Fidelity of delivery and contextual factors influencing children's level of engagement with an online behavioural intervention for tics: Process evaluation of the ORBIT trial

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Abstract

Background: The Online Remote Behavioural Intervention for Tics (ORBIT) study was a multicentre randomized controlled trial of a complex intervention that consisted of an online behavioural intervention for children and young people (CYP) with tic disorders. In this first part of a two-stage process evaluation, we conducted a mixed-methods study exploring reach, dose, and fidelity of the intervention and contextual factors influencing engagement with the intervention.

Objective: This study aims to explore the fidelity of delivery and the contextual factors underpinning the ORBIT intervention.

Methods: Baseline study data and intervention usage metrics from participants in the intervention arm were used as quantitative implementation data (n=112). The experiences of being in the intervention were explored by semi-structured interviews with children (n=20) and parent (n=20) participants, therapists (n=4), and referring clinicians (n=6). A principal components analysis was used to create a comprehensive, composite measure of CYP's engagement with the intervention. Engagement factor scores reflected relative uptake as assessed by a range of usage indices including chapters accessed, number of pages visited and number of logins.

Results: The intervention was implemented with high fidelity, and participants deemed the intervention acceptable and satisfactory. Engagement and adherence were high with child participants completing an average of 7.5/10 chapters and 100/112 (89.3%) participants completed a minimum of 4 chapters: the pre-defined threshold for effective dose. Compared to the total population of children with tic disorders, the sample tended to have more educated parents and live in more economically advantaged areas but socioeconomic factors were not related to engagement factor scores. Factors associated with higher engagement factor scores included participants enrolled at the London site vs. the Nottingham site (P=.011), self-referred vs. clinic-referred (P=.041), higher parental engagement as evidenced by number of parental chapters completed (?=0.73, n=111, P<.001) and more therapist time for parent (?=0.46, n=111, P<.001). A multiple linear regression indicated that parents' chapter completion (?=.69, t110=10.18, P<.001) and therapist time for parent (?=.19, t110=2.95, P=.004) were the only significant independent predictors of engagement factor scores.

Conclusions: Overall, the intervention had high fidelity of delivery and was evaluated positively by participants, although reach

may have been constrained by the nature of the randomized controlled trial. Parental engagement and therapist time for parent were strong predictors of intervention implementation which has important implications for the design and implementation of digital therapeutic interventions into Child and Adolescent Mental Health Services. Clinical Trial: International Standard Randomized Controlled Trial Number (ISRCTN) 70758207; https://doi.org/10.1186/ISRCTN70758207 and ClinicalTrials.gov NCT03483493; https://clinicaltrials.gov/ct2/show/NCT03483493

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Abstract

(396 words (*max 450 words*))
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Conclusions: Overall, the intervention had high fidelity of delivery and was evaluated positively by participants, although reach may have been constrained by the nature of the randomized controlled trial. Parental engagement and therapist time for parent were strong predictors of intervention implementation which has important implications for the design and implementation of digital therapeutic interventions into Child and Adolescent Mental Health Services.

Trial registration: International Standard Randomized Controlled Trial Number (ISRCTN) 70758207; https://doi.org/10.1186/ISRCTN70758207 and ClinicalTrials.gov NCT03483493; https://clinicaltrials.gov/ct2/show/NCT03483493

Keywords: process evaluation; implementation fidelity; Tourette syndrome; chronic tic disorders; online behavioural intervention; mixed methods; children and young people.

Background

Tics are sudden, brief, rapid, and recurrent non-rhythmic movements or vocalizations that are more common in children and young people (CYP) than in adults [1]. Tic onset typically occurs between the ages of 3 and 8 years (mean onset is 6 and 7 years of age) [2] with the reported average age of greatest tic severity by age 10 years [3]. Although most CYP with tics only require educational support as the main form of treatment [4], there are interventions available for severe or disabling tics, as in Tourette syndrome (TS) or chronic tic disorders. Historically, pharmacotherapy, such as antipsychotics, have been the first line of treatment for severe tics; however, they often have undesirable side effects, such as weight gain and sleepiness [5]. Behavioural interventions are appealing and effective alternatives to pharmacotherapy. However, they require the patient to invest time and energy in practicing demanding behavioural techniques such as tic control or habit reversal. Despite the benefits and evidence-based effectiveness of behavioural therapies for tic disorders [6–8], there is great difficulty in patients accessing behavioural treatments due to a shortage of trained therapists [9]. One promising development in increasing accessibility to behavioural treatments is the use of Digital Health Interventions (DHIs) [10]. There is preliminary evidence that DHIs are efficacious for CYP with tic disorders in pilot randomized controlled trials (RCT) [11–13]. A study that has assessed DHIs for tic disorders is the ‘Online Remote Behavioural Intervention for Tics’ (ORBIT) trial, which has been described in detail previously [14] (see Figure 1 for brief description).

Design: A 10-week, two-armed, parallel group, single blind, randomized controlled trial (RCT) with an embedded process evaluation.

Aim: To evaluate the effectiveness of an online, remote, therapist-supported and parent-guided behavioural intervention for tics, initially developed and piloted in Sweden called BIP TIC.

Intervention group: 112 children and young people received 10 modules (called 'chapters') of behavioural therapy following the principles of Exposure and Response Prevention (ERP) via a secure online platform, with access to a therapist, delivered over a period of 10-12 weeks.

Control group: 112 children and young people received 10 chapters of psychoeducation via a secure online platform, with access to a therapist, delivered over a period of 10-12 weeks.

Primary outcome: Total Tic Severity Score (TTSS) on the Yale Global Tic Severity Scale (YGTSS) at 3-months post randomization.

Therapist role: Both children and parents had regular contact with a therapist during the 10-12 weeks via messages that were sent within the treatment platform (resembling an email) or telephone if required. The therapist was also able to directly comment on exercises that the participant had been working on, and give specific feedback to motivate participants. All participant contact with the therapist was asynchronous.

Parent role: One or both of the child's parents received a separate login to the online treatment where they could access their own chapters. The parent chapters contained information regarding parent coping strategies, how to support their child in working with BIP TIC and functional analysis relating to tics. They also had access to the assigned therapist.

Figure 1. Brief description of the Online Remote Behavioural Intervention for Tics (ORBIT) trial.

The population impact of any given intervention depends on both its effectiveness and its reach, defined as the proportion of the target population who access the intervention [15]. Although RCTs are the "gold standard" method for determining efficacy, additional data are needed before a decision as to whether an intervention should be adopted into mainstream healthcare can be reached. These additional data include understanding the reach of the intervention, and the extent to which the data from an RCT, where the delivery of the intervention is often tightly controlled and monitored, can be extrapolated to use in routine healthcare. It has been argued that studies addressing questions about

reach and effectiveness in routine care are needed [16,17]. However, like all research, such studies are expensive, and a process evaluation conducted alongside an RCT is an efficient method of maximising the information yielded by the trial.

The Medical Research Council (MRC) has developed specific guidelines for conducting process evaluations of complex interventions [18]. The MRC outline three essential components for evaluating complex interventions: implementation, mechanisms of impact, and context. Implementation can refer to how an intervention will be delivered within routine clinical practice, having shown efficacy in an outcome evaluation. However, this paper is concerned with another aspect of implementation: the extent to which the delivery of an intervention is achieved within the context of an RCT and the structures and processes through which an intervention is delivered as intended (i.e. fidelity) [18]. For complex interventions like DHIs, an important component of implementation fidelity is the degree to which participants engage with the intervention, and use it as intended. Effective engagement requires participants to register with the programme, and then continue to use it and apply the recommended behavioural techniques over time. Non-use of DHIs is a well-recognised challenge (e.g. Eysenbach's Law of Attrition [19]), and can be considered in two parts: initial uptake (e.g. registration/onboarding) and ongoing adherence or engagement.

In order to evaluate intervention implementation, MRC guidelines for process evaluations suggest researchers assess: i) reach – the extent to which a target audience comes into contact with the intervention; ii) dose – how much intervention is delivered and received; iii) fidelity – the quality of what was delivered; iv) adaptations – any modifications made to an intervention in order to achieve better contextual fit. The intended target audience for ORBIT was CYP with tic disorders; however, there were pertinent questions that could be asked, such as whether there were socioeconomic biases in who was reached. In terms of dosage, the ORBIT protocol [14] states the intervention should consist of 10 individual intervention chapters following a suggested frequency and total duration of 10-12 weeks. There were 4 core chapters, and this was deemed the minimum requirement for

treatment completion. There were 6 additional chapters offering reinforcement, further practice, and relapse prevention. For DHIs, the fidelity of delivery of the intervention is assured by the online delivery platform. However, the intervention that is experienced by the user is highly dependent on the extent to which they engage with the intervention and use it as intended. Hence in this paper, we look at usage and the proportion of participants receiving the pre-defined "minimum effective dose" of four or more chapters. Finally, understanding adaptations to the intended intervention involves exploring whether these improve its contextual fit or compromise its functioning [20], or whether they represent innovation, or intervention drift [21]. Participants were able to make modifications to various components of the intervention, such as the "tic stopwatch" which was used to self-time the length of tic control.

The aim of this study was to conduct the first part of a two-stage process evaluation of the ORBIT trial as outlined in the study protocol [22]. Part 1 focuses on intervention implementation by exploring the fidelity of delivery experienced by participants using usage statistics, reach, and the acceptability of the intervention. It also investigates contextual factors associated with the observed variation in uptake and usage by examining the components specified in MRC guidelines [18]. Mechanisms of impact will be explored in part 2. Table 1 shows the two parts of the process evaluation, areas of research, explanatory data, and outcomes.

Table 1. Process evaluation parts, areas of research, explanatory data, and outcomes

Process evaluation components	**Research questions**	**Explanatory data**	**Outcomes**
Part 1. Intervention implementation (What is implemented and how?)	➢ Fidelity of implementation ➢ Dose of intervention delivered ➢ Adaptations ➢ Reach	➢ Therapist contact/time (n=112) ➢ Intervention adherence (n=112) ➢ Usage metrics (n=112) ➢ Clinician (n=6), children and parent (n=20), therapist (n=4)	➢ Engagement and satisfaction with the intervention

		interviews	
Part 2. Mechanisms of impact (How does it produce change?)	➢ Mediators and moderators ➢ Unexpected pathways and consequences	➢ Usage metrics ➢ Therapist contacts ➢ Clinician, children and parent, therapist interviews	➢ Relationship between engagement with intervention and change in the severity of tics
Part 1 & 2. Context (How do factors external to the intervention affect intervention implementation and change?)	➢ Factors related to fidelity of delivery (part 1) and improvement in tics (part 2)	➢ Demographic data ➢ Clinician, children and parent, therapist interviews ➢ Service use ➢ Comorbidities ➢ Baseline severity of tics	➢ Engagement with the intervention (part 1) ➢ Change in tic severity (part 2)

Methods

Study design

This study followed MRC guidelines [18] for the process evaluation of complex interventions and used a mixed-methods, longitudinal design to explore the implementation fidelity of an online intervention for CYP with tics [14] and the contextual factors that influenced level of engagement.

Participants

The sample included in the quantitative phase of the process evaluation consisted of key information from all participants (n=112) from the intervention arm of the RCT. The sample included in the qualitative component of the process evaluation consisted of interviews with child and parent participants (target n=>20), interviews with all therapists delivering the intervention or supervising the therapists, and interviews with referring clinicians (target n>5).

Quantitative data collection

Quantitative process data were collected simultaneously along with enrolment, intervention delivery, and outcome data collection in the main RCT.

Demographic and clinical data

Demographic and clinical information was recorded from a baseline demographics questionnaire. These data included the child's age, residence (full postcode), gender, ethnicity, parental education level and occupation, all current suspected or confirmed diagnoses and interventions, and medication use.

Index of Multiple Deprivation

Index of Multiple Deprivation (IMD; 2019) is a relative measure of deprivation across seven different domains: income deprivation; employment deprivation; education, skills and training deprivation; health deprivation and disability; crime; barriers to housing and services, and living environment deprivation [23]. Based on the six-digit postcode, a rank of deprivation associated with participants' area of residence was calculated (https://www.gov.uk/government/statistics/english-indices-of-deprivation-2019) from 32844 small areas or neighbourhoods in England, with higher ranks indicating greater deprivation. Ranks were re-coded into quintiles with 1 being most deprived and 5 being least deprived.

Yale Global Tic Severity Scale

The primary outcome measure used in the ORBIT intervention was the Total Tic Severity Score (TTSS) as measured by the Yale Global Tic Severity Scale (YGTSS). The YGTSS is a valid and reliable, clinician-rated scale [24], which scores the severity of motor and vocal tics separately by an evaluation of the number, frequency, intensity, complexity, and interference of tics. Each domain is scored on a 0-5 scale. Two tic severity scores are given: total motor (0-25) and total vocal (0-25), which when combined give the TTSS (0-50).

Mood and Feelings Questionnaire

The Mood and Feelings Questionnaire (MFQ) [25] is a 33-item measure evaluating depressive symptoms rated on a 3-point scale: 0 is "not true"; 1 is "sometimes" and 2 is "true". Total scores range from 0 to 66 with higher scores reflecting more severe depression. A cut-off score of ≥29 is

generally used to suggest clinically significant depression [26].

Usage metrics

Online usage data was collected and recorded from participants throughout the trial. This included the following measures: number of chapters completed per child and per parent; total therapists' time per child and per parent; individual therapist's telephone time with participants; volume of written communication (total number of characters) submitted by child and parent via the online system; total number of logins for child and for parent; average time between each login (in days) for child and for parent; and average pages visited per login for child and for parent.

Satisfaction and treatment credibility

At the 3-week post-randomization point of treatment, participants were asked to rate treatment credibility. Two questions were asked: one relating to how well suited the participant felt the intervention was for helping CYP to manage their tics and the other question was about how much better they expected to feel as a result of the intervention. The responses were on a Likert-scale of 0 to 4 for each question with higher scores indicating higher treatment credibility. At the primary endpoint, participants were asked to rate their satisfaction with the intervention. Eight satisfaction questions were asked with responses rated on 0 to 4 scales meaning the overall satisfaction score was out of 32.

Qualitative data collection

Interviews with therapists and therapist supervisors involved in the ORBIT trial were conducted early in the study and near the end of recruitment in order to gain an understanding of their experience at different time points. Interviews with referring clinicians were conducted at the end of recruitment. Interviews with children and parent participants were conducted following completion of the intervention at the three-month (primary end-point) follow-up assessment in the main RCT in order to minimize the risk of bias in the outcomes. Recruitment for the interviews began in August 2018 and ended in October 2019.

All interviews were conducted either face-to-face, by telephone, or via videoconferencing (WebEx or Skype). Younger children were interviewed together with their parents, while older children (>13 years old) were interviewed separately. Participants were purposively sampled so that a diverse range of views on the intervention were voiced. This included ensuring perspectives were heard from participants with a range of ages, gender, ethnicity, and level of interaction with the intervention. The overall sample enabled a diversity of views of the intervention and ensured that data reached a level of saturation. In addition to the interviews, at the end of treatment, all participants were asked to give their overall feedback on the intervention to which they could provide open-ended responses. Table 2 demonstrates how the various data sources contribute to different components of implementation fidelity.

Table 2. Implementation fidelity components and data sources

	Reach	Dose	Fidelity	Adaptations	Context
Quantitative data sources					
Demographic and clinical data	✓				✓
Usage metrics		✓			
Treatment credibility and satisfaction			✓		
Qualitative data sources					
Child interviews		✓	✓	✓	✓
Parent interviews		✓	✓		✓
Therapist and clinician interviews	✓		✓	✓	✓
End of treatment feedback questionnaire			✓		

Data analysis

The quantitative data set were presented with total numbers and percentages and mean with standard deviation (SD) or median (range), if not normally distributed. Data were tested for normality using the Kolmogorov–Smirnov test. A principal components analysis was run to determine a composite measure of level of engagement. Correlations between variables were examined using bivariate

Spearman correlations, and a *t*-test was calculated to explore any significant differences between groups. A multiple linear regression was calculated to identify predictors of engagement with independent variables. All statistical analyses used a significance level of $P<0.05$ and were conducted using IBM SPSS Statistics 25.

All interviews were recorded either by videoconferencing software or by dictaphone and were then transcribed verbatim. Transcripts were checked for accuracy against the recordings with any corrections made as appropriate and anonymized for confidentiality purposes. As the process evaluation was a combination of exploration and description, the Framework Method [27] of analysis was used to identify, analyze, and report patterns within the transcribed interviews. Moreover, the steps outlined by Gale et al. [28] were systematically followed to create an overall framework matrix using categories of adherence and potential moderators. Consistency of analysis was ensured throughout by the use of a codebook and through frequent meetings between researchers. Researcher bias was minimized through regular cross-checking of data and outcomes by members of the research team.

The software package QSR NVivo 12 was used to analyze the interview data. In addition, the end of treatment feedback questionnaire was exported to an Excel spreadsheet and quantitative content analysis [29] was performed. Overall, the findings from the qualitative analysis were linked to relevant quantitative adherence outcomes and contextual factors to assess which potential moderators may have influenced implementation fidelity and in what way, in an approach termed ‘triangulation’ [30].

Ethical considerations

Ethical approval for the process evaluation was obtained from North West - Greater Manchester Central Research Ethics Committee as part of the ORBIT trial (REC: 18/NW/0079). All child and parent participants provided written informed consent and all interview participants provided oral consent for audio-recording.

Results

Semi-structured interviews were conducted with children (n=20) and parents (n=20), therapists (n=4) and clinicians (n=6). The average age of child interviewees was 12 years (range 9-16 years) with 16/20 (80%) of the sample being male and 4/20 (20%) females. The majority of the sample was white (18/20, 90%). The mean TTSS was 28.8/50 (SD 7.2) with a range of 13-45 for child interviewees. All 20 of the interviews with the parents were with the CYP's mother with all 20 having completed at least further education. One of the therapist interviewees was a therapist's supervisor and the majority of clinicians were consultant psychiatrists (3/6, 50%).

Reach

Participants were eligible for the study if they were aged 9–17 years, with a suspected or confirmed tic disorder, competent to provide written, informed consent (parental consent for a child aged <16 years) and had broadband internet access and regular use of a computer, with mobile phone text messaging facilities. Patients were excluded from the study if they had received any form of structured behavioural intervention for tics within the preceding 12 months, had a change of medication for tics within the previous two months, any diagnoses of alcohol/substance dependence, psychosis, suicidality, or anorexia nervosa or moderate/severe intellectual disability, were an immediate risk to self or others, and/or parent or child was not able to speak, or read and write English.

Four hundred and forty-five families expressed an interest in taking part in the study either through self-referral via Tourettes Action charity website (n=251) or via clinic referral (n=194); however, 47 were subsequently uncontactable and 90 were ineligible to take part. Of the 308 potentially eligible CYP, 84 families (27.3%) declined to take part and 112/224 CYP (90 male, 22 female) with an average age of 12.2 years (range 9-17; Table 3) were randomized to the intervention arm of the ORBIT trial and were included in the process evaluation. The sample was predominantly white (96/112, 85.7%) and well-educated with just over half (60/112, 53.5%) of the participants mothers

having completed university/higher education.

The median IMD rank was 19318 with a range of 147 to 32668 (out of 32844). Of the 112 participants, 8 (7.1%) were in the most deprived quintile (1), 31 (27.7%) in quintile 2, 18 (16%) in quintile 3, 26 (23.3%) in quintile 4, and 29 (25.6%) were in the least deprived quintile (5). Although the reach of the intervention was not limited geographically, for the purposes of the research participants did have to attend a baseline screening assessment at either the Nottingham study site (57/112, 50.9%) or the London study site (55/112, 49.1%) depending on personal preference and/or location of residence. All participants were based in England with 63/112 (56.3%) participants living in towns, 30/112 (26.7%) in cities, and 19/112 (17%) living in villages.

In terms of clinical characteristics, the intervention reached a moderately severe symptomatic sample with a mean TTSS of 28.5 (SD 7.7) out of a maximum of 50, with a range of 12-50. The majority of participants (100/112, 89.3%) were not on any medication for their tics and just under half of the overall intervention sample had no diagnosed or suspected comorbidities (51/112, 45.5%). Of those who did have a comorbid diagnosis, the most common was attention-deficit/hyperactivity disorder (ADHD; 26/112, 23.2%). An assessment of depressive symptoms by the MFQ showed a mean score of 16.2 (SD 11.3) out of 66 with 14/112 (12.5%) participants scoring above the cut-off (≥29) suggesting clinically significant depression [26].

Table 3. Demographic and clinical characteristics of participants in the ORBIT trial intervention group (n=112)

	n (%)
Mean age, years (range)	12.2 (9-17)
Gender	
Male	90 (80.4)
Female	22 (19.6)
Study site	
Nottingham	57 (50.9)
London	55 (49.1)
Ethnicity	
White	96 (85.7)

Asian	7 (6.2)
Mixed race	3 (2.7)
Other	6 (5.4)
Supporter	
Mother	93 (83.0)
Father	16 (14.3)
Other	3 (2.7)
Highest level of education (Mother)	
Did not complete compulsory education	3 (2.7)
Completed compulsory secondary education	16 (14.3)
Completed further education	33 (29.5)
Completed university/higher education	43 (38.4)
Completed postgraduate taught degree	11 (9.7)
Completed doctorate/medical degree	6 (5.4)
Highest level of education (Father)	
Did not complete compulsory education	2 (1.8)
Completed compulsory secondary education	29 (25.9)
Completed further education	35 (31.2)
Completed university/higher education	29 (25.9)
Completed postgraduate taught degree	10 (8.9)
Completed doctorate/medical degree	7 (6.3)
Method of referral	
Self	69 (61.6)
Clinic	43 (38.4)
IMD rank, median (range)	19318 (147-32668)
No tic medication	100 (89.3)
On tic medication	12 (10.7)
Comorbidities	61 (54.5)
No comorbidities	51 (45.5)
TTSS baseline score, mean (SD)	28.5 (7.7)
MFQ, mean (SD)	16.2 (11.3)

Note *IMD – Index of Multiple Deprivation; TTSS – Total Tic Severity Score; MFQ – Mood and Feelings Questionnaire.

It was not possible to interview people who had not taken part in the study so the qualitative data threw little light on reach; however, one clinician identified the following issue under the theme

clinician perceptions of and contribution to recruitment (see Multimedia Appendix 1 for full list of framework categories and themes):

"So children quite often with autism umm other kind of family reasons where I think they were just worried about the level of that kind of commitment to umm an intervention to be able to kind of travel to Nottingham or London for the initial assessment" (Clinician 3, Psychiatrist).

Another clinician highlighted the lack of access to children with intellectual disabilities:

"So say for example they've got severe intellectual inabilities so they're non-verbal you know so clearly they're not gonna be able to access the trials and things. I mean even somebody with a mild umm intellectual disability to be honest if it was on the low end of the mild so kind of like between 50 to 60 in the IQ kind of thing…you would struggle to, you know, to access it" (Clinician 1, Psychiatrist).

Finally, one of the clinicians struggled to gain her colleagues' interest in the intervention despite numerous attempts:

"So but the interesting thing is to get clinicians interested in it and thinking about the children because we have a big Trust with three areas and I have sent it out over and over and over and over again and I think the uptake has been really low from the other err professionals" (Clinician 2, Psychiatrist).

Dose

Child participants completed an average of 7.5 (SD 2.7; Table 4) and their parents completed an average of 7.6 (SD 2.8; Table 5) out of 10 chapters of the intervention indicating high engagement. Only 12/112 (10.7%) child participants and 13/112 (11.6%) parents failed to meet the criteria for treatment completion (i.e. minimum of 4 chapters completed as per protocol) with a total of 100/112 (89.3%) child participants and 99/112 (88.3%) parents completing their treatment, meaning that adherence to the intervention was high. Indeed, 46/112 (41%) CYP and 52/112 (46.4%) parents completed all 10 chapters of the intervention and only 1 child participant failed to complete any

chapters. Participants were given 10 weeks of supported therapeutic input in order to complete their treatment chapters. In some circumstances, such as holidays or particularly busy periods, 1 or 2 weeks were added on to supplement this time. Although the majority of families (73/112, 65%) finished their therapy within 10 weeks, 39/112 (35%) required extra time to complete treatment. Child participants logged onto the online treatment platform an average of 19.8 (SD 10.9) times throughout the 10-12 weeks with an average of 4.2 (SD 2.6) days between logins. In terms of total interactions with their assigned therapist, child participants required their therapist's online assistance for an average of 59 minutes 14 seconds (SD 00:29:08) over the course of treatment, which results in around 6 minutes per child per week. Whereas, parents interacted online with their assigned therapist an average of 1 hour 23 minutes 55 seconds (SD 00:42:45), which results in around 8 minutes per parent per week. Of 112 CYP, only 2 (1.8%) were contacted by telephone by their assigned therapist. Of 112 parents, 49 (43.7%) were contacted by telephone by their assigned therapist.

Table 4. Usage data for child participants in the ORBIT trial intervention group (n=112)

	Median (Range)	**Mean (SD)**
Chapters completed	8 (0-10)	7.5 (2.7)
Total therapist time, hh:mm:ss	00:53:57 (00:07:27 - 03:11:08)	00:59:14 (00:29:08)
Telephone time with therapist, hh:mm:ss	00:00:00 (00:00:00-00:18:44)	00:00:10 (00:01:46)
Number of logins	19 (3-57)	19.8 (10.9)
Number of days between logins	3 (1-16)	4.2 (2.6)
Number of pages visited per login	15 (7-38)	16.9 (5.8)
Total number of characters submitted	2507 (238-8749)	2784 (1608)

Table 5. Usage data for parents in the ORBIT trial intervention group (n=112)

	Median (Range)	**Mean (SD)**
Chapters completed	9 (1-10)	7.6 (2.8)
Total therapist time, hh:mm:ss	01:15:33 (00:22:01 -	01:23:55 (00:42:45)

	04:48:19)	
Telephone time with therapist, hh:mm:ss	00:00:00 (00:00:00-00:49:00)	00:04:06 (00:07:41)
Number of logins	18 (3-50)	20.4 (11.4)
Number of days between logins	4 (0-19)	4.2 (2.7)
Number of pages visited per login	17 (9-36)	17.4 (5.2)
Total number of characters submitted	6533 (346-29631)	7286 (5093)

Interview data relating to participants' *perceptions of ORBIT organization* covered the implementation component of dose. Although the majority of participants felt that the intervention was just the right length, some CYP wished to have a longer period of time in which to access their therapist:

"I just liked doing the whole bit of ORBIT and chatting to my therapist but I think it was too short. Cause I could only chat to my therapist for 10 weeks, but then we had a full year logging on to ORBIT but we could not chat to our therapist which I found a bit annoying" (Child 20, 12 years old).

One child felt that the intervention could have been condensed to make it shorter:

"9 weeks with 12 chapters. Make the chapters shorter. Some of them are like 13 pages like you have to do the questions. Like those pages questions" (Child 26, 9 years old).

On the whole, parents agreed with their child that the dose received was just right with one parent claiming if it was longer it would have affected engagement:

"Just the right length. I think if it'd been any longer he'd have got he wouldn't have engaged as much" (Parent 26, Mother).

Fidelity

At the 3-week point of post-randomization, participants were asked to rate treatment credibility. Treatment credibility was rated highly by child participants with a mean score of 6.4 (SD 1.5) out of

8. Furthermore, at the primary end-point, participants were asked to rate their overall satisfaction with the intervention. Child participants were highly satisfied with the intervention with a mean score of 24.8 (SD 5.2) out of a total of 32. At the end of treatment, participants were asked to give their feedback on the intervention within the online platform and they were able to give open-ended responses. Only 67/112 (59.8%) child participants provided this feedback. From the quantitative content analysis conducted, four categories were generated relating to implementation fidelity, namely, 'limitations of ORBIT' (51/67), 'ORBIT as a suitable treatment' (49/67), 'problems with using ORBIT' (20/67), and 'feeling supported' (19/67). The main code relating to 'limitations of ORBIT' centred on *improvement required* (n=33). This code captured anything related to the intervention being unhelpful or inappropriate. Examples included repetitiveness of treatment, the treatment being too short or too long, unhelpful aspects, and suggested improvements. Two child participants reported technical issues with the ORBIT platform, which related to intermittent problems with connectivity. Despite this, many participants felt the intervention was acceptable as a treatment with the largest number of participants being coded at *positive experience of ORBIT* (n=42), which related to being pleased to have taken part and finding it enjoyable whilst recommending the treatment to other CYP with tic disorders.

Although satisfaction was rated highly, some participants felt that the role of the therapist was somewhat misleading. This was captured by the theme *expectations of role of the therapist*. Some felt that a therapist was not needed for the delivery of the intervention:

"Like that just I don't like emailing so I think I felt a bit awkward cause I didn't really know how to write back but I felt most of the comments were quite generic…I don't know just I'd say something and [therapist] be like 'oh well done'…Um but I don't think [therapist] necessarily has to be there. I think you could have done it on your own" (Child 21, 15 years old).

Some parents agreed with this sentiment:

"I probably could have done without the therapist because I would want a therapist to advise me

about [child's name] tics I didn't need advising about using the therapy, does that make sense?" (Parent 25, Mother).

The term 'therapist' itself was felt to be somewhat misjudged as a label:

"I don't know that the therapist was of any use. We didn't utilise the therapist I don't think. It was more sort of it felt like they were cheering you on…they are more like a motivator than a therapist I think. I kind of maybe expected a little too much from the ORBIT study" (Parent 30, Mother).

The therapists themselves concurred with this:

"I think part of it would come down to whether we would want to use the word 'therapist' within ORBIT because there's a lot of semantics and meaning about that word and I'm not sure off the top of my head if therapist or…what's the lay meaning of therapist basically? Does that mean psychotherapist, does that mean someone who's got a doctorate, who knows? So, everyone could… participants come into that with their own meaning and it also assumes that I…they've got expectations about what a therapist is, it assumes that I'm the expert and I really felt like I wasn't in this. My supervisors were experts" (Therapist 1).

At the end of the interviews, participants were asked if they had any recommendations in order to improve the intervention and the overriding majority felt that a mobile application was needed in future iterations of the intervention:

"I mainly focused on um wanting to beat my score and like I couldn't actually put that on when I was like…I couldn't actually put it online when I was um just like in lesson or when I was like doing it… watching TV, just like do the stop clock on my phone. So I think like if they had an app or something" (Child 27, 13 years old).

Whilst some of the older CYP felt that the content and presentation of the intervention was childlike and aimed more towards the younger participants. Therefore they felt there could be two separate versions:

"The layout and stuff was very much directed to younger kids. Um and I think if there was like a

separate part of ORBIT that was for more like teenagers and stuff um and um the videos were a bit more um accustomed to young children. And um I think if there was just a bit there that was more directed to teenagers I think it would be better in that way" (Child 14, 13 years old).

Adaptations

Regarding adaptations, the intervention did not appear to evolve in any way from the original plans. There appeared to be consistency in the way the intervention was delivered and received. Interviews with therapists confirmed how consistency was maintained in delivery:

"We had standardized documents, of like a collection of standardized responses so any time we'd come across something unique or difficult or not immediately obvious to answer, after sort of emailing around and reviewing potential answers we'd obviously say how to come up with an answer to send to the participant and once I'd done so, I'd add a section into the collection of responses and add it in. So basically, we had something we could look at and call upon when we see someone and go 'look, we're not sure how to answer that, let me check this document' and then you can see if there was anything similar, or it's been answered before umm, that was very useful…" (Therapist 2).

Parts of the intervention were designed to be adapted by the user and tailored to their needs and preferences, such as the 'tic stopwatch' and 'tic ladder' (hierarchy of exposure exercises):

"I had to answer questions in the chapters and when I finished it I could go back and change it and I could change my ladder when I do my tics and where I do my tics most often and my tic list of what I have. I liked the idea that I could change it. And it helped me" (Child 20, 12 years old).

Another participant adapted the intervention to make it easier to complete:

"We um changed some of the activities that like um so one of them was like um doing trying to suppress your tics whilst focussing only on your tics. But I really wasn't able to do that one at all really so we did that while I was watching TV or like being on my phone. So we changed some bits" (Child 22, 15 years old).

Contextual factors influencing intervention implementation

In order to establish a measure of intervention implementation that captured both the breadth and depth of participants' usage, a principal components analysis with varimax (orthogonal) rotation was conducted on the 7 items relating to the dose of intervention received. The analysis suggested a two-factor model. The strongest factor accounted for 47% of the variance (Eigenvalue 3.3) (see Table 6) and seemed to capture strength of engagement with the intervention. Factor scores ranged from -2.65 to 2.26 with a mean of 0.001 (SD 0.99) and these scores were used as the engagement measure.

Table 6. Summary of principal components analysis for child's usage data for the ORBIT intervention (n=111)

	Factor Loadings	
Item	**Factor 1 - Engagement**	**Factor 2 – Sporadic use**
Number of logins	.90	
Chapters completed	.79	
Total therapist time for child	.76	
Total number of characters submitted	.74	
Number of days between logins	-.63	.54
Number of pages visited per login	-.41	.80
Telephone time with therapist	-.44	-.46
Eigenvalue	**3.3**	**1.5**
% of variance	**47**	**21**

A 2-tailed *t*-test found that participants who were enrolled at the London site (Mean 0.25, SD 0.90) scored significantly higher on engagement compared to those enrolled at the Nottingham site (Mean -0.22, SD 1.03), t_{109}=-2.58, *P*=.011. Moreover, those who were self-referred (Mean 0.16, SD 0.94) scored higher on engagement than those who were referred through clinics (Mean -0.24, SD 1.04), t_{109}=-2.06, *P*=.041. Spearman's rho correlations were run to determine the association between engagement and various contextual factors. CYP's engagement factor score was strongly correlated with parents' chapter completion (ρ=0.73, n=111, *P*<.001) and moderately correlated with therapist

time for parent (ρ=0.46, n=111, *P*<.001). There were no significant relationships between CYP's engagement factor score and age, parental education, IMD, TTSS at baseline, or MFQ baseline score. There were also no statistically significant relationships between child's gender, comorbidities, or use of tic medication and CYP's engagement.

A multiple linear regression was conducted with CYP's engagement factor score as the dependent variable, and site, child's age, child's gender, IMD, TTSS, method of referral, parental education, therapist time for parent, and parents' chapter completion as the independent variables. The results of the simultaneous regression indicated that collectively the independent variables had a significant amount of variance on the CYPs engagement factor score, $F_{10,100}$=20.84, *P*<.001, R^2=.64. There was no evidence of multi-collinearity, with all tolerances above 50%, and all variance inflation factors below 2. Only parents' chapter completion (β=.69, t_{110}=10.18, *P*<.001) and therapist time for parent (β=.19, t_{110}=2.95, *P*=.004) were significant independent predictors in the model.

Under the framework category 'participant contextual factors', the theme of *parental persuasiveness* was generated. Many of the parents interviewed outlined that they were often the main motivating force behind their child's level of engagement:

"If he's got a really bad tic and I'll say to him you know, [child's name] use your tic timer in your head, try and see how long you can do he will then do it. Um but he doesn't really use the techniques himself without being reminded to… So I suppose that was a little bit of a disappointment" (Parent 15, Mother).

Some parents found motivating their child to engage very challenging:

"Obviously for me trying to keep [child's name] engaged um on the computer and with the time aspect, um you know that was the challenging part" (Parent 28, Mother)

This was even more challenging for those with children who have comorbidities:

"I knew I'd have to help motivate him um cause he has ADHD…he's got easily distracted and um he hasn't got a great attention span but that was fine because I knew the importance of it so I was fully

aware when I went into it" (Parent 8, Mother).

Some parents found it difficult to support their child due to hectic schedules, which was captured by the theme of *busy lives*:

"It was a challenge as I said because I work 4 days a week um ideally it would have been better to do it after school when we had plenty of time. It was a bit sort of frantic at times um you know trying to fit cooking tea in and um try and fit it in before bedtime so from that point of view…as I said I knew that would be our biggest challenge was the time aspect…Um so yeah it was a challenge" (Parent 28, Mother).

Although under the theme *high motivation levels*, this found highly engaged CYP without their parents' persuasion:

"[Child's name] was fully engaged and I think the whole thing made him feel quite special. I think the fact it was targeted. The fact it was all about tics and it was educational and he was seeing other kids with it. It was all positive" (Parent 6, Mother).

Discussion

This process evaluation used a mixed-methods approach to investigate the extent to which the ORBIT intervention was implemented as planned within the randomized controlled trial and to explore participants' experiences with the intervention and the contextual factors influencing children's engagement. In doing so, this made it possible to identify reasons for variation in uptake, usage, and engagement, to reflect on how implementation may ultimately give us greater confidence in the outcomes, and to outline lessons for potential future implementation within routine care. Uptake of the intervention was high with nearly 90% of participants receiving the pre-defined minimum effective dose of four chapters completed. The median uptake was eight chapters and only one child failed to access any chapters. Fidelity of delivery was also excellent with participants reporting high levels of satisfaction and acceptability.

The intended sample of CYP with a diagnosed tic disorder was reached, with 7.1% of families

residing in the most deprived areas (IMD quintile 1) and over a quarter (25.6%) of the families residing in the least deprived areas (IMD quintile 5). As over half (53.5%) of the CYP's mothers had completed graduate-level education, against a UK average of 42% [31] it seems that more advantaged families may have been over-represented. This is a concern, as one of the aims of ORBIT was to increase access to evidence-based therapeutic interventions for CYP with tic disorders. Particularly as access to services is generally limited for those from lower economic backgrounds [32]. However, the initial baseline visit with associated travel may have been a disincentive to more disadvantaged families: a limitation which would not be relevant if ORBIT was delivered entirely remotely in routine care rather than as part of an RCT. Moreover, there was no evidence that socioeconomic factors influenced CYP's engagement with ORBIT. Furthermore, child's age, severity of tics, well-being and comorbidities did not appear to influence the child's level of engagement with the intervention providing further evidence that the intervention would have a wide reach within routine clinical care.

London study site, self-referral, and higher parental engagement were all associated with higher levels of engagement. The London site is a world-renowned centre of excellence for paediatric care which may have increased parents' motivation for treatment. However, the only independent predictors of child engagement in the multivariate analysis was level of parental engagement with intervention as measured by their chapter completion and by parent time with therapist. This is consistent with previous literature [33–36] which found that parental involvement was particularly key for younger CYP to assist with their engagement with therapeutic interventions, which in turn leads to better outcomes [37–39]. It has been shown in the literature that parental engagement may impact a provider's ability to implement parent- and family-focused evidence-based treatment with fidelity [37]. Therefore, it is crucial to understand the role of parental support for the implementation of DHIs for children, as without attention to the key processes of child and family engagement, efforts to improve the effectiveness and efficiency of the treatment are less likely to succeed.

Furthermore, it will be crucial to assess whether parental support also predicts intervention efficacy and the mechanisms through which its impact is achieved.

An interesting finding is the usage and interactions with the therapist within this study. Therapists interacted online with their assigned child participants an average of about six minutes per child per week, which is lower than the 24 minutes average time per week participants interacted with their therapist in the Swedish pilot trial of BIP TIC [13]. However, in the UK study therapists were encouraged to use pre-prepared scripts to respond to participants. Their responsibilities involved reinforcing the ORBIT treatment material with the aim of spending around six minutes a week responding to each child. Detailed analysis of the content of therapists' interactions is outside the remit of this study, but it is apparent from qualitative interviews that many participants felt that the term 'therapist' was somewhat misleading. Some participants felt that 'therapist' had connotations of a clinically-trained individual delivering an intervention. This may have limited their reliance on the therapist. Therefore in any implementation of this intervention within routine healthcare, it would be sensible to alter the title to 'coach', 'guide', or 'mentor' as this better reflects the role of the therapist.

Strengths and limitations

To the best of our knowledge, this study is one of the first studies to have conducted an in-depth mixed-methods process evaluation of a complex intervention aimed at CYP with TS and chronic tic disorders. A number of important findings emerged from the process data which help us to characterise the implementation of the intervention within an RCT and provide lessons for potential future implementation within routine care. However, these lessons can only be fully realized once the main RCT outcome data has been analyzed. Furthermore, a principal components analysis of participants' usage data provided an objective, reliable, and comprehensive measure of engagement with which to explore the role of contextual factors.

However, this study has some limitations. Firstly, there was the issue of potential recruitment bias. It may have been that the more motivated families self-referred to the trial and that recruitment from

clinics was skewed towards punctual, frequent attenders, in contrast to patients with multiple missed appointments. This may have limited the power of this process evaluation to detect socioeconomic biases in engagement. Secondly, the information on uptake, although comprehensive, cannot fully capture the quality and quantity of adherence to ORBIT. For example, indices such as chapter completion, number of pages visited and number of logins may not fully capture factors such as level of attention or adherence to practice exercises. Finally, and perhaps most crucially of all, a major limitation was that it was not possible to interview those who had not taken part in the RCT or to reach those who had withdrawn early from the study. Their perspective is obviously vital to fully understanding factors influencing engagement with DHIs.

Conclusions

We conclude that the intervention had high fidelity of delivery and was evaluated positively by CYP, although reach may have been constrained by the nature of the RCT. Parental engagement was a strong, independent predictor of intervention implementation, which has important implications for the design and implementation of digital therapeutic interventions into Child and Adolescent Mental Health Services.

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List of abbreviations

ADHD: attention-deficit/hyperactivity disorder
CYP: Children and young people
DHI: Digital health interventions
IMD: Index of Multiple Deprivation
MFQ: Mood and Feelings Questionnaire
MRC: Medical Research Council
ORBIT: Online Remote Behavioural Intervention for Tics trial
RCT: Randomized controlled trial
TS: Tourette syndrome
TTSS: Total Tic Severity Score
YGTSS: Yale Global Tic Severity Scale

Declarations

Ethics approval and consent to participate

Ethical approval for the conduct of the study was gained from the North West - Greater Manchester Central Research Ethics Committee (REC: 18/NW/0079). We sought written parental consent, and written informed assent/consent to participate in the study from children and young people. This covered process evaluation measures. All participants provided verbal consent to be audio-recorded for all interviews.

Consent for publication

Not applicable.

Availability of data and materials

Part of the data generated or analysed during this study is included in this article and its supplementary information files or is available from the corresponding author on reasonable request. The full datasets generated or analysed during the current study are also available from the corresponding author on reasonable request.

Competing interests

The authors declare no competing interests.

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The views represented are the views of the authors alone and do not necessarily represent the views of the Department of Health in England, the NHS, or the National Institute for Health Research.

Authors' contributions

CG, KK, CLH, and EBD contributed to the conception of the mixed methods process evaluation and developed the interview guides. KK conducted the data collection and initial analysis of both the quantitative and qualitative data. KK wrote the initial draft and subsequent revisions. CG, CLH and EBD provided the critical review and editing of the initial and subsequent drafts of the manuscript. All authors critically revised the manuscript, read, and approved the final manuscript. CH is PI on the ORBIT trial, designed the trial and wrote the original grant application.

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Supplementary Files

Figures

Brief description of ORBIT.

Design: A 10-week, two-armed, parallel group, single blind, randomized controlled trial (RCT) with an embedded process evaluation.

Aim: To evaluate the effectiveness of an online, remote, therapist-supported and parent-guided behavioural intervention for tics, initially developed and piloted in Sweden called BIP TIC.

Intervention group: 112 children and young people received 10 modules (called 'chapters') of behavioural therapy following the principles of Exposure and Response Prevention (ERP) via a secure online platform, with access to a therapist, delivered over a period of 10-12 weeks.

Control group: 112 children and young people received 10 chapters of psychoeducation via a secure online platform, with access to a therapist, delivered over a period of 10-12 weeks.

Primary outcome: Total Tic Severity Score (TTSS) on the Yale Global Tic Severity Scale (YGTSS) at 3-months post randomization.

Therapist role: Both children and parents had regular contact with a therapist during the 10-12 weeks via messages that were sent within the treatment platform (resembling an email) or telephone if required. The therapist was also able to directly comment on exercises that the participant had been working on, and give specific feedback to motivate participants. All participant contact with the therapist was asynchronous.

Parent role: One or both of the child's parents received a separate login to the online treatment where they could access their own chapters. The parent chapters contained information regarding parent coping strategies, how to support their child in working with BIP TIC and functional analysis relating to tics. They also had access to the assigned therapist.

Multimedia Appendixes

Analytic Framework.
URL: https://asset.jmir.pub/assets/53e8f87b1414382e58698937966b830e.docx

TOC/Feature image for homepages

TOC+placeholder.