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Protocol for a UK-based feasibility randomised controlled trial evaluating the BabblePlay app intervention to encourage vocalising in infants with Down syndrome

Laura Boundy

laura.boundy@york.ac.uk

University of York https://orcid.org/0000-0001-8922-9626

Tamar Keren-Portnoy

University of York

Mona Kanaan

University of York

Kelly Burgoyne

The University of Manchester

Helena Daffern

University of York

Sue Buckley

Down Syndrome Education International

Sab Arshad

University of York

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Abstract

Background:

Down syndrome affects around 1 in 893 live births in the UK and is the leading cause of intellectual disabilities. Children with Down syndrome experience delays in speech, language and communication, which increase with age. This delay is associated with negative social, emotional and economic outcomes in adulthood. Previous research has found that early, frequently delivered language intervention in infants with Down syndrome may be effective at improving early language skills. However, there are limited high quality, evidence-based interventions for early speech development. This study is a feasibility randomised controlled trial (RCT) for evaluating the feasibility of an app, BabblePlay, as an intervention tool for increasing vocalisations in infants with Down syndrome.

Method:

The study is a UK-based, remotely-delivered feasibility RCT. Online recruitment will take place July 2024 to January 2025. Children will be randomly allocated to the BabblePlay intervention group or a control group. The intervention group will be asked to use BabblePlay with their child for 5 minutes twice a day for 6 weeks (split into two 3-week periods). The control group will be asked to use a mirror with their child for 5 minutes twice a day for 3 weeks, followed by 5 minutes twice a day with the app for 3 weeks. We aim to recruit 40 children with Down syndrome aged 7–15 months. Data will be collected by BabblePlay, questionnaires and 2-day-long audio recordings at baseline and after the first and second 3-week activity periods. We will run focus groups with caregivers to explore the acceptability of the app. Feasibility (including recruitment and retention) will be assessed through progression criteria using a traffic light system.

Discussion:

This feasibility RCT will allow us to evaluate progression to a definitive RCT to examine whether use of the app results in changes to infant babbling and word production. This study will highlight any necessary adjustments to the protocol. It will also add to the limited research on early speech and language intervention for children with Down syndrome.

Trial registration:

This trial is registered with the OSF registry (registration DOI: https://doi.org/10.17605/OSF.IO/V2DNH) registered on 10 August 2024.

Key messages regarding feasibility

What uncertainties existed regarding the feasibility?

Results of a one-week pilot trial showed that infants with Down syndrome were able to use the BabblePlay app. However, there were uncertainties regarding the feasibility of recruiting and retaining a sufficient number of families with young infants with Down syndrome for a larger trial, adherence to BabblePlay app use over a longer intervention period (3-6 weeks), and completion of intensive home audio recordings.

What are the key feasibility findings?

The feasibility trial will assess recruitment and retention rates, as well as adherence to the intervention and study procedures, including app use over a longer period (3 vs. 6 weeks) and completion of home audio recordings. The acceptability of the intervention will also be explored through parent feedback questionnaires and optional focus groups.

What are the implications of the feasibility findings for the design of the main study?

The findings from this feasibility study will inform the protocol of a future definitive randomised controlled trial to test the effectiveness of the intervention for increasing vocalisations and supporting later language development in infants with Down syndrome. This may include refinement of study procedures and outcome measures, to ensure they are acceptable and feasible for families.

Background

Down syndrome affects around 1 in 893 live births in the UK [1] and is the leading genetic cause of intellectual disabilities [2]. Individuals with Down syndrome often have difficulties with their language development and many experience delays in their early language skills, particularly their spoken language [3] and phonological awareness [4]. This delay in language abilities continues into adulthood [5]. In a study with people with Down syndrome aged 5 to 21 years, it was found that speech impairment predicts several different measures of adaptive behaviour, including conceptual skills (e.g. ability to give multi-step instructions and follow common signs) and general life skills (e.g. self-care, asking for assistance in a shop) [6]. Therefore, establishing early language intervention to target these delays may help to improve both language skills and these later outcomes.

For infants with Down syndrome, early targeted speech and language intervention has been found to be beneficial for language development [7, 8]. Furthermore, language interventions that provide higher frequency sessions (i.e. 5 days a week) significantly improve vocabulary outcomes compared to lower frequency [9]. However, Speech and Language Therapy (SLT) provision and frequency in the UK is varied, often with clear geographical inequalities [10]. A survey by the Royal College of Speech & Language Therapists [11] found that within the SLT profession, vacancy rates for children's SLT services were high: In 2023 there was an average vacancy rate of 25% in England (11% in Wales and 14% in Scotland), reaching up to 36% in some regions. This limited access to services is also apparent for SLT provision in

older children with Down syndrome. A report by Hargreaves et al. [12] found that only 37% of children attending a mainstream primary school and 45% of children attending a specialist school received SLT once a month or more. Other children either received SLT less than once a month, or not at all (10% of children in mainstream and 13% in specialist primary schools). This is despite parents ranking communication difficulties as the most challenging area developmentally for their children.

Therefore, there is a need for more evidence-based interventions which deliver frequent, targeted language support to these children but do not rely on practitioners and the already overstretched SLT services in the UK. An alternative approach is to develop caregiver-led interventions that can be easily accessed and completed at home. Home-based interventions provide several advantages for families with young children: they eliminate the need to travel which reduces costs for families, the interventions can be delivered daily by caregivers, which makes high dosage of treatment possible, and they are more accessible to families who may not have the opportunity to receive clinic-based services [13].

Previous studies have highlighted the efficacy of interventions, some of them caregiver-led, aimed at increasing communication skills in the pre-linguistic stage of development for infants with Down syndrome [9, 14–16]. At this stage babbling (i.e. production of language-like syllables in consonant-vowel combinations) begins to emerge, usually between 6 and 8 months in typically developing children [17]. Earlier mastery of consonant production through the vocal practice afforded by babble correlates with earlier word production in typically developing children [18, 19] and the onset of babble correlates with earlier social communication skills in children with Down syndrome [20]. A more recent study by Locatelli and colleagues [21] found that babbling in infants with Down syndrome is a significant predictor of their verbal cognitive development (measured using a Verbal index of the Wechsler Preschool and Primary Scale of Intelligence, which included subtests of information, receptive vocabulary and picture naming) [22].

However, infants with Down syndrome generally vocalise less than their typically developing peers [23] and the onset of babble is delayed, with the average age of onset around 9 months [20]. Their first recognisable words appear around 21 months, compared to 14 months for typically developing infants [24]. Yoder et al. [9, 25] demonstrated that increasing syllable (babble) production in infants with Down syndrome aged 18–27 months through almost-daily intervention led to larger spoken vocabularies at post-treatment, relative to the vocabularies of infants given the intervention just once a week. Recently, a small-scale feasibility study was carried out for a caregiver-led intervention (Babble Boot Camp: [15]), targeted at increasing vocalisations and communicative turns in young 4–16-month-old infants with Down syndrome and caregiver verbal responses to them, and implemented through frequent coaching from a speech and language professional. The results also suggest an increase in babble complexity and earlier word production. Both these interventions are implemented partly through guiding caregivers to provide verbal responses to their infants' initiation of communication or to their vocalisations. Such caregiver responsiveness is used to reinforce more vocalisations by the infants with Down syndrome. Reinforcing infant vocalisations through mothers imitating them was the focus of another small-scale study [26]. Fiani et al. found that such imitation led to an increase in speech-like sounds even with 4-

month-old infants with Down syndrome. Notably, caregiver responsiveness has been found to predict later lexicon size in typically developing infants [27] and in general, caregiver input speech quantity has been linked to typically developing children's lexical development rate (e.g. [29]). Interestingly, other research has also shown the reverse relationship, that for typically developing children, child vocalisations elicit verbal parental responses: Fagan and Doveikis [28] found that infant vocalisations were followed by a vocal response from the mother more frequently than other infant vocal behaviours (e.g. hiccups, sneezes or crying).

The Yoder et al. [9, 25], Fiani et al. [26] and Peter et al. [15] studies all suggest that speech-sound intervention (achieved partly through increased caregiver responsiveness, and perhaps input speech quantity), is effective at facilitating early language development in children with Down syndrome, but there is limited evidence-based support for this. A review [30] found that only 11 studies focused on language interventions for children with Down syndrome under 6 years of age, and 7 of these studies did not meet the minimum criteria for quality (assessed through the speechBITE Physiotherapy Evidence Database quality rating scale, PEDro-P) [31]. The authors therefore highlighted a need for higher quality studies examining early language intervention in children with Down syndrome.

The current study:

The current paper outlines the protocol for a feasibility randomised controlled trial (RCT) to examine the feasibility of an app, BabblePlay, as an intervention tool for increasing vocalisations in infants with Down syndrome. The BabblePlay app [32, 33] encourages vocalisations by responding to infant sounds with colourful moving shapes on an iPad screen. If the infant learns that their vocalisations make shapes appear, they may vocalise more to see the shapes. The intervention is caregiver-led and can be delivered daily at home. A pilot study [34, in prep] exploring the usability of the app with 26 infants with Down syndrome observed a higher mean rate of infants' vocalisations when playing with BabblePlay over a week (4.99 vocalisations per minute) compared with playing with a mirror in the preceding week (3.38 vocalisations per minute). Eighteen infants vocalised more during BabblePlay than during the mirror play, while eight showed the opposite pattern. Adherence was high over the one-week period, with 89% of families completing five mirror sessions and, of these, 92% completing the requested five app sessions. In addition, 15 of 24 families reported that, if made available they would use the app at least once per day. This preliminary work suggests that infants with Down syndrome and their families can engage with the BabblePlay app, and it may increase infants' vocalisations. Additional research is needed to assess whether the app can be used over longer periods of time and result in longer-term changes to the quantity of infant vocalisations and babble, and later words.

Aims and objectives:

The aim of the current feasibility study is to establish a protocol for future use in a definitive RCT which will explore whether BabblePlay, when used over longer periods of time, leads to longer-term, more generalised changes in (1) the quantity of infant babbling (2) the quantity of vocal input from their caregiver and (3) earlier word production. To meet these aims, this study has the following objectives:

1. To determine the recruitment, retention and adherence rates and acceptability of the intervention and trial for families.
2. To explore the suitability of assessment measures and establish the sample size needed for a definitive trial.

Methods/Design

Trial design:

This study is a two-arm feasibility RCT in which children and their caregivers will be randomly allocated to the BabblePlay intervention group or to a control group (who receive a delayed intervention for a shorter period). Recruitment will take place between July 2024 and January 2025. Caregivers in the intervention group will be asked to interact with the app with their child twice a day for 5 minutes at a time for 6 weeks (split into two 3-week activity periods). Caregivers in the control group will interact with a mirror with their child twice a day for 5 minutes at a time for 3 weeks and then with the app for 3 weeks (in two 3-week activity periods). We have chosen to use a mirror as the control condition, because like BabblePlay it is responsive to an infant's actions (i.e., the infant sees a change in their reflection when they move), but it does not respond specifically to sound. It therefore allows the control activity to be interactive and responsive, like the intervention, but not focused in any way on language sounds. The protocol has been developed in line with the Standard Protocol Items: Recommendations for Interventional Trials (SPIRIT) 2013 checklist (see additional file 1). See Fig. 1 below for a flow chart of the study.

Setting:

We will recruit families from across the UK. The intervention will be delivered by caregivers in their own homes, with all interactions with the research team taking place remotely. Intervention materials will be delivered to the homes of participating families by post. Assessments and questionnaires will be completed online by the child's caregivers, using Google forms and secure, assessment-specific links (e.g. individual Vineland Adaptive Behaviour Scales links will be sent to each caregiver). The study will take place from July 2024 to July 2025.

Participants:

Eligibility criteria:

Inclusion criteria are as follows:

1. Infant has a diagnosis of Down syndrome
2. Infant is aged between 7–15 months at start of project (when caregivers sign the consent form)
3. Infant is growing up in a monolingual English-speaking household (or exposed to less than 10% of another language[1] based on caregiver report)

4. Infant has eyesight good enough to be able to see shapes on an iPad screen (based on caregiver report)
5. Twins can also take part
6. Family lives in the UK

No additional exclusion criteria will be applied. Infants will only be excluded if they did not meet the inclusion criteria listed above.

[INSERT FIGURE 1 **HERE]**

Figure 1 Down Syndrome BabblePlay Feasibility Randomised Controlled Trial Design

Intervention design:

The BabblePlay intervention is described in accordance with the Template for Intervention Description and Replication (TIDieR) checklist [35].

BabblePlay is a preventative, caregiver-led, app-based intervention designed to encourage early vocalisations in infants with Down syndrome. Given that Down syndrome is diagnosed at birth (or prenatally), the intervention can start within the first year of life. Early interventions can target areas known to be developmental challenges (e.g. speech and language) before any delay is apparent, to help improve early outcomes and potentially minimise delays [36]. BabblePlay is intended to be implemented before any speech and language difficulties are evident and does not require screening or diagnosis by a clinician.

Materials and delivery

BabblePlay will be delivered by the child's caregivers in their home. To deliver the intervention, caregivers will be sent an iPad with the BabblePlay app pre-installed, along with a paper-based instruction booklet. All communication with the research team will be conducted remotely using email, video conferencing (Zoom, version 6.3.10 or later), text messages, and phone calls.

Intervention procedures

To start a BabblePlay session, caregivers will be asked to place the iPad in front of their child, open the app and start a session. The app responds to sounds by producing colourful, moving shapes on an iPad screen. Caregivers will be asked to remain quiet during the sessions so that infants learn that the app response is contingent on their vocalisations. Each session lasts 5 minutes, after which the app automatically turns off, although caregivers can end the session earlier if needed.

Intervention schedule and intensity

Caregivers will be asked to complete the 5-minute app sessions twice a day over a 6-week intervention period. Sessions can be completed at times that suit the child and caregiver. Full details of the intervention schedule are provided in the 'Intervention group procedure' section.

Tailoring and modification

Participating families will be asked to follow the intervention schedule to allow the research team to assess duration and intensity feasibility. Families will be told that this represents the optimal schedule, however it may not always be possible to adhere strictly to it. Flexibility will therefore be emphasised, and sessions can be completed at times that suit the family and infant.

Adherence and fidelity

At the end of a session the app automatically generates an email, which contains a sound recording of the session, the session date, time, and duration (in seconds), and the quantity and timestamps of vocalisations identified. Caregivers are asked to send these emails to the research team, to assess intervention adherence and fidelity. Alternatively, families can choose to delete these emails; data from deleted sessions will not be used in the study. If monitoring indicates that a family is struggling with adherence (e.g. they have missed three consecutive days of app use), a member of the research team will contact the family to discuss any barriers and offer advice where appropriate.

Comparator

The comparator is a waiting control group who will interact with a mirror during their first 3 weeks of participation and then receive the intervention for the second 3-week period.

Sample size

We aim to recruit 40 families over 12 months, with children with Down syndrome aged between 7 and 15 months. This sample size is realistic based on our pilot study, which ran over 8 months and had 38 eligible families consenting to take part. Of these, 30 engaged with the main training session and 26 were retained to the end of the study [34]. Therefore, we anticipate that 57 eligible families will consent to be part of the study over a 12-month period, of which 39 families will complete the study, assuming the same rate of attrition.

Recruitment

We will advertise the study through social media and local and national Down syndrome support groups. Interested families will complete an initial sign-up form and will be screened according to the eligibility

criteria. All eligible families will be sent an information sheet over email with a link to the online consent form attached. Families will also be invited to an online information event where they will have the opportunity to hear more about the project and what they would be asked to do if they are involved, including the process of randomisation and completing assessments and recordings. If eligible families wish to take part, they will fill out and submit the online consent form, at which point they will complete the baseline assessments (see Table 1 for the timings of the different activities to be undertaken in the study). We will use a waiting list system. Families who stop engaging with us (i.e., stop responding to any communications from the research team) or actively request to withdraw from the study will be replaced with families on the waiting list, but only prior to randomisation. Interested families with a child who is too young to participate when they first contact us will also be added to a waiting list system and will be contacted once the child is the right age and if we still require participants.

Once the questionnaires are complete, we will send the family an audio recorder to be worn by the child. A member of the research team will conduct a Zoom call with the families to explain how to use the recorder and how to conduct the recording. Families will be asked to record the sounds produced by and around the child over a minimum of 8 hours and up to 15 hours on two consecutive days and to fill in daily activity logs to accompany the recording, including the time that their child took a nap. This log will allow us to systematically sample an event (i.e. the hour after a nap) that will occur in each child's recording and manually transcribe this hour. Families who send the recording back to the research team will be randomised.

Randomisation:

Following completion of baseline assessments and recordings, families will be randomly allocated in a 1:1 ratio to either the intervention group or the control group. The randomisation will be stratified by chronological and developmental age of the infant, established using the Vineland Adaptive Behaviour Scales, Vineland-3 [37]. We will use block randomisation with randomly permuted block sizes. Randomisation code will be generated by the statistical software STATA (Version 18 or higher) [38]. Sealed, opaque envelopes, each marked by an ID number on the outside and containing the group allocation on a slip of paper, will be prepared by someone independent to the research team. A member of the research team, who has no access to the sealed envelopes, will run the randomisation code to generate an ID number that corresponds to one of the ID numbers on the sealed envelopes. A different team member, with no access to the randomisation code, will then open the corresponding envelope to reveal the group allocation for each participant.

Intervention group procedure:

Participants randomised to the intervention group will be sent an iPad with the BabblePlay app installed. They will also be sent two additional audio devices for the post-activity recordings and a paper-based instruction booklet. Over a Zoom call, intervention group families will be shown how to register their child on the app and how to use the app and iPad, and will be asked to interact with the app with their child for 5 minutes at a time twice a day, for 6 weeks, or as close to this as possible. Participation is split into two

3-week activity periods. At the end of the first 3-week activity period (activity period 1), families will complete a 2-day audio recording of their child's day at home (with a minimum duration of 8 hours and up to 15 hours). After completion of the recording, a research team member will conduct another Zoom call with the family, to change the ID code on the app and answer any questions they might have. Following that, families will start their next 3-week activity period (activity period 2) with the app. At the end of each session the app generates an email to be sent to the research team. This will allow us to monitor adherence (detailed below), and we will contact families who do not appear to be completing sessions to check reasons for non-adherence and offer support where necessary. Information and guidance will also be provided through the instruction booklet, and the research team will offer support through phone calls and emails. At the end of the 6 weeks, families will complete a final audio recording (8 to 15 hours) at home over 2 days. Once the recording is received by the research team, a feedback form will be filled by the caregivers. Participants will also be invited to take part in an optional focus group.

Control group procedure:

Participants randomised to the control group will be sent a mirror and a paper-based instruction booklet. The mirror the participants will be using has been specifically chosen based on the similarities of its dimensions to the iPad. The mirror face measures 7.5 inches x 9 inches compared to the iPad screen measuring 6 inches x 8.5 inches. The mirror also has handles on either side matching the shape and handles of the iPad case. The dimensions of the mirror including handles are 8.5 inches x 14 inches as compared to the iPad (inside the case) measuring 8.5 inches x 12.5 inches. A member of the research team will conduct a Zoom call with the family. The family will be asked to interact with the mirror with their child for 5 minutes at a time, twice a day, for 3 weeks (or as close to that as possible), during activity period 1. Adherence will be assessed through a short online form which will be accessible through a QR code taped to the back of the mirror (see details below). We will ask caregivers to fill in the form every time they complete a session, to confirm that they have done this. In the last week of this activity period, we will send control group families the iPad and two audio recording devices for the post-activity recordings and a hard-copy information booklet. After the first 3 weeks, families will complete a 2-day audio recording of their child's day at home (8 to 15 hours). Once the recording reaches the research team, the second 3-week activity period (activity period 2) will commence, this time with the BabblePlay app. During a Zoom call a research team member will assist the family with registering their infant on the app and explain how to use the app and iPad. The family will be asked to interact with the app with their child for 5 minutes at a time, twice a day for 3 weeks (or as close to this as possible). Adherence will be monitored through the emails sent to the research team after every app session, and we will contact families who do not appear to be completing sessions to find out why and offer support (see details in the next section). Information will also be provided through an instruction booklet, and the research team can offer support where needed through phone calls and emails. At the end of the 6 weeks, families will complete a final audio recording (8 to 15 hours) at home over 2 days. Once that is returned to the research team, a feedback form will be filled by the caregivers. Participants will be invited to take part in an optional focus group.

See Table 1 for the timeline of the study.

Adherence to the intervention and contamination:

Adherence to the intervention will be assessed through monitoring the frequency of use by each family for both the app sessions (measured through the app emails sent to the research team at the end of a session) and the mirror sessions (measured through the online form filled out by families during a session). If we do not receive an app email or mirror form response from families for more than 3 consecutive days during the activity period, a member of the research team will get in contact with the family to check reasons for non-adherence. If adherence is low due to hospitalisations, illnesses etc., we will extend the relevant activity period for that family.

To reduce the risk of contamination, when caregivers are contacted to inform them of their group allocation, they will be reminded that they should not share the app or details of their group allocation with any other families. Families will only have access to the app for the time allocated to them (3 weeks or 6 weeks depending on their group). The app can only be accessed through the iPad sent to participating families (which is returned to the research team after the end of the second activity period) and so risk of contamination is deemed low.

Table 1
Participant timeline of the BabblePlay feasibility randomised controlled trial

1	Timepoints	Group	Rcrtmt	BL	Post-BL	AP1	PAP1	AP2	PAP2
	Developmental level (using Vineland Adaptive Behaviour Scales (VABS)) [37]	**All eligible**	**x**						
2	Child health questionnaire	All eligible	x						
3	Demographic questionnaire	All eligible	x						
4	Audio recording	All eligible		x					
5	Randomisation	All eligible			x				
6a	Activity Period 1 BabblePlay	Int				x			
6b	Activity Period 1 Mirror	Ctrl				x			
7	Audio recording	Both					x		
8	Activity Period 2 BabblePlay	Both						x	
9	Audio recording	Both							x
10	Feedback questionnaire	Both							x
11	Focus group (optional)	Both							x

NOTE:GP = Group, Int = Intervention, Ctrl = Control, Both = both groups, Rcrtmt = Recruitment stage, BL = Baseline, AP1 = Activity Period 1, AP2 = Activity Period 2, PAP1 = Post-Activity Period 1, PAP2 = Post Activity Period 2

Change in allocated intervention:

We will analyse families on an intention-to-treat basis. This means that families will be analysed in the group that they were allocated to at randomisation irrespective of the intensity of using the BabblePlay app or the mirror or the frequency of the play sessions, in as far as they complete the two post-activity audio recordings and feedback questionnaire.

Data collection:

At baseline and prior to randomisation, caregivers who have signed a consent form will be asked to complete the following:

1. The Vineland Adaptive Behaviour Scale (VABS, Vineland-3) [37] to measure general cognitive and adaptive functioning. This will be completed online by caregivers and will help to establish the child's developmental age for randomisation stratification.
2. An online contact detail and child health and development questionnaire (e.g. vision/hearing issues, access to/provision of speech and language therapy), for descriptive background information.
3. An optional online anonymous questionnaire to measure demographic information and representativeness of the sample to the general population (e.g. caregiver ethnicity, education, employment, financial situation).
4. A 2-day audio recording will supply the team with information about the sounds infants make and hear in their daily lives.
5. A hard-copy hourly activity log will be filled in by caregivers to accompany the 2-day audio recording.

Following randomisation, once families are allocated to the intervention or control groups, data will be collected using the following methods and procedures (as described in Table 2):

6. BabblePlay app emails (from both the intervention and control groups) will supply the data regarding the number of sessions conducted and the number of vocalisations produced in each, as well as sound recordings from the session.
7. The electronic questionnaires filled in by caregivers in the control group will record the number of sessions conducted with the mirror.
8. Hard-copy activity logs will be filled in by caregivers whenever they conduct a 2-day audio recording.
9. The 2-day audio recordings will record speech sounds infants make and hear in their daily lives.

    a. The recordings will be processed through the software ALICE (Automatic Linguistic Units Counter) [39], which will provide automatic analysis of the estimated duration (in seconds) of child vocalisations and the duration (in seconds) and number of words of adult speech input to the child.
    b. We will manually transcribe an hour after a nap (identified using the activity log) from each of the children's 2-day recordings, to get an accurate measure of child vocalisations and consonant use across the three timepoints. This information is qualitatively different from that supplied by ALICE and is not intended to be used for reliability checks on ALICE's estimates. Manual transcription is needed to accurately count child vocalisations (rather than estimate their durations), to distinguish them from vegetative sounds (e.g., coughs and sneezes), and to identify consonants produced by the child.

10. Caregivers will fill in feedback forms about the study and the intervention.
11. Participants will be invited to give open feedback on the study through participation in an optional focus group.

Qualitative evaluation:

At the end of the second activity period, caregivers will be asked to fill out an online feedback questionnaire to report on their experience of using the app and completing the assessment measures (e.g. the 2-day audio recording). We will also invite caregivers to take part in an online focus group via video conferencing, to discuss their views on the app and their experience of taking part in the trial. This will allow for a more detailed exploration of the acceptability and usability of the app and trial procedure.

Outcomes:

The outcome measures will assess the success of the feasibility RCT and determine if it is viable to progress to a definitive RCT. The measures in this feasibility study will also highlight any adjustments to the intervention delivery, materials or data collection procedure that may be necessary.

Primary outcome measures:

The following are the primary outcome measures to be collected:

1. Feasible recruitment: recruitment of 40 families (who consent and are randomised) from the time that advertising begins.
2. Feasible retention: retention of a minimum of 30 families who will complete the study. A participating family will be regarded as having completed the study if, following randomisation, they send the team both post-activity audio recordings and fill in the final feedback form.

We will follow Avery et al.'s 'traffic light system' for criteria for progression to a definitive RCT [40]. The criteria have been developed in consultation with the study Steering Group, which is comprised of three independent academic experts with experience conducting randomised controlled trials and research with children with Down syndrome. The group advised on the feasibility targets and progression thresholds. Thresholds were informed by our pilot study and what was considered achievable for the current trial. Progression will be informed by two main criteria (recruitment and retention) with predefined thresholds set in consultation with the Steering Group (see Table 3)

Table 3
Progression criteria for primary outcomes (recruitment and retention) as agreed by the trial Steering Group.

1	Outcome	Green (go)	Amber (review)	Red (stop)
	Recruitment (target N = 40)	**≥ 36 recruited, consented and randomised (≥ 90%)**	**30–35 recruited (75–89%)**	**< 30 recruited (< 75%)**
2	Retention (target N = 30)	≥ 24 complete trial (≥ 80%)	18–23 complete trial (60–79%)	< 18 complete trial (< 60%)

If both progression criteria are in green, we will proceed to a definitive RCT as per the current protocol. If there is a mix of green and amber, then the team will propose amendments to the current protocol to address issues highlighted by amber items and discuss these with the Steering Group. Any criteria in the

red bracket will be discussed with the Steering Group to consider next steps, which might include not proceeding to the definitive trial.

Secondary outcome measures:

Adherence measures:

1. Intervention adherence: Families complete a minimum of one session, mirror or app, per day for 14 days out of the total 3 weeks. This amounts to 14 sessions per activity period.
2. Adherence to the study procedure: Conducting the 2 post-activity audio recordings, submitting audio recordings of a sufficient length (8 hours or more), and submitting the 2 post-activity hourly activity logs. For the control group only, submitting the electronic forms that record mirror sessions.

Speech outcome measures:

We will collect these measures through the 2-day-long audio recordings completed by all families at baseline, post-activity period 1 and post-activity period 2. These findings will help to identify suitable outcome measures for the definitive RCT.

1. Average rate of child vocalisations per minute (based on automatic analysis of the 2-day audio recordings by ALICE).
2. Average rate of child vocalisations per minute (based on manual phonetic transcription of one hour of the audio recordings following a nap).
3. Average rate of child consonant use per minute (based on manual phonetic transcription of one hour of the audio recordings following a nap).
4. Average rate of adult speech input to child per minute (based on automatic analysis of the 2-day audio recordings by ALICE).

Acceptability measures:

1. Responses from the feedback questionnaire. The feedback questionnaire will gather information on caregivers' experiences of study participation, including recruitment, data collection measures, and randomisation. It will also assess the acceptability of both the intervention and control activities (e.g. app usability, burden to families, barriers to participation, and perceived infant engagement).
2. The number of sessions and vocalisation with BabblePlay in activity period 1 relative to activity period 2 (within participant, in the intervention group only). This will allow us to see if fatigue sets in with a longer period of use (expressed by a decline in the numbers with time) or whether both caregivers and infants find the longer period acceptable (with numbers stable or rising from activity period 1 to activity period 2).

Data analysis and presentation:

The objectives of the current feasibility RCT will be assessed using descriptive statistics rather than formal hypothesis testing, to explore potential effects of the intervention and determine whether it would be viable to progress to a definitive RCT. Analysis is therefore as follows, we will:

1. Calculate recruitment and retention rates and compare these to progression criteria.
2. Assess adherence to the intervention and procedure using descriptive statistics to summarise the number and length of completed app sessions, and the number and length of required audio recordings and logs.
3. Calculate descriptive statistics to use for any future sample size calculations.
4. Assess the acceptability of the intervention by summarising caregiver responses on the feedback questionnaire using descriptive statistics (e.g. means, frequencies and percentages). Acceptability of the intervention duration will be additionally assessed within the intervention group by descriptively comparing 3 vs. 6 weeks of app use (e.g. number and duration of app sessions), to see whether engagement is sustained over a longer intervention period.
5. Descriptive statistics (means and standard deviations or frequencies and percentages where appropriate) and estimated effect sizes will be calculated for speech outcomes:

    a. Average rate of child vocalisations (based on ALICE counts) and average rate of consonants per minute (based on manual transcriptions) when using the app for 3 weeks vs. not using the app (comparing between subjects: intervention group vs. control group at post-activity period 1, and within subjects: control group at post-activity period 1 vs. control group at post-activity period 2).
    b. Average rate of child vocalisations and average rate of consonants per minute when using the app for 3 weeks vs. 6 weeks (comparing between subjects: intervention group vs. control group at post-activity period 2, and within subjects: intervention group at post-activity period 1 vs. intervention group at post-activity period 2).
    c. Average rate of adult input to child per minute when using the app for 3 weeks vs. not using the app (comparing between subjects: intervention group vs. control group at post-activity period 1).

Data management and security:

Data transfer, storage and archiving:

Prior to running the study, we will complete a Data Protection Impact Assessment (DPIA) as per requirements of the Data Protection Officer at the University of York. All study data, including the personal information stored in the app on the iPads and the audio device recordings which contain hours of everyday interactions for each family, will be transferred and deleted off the devices, pseudo-anonymised (by use of a unique participant ID) and stored on a secure University of York Google Drive and Storage filestore as soon as possible. The iPads are password protected and can be wiped remotely if an iPad is lost or stolen. The audio recording devices contain no identifying information on them. iPads and recording devices will be delivered to and collected from families using special delivery pre-paid

envelopes. All laptops and PCs used to transfer and upload data will be managed by the University of York and are automatically encrypted. Files containing personal information will be kept in a separate folder from the study data. External collaborators on the research team will be given University of York associate status and so will have access to the Google Drive and Storage. The University of York, University of Manchester and Down Syndrome Education International (DSEI) have a data sharing agreement.

Participating families will be fully informed of the storage and use of data and recordings before providing consent and will have the option to ask the research team to delete sections from the 2-day recordings that they do not wish to be included in the transcriptions or analysis by giving us the time interval they want deleted from the recording. The emails generated by the app contain a sound file. Participating families will have control over whether to send these emails to the research team or delete them. Data will be stored and retained in line with the University of York's Data Management and Retention Policy.

The PL (Professor Keren-Portnoy) will have overall responsibility for data management and archiving at the University of York.

Table 2
Outcome measures used for BabblePlay feasibility randomised controlled trial

	Measurement	Group	BL	AP1	PAP1	AP2	PAP2
	Adherence measures						
1	Number of play sessions emailed by BabblePlay (minimum of 14 sessions per activity period)	Int		x		x	
		Ctrl				x	
2	Number of play sessions conducted with mirror (measured by online form filled in each time, minimum of 14 sessions)	Ctrl		x			
3	Number of 2-day audio recordings sent by families for both post-activity periods	Both			x		x
4	Number of 2-day audio recordings of sufficient duration (8 hours or more) for both post-activity periods	Both			x		x
5	Number of completed activity logs to accompany audio recordings	Both			x		x
	Speech measures						
6	Rate of child vocalisations per minute (based on ALICE automated analysis)	Both	x		x		x
7	Rate of child vocalisations per minute in a single hour of audio recording following a nap (based on manual phonetic transcription of 1 hour of the 2-day recording)	Both	x		x		x
8	Rate of consonants produced by the child per minute in a single hour of audio recording following a nap (based on manual phonetic transcription of 1 hour of the 2-day recording)	Both	x		x		x
9	Rate of adult speech input to child per minute (based on ALICE automated analysis)	Both	x		x		x
	Acceptability measures						
10	Responses from feedback questionnaire	Both					x
11	Number of play sessions conducted, and vocalisations produced with BabblePlay in activity period 1 vs. 2 (intervention group only)	Int		x		x	

NOTE:GP = Group, Int = Intervention, Ctrl = Control, Both = both groups, BL = Baseline, AP1 = Activity Period 1, AP2 = Activity Period 2, PAP1 = Post-Activity Period 1, PAP2 = Post Activity Period 2

Study governance:

The project management team consists of the Project Lead (PL), Project Co-Leads (PcL), Research and Innovation Associate (RaIA), Grant Manager, and Project Partner. The PL will meet with the RaIA and Grant Manager weekly, and the whole team will meet once a month (or as needed) to discuss the progress of the project.

A Trial Management Group, which will consist of clinicians, caregivers of children with Down syndrome, and adults with Down syndrome, will be involved in the shaping of the trial procedures and public facing materials, evaluating the running of the study and disseminating findings to relevant users and policy makers.

In addition, a Steering Group, comprised of independent academic experts who developed the progression criteria for the project, will be consulted in evaluating whether the feasibility RCT outcomes merit progression to a definitive trial.

We will meet with these groups 3 times over the course of the project, to seek advice and guidance.

Dissemination:

The findings of the study will be published in peer reviewed journals and publications aimed at practitioners. We will also present results at academic and clinical conferences concerned with atypical language development, including the Down Syndrome Clinical Excellence Network and the annual Down Syndrome Research Forum. All participating families will receive updates on the project through an online newsletter, which will be emailed to them periodically and will include a lay summary of the research findings. These newsletters will also be shared with relevant support groups for dissemination to the Down syndrome community.

Discussion

There is currently a limited evidence-base for early language intervention in children with Down syndrome and geographical disparities in the provision of SLT for these children within the UK. This study will help to address the need for these interventions by evaluating the feasibility of an app, BabblePlay, as a tool for increasing vocalisations in children with Down syndrome. This will form the foundation for a definitive RCT. By providing a tool that caregivers can use at home and is easily accessible regardless of geographical location within the UK, the intervention can help to mitigate place and wealth- related inequalities regarding access to speech and language intervention and therapy. This feasibility trial will allow us to assess the current protocol, including the outcome measures and overall feasibility of running a larger scale trial, in addition to exploring the acceptability of the trial for the families involved. Allocation concealment will be achieved using sealed envelopes, a method that has potential risks of allocation bias if treatment assignment could be foreseen. To mitigate this risk, clear separate roles will be implemented: envelopes are prepared independently, the randomisation sequence is generated by a separate team member and envelopes are opened by an individual with no access to the randomisation

code. If rates of recruitment and retention are of an acceptable level, we will seek funding to conduct a definitive RCT.

Trial status:

This paper refers to protocol version 1 dated 10th August 2024. Recruitment began in July 2024 until January 2025. All participants are expected to have completed the intervention and required tasks in July 2025.

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Declarations

Ethical approval and consent to participate:

This study received ethical approval from the University of York Language and Linguistic Science Departmental Ethics Committee (reference: Keren-Portnoy_030724_Staff_2.)

Consent for publication:

Not applicable

Funding:

The study is funded by the UKRI ESRC, Grant Ref: ES/Z502571/1. The funder did not play a role in the design of the trial.

Authors' contributions:

TKP and HD led the development of the app that is used for the intervention. TKP, KB, MK and SB conceptualised the research. All co-authors are co-investigators on the ESRC grant and constituted the core team that developed the feasibility trial protocol, led by TKP, KB, and MK. LB coordinated the development of the trial protocol, with input from TKP and SA. MK led on the statistical plans for analysis. LB led the development of the protocol manuscript. All authors contributed to the final manuscript version and approved it for submission.

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Availability of data and materials:

The registered protocol is available on the OSF registry (registration DOI: https://doi.org/10.17605/OSF.IO/V2DNH)

Competing interests:

The authors declare that they have no competing interests

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Footnotes

1. The criterion was taken from the ManyBabies1 definition of who are bilingual or monolingual

Figures

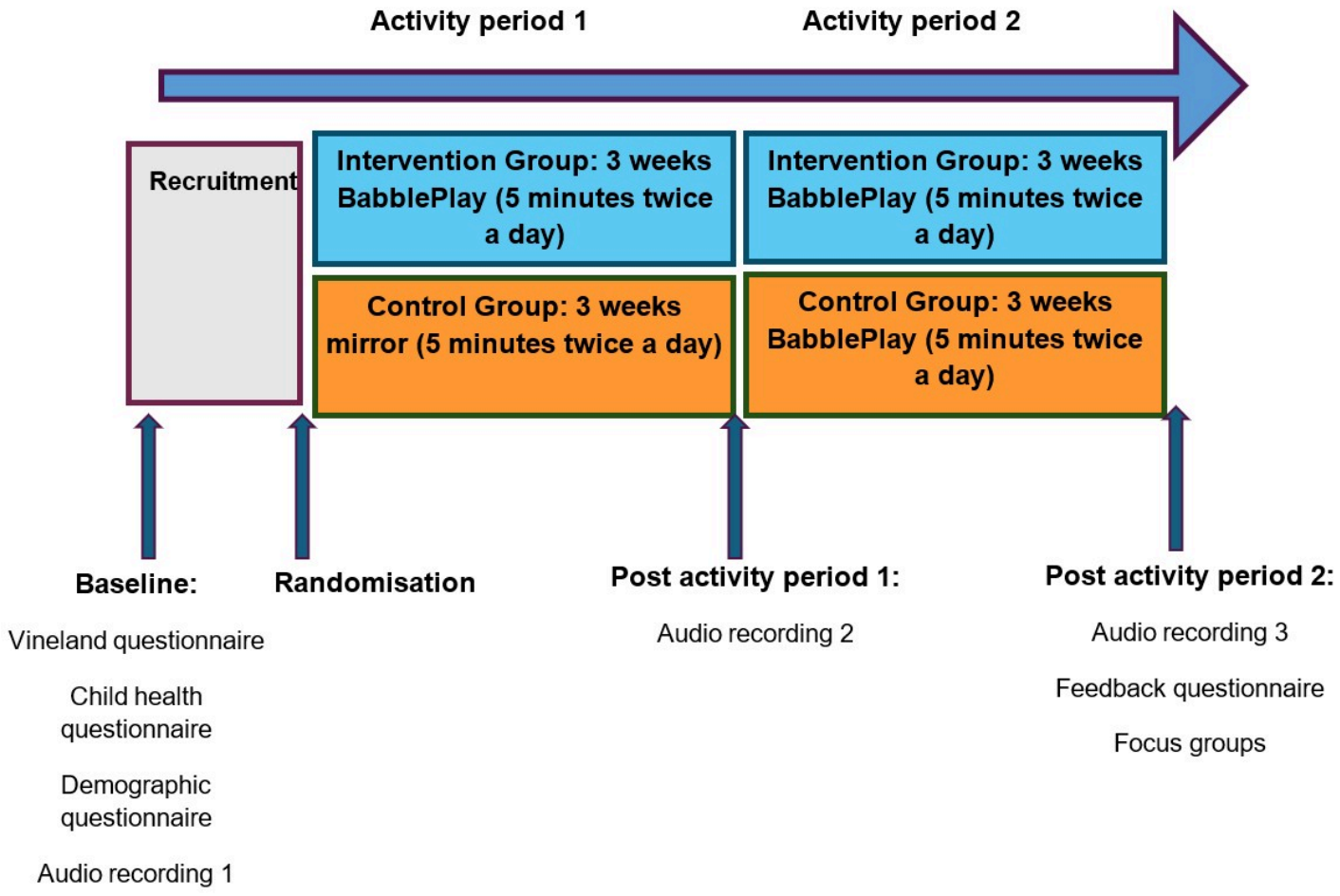


Figure 1

Down Syndrome BabblePlay Feasibility Randomised Controlled Trial Design

Supplementary Files

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- Additionalfile1SPIRITFillablechecklistBabblePlayDS.docx
- Appendix1OnlineParticipantConsentFormGoogleForms.pdf
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