Online remote behavioural intervention for tics in 9- to 17-year-olds: the ORBIT RCT with embedded process and economic evaluation

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Disclosure of interests

Full disclosure of interests: Completed ICMJE forms for all authors, including all related interests, are available in the toolkit on the NIHR Journals Library report publication page at https://doi.org/10.3310/CPMS3211.

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Scientific summary

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Scientific summary

Background

Tic disorders including Tourette syndrome and chronic tic disorders are common conditions that affect approximately 1% of the population in the UK. Young people with tics often report substantial impairment, thus it is important that they have access to evidence-based treatment. Face-to-face behavioural therapy (BT) such as exposure and response prevention (ERP) may be offered to some young people. However, due to a lack of trained therapists, there are often difficulties accessing BT, and there is a better need to understand the clinical and cost-effectiveness of the online delivery of such therapy.

Objectives

The primary objective of this study was to evaluate the clinical effectiveness of therapist-guided, parent-assisted, internet-based ERP BT for tics in young people with tic disorders compared to online psychoeducation.

Secondary objectives included (1) optimising the design of the intervention, (2) undertaking an internal pilot, (3) evaluating cost-effectiveness, (4) establishing whether the efficacy is maintained longer term, (5) understanding the mechanisms of impact of the intervention and (6) identifying barriers to implementation.

Methods

We conducted an individually randomised (1 : 1 ratio), multicentre trial, with an internal pilot and embedded process evaluation. Participants were assigned to either receive online, remotely delivered, therapist- and parent-supported ERP for tics or online, remotely delivered, therapist- and parent-supported psychoeducation for tics.

Participants were recruited from the two study sites, 16 patient identification centres in England or could self-refer online via the study webpage or via Tourettes Action (a national charity for tics).

The inclusion criteria were age between 9 and 17 years, with tics assessed on the Yale Global Tic Severity Scale (YGTSS), able to provide written informed consent (parental consent for children aged < 16 years) and with a suitable device they could use to access the internet. Exclusion criteria included receiving a therapy for tics in the past 12 months, starting or stopping tic medication within the past 2 months and intellectual disability/substance use/anorexia nervosa/psychosis/suicidality, moderate/severe intellectual disability, risk to self or others or parent or young person unable to speak or read/write English.

All potential participants attended a screening/baseline appointment at one of the two study centres. Participants who were eligible and consented were randomised into one of two study groups. In the intervention group participants received 10 weeks of the remotely delivered, therapist-guided ERP behaviour therapy. In the control group participants received 10 weeks of remotely delivered, therapist-guided psychoeducation about tics.

Participants completed measures at the mid-treatment point (5 weeks) and at 3 and 6 months (this formed phase 1, per-protocol design). For phase 2 (a naturalist design), follow-up measures were

obtained at 12 and 18 months. The primary outcome (at 3 months) was the total tic severity score (TTSS) on the YGTSS. Secondary outcomes included measures of tics (parent tic questionnaire), general difficulties (strengths and difficulties questionnaire), mood and anxiety (moods and feelings questionnaire and Spence Child Anxiety Scale), global functioning (Children's Global Assessment Scale, Clinical Global Impressions-Improvement), adverse events, need for further treatment, treatment credibility and satisfaction and the Child and Adolescent Version of the Gilles de la Tourette Syndrome Quality of Life Scale. Quality of life [child health utility 9D (CHU9D)] and resource use (modified child and adolescent service use schedule) data were also collected for the economic evaluation. Follow-up assessments were completed online or via telephone/Webex videoconferencing (YGTSS).

The trial internal pilot evaluated recruitment rate, engagement with the intervention and retention to the primary outcome at 9 months into the trial and the results were reported to the relevant oversight committees (Trial Steering Committees and Data Monitoring Committee). A sub-sample of parents and young people in the intervention arm, clinicians and therapists were interviewed to explore barriers/facilitators to implementation and refine the intervention for future use, which formed part of the mixed-methods process evaluation. The quantitative data for the process evaluation included intervention usage metrics, clinical and demographic trial data and therapist contacts.

Intervention

The intervention was delivered via Barninternetprojektet (Child Internet Project; Swedish digital platform) (BIP), a Swedish web-based digital platform. The BT intervention (ERP) was translated from the Swedish original intervention (BIP TIC), refined and adapted for UK use by our trial team. The active control intervention (psychoeducation) was created by our trial team.

Both interventions consisted of 10 web-based chapters, designed to last 10 weeks. Participants had regular contact with a therapist during this time via messages that could be sent inside the treatment platform (resembling an email). The therapist's role was to give specific feedback to motivate the patient and not to deliver therapeutic content.

The young person and the parent/carer were provided with their own separate logins to the BIP platform. For both the intervention and the comparator, treatment completion was defined as completion of the first four child chapters.

Results

The trial recruited and retained participants exceeding the pre-specified criteria for the internal pilot and therefore proceeded to the full trial.

In total 445 candidates signed up to the study and were assessed for eligibility, of which 221 potential participants were excluded (90 did not meet inclusion criteria, 84 declined to participate and 47 were unable to contact family). This meant 224 participants were enrolled and randomly assigned (1 : 1) to either the ERP BT group (n = 112) or psychoeducation group (n = 112).

The characteristics of the two groups were similar at baseline. The enrolled patients were mostly male (n = 177; 79%) and of white ethnicity (n = 195; 87%). Only 13% of participants were receiving tic medication.

Adherence to the intervention was good in 99 (88%) of the 112 participants in the ERP group, and 105 (94%) of the 112 participants in the psychoeducation group were classified as treatment completers (i.e. completing at least the first four chapters). Retention to the primary outcome at the 3-month primary

end point (90%) and 6-month follow-up (> 80%) was excellent. Retention to the primary outcome measure remained high at 12 months (81% in both arms) and 18 months (> 79% in both arms).

The primary analysis showed that participants in the ERP group [16% reduction, standard deviation (SD) 1.1] had a greater decrease in tics than those in the psychoeducation group (6% reduction, SD 1.0) at 3 months (primary end point). The estimated mean difference in YGTSS-TTSS change between the groups adjusted for baseline and site was −2.29 points [95% confidence interval (CI) −3.86 to −0.71] in favour of ERP, with an effect size of −0.31 (95% CI −0.52 to −0.10). This effect was sustained at 6 months, with a mean decrease of 6.9 points (24%, SD 1.2) in the ERP group versus 3.4 points (12%, SD 1.0) in the psychoeducation group.

For phase 2, participants in the ERP group continued to have a greater decrease in tics than the control group. The estimated mean difference in YGTSS-TTSS between groups adjusting for baseline and site at 12 months was −2.64 points (95% CI −4.48 to −0.79), with an effect size of −0.36 (95% CI −0.61 to −0.11), at 18 months it was −2.01 points (95% CI −3.86 to −0.15), with an effect size of −0.27 (95% CI −0.52 to −0.02), in favour of the ERP group.

In addition, extended follow-up showed those receiving online ERP compared with online psychoeducation had reduced scores for low mood and anxiety at 12 and 18 months and superior tic-specific quality of life, with the largest effects seen at 18 months.

The direct cost of the intervention was £155 per person, including £104.57 for the online platform, supervision and training and a mean variable cost of £50.43 per participant for therapist time in the trial. At 18 months, using proxy parent-completed CHU9D responses, there were a mean additional 0.040 (95% CI −0.004 to 0.083) quality-adjusted life-years (QALYs) per participant in the ERP group compared with psychoeducation, with an addition mean cost per participant of £662 (95% CI −£59 to £1384). The incremental cost-effectiveness ratio in the primary analysis was £16,708 per QALY gained from a health and social care cost perspective at 18 months. In the 10-year long-term decision model, online ERP cost £537 less per participant than face-to-face BT and resulted in 0.02 fewer QALYs.

Two serious adverse events (SAEs) occurred (hospital attendance due to one ‘collapse’ and one ‘tic attack’), both in the active control psychoeducation group, neither of which were related to the study intervention.

The process evaluation found that the ERP intervention was implemented with high fidelity, and participants found the intervention acceptable and satisfactory. Engagement was high, with child participants completing an average of 7.5/10 chapters and 99/112 (88.4%) participants completing the minimum of the first four chapters (the predefined threshold for effective dose). Parental engagement was the only significant independent predictor of child engagement. Improvement in tic severity and overall clinical condition was not moderated by the relationship between demographic or baseline clinical factors and engagement and no mediators were found. However, level of parental engagement was associated with overall clinical improvement, and this relationship was illuminated by the qualitative data.

Conclusion

Implications for health care

- The findings demonstrate that online, therapist-supported ERP for young people with chronic tic disorders is clinically effective at reducing tic severity. Therefore, this is an efficient public mental health approach to supporting young people with tics.

- The intervention can be delivered at lower cost than standard face-to-face BT and may also result in improved service efficiencies, allowing a greater number of young people to access evidence-based care.

Future research implications

- Further 'field trials' should be conducted to explore the clinical and service implications of delivering the intervention in real-world settings.
- Given that online interventions are context dependent, exploring the validity of these findings in different cultures/countries is important.

Future research should explore where online, therapist-supported ERP best fits in the tic disorder care pathway and how online and face-to-face therapy can be best combined (e.g. non-responders to online ERP are 'stepped up' to face-to-face therapy).

Trial registrations

This trial is registered as ISRCTN70758207 and ClinicalTrials.gov (NCT03483493). The trial is now complete.

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