by 8.2 points for each time period. With an intercept of 29.8, the overall reduction was from 29.8 to 13.4 at 1-year follow-up. Though the slope of YGTSS Impairment Score was comparable to the slope of TTSS, the other effect sizes were substantially smaller, which might be explained by the larger variation in outcome ($SE = 2.0$ versus $SE = 1.1$). Self-reported tic-related quality of life (GTS-QoL) also improved significantly, with a reduction of 6.6 points for each time period ($F = 12.0$, $p < 0.001$).

We conducted the same set of analyses on data in which missing values were imputed by carrying the last observation forward and compared these results with the results from the main analyses. The slope for TTSS increased slightly, 0.8 points (from –6.7 to 5.9), indicating a somewhat smaller change estimate. The other estimates for TTSS did not change notably ($F = 37.6$, $t = -6.1$, $p < 0.001$, Cohen's $d = 1.20$). For YGTSS Impairment Score, this pattern was similar. The slope estimate increased by 0.6 points (from −8.2 to −7.6), and the other estimates did not change ($F = 18.4$; $t = -4.3$; $p < 0.001$; Cohen's $d = 0.84$). The estimates for GTS-QoL also showed only minimal change, i.e., slope = −5.9 (reduction of 0.7 points); ($F = 12.4$; $t = -3.5$; $p < 0.001$; Cohen's $d = 0.69$).

Linear change for YGTSS motor and vocal severity score

The TTSS was split into motor and vocal severity scores. Results showed a significant change in both scores. The change rate for motor tic severity was somewhat larger than for vocal tic severity, i.e., −3.6 versus −3.1, respectively. Cohen's d was substantially larger for motor tic severity than for vocal tic severity. Note that the intercept was substantially larger for motor tics than for vocal tics, illustrating that pretreatment scores for motor tics were much larger for motor tics.

Linear change of the GTS-QoL subscales

In the next step, the four GTS-QoL subscales (i.e., the psychological, physical, obsessive-compulsive, and cognitive subscales) were analyzed separately. There was significant change for the psychological subscale ($F = 17.0$, $p < 0.001$) and the physical subscale ($F = 11.2$, $p = 0.002$). However, no significant change occurred for the obsessive-compulsive subscale ($F = 1.5$, $t = -1.2$, $p = 0.233$) and cognitive subscale ($F = 0.39$, $t = -0.63$, $p = 0.534$).

TTSS change scores and CGI-I

We found that 83% of our patients had a reduction of 25% or more in TTSS from pretreatment to posttreatment. According to Jeon *et al*. (**2013**) and Cavanna (**2020**), a 25% reduction can be considered an indicator of clinical treatment response.

These results resonate with the results from the CGI-I, i.e., therapists' improvement ratings based on patients' feedback at 1-year follow-up. According to this assessment, 78.3% were rated as “much improved” or “very much improved,” and 21.7% as “minimally improved.” None of the patients were rated as “no change” or “deteriorated.”

Piecewise change: Linear spline model

The results of the linear spline model showed that there was significant change of TTSS during the first treatment episode ($F = 35.4$, $p < 0.001$, 11.9 points reduction) but not during the second episode ($F = 0.25$, $p = 0.62$, 1.1 points reduction). Thus, overall treatment gain was obtained during the first treatment episode (8-week treatment phase and the three booster sessions) and maintained from posttreatment to 1-year follow-up. These results are demonstrated in Fig. **3**, where it can be observed that all changes occurred from pretreatment to posttreatment.

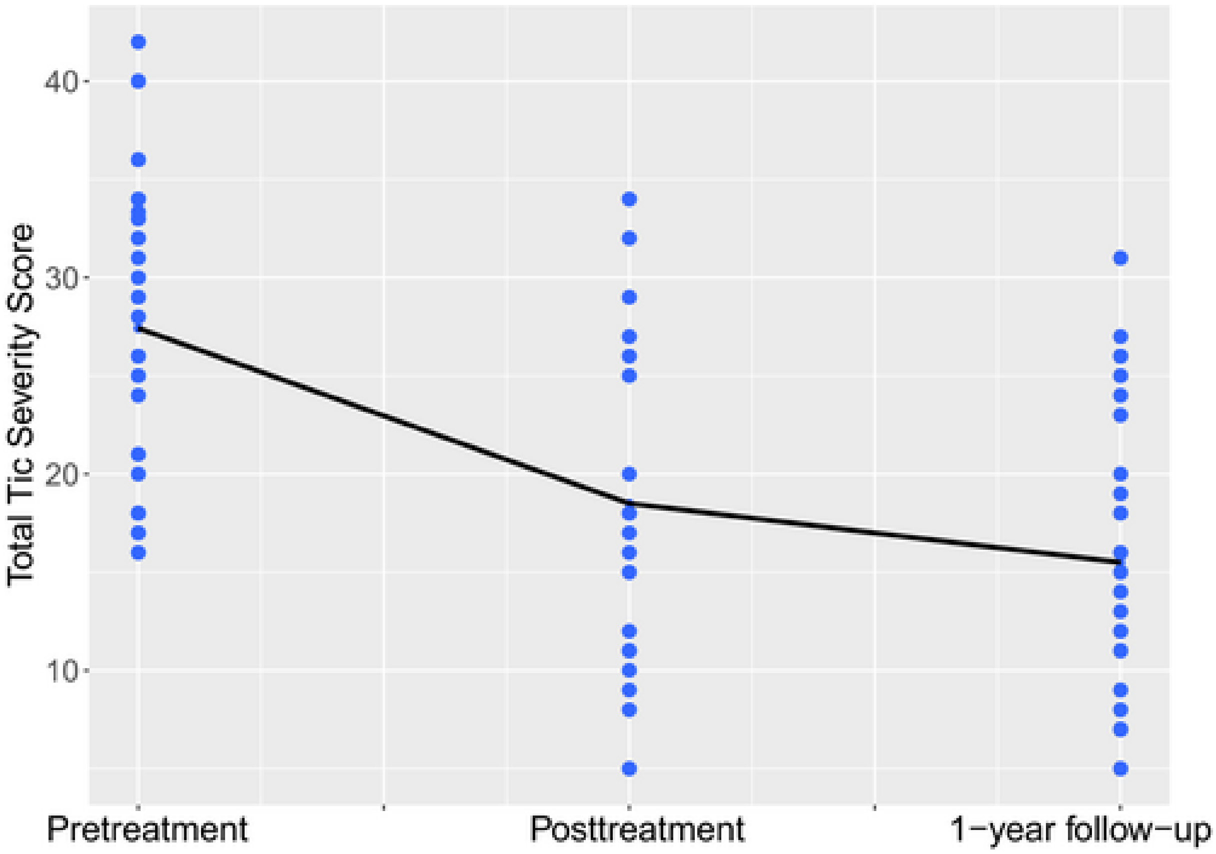

Fig. 3

Open in figure viewerPowerPoint

Piecewise change of Total Tic Severity Score (linear spline model).

This is also apparent with respect to tic-related impairment. There was significant change of the YGTSS Impairment Score during the first time period, i.e., a reduction of 13.3 points from pretreatment to posttreatment ($F = 11.6$, $p < 0.001$), and a non-significant reduction of 2.6 points from posttreatment to 1-year follow-up ($F = 0.40$, $p = 0.53$).

As with the other two measures, the results of the linear spline model revealed that all improvement of tic-related quality of life (GTS-QoL) occurred during the first time period, i.e., a reduction of 16.1 points from pretreatment to posttreatment ($F = 21.7$, $p < 0.001$) and a non-significant reduction of 3.1 points from posttreatment to 1-year follow-up ($F = 0.76$, $p = 0.39$).

DISCUSSION

The main purpose of the current pilot study was to examine the effectiveness of CBIT administered in groups for adults with TS or CTD. The results showed a significant reduction in TTSS from pretreatment to 1-year follow-up, with large effect sizes. The YGTSS Impairment Score and tic-related quality of life (GTS-QoL) also improved significantly during the study period, although the effect sizes were smaller. Control analyses in which missing values were imputed using the last observation carried forward did not change the conclusions based on the main analyses. Motor tic severity showed a stronger reduction than vocal tic severity. With respect to the four subscales of the GTS-QoL, only the psychological and physical subscales changed significantly during treatment. No significant improvements were observed for the obsessive-compulsive and cognitive subscales. The linear spline model showed that all change was achieved during treatment and that this effect was maintained from posttreatment to 1-year follow-up. The dropout rate was low (3.8%), providing support for the acceptability of this treatment approach.

There is good empirical evidence for the efficacy of individually administered behavioral therapies like CBIT and ERP (McGuire *et al*., **2014**). The current study indicates that CBIT delivered in groups is a viable alternative to individual therapy. The large effect sizes of our study are remarkable when taking into account the relatively long follow-up period. However, factors other than the group format itself could have influenced the results. Besides the treatment format itself, we could identify two main factors that may have contributed to the good treatment outcomes. Selection bias could be one plausible explanation since our study included patients who personally searched for and took the initiative for referral to treatment, suggesting high readiness for change. It should also be noted that the mean age of our patients was 36 years, higher than in most previous treatment studies. According to the meta-analysis by McGuire *et al*. (**2014**), older mean participant age was associated with larger effect sizes, which may be explained by a better ability to implement behavioral strategies in HRT. Another possible reason for the good results of our study could be related to treatment duration and treatment intensity. Duration of the treatment in our study differs from a standard individual treatment (i.e., 9 h treatment). Our treatment program consisted of eight 3-h treatment sessions and three booster sessions following the initial eight treatment sessions. Patients were also offered a meeting with the therapists and family members or other significant persons, as well as two meetings with the Norwegian Tourette Association. These meetings may have affected the treatment outcomes positively, for instance, by correcting possibly erroneous beliefs about the condition among relatives and friends, and thereby changing dysfunctional interaction patterns that maintain or exacerbate tics. McGuire *et al*. (**2014**) recommend further studies to determine the role and impact of factors such as duration and intensity of treatment and amount of therapeutic contact on treatment outcome.

The good treatment results of our study could also be explained by the treatment modality itself, i.e., the group therapy setting. Group factors, such as receiving and giving feedback and encouraging others to engage in the treatment program, may have increased engagement to carry out homework assignments such as competing response training. The group setting also provides a context in which patients can experience and practice how awareness training and competing response training can be performed in natural situations, for instance while directing attention to the other patients and following along with conversations. As such, it might have eased generalization of competing responses to real-life contexts. Finally, meeting other people with similar experiences provided a context for recognition, mutual understanding, and learning through sharing experiences and feelings. This may have helped to normalize feelings and facilitate being open to others, and it suggests a positive impact on tic severity by alleviating triggers to tics such as self-focused attention, tension, and anxiety in social situations.

We also suggest that participating in treatment groups augmented the effect of CBIT on the reduction of tic-related impairment. For instance, functional interventions provided in the group setting opened participants up for an exchange of ideas about and experiences with strategies to deal with challenges of living with tics, or how to modify tic-exacerbating situations, thereby helping to reduce tic-triggering situations. Our clinical experience showed that the patients became active participants in elaborating functional interventions such as new ways to deal with such tic-prone situations, for instance what to do or say, or challenging assumptions and beliefs about such situations, and thereby helping alter emotions coupled with such situations. In this way, functional interventions contributed to coping and suggest experienced empowerment. This may have positively affected social functioning such as increased engagement in social and daily life, boosting self-expression, and accordingly opened up opportunities for new and positive experiences.

Our results are in accordance with the study by Wilhelm *et al*. (**2012**), since this study also found significant improvement in tic-related impairment. Learning and mastering competing responses enables an increased control over tics. Experiencing such control over tics implies being less disturbed by the tics whenever they occur, with subsequent improvements in daily and occupational functioning. In addition, experiencing having better tic control may relieve stress and anxiety in tic-triggering situations and thus increase participation in daily social life and activities (McGuire *et al*., **2020**). Our clinical experience suggests that the patients differed in their use of competing responses. Some patients used competing response as a coping strategy only in situations in which they felt the need to control the tics, whereas other patients engaged in the use of competing responses more regularly and across different situations. According to McGuire *et al*. (**2014**), experience of significant overall improvement may be related to using competing response in situations in which tics cause impairment.

In line with the study of Wilhelm *et al*. (**2012**) and Yates *et al*. (**2016**), we found that motor tic severity showed a stronger reduction than vocal tic severity. Wilhelm *et al*. (**2012**) proposed that this finding could be explained by the fact that most patients chose to focus on motor tics rather than vocal tics. It should be noted, however, that the mean pretreatment level of motor tics was substantially higher than the mean level of vocal tics, in both Wilhelm's study and our study. Higher scores at baseline may facilitate larger change during treatment.

We found significant gains on the GTS-QoL physical and psychological subscales, but not on the obsessive-compulsive and cognitive subscales. The physical subscale overlaps with the YGTSS Impairment Score, and the significant gains we found on the YGTSS Impairment Score may explain the outcome on this subscale. Reduced tic severity also suggests less physical discomfort or pain. The positive outcome on the psychological subscale may be attributed to both effects of CBIT and group participation. For instance, CBIT may be beneficial for specific psychiatric symptoms and self-esteem (Weingarden *et al*., **2018**), symptomatology included in the psychological subscale. Moreover, CBIT is found to impact perception of quality of life in adults with TS (Cavanna *et al*., **2013**; Huisman-van Dijk, Matthijssen, Stockmann, Fritz & Cath, **2019**; Isaacs, Riordan & Claassen, **2021**). Besides these possible beneficial effects of CBIT, group participation may have been beneficial to well-being and quality of life as well. The group setting provided a safe and non-judgmental context, facilitating disclosure and support. Group processes such as identifying and sharing experiences and feelings with the other patients may have been helpful in gaining relief from shame and stigma and further improved self-esteem. Support and acceptance from the others is useful in helping to cope with negative feelings and beliefs. The group also became an arena in which to discuss and test out new ways to deal with bothersome situations and to enhance empowerment and coping.

CBIT, on the other hand, does not aim to treat obsessive-compulsive symptoms, which may explain the lack of significant gain on the obsessive-compulsive subscale. Symptoms of

ADHD are found to be associated with the cognitive subscale (Isaacs, Riordan & Claassen, 2021). Reduction in tic severity and increased tic control could suggest improvement in attention and concentration. However, the subscale includes additional factors that are not targeted in CBIT or that may not be responsive to CBIT, which may explain the lack of significant gains on the GTS-QoL cognitive subscale.

Limitations

Potential selection bias may reduce generalizability of the results. However, the sample appears to be representative for clinical samples of patients with TS or CTD exemplified by a similar co-occurrence pattern of OCD and ADHD as in other samples, e.g., Robertson *et al*. (2017).

The YGTSS was assessed by therapists participating in the treatment, and the effect sizes could have been inflated due to assessment bias by the therapists (researcher allegiance) or by the patients (social desirability bias). Moreover, the lack of a control condition implies that the results must be considered as preliminary. In future studies on group therapy for tics or TS, a control condition should be used and the YGTSS should be administered by independent raters. We do not know whether medication for tics might have influenced the results. Although we asked patients explicitly not to change their tic medication from pretreatment to 1-year follow-up, we did not track whether they really adhered to this no-change regime. Only one patient spontaneously told us that she did not need injections with botulinum toxin anymore because of the good effect of psychological treatment. Finally, with regard to administering the CGI-I, it should be mentioned that we deviated slightly from traditional procedures, and we do not know whether our procedures are as reliable as the traditional procedures.

Summary

The results of this pilot study indicate that CBIT administered in a group setting is a promising treatment for tics. Different patients may have benefited from the different components of CBIT and participation in a group per se. Group treatment may augment the effect of CBIT by interpersonal learning, normalization of feelings, and social support, as well as increased empowerment and belief in control of relevant techniques. The benefit of group participation as such was highlighted by all patients, emphasizing that it was of significant importance. The group represented a safe context for freely sharing experiences, thoughts, and feelings with others with similar lived experiences. Patients mentioned that being accepted and understood and sharing experiences were helpful in managing feelings, reframing problems, and working out strategies to cope with tic-related difficulties. Group participation thus helped ease shame and stigma, enhance self-esteem, and give new perspectives on one's own life, such as new insights and a more nuanced view about oneself.

FUNDING INFORMATION

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CONFLICT OF INTEREST

The authors do not have anything to disclose.