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Therapist-guided and parent-guided internet-delivered behaviour therapy for paediatric Tourette's disorder: a pilot randomised controlled trial with long-term follow-up

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ABSTRACT

Objective Behaviour therapy (BT) for Tourette's disorder (TD) and persistent (chronic) motor or vocal tic disorder (PTD) is rarely available. We evaluated the feasibility of adapting two existing BT protocols for TD/PTD (habit reversal training (HRT) and exposure and response prevention (ERP)) into a therapist-guided and parent-guided online self-help format.

Design A pilot, single-blind, parallel group randomised controlled trial.

Setting A specialist outpatient clinic in Sweden.

Participants Twenty-three young people with TD/PTD, aged 8–16.

Interventions Two 10-week therapist-guided and parent-guided internet-delivered programmes (called BIP TIC HRT and BIP TIC ERP).

Outcome The primary outcome measure was the Yale Global Tic Severity Scale. Blinded evaluators rated symptoms at baseline, post-treatment and 3-month follow-up (primary endpoint). All participants were naturalistically followed up to 12 months after treatment.

Results Patients and parents rated the interventions as highly acceptable, credible and satisfactory. While both interventions resulted in reduced tic-related impairment, parent-rated tic severity and improved quality of life, only BIP TIC ERP resulted in a significant improvement on the primary outcome measure. Within-group effect sizes and responder rates were, respectively: d=1.12 and 75% for BIP TIC ERP, and d=0.50 and 55% for BIP TIC HRT. The therapeutic gains were maintained up to 12 months after the end of the treatment. Adverse events were rare in both groups. The average therapist support time was around 25 min per participant per week.

Conclusions Internet-delivered BT has the potential to greatly increase access to evidence-based treatment for young people with TD/PTD. Further evaluation of the efficacy and cost-effectiveness of this treatment modality is warranted.

Trial registration number NCT02864589; Pre-results.

Strengths and limitations of this study

- ► Strengths include the randomised controlled design, high participant retention and few missing data points.
- ► Assessors were blind to treatment allocation at the primary endpoint (3-month follow-up).
- ► Participants were followed up to 12 months after treatment, with the 6-month and 12-month follow-ups being unblinded.
- ► Because all participants received an intervention, there was no control for the natural passage of time.

INTRODUCTION

In recent years, there has been a renewed interest in behavioural treatments for Tourette's disorder (TD) and persistent (chronic) motor or vocal tic disorder (PTD).[1] Several randomised controlled trials (RCTs) have shown that both habit reversal training (HRT) and exposure and response prevention (ERP) are effective in reducing tic severity and associated impairments.[2 3] Both treatments are currently recommended in European and Canadian guidelines[4 5] but are rarely available.[1] Reported barriers include a lack of information about tic disorders among service users and providers, a shortage of trained therapists and long travel distances to specialist treatment providers.[6]

One possible way to make behaviour therapy (BT) more accessible is to deliver it remotely. To date, two small RCTs (N=20) have evaluated HRT delivered through video-conference calls, compared with wait-list[7] and face-to-face BT.[8] These studies showed that video-conferencing is a feasible and acceptable format for the delivery of BT for TD/PTD. However, although video-conferencing may solve the issue of long travel distances, it does not remedy the shortage of trained therapists or the high costs associated with treatment.

Another approach to make BT more accessible is to deliver the treatment remotely as an online self-help programme, briefly supported by a therapist. A major advantage of this treatment format is that it requires considerably less therapist time than traditional face-to-face BT, thereby reducing overall costs.[9] Additionally, the majority of the treatment content is delivered online in a standardised format which minimises the risk of therapist drift and makes it less dependent on expert clinicians delivering the treatment. Therapist-guided internet-delivered self-help programmes have proven to be effective for a range of different mental health problems in both children and adults,[9 10] but have yet to be evaluated in TD/PTD. To our knowledge, two unguided internet-delivered programmes[11 12] are currently being evaluated in children and adults with TD/PTD (both based on HRT protocols). While pure self-help approaches are attractive due to the low costs of implementation, concerns have been raised about low adherence rates.[13]

With the aim to increase availability of BT for young people with TD/PTD, we developed two novel therapist-guided and parent-guided internet-delivered interventions based on existing HRT and ERP protocols.[14 15] Because we had no previous knowledge of whether both of these treatments would lend themselves equally well to a guided self-help format, we evaluated the feasibility, credibility, acceptability, potential efficacy and durability of both treatments in a pilot RCT. It is hoped that this study will help gather the necessary knowledge to optimise the interventions and design a definitive RCT.

METHODS

The study is reported according to the Consolidated Standards of Reporting Trials (CONSORT) statement.

Trial design

This was a pilot, single-blind, parallel group RCT of two novel therapist-guided and parent-guided internet-delivered behavioural treatments for children diagnosed with TD/PTD, called BIP TIC HRT and BIP TIC ERP. Participants were randomly assigned at a 1:1 ratio. The study did not aim to directly compare the two interventions, but to evaluate the feasibility and optimise the procedures of a future definitive RCT. No changes in the methods were made after the registration and subsequent start of the trial.

Participants

The study was carried out at the Child and Adolescent Psychiatry Research Center in Stockholm, Sweden. Inclusion criteria were: children and adolescents aged 7–17; a diagnosis of TD or PTD based on the fifth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-5)[16]; a Total Tic Severity Score >15 (or >10 if only motor or vocal tics had been present the last week) on the Yale Global Tic Severity Scale (YGTSS)[17]; a minimum of one available parent to support the child/adolescent throughout the treatment; child/adolescent and parent fluency in Swedish; and access to a computer connected to the internet.

Exclusion criteria were: acute psychiatric problems such as severe depression, suicidal risk, substance abuse or another psychiatric disorder that could interfere with treatment; a lifetime history of organic brain disorder, intellectual disability (based on whether the child attended a special needs school, according to parental report), pervasive developmental disorder, psychosis or bipolar disorder; severe tics causing immediate risk to the patient or others (ie, self-injurious tics, such as eye damage caused by repeated poking) and requiring urgent medical attention; previous BT (HRT or ERP) for a minimum of eight sessions with a qualified therapist within the 12 months prior to assessment; simultaneous psychological treatment for TD/PTD; or initiation or adjustment of any psychotropic medication for TD/PTD within the 6 weeks prior to assessment.

Patient involvement

Prior to the development of the BIP TIC interventions, a focus group was convened at our clinic in Stockholm, including five children with TD (and their parents). Families were asked a series of questions regarding the acceptability, convenience, ease of use and perceived efficacy of the proposed internet-delivered approach. In sum, we learnt that young people and their parents were enthusiastic about digital interventions for tics. The group was however not directly involved in the development of the treatment content.

Interventions

Treatments were delivered through an encrypted purpose-tailored online platform called BIP (*Barninternetprojektet* (Child Internet Project); www.bup.se/bip). The content of BIP TIC HRT and BIP TIC ERP is based on previously published evidence-based treatment manuals[14 15] and adapted to an online self-help format. Each treatment consists of 10 chapters (modules) of age-appropriate texts, animations, films and various exercises, delivered over 10 weeks (online supplementary table S1 and figure S1). In BIP TIC HRT, participants focus on one tic at a time, learn to become more aware of this tic, and to prevent the tic from occurring using a competing response. Eleven specifically created films, featuring a clinical psychologist (the first author), are used to illustrate a wide range of competing responses corresponding to specific muscle groups. In BIP TIC ERP, participants practice to suppress all tics at the same time (response prevention), and then to (with the help of the parent) gradually provoke bodily sensations (premonitory urges) to make tic suppression more challenging (exposure). BIP TIC ERP includes an inbuilt stopwatch to help practice tic suppression, and children are encouraged to suppressing tics for increasing periods of time.

Parents have separate logins to the online platform and can access extended versions of the treatment content (online supplementary table S1). Specifically, parents

learn about parental coping strategies, social support and functional analysis (the latter as in Woods *et al*[15]).

Throughout the treatment, a therapist supports each family via written messages within the platform and occasional phone calls. Therapists logged in to the online platform at least once a day during working hours to provide guidance and feedback, answer questions and prompt participants to login in case of inactivity. Therapists were two clinical psychologists with considerable clinical experience in treating TD/PTD and two clinical psychology trainees with no previous experience or contact with this patient group. The trial coordinator (clinical psychologist) trained and supervised the trainees regularly to ensure adherence to treatment protocols. Families continued to have access to the treatment for the entire duration of the follow-up, but without therapist support. All therapist time was logged, either automatically (platform logins) or manually (phone calls).

Outcome measures and assessment points

Indicators of feasibility and acceptability included the technical aspects of successfully adapting the original HRT/ERP face-to-face protocols to a therapist-guided and parent-guided internet-delivered format, ease of participant recruitment, attitudes from referring clinicians, treatment credibility and participant retention.

The primary outcome measure was the Total Tic Severity Score of the YGTSS, a semistructured clinician-administered interview with two independent ratings of tic symptom (motor and vocal) severity and tic-related impairment, each scored on a 0–50 scale.[17] All study assessors were trained in the YGTSS and watched a total of five video-recorded cases. Assessors achieved excellent inter-rater reliability (intraclass correlation=0.98).

Secondary clinician-rated outcome measures were the YGTSS Impairment score, the Children's Global Assessment Scale (CGAS),[18] and the Clinical Global Impression–Severity (CGI-S) and Improvement (CGI-I).[19] Following previous work in the field,[2] treatment response was defined as a rating of 1 (*very much improved*) or 2 (*much improved*) on the CGI-I.

Secondary self-rated outcome measures were the Premonitory Urge for Tics Scale (PUTS);[20] the Gilles de la Tourette Syndrome-Quality of Life Scale (GTS-QOL),[21] an adapted child version of the Work and Social Adjustment Scale (WSAS-Y (child)),[22] the Obsessive Compulsive Inventory-Child version[23] and the Children's Depression Inventory-Short version (CDI-S),[24] with an additional item screening for suicidal ideation.

Secondary parent-rated outcome measures were the Parent Tic Questionnaire (PTQ),[25] an adapted parent version of the Work and Social Adjustment Scale (WSAS-Y (parent)),[22] and the Short Mood and Feelings Questionnaire-Parent version (SMFQ).[26] All self-rated and parent-rated outcome measures were administered over the internet.

All outcome measures were administered at baseline, post-treatment and at 3, 6 and 12-month follow-up. The 3-month follow-up was pre-specified as the trial's primary endpoint. In addition to the above-mentioned assessment points, the PTQ, PUTS, CDI-S and SMFQ questionnaires were also administered 5 weeks into treatment (mid-treatment).

Adverse events were registered using an adapted version of the Safety Monitoring Uniform Report Form (SMURF)[27] at mid-treatment (via telephone) and post-treatment (at the clinic).

Recruitment and procedures

Information about the study was primarily distributed to health services, patient organisations and advertised on the study website (www.bup.se/bip). Participants could either be referred by a mental health professional or self-refer through the website. After a brief telephone screening, families were given an appointment with a clinical psychologist at our specialist obsessive-compulsive disorder and related disorders clinic. The aim of this visit was to confirm the TD/PTD diagnosis using DSM-5 criteria and to rate symptom severity using the YGTSS. Comorbid psychiatric diagnoses were established using the Mini International Neuropsychiatric Interview for Children and Adolescents (MINI-KID).[28] If eligible, families received additional information about the study and both children and parents signed the informed consent. Participants were informed they would be randomised to one of two evidence-based BT protocols for TD/PTD administered over the internet. After being enrolled by the assessor, participants were randomised by the trial coordinator and asked to fill in the baseline self-rated and parent-rated online questionnaires within a few days. Treatment started within 1 week of inclusion.

Post-treatment and 3-month follow-up assessments were mainly conducted face-to-face at the clinic, except for four 3-month follow-up assessments which were done via video-conference software (VSee). Six-month and 12-month follow-up assessments were conducted face-to-face (n=29), via video-conference (n=14) or via telephone (n=3), depending on travel distances, personal preferences or technical complications.

Randomisation and allocation concealment

Participants were allocated using a randomisation sequence in blocks of two stratified by Attention-Deficit/Hyperactivity Disorder (ADHD)-status (according to the MINI-KID assessment). The sequence was created by an independent researcher using an online service (www.sealedenvelope.com) and then concealed in sequentially numbered opaque sealed envelopes. Post-treatment and 3-month follow-up assessments were conducted by one clinical psychologist and two clinical psychology trainees who were blind to treatment allocation. Participants were explicitly informed not to disclose information about treatment content during the assessments. To measure blinding integrity, all assessors guessed each participant's treatment allocation at each assessment point. As per protocol, blinding was broken after the 3-month follow up

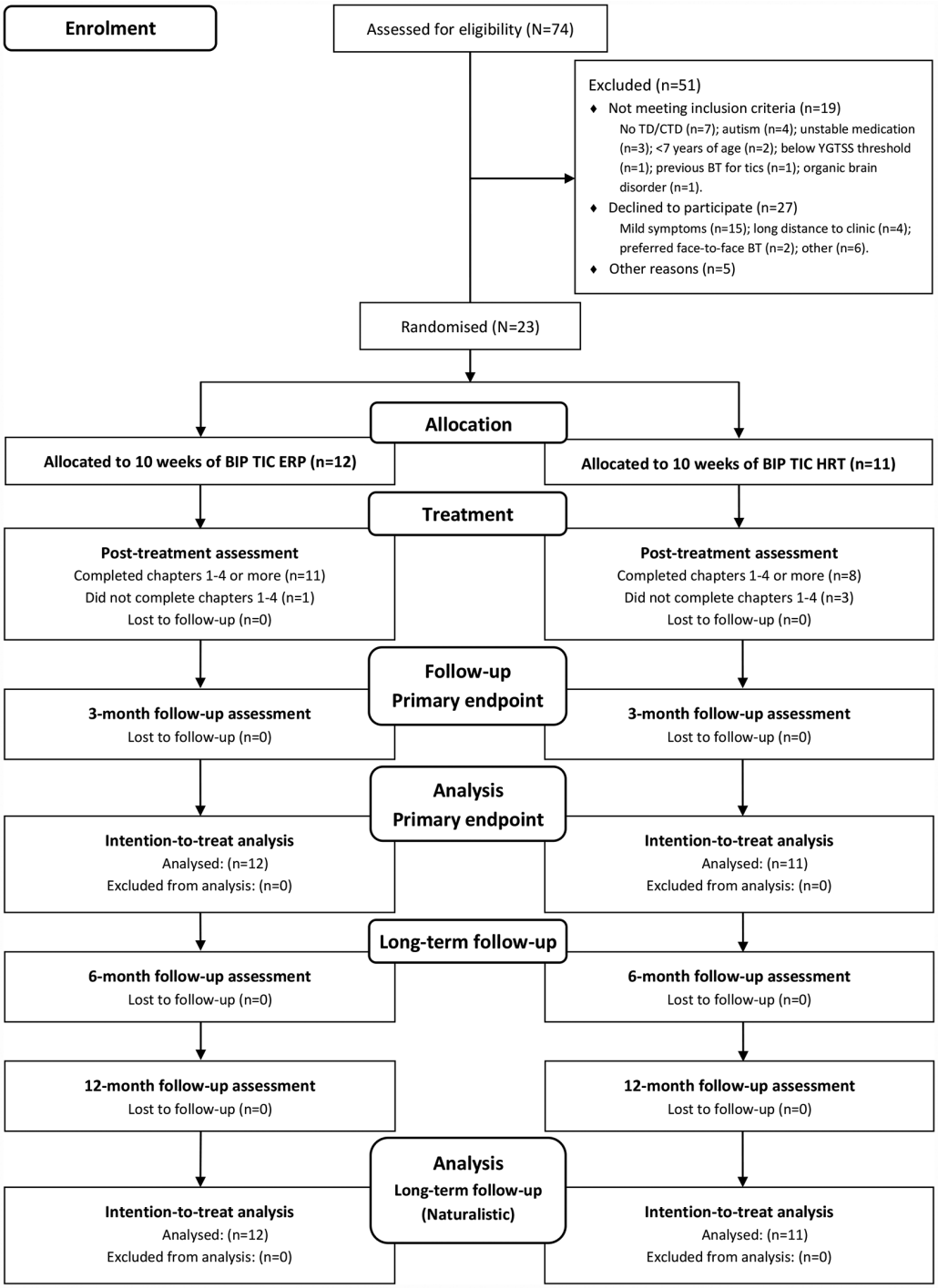

Figure 1 Consolidated Standards of Reporting Trials flow diagram. BIP TIC ERP, internet-delivered exposure and response prevention; BIP TIC HRT, internet-delivered habit reversal training; BT, behaviour therapy; PTD, persistent (chronic) motor or vocal tic disorder; TD, Tourette's disorder; YGTSS, Yale Global Tic Severity Scale, Total Tic Severity Score.

assessments, and the naturalistic 6-month and 12-month follow-up assessments were thus unblinded.

Power calculation

Power calculations were done using G*Power V.3.1. The trial was powered to detect significant within-group changes in tic severity in each treatment arm. Given a standardised Cohen's *d* effect size of 1.0,[2 29] 10 participants would be required in each group (paired t-test, 80% power, p<0.05, allowing for 20% dropout). The trial did not aim, and was hence not powered, to detect superiority or equivalence of BIP TIC HRT and BIP TIC ERP.

Statistical analyses

Intention-to-treat analyses were performed using mixed-effects regression models for repeated measures on all continuous outcome measures. The models included fixed effects of time, and random intercepts and slopes for the participant effects, and used all available data. The main analysis included baseline, post-treatment

Table 1 Demographic and clinical characteristics of the sample, by treatment condition

	BIP TIC ERP (n=12)	BIP TIC HRT (n=11)
Age, mean (SD); min–max	11.80 (2.51); 8–15	12.79 (2.62); 8–16
Males, n (%)	8 (67)	7 (64)
Tic disorder, n (%)		
TD	12 (100)	10 (91)
PTD	0 (0)	1 (9)
Comorbidity, n (%)		
Anxiety disorder	2 (17)	4 (36)
Attention-deficit/hyperactivity disorder	5 (42)	4 (36)
Depression	0 (0)	1 (9)
Obsessive-compulsive disorder	2 (17)	1 (9)
Previous contact with child/adolescent mental health services		
Yes, n (%)	12 (100)	10 (91)
Previous psychological treatment for TD/PTD, n (%)		
None	11 (92)	8 (73)
Behaviour therapy	1 (8)	2 (18)
Other	0 (0)	1 (9)
Baseline medication status		
None	11 (92)	8 (73)
Antipsychotic	0 (0)	2 (18)
Stimulant	0 (0)	1 (9)
SSRI	0 (0)	1 (9)
Melatonin	1 (8)	0 (0)
Distance from home to clinic, km, mean (median); min–max	51 (23.5); 7–212	84 (37.6); 5–416
Education, main parent involved in treatment, n (%)		
Primary school	0 (0)	1 (9)
Secondary school	1 (8)	3 (27)
College/university	9 (75)	6 (55)
Doctoral degree	2 (17)	1 (9)

BIP TIC ERP, internet-delivered exposure and response prevention; BIP TIC HRT, internet-delivered habit reversal training; PTD, persistent (chronic) motor or vocal tic disorder; SSRI, selective serotonin reuptake inhibitor; TD, Tourette's disorder.

and 3-month follow-up (primary endpoint) data. In order to investigate the durability of the treatment outcomes, we also fitted separate models including the primary endpoint, and the 6-month and 12-month follow-ups. All alpha levels (two-tailed) were set to p<0.05. Within-group effect sizes were calculated using the Cohen's *d* formula.[30] Logistic regression was used to evaluate the integrity of the blinding procedures. All analyses were performed using Stata V.14.2 (StataCorp).

RESULTS

Study flow and participants

A total of 23 participants were recruited between 18 August and 14 October 2016 and randomised to BIP TIC HRT (n=11) or BIP TIC ERP (n=12) (figure 1). Follow-up assessments were performed up to 12 months after the end of treatment, with the last data collected on 2 January 2018. There were no missing data points for any measure in the BIP TIC ERP group. In the BIP TIC HRT group, one participant had missing data on four secondary measures at mid-treatment. Table 1 summarises the characteristics of the sample.

Study take-up, credibility, module completion and satisfaction

The 23 participants were recruited during an approximate period of 8 weeks. Only 2 out of 74 assessed families declined participation because they preferred face-to-face treatment (figure 1). Three weeks into treatment, children and parents rated both treatments as credible (online supplementary table S2).

The average number of completed chapters was 7.92 (for both children and parents; SD=2.47) in the ERP group, and 7.36 (children; SD=3.04) and 7.09 (parents; SD=2.91) in the HRT group. Six children (50%) and five parents (42%) in the ERP group, and five children and parents (45%) in the HRT group completed all 10 chapters. Treatment satisfaction at post-treatment was high in both groups (online supplementary table S3).

Primary outcome measure

Table 2 shows the means and SDs for each assessment point and group. Mixed-effects regression analyses at the primary endpoint (3-month follow-up) showed a significant reduction on the YGTSS Total Tic Severity Score for BIP TIC ERP, but not for BIP TIC HRT (figure 2). Within-group Cohen's *d* was 1.12 for BIP TIC ERP and 0.50 for BIP TIC HRT. Post-hoc analyses of the YGTSS Motor and Vocal Tic Severity scales showed significant reductions in motor tic severity in both groups and a significant reduction in vocal tic severity in the BIP TIC ERP group, but not in the BIP TIC HRT group.

Treatment response

At the primary endpoint, nine participants (75%) in the BIP TIC ERP group and six participants (55%) in the BIP TIC HRT group were classified as treatment responders according to the CGI-I. Online supplementary figure S2 shows CGI-I ratings at all assessment points.

Secondary outcome measures

All secondary outcome measures are reported in table 2 and in online supplementary table S4. At the primary endpoint, significant reductions were found in both treatment groups on YGTSS Impairment, PTQ, PTQ Motor Tic Severity subscale (post-hoc analysis) and GTS-QOL, as well as a significant increase in global functioning on CGAS. Additionally, significant reductions were found on PTQ Vocal Tic Severity subscale (post-hoc analysis) in the BIP TIC ERP group and on WSAS-Y (child) in the BIP

Table 2 The YGTSS for the BIP TIC ERP and BIP TIC HRT groups at all measure points

	BIP TIC ERP (n=12)			BIP TIC HRT (n=11)		
		Within-group difference†	Within-group effect size‡		Within-group difference†	Within-group effect size‡
YGTSS	Mean (SD)*	Coefficient (95% CI); p value	Cohen's *d* (95% CI)	Mean (SD)*	Coefficient (95% CI); p value	Cohen's *d* (95% CI)
Total Tic Severity Score						
Baseline	23.75 (5.26)	–	–	23.45 (6.88)	–	–
Post-treatment	19.00 (7.48)	−4.75 (−8.18 to −1.32); p=0.007¶	0.73 (-0.10 to 1.56)	21.18 (6.19)	−2.27 (−5.65 to 1.11); p=0.187	0.35 (−0.50 to 1.19)
3-month follow-up§	18.25 (4.54)	−5.50 (−8.93 to −2.07); p=0.002¶	1.12 (0.24 to 1.97)	20.18 (6.21)	−3.27 (−6.65 to 0.11); p=0.058	0.50 (−0.36 to 1.34)
6-month follow-up	15.00 (7.01)	−3.25 (−6.37 to −0.13); p=0.041¶	0.55 (−0.27 to 1.36)	19.45 (7.47)	−0.73 (−3.71 to 2.26); p=0.633	0.11 (−0.73 to 0.94)
12-month follow-up	16.92 (5.55)	−1.33 (−4.46 to 1.79); p=0.403	0.26 (−0.54 to 1.06)	19.36 (8.48)	−0.82 (−3.80 to 2.17); p=0.591	0.11 (−0.73 to 0.95)
Motor Tic Severity						
Baseline	14.42 (1.73)	–	–	15.27 (1.85)	–	–
Post-treatment	11.42 (4.17)	−3.00 (−4.85 to −1.15); p=0.001¶	0.94 (0.08 to 1.78)	13.18 (2.27)	−2.09 (−3.65 to −0.53); p=0.009¶	1.01 (0.11 to 1.89)
3-month follow-up§	12.33 (1.97)	−2.08 (−3.93 to −0.24); p=0.027¶	1.12 (0.25 to 1.98)	12.55 (2.70)	−2.73 (−4.29 to −1.17); p=0.001¶	1.18 (0.26 to 2.08)
6-month follow-up	9.67 (5.05)	−2.67 (−4.69 to −0.64); p=0.010¶	0.70 (−0.14 to 1.51)	12.55 (2.58)	0.00 (−1.25 to 1.25); p=1.000	0.00 (−0.84 to 0.84)
12-month follow-up	10.58 (1.73)	−1.75 (−3.77 to 0.27): p=0.090	0.94 (0.09 to 1.78)	12.36 (3.17)	−0.18 (−1.43 to 1.07); p=0.775	0.06 (−0.77 to 0.90)
Vocal Tic Severity						
Baseline	9.33 (4.98)	–	–	8.18 (6.19)	–	–
Post-treatment	7.58 (4.01)	−1.75 (−4.17 to 0.67); p=0.156	0.39 (−0.43 to 1.19)	8.00 (5.37)	−0.18 (−2.71 to 2.34); p=0.888	0.03 (−0.80 to 0.87)
3-month follow-up§	5.92 (4.72)	−3.42 (−5.84 to −1.00; p=0.006¶	0.70 (−0.13 to 1.52)	7.64 (4.76)	−0.55 (−3.07 to 1.98); p=0.672	0.10 (−0.74 to 0.93)
6-month follow-up	5.33 (4.68)	−0.58 (−2.57 to 1.40); p=0.564	0.12 (−0.68 to 0.92)	6.91 (5.68)	−0.73 (−2.85 to 1.40); p=0.503	0.14 (−0.70 to 0.97)
12-month follow-up	6.33 (5.42)	0.42 (−1.57 to 2.40); p=0.680	−0.08 (−0.88 to 0.72)	7.00 (6.08)	−0.64 (−2.76 to 1.49); p=0.558	0.12 (−0.72 to 0.95)
Impairment						
Baseline	16.67 (6.51)	–	–	17.27 (7.86)	–	–
Post-treatment	6.67 (7.78)	−10.00 (−13.83 to −6.17); p<0.001¶	1.39 (0.48 to 2.28)	10.00 (7.75)	−7.27 (−12.18 to −2.37); p=0.004¶	0.93 (0.04 to 1.81)
3-month follow-up§	4.17 (5.15)	−12.50 (−16.33 to −8.67); p<0.001¶	2.13 (1.10 to 3.13)	8.18 (9.82)	−9.09 (−14.00 to −4.19); p<0.001¶	1.02 (0.12 to 1.90)
6-month follow-up	1.67 (3.89)	−2.50 (−5.25 to 0.25); p=0.075	0.55 (−0.27 to 1.36)	7.27 (9.05)	−0.91 (−3.39 to 1.57); p=0.473	0.10 (−0.74 to 0.93)
12-month follow-up	1.67 (3.89)	−2.50 (−5.25 to 0.25); p=0.075	0.55 (−0.27 to 1.36)	7.27 (10.09)	−0.91 (−3.39 to 1.57); p=0.473	0.09 (−0.75 to 0.93)

*Observed means.
†Coefficients at post-treatment and at the 3-month follow-up compare with baseline, while coefficients at the 6-month and 12-month follow-ups compare with the 3-month follow-up.
‡All Cohen's *d* effect sizes are calculated from observed data. Post-treatment and the 3-month follow-up effect sizes compare to baseline, while the 6-month and 12-month follow-up effect sizes compare to the 3-month follow-up. Effect sizes of 0.2, 0.5 and 0.8 are considered small, moderate and large, respectively.
§Primary endpoint.
¶Significant at an alpha level of 0.05.

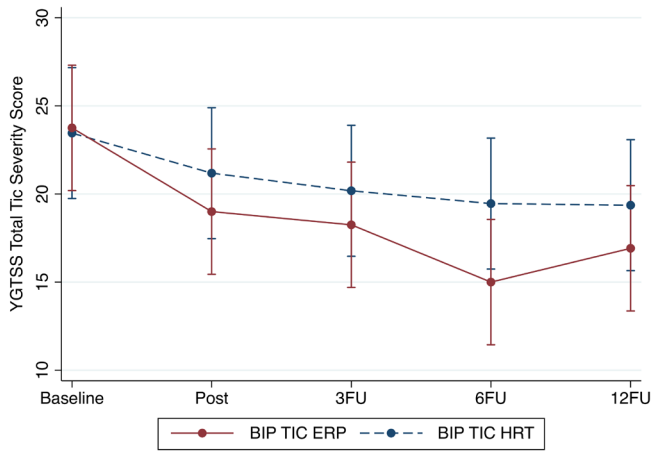

Figure 2 Graphical representation of the YGTSS Total Tic Severity Score across the five assessment points. Primary endpoint is the 3-month follow-up; after this point, assessments are not blinded. Error bars indicate 95% CIs. 3FU, 3-month follow-up; 6FU, 6-month follow-up; 12FU, 12-month-follow-up; BIP TIC ERP, internet-delivered exposure and response prevention; BIP TIC HRT, internet-delivered habit reversal training; Post, post-treatment; YGTSS, Yale Global Tic Severity Scale.

TIC HRT group. Online supplementary figure S2 shows CGI-S ratings for all assessment points.

Therapist support time

The average therapist time per participant and week (children and parents combined) was 23.84 (SD=7.99) min in the BIP TIC ERP group and 26.90 (SD=10.96) min in the BIP TIC HRT group. This includes both messages in the platform and occasional telephone calls of short duration (the latter representing a mean of 1.34 (SD=1.34) min per participant and week for the two groups).

Long-term follow-up

As shown in table 2 and online supplementary table S4, patients in both groups largely maintained their therapeutic gains on primary and secondary measures between the 3-month (primary endpoint) and the 12-month follow-ups. Patients randomised to BIP TIC ERP improved further on the YGTSS Total Tic Severity Score between the 3-month and the 6-month follow-ups (coefficient (95% CI)=−3.25 (−6.37 to −0.13), p=0.041), but this reduction was temporary.

Blinding concealment and protocol deviations

Treatment allocation was unintentionally revealed to blind assessors at two occasions (one in each treatment condition, both at the 3-month follow-up). Besides these two deviations, the assessors' guesses were not better than chance (45% and 30% correct guesses in BIP TIC ERP and BIP TIC HRT groups, respectively; p=0.262).

One participant in the BIP TIC HRT group increased the dosage of risperidone from 0.50 to 0.75 mg between the pre-treatment and mid-treatment assessments. Another participant, also in the BIP TIC HRT group, started risperidone (0.50 mg) between the post-treatment assessment and the primary endpoint. Mixed-effects regression analysis of the BIP TIC HRT group excluding these affected data points showed similar results (data not shown). No other protocol deviations were recorded during the controlled phase of the trial.

During the naturalistic follow-up period, no participants received additional TD/PTD-specific psychological treatment. Four participants, all in the BIP TIC HRT group, altered their medication for TD/PTD during the follow-up: one initiated and later terminated treatment with guanfacine between the 3-month and 6-month follow-ups; one increased and later decreased (to the former level) the dosage of aripiprazole between the 3-month and 6-month follow-ups; and two decreased their dosage of risperidone (from 0.75 to 0.50 mg, and from 0.50 to 0.25 mg, respectively) between the 6-month and 12-month follow-ups.

Adverse events

Twelve participants self-reported adverse events between baseline and mid-treatment and five participants between mid-treatment and post-treatment, as assessed by the SMURF. None of the adverse events were rated as serious (online supplementary table S5).

DISCUSSION

We evaluated the feasibility of adapting two existing evidence-based BT protocols for TD/PTD[14 15] into a parent-guided online self-help format, with brief therapist support. Patients and parents rated the interventions as highly credible and satisfactory. It was relatively easy to recruit to the study—reflecting the shortage of specialist care for this patient group—and participant retention was high (100% complete data at 12-month follow-up). While both interventions resulted in reduced tic-related impairment, parent-rated tic severity and improved quality of life, only BIP TIC ERP resulted in a significant improvement on the primary outcome measure at the primary endpoint. The therapeutic gains were maintained up to 12 months after the end of treatment. Adverse events were rare in both groups. Therapist support time was only a fraction of that required in conventional face-to-face BT, indicating potential cost-effectiveness.

The trial did not aim, or was powered, to compare the relative efficacy of the two interventions. However, within-group effect sizes for the primary outcome measure and some secondary measures were approximately twice as large for BIP TIC ERP as for BIP TIC HRT, and the number of treatment responders was 75% vs 55%, respectively. Interestingly, post-hoc analyses of the YGTSS and PTQ subscales showed that both BIP TIC ERP and BIP TIC HRT resulted in significantly improved motor tic severity, but only BIP TIC ERP improved vocal tic severity. The remote delivery of HRT may be more technically demanding, particularly for vocal tics, as finding a suitable competing response for specific tics may require

more therapist guidance, whereas it may be easier to teach patients to suppress all tics at once, as in ERP. In keeping with the original protocols, HRT required considerably more content to deliver (208 compared with 152 online pages/screens in ERP), and thus may have been harder to grasp. Despite this, the rate of treatment responders in the BIP TIC HRT group (55%) was still comparable with that obtained in the largest paediatric face-to-face HRT trial to date (53%).[2] Our results should not be taken as evidence of superiority of ERP over HRT per se but, rather, of a more successful technological transfer into the online self-help format.

This trial had several strengths, including its novel approach, a randomised controlled design following CONSORT statement guidelines, the use of blind assessors, high participant retention, few missing data points and the long-term follow-up. The latter is a rare feature in TD/PTD trials but is particularly important, as the severity of tics is known to fluctuate naturally over time. The study also had limitations. First, while the trial was powered to detect large within-group effect sizes, more variability in the data may have resulted in insufficient power to detect a statistically significant improvement at the primary endpoint for the BIP TIC HRT group. Second, our design did not control for the natural passage of time. Third, the 6-month and 12-month follow-up assessments were unblinded. Finally, it is worth noting that the participants were largely self-referred, highly motivated and possibly less complex than other clinically recruited samples. However, nearly all participants had previously been in contact with child and adolescent mental health services, and the baseline YGTSS Total Tic Severity scores were similar to previous BT trials.[2 3 7 8] Future evaluations of BIP TIC should aim to recruit more broadly to ensure adequate representation of the full tic disorder population.

CONCLUSION

To summarise, it is feasible, acceptable and safe to deliver BT for paediatric TD/PTD remotely via the internet, with minimal therapist support. ERP is possibly easier to deliver using this format. We also provide preliminary data suggesting that BIP TIC may be efficacious and cost-effective, and that its effects may be maintained long-term. The results of this pilot study will inform the design of a definitive RCT to evaluate the efficacy and cost-effectiveness of BIP TIC ERP, compared with either a credible control condition (eg, a therapist-guided internet-delivered version of the supportive psychotherapy and education used by Piacentini *et al*[2]) or other treatment modalities.

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Contributors The trial was designed by DM-C, ES, EA and PA. PA, with assistance from TM, developed the treatment content. PA was the trial coordinator. Treatment was provided by PA, KA, PW and SR. Statistical analyses were performed by PA, in collaboration with LFC, EA and KI. PA drafted the manuscript, in collaboration with DM-C. The manuscript was reviewed and revised by all authors who have also read and approved the final version.

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