



Title: Evaluating ‘An Early Social Communication Intervention for Young Children with Down Syndrome’ (ASCEND): Results from a Feasibility Randomised Control Trial

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Abstract

Background: This paper reports the results from a feasibility trial of an early parent-delivered social communication intervention for young children with Down syndrome ('ASCEND'). The intervention focuses on developing children's early social communication skills, in particular responding to shared attention. The aim was to inform the feasibility of running a full-scale trial through National Health Service (NHS) Speech and Language Therapy (SaLT) Services, to assess whether the intervention is effective in improving language skills before children with Down syndrome start school.

Methods: This was a two-arm feasibility randomised controlled trial (RCT), with 1:1 randomisation stratified by trial site, comparing the intervention plus standard NHS SaLT provision with standard NHS SaLT alone. We recruited 20 children with Down syndrome aged between 11 and 36 months through three NHS SaLT services, 19 of whom were randomised (10 - intervention group, 9 - control group). Pre and post intervention and 6-month follow-up assessments included language, social communication skills, adaptive behaviour, quality of life (parents and children), parental anxiety and depression. The intervention was parent-delivered with parents having access to SaLT services and the research team during the intervention. Data were collected on recruitment and retention, standard care, treatment fidelity, acceptability of the intervention by the parents and speech and language therapists, feasibility of collecting health economic measures and suitability of the primary outcome measure.

Results: The sample was sufficient for a feasibility study. The intervention (manual, support, materials) was positively received by the participating parents. Speech and language therapists also evaluated the acceptability of the intervention positively. Treatment fidelity and retention were acceptable. The preliminary health economic data suggest that this intervention will be low cost. The sample size calculation suggests that 290 participants would need to be recruited, with 228 having a complete data set, for a full RCT.

Conclusion: Based on recruitment, retention and treatment fidelity, as well as the acceptability of the intervention to parents and speech and language therapists, a full-scale trial would be feasible in order to assess the effectiveness of the intervention.

Keywords: Down syndrome, intervention, social communication, language, randomized control trial

Trial registration: ISRCTN13902755. Registered on 25th August 2020.

<http://www.isrctn.com/ISRCTN13902755>

Key messages

- What uncertainties existed regarding the feasibility?

We had uncertainties regarding study recruitment and retention of participants, and whether parents would be willing for their child to be randomised to a control group, how acceptable the intervention would be for parents and for speech and language therapists, whether the intervention would be delivered as described in the protocol and how many participants we would need for a fully powered randomized control trial.

- What are the key feasibility findings?

The recruitment and retention rates are acceptable, recruitment was undertaken mainly through NHS SaLT services and through local charities, over 90% of parents were willing for their child to be randomized to the control group, the intervention package was positively evaluated by parents and speech and language therapists and 290 participants with Down syndrome (with a view to having 228 complete the trial) are required, for a fully-powered RCT.

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

Based on our feasibility study outcomes, we conclude that a full-scale RCT is possible to assess the effectiveness of an early parent-delivered social communication intervention for young children with Down syndrome in optimizing language development. The intervention is well suited for

parents to deliver at home given the young age of the children and this mode of delivery is in line with paediatric SaLT services.

Evaluating ‘An Early Social Communication Intervention for Young Children with Down Syndrome’ (ASCEND): A Feasibility Randomised Control Trial

Background

Down syndrome is a genetic condition which results from an extra chromosome 21. Recent reports estimate a prevalence of 25.4 per 10,000 total births in England [1]. Down syndrome is the most common genetic cause of learning disability [2]. Most children with Down syndrome have difficulties acquiring speech and language, which often has adverse effects on communication skills. Evidence suggests that language ability at school entry can predict later psycho-social, educational and academic outcomes [3,4] and that early language skills are primary indicator of child well-being [5]. It is therefore crucial that children with Down syndrome are provided with opportunities to advance their language and communication skills as early as possible. There is evidence from other clinical populations that shows early interventions can optimize language and communication outcomes [6,7]. Given that Down syndrome can be diagnosed at birth, or even prenatally, interventions to optimize children’s language and communication outcomes can start very early. Children acquire language in the context of social interactions with others. Before children produce their first words, they acquire early social communication skills which are precursors for language [8,9,10]. Shared attention skills are fundamental early social communication skills acquired between 6 and 12 months of age, which allow the child and parent/caregiver to simultaneously focus on the same object or event and this in turn provides an opportunity for the parent/caregiver to label the object or event. The child may choose the object or topic/event upon which to focus, in which case the child has initiated shared attention. When the parent/caregiver chooses an object/topic to which the child’s attention is directed, the parent/caregiver provides an opportunity for the child to respond to shared attention. When the child responds to a bid for shared attention, the child receives language input from carers. The more a child responds to bids for shared attention, the more language input the child receives [11]

and successful language acquisition relies on good quality and quantity of input [12,13,14,15]. Evidence suggests that how well a child responds to the parent/caregiver's bids for shared attention may be an important predictor of later language outcomes for children with Down syndrome [9,16].

Reviews of interventions available which focus on the development of early social communication skills in young children with Down syndrome [17,18] conclude that the current evidence base is of low quality due to low number of studies, heterogenous data and outcome measures, and moderate to high risk of bias across studies. Our preliminary work [19,20] shows that an early intervention focusing on social communication skills, and particularly on responding to shared attention, can lead to better language outcomes in young children with Down syndrome. The children, aged between 17 and 23 months with Down syndrome in the intervention group (n=16), who had a 10 week intervention delivered jointly by a researcher and a parent, had significantly higher receptive vocabulary scores on the Reading Communicative Development Inventory (R-CDI) [21] 12 months after the intervention, compared to an age matched control group of children with Down syndrome who did not receive this type of intervention.

The aim of the current feasibility study was to estimate the parameters to inform a future randomised controlled trial that will evaluate whether the early social communication intervention plus standard care is more effective than standard care alone for enhancing the language and early communication skills and family health outcomes for young children with Down syndrome aged 11 to 36 months.

The feasibility study's **objectives** were to:

- Determine whether parents of children with Down syndrome are willing to be randomized
- Determine the acceptability of the intervention to speech and language therapists (SaLTs)
- Determine the effectiveness of recruitment of children with Down syndrome by SaLTs
- Identify different routes to identifying eligible children with Down syndrome (pediatricians, health visitors, SaLTs, charities)

- Estimate follow up rate and adherence to intervention.
- Inform the measurement of health economic outcomes and resource implications of a parent-delivered intervention
- Estimate the standard deviation of the primary outcome measure to inform a sample size calculation for a full trial

Method

Feasibility trial design

The current study was a two-arm randomised controlled trial (RCT) which investigated the feasibility of carrying out a definitive RCT to evaluate the effectiveness of an early social communication intervention in addition to standard NHS speech and language therapy (SaLT) (compared with standard NHS SaLT alone) for young children with Down Syndrome. The protocol was developed in line with the Standard Protocol Items: Recommendations for Interventional Trials [22] and published [23]. The results are reported in line with the CONSORT extension to pilot and feasibility trials [24].

Setting

The study was conducted in 3 NHS sites in England, providing SaLT services across three geographical regions: Berkshire Health NHS Foundation Trust (BHFT), Oxford Health NHS Foundation Trust (OHFT) and North-East London NHS Foundation Trust (NELFT) and each site had a Principal Investigator. All assessments were conducted remotely using either online or paper questionnaires, with support by telephone. This was in response to the COVID-19 pandemic when most face-to-face services stopped. The protocol was amended so that there was no face-to-face contact between the research team and the participants and their families.

Public and patient involvement (PPI)

The study protocol was developed with the help of our PPI group, who contributed to the finalising of the procedure, improving the readability of the parent manual and actively contributing to all decision making regarding the feasibility trial as members of the Trial Steering Group.

Participants

a) Children with Down syndrome and their parents/carers

Children with Down syndrome and their families were recruited through SaLT services in BHFT, OHFT and NELFT who distributed information sheets and consent forms about the study to the parents/carers of every child with Down syndrome between the ages of 11 and 36 months on their caseloads. It was up to the parents/carers to decide whether one parent or both parents/carers participated and delivered the intervention. Parents who either declined to participate or did not engage with the research project, were invited by their child's SaLTs to give their reasons for not participating.

Eligibility criteria

Inclusion Criteria

1. Parent or guardian willing and able to provide informed consent on behalf of participant.
2. Confirmed diagnosis of trisomy 21 (Down Syndrome).
3. Male or female child, 11 to 35 months old at study entry.
4. Parent/guardian has the literary and language skills needed to use the parent intervention manual.
5. The participant is not currently taking part or due to take part in a language-based intervention study.

Exclusion Criteria

1. Children with co-morbid conditions (for example Autism Spectrum Disorder) as determined by the Principal Investigator for each NHS site.

2. Any reason that may hinder participation, such as complex health issues requiring repeated hospital admissions.
3. Prior knowledge of the intervention as specified in the parent manual.

There was a change in the age range in the inclusion criteria after the feasibility trial had commenced. In the original protocol, children with Down syndrome aged 12 to 24 months were to be included. This age range was extended to be 11 to 35 months in order to improve recruitment rates as well as based on the recommendations of the SaLT services that children with DS who were slightly younger or old would still benefit from this type of intervention.

b) SaLTs

SaLTs were recruited through clinical and professional networks and current NHS sites to take part in an interview on parent-delivered interventions, the acceptability of the intervention from a SaLT service delivery point of view and their views on clinical trials.

Inclusion criteria

1. Currently practising SaLT in the UK with a paediatric caseload
2. Currently working within the NHS or having recently worked within the NHS (not more than 2 years have passed since last NHS post)

Procedure and Intervention

The SaLTs supporting the children who took part in the intervention attended a one-hour training session delivered by the research team on the main goals of the intervention and the different stages so that they could support the parents if needed. The SaLTs had the opportunity to ask any questions and to feed into the intervention materials prior to the participant recruitment. The SaLTs acted as a point of contact for the families, to support the parents/carers with delivering the intervention when needed.

The intervention is focused on promoting and supporting the development of early social communication skills, and in particular, the child's ability to respond to shared attention. During the sessions, the parent used the toys provided to encourage their child to engage in shared attention. A shared focus of attention can be achieved through 7 levels, depending on the child's developmental stage. During the sessions and over the course of the 10 weeks, parents/carers progressed through different levels of responding to joint attention from level 1 (the adult gently put the child's hand on object to signal that the specific object is the focus of attention) to level 7 (the adult placed a toy outside the child's visual field and the child then followed the adult's gaze to establish shared attention with the object) (see Appendix 6 for a full description of the different levels based on [25]). Once shared attention was established (through any of the levels), the adult used this as an opportunity to provide rich language input to the child by: a) labelling the object/labelling activities around the object (e.g., it's a bus, it drives around); b) describing the toys/objects in terms of colour, shape, size noise they make, how we can play with the toy; c) inviting the child to play/interact with the object. The approach is based on the social-pragmatic account of language acquisition, which assumes that shared attention and understanding the intentions of another person are the prerequisite skills for language development [26]. The parents recorded every session they had with their child on the diary form provided (see Appendix 1) and sent weekly to the trial manager.

Recruitment of children with Down syndrome and their families

SaLTs from participating NHS Trusts identified potential participants (children with Down syndrome) and their parents by reviewing their caseloads against the inclusion criteria. They then introduced the study to the parents of all potentially eligible children at a routine appointment, or via email/telephone call and provided the parent/caregiver with a participant information sheet giving details of what study participation would involve. They invited them to contact the research team if interested in participating, or if they had questions. Consenting was done by the research team. Parents/carers took the time they needed to consider participation and were able to delay

study entry by a few weeks to fit in with other family commitments. Where possible, reasons for non-participation were gathered by the child's SaLT.

Sample size

The target sample size was 25 children with Down syndrome based on literature recommendation of a minimum of 24 participants for feasibility studies [27, 28, 29] in order to estimate a standard deviation (SD) for the purposes of informing a subsequent sample size calculation.

Randomisation and allocation

The participants were enrolled to the study by the research assistant who took consent, administered the assessments and who was blind to participant allocation. After written consent was given by the children's parents/carers and baseline assessments completed, the children with Down syndrome were randomised by an external provider Sortition® (a secure web-based clinical trial randomisation software developed by the University of Oxford) using block randomisation to receive standard care (Control) or standard care plus the intervention (Intervention) in a 1:1 ratio, stratified by site, to account for regional differences in standard care. The external company communicated the allocation to a group for each participant to the clinical trial manager. Following randomisation, the parents/carers were contacted by the research team, who explained their child's study allocation, what that meant and what would happen next.

Blinding

Due to the nature of the intervention (parent-delivered), it was impossible to blind the parents and the children's SaLTs to group allocation. The research assistant, who administered the pre- and post- intervention assessments and who entered all the data was blind to group allocation and also did not have access to the intervention materials until the end of the project.

Intervention group

Once randomisation was complete, the intervention manual (paper based and printed using non-tear paper), blank diary forms (Appendix 1), a bag of age-appropriate toys and links to short video demonstrations of the intervention were posted to the parents/carers in the intervention group. The intervention was designed to be delivered by parents/carers over a period of 10 weeks. The intervention was delivered in the child's home by one or both of their parent(s)/carer(s) over 10 weeks. Parents/carers were advised to deliver it for one hour in total each week, over 3 to 6 individual sessions. The sessions could last between 10 and 20 minutes and parents/carers chose how to allocate the time.

Support to deliver the intervention from the child's SaLT was available at the request of parents by telephone/email. Parents also had access to the principal investigator of each site and to the Chief investigator. The SaLTs and investigators recorded all contacts from parents including duration and content of each contact.

Families continued to access standard NHS SaLT for the duration of the project. All contacts related to standard care were recorded, including the duration, number of contact points and activity type (assessment, advice, intervention/review).

Control (comparator) group: standard care

The control group received standard NHS SaLT care for this patient group. Standard care varied depending on each individual child's needs and on the pathway specific to each NHS site, ranging from two contacts per year, to monthly contacts. This typically included assessment, advice, intervention and review on feeding, use of baby sign Makaton, or communication. SaLTs recorded all contacts with the family for the duration of the study.

At the end of the 6-month follow up period, all families in the control group were provided with the intervention manual, accompanying materials and video links and had access to their child's SaLT and/or members of the research team for support with delivering the intervention.

Measures (assessments)

Assessments were administered at baseline, immediately post intervention (10-14 weeks after baseline) and follow up (6 months later) and scored by a research assistant blind to group allocation.

a) Language and communication

Primary outcome measure

- i) Reading Communicative Development Inventory (R-CDI)** [21], is a widely used parental checklist which assesses receptive and expressive language. Parents are asked to tick the words their child understands, understands and says, or understands and signs. We computed children's understanding of words and expressive language. Expressive language was measured by adding together all spoken words and signs the child was reported to use. For the bilingual children, a total vocabulary was computed which included a sum of all the words (spoken, signed and understood) in both/all of their languages. The Communicative Development Inventory (CDI) was chosen as the primary outcome measure because the aim of the intervention is to increase children's language by increasing their vocabulary and this measure directly assesses expressive and receptive vocabulary. Parental reports of language (such as the CDI) are widely used because parents have extensive experience with their children in a variety of naturalistic settings [30]. The MacArthur-Bates Communicative Development Inventories have been widely used in theoretical studies and in studies of importance for public health [31, 32, 33]. This measure has a reasonable predictive and concurrent validity. For example, children's scores on the CDI at ages 2 and 3 correlate significantly positively with standardised receptive language measures [30]. Importantly, the concurrent validity of the CDI has been established for children with Down syndrome [34]. The measures of children's language obtained on the CDI and standardised measures of language correlated strongly (between 0.70 and 0.82).

Secondary outcome measure

- ii) **Communication and Symbolic Behaviour Scale (CSBS)** [35]. This is a norm-referenced instrument available as an online or paper questionnaire, completed by parents/carers. It assesses communicative functions, gestural communicative means, vocal communicative means, verbal communicative means, reciprocity, social-affective signalling, and symbolic behaviour. This measure was chosen as a secondary outcome measure because the intervention focuses on increasing children's early social communication behaviours and these can be assessed using this scale. The measure has reasonable reliability and validity [35].

b) Quality of life

- i) Infant Toddler Quality of Life (ITQOL-SF47) [36] is completed by parents/carers and is a measure of infant quality of life
- ii) Adult Quality of Life Questionnaire [37] is completed by parents/carers and is a measure of parent/carer quality of life.
- iii) Hospital Anxiety and Depression Scale [38] is completed by parents/carers and is a self-assessment measure of symptoms of anxiety and depression.

In addition, all parents/carers of participating children completed a demographic questionnaire devised by the research team at baseline which asked questions about parental age, employment status, education and also contained the **Vineland Adaptive Behaviour Scale** [39], which is a standardised measure and was used to assess the children's general cognitive and adaptive abilities.

All questionnaires were completed by parents/carers using online links, or using paper copies posted to the participants, with support from a member of the research team if required. The **Communication and Symbolic Behaviour Scale (CSBS)** was administered over the phone.

Adherence to the Intervention and Contamination

Adherence to the intervention was monitored by asking the parents/carers to complete a weekly diary (see Appendix 1). The diary provided information on how many sessions the parent/carer carried out with their child, their duration, the number of different toys used at each session, the level at which they were working, and additional comments. The Principal Investigators contacted the parents/carers by telephone in weeks 4 and 8 to check adherence, with a window of +/- 7 days. They asked a standard set of questions specifically designed to obtain information on how closely the parents were following the manual, how often they carried out the intervention sessions and their duration, the range of toys they used, what they usually did in order to engage their child (see Appendix 2 for a standard set of questions asked).

Contamination was assessed at: 1) study entry for those who were randomised to the intervention group and 2) before the final follow up for those randomised to the control group. Parents/carers were sent a short questionnaire (Appendix 3) asking them whether they were familiar with the social communication intervention, whether they had seen the materials, or whether they had seen the intervention being carried out. There was no evidence of contamination during the trial based on the data provided by the questionnaires. Given that the trial was ongoing during the height of the COVID-19 pandemic, the opportunities for parents/carers who live in different regions, to share the hard copies of the manual and toys provided were minimal. Importantly, none of the participating families reported having seen the intervention manual and materials prior to being provided with their own set of the study materials.

Parent/carer satisfaction with the intervention

Parent/carer satisfaction with the intervention was evaluated via a brief questionnaire comprising 6 questions (see Appendix 4), the first 5 of which were closed questions and the 6th, asking for comments on the intervention, was open-ended. Only parents/carers who completed the intervention were sent the questionnaire.

Acceptability of the intervention to SaLTs

We invited all the SaLTs who had been involved with the current study to take part in an interview with a member of the research team. We also opened the call to a wider group of SaLTs who had not been involved in the current study across the three NHS sites, and SaLTs in other areas of the UK working in paediatric services for children with complex needs to take part in an interview as we wanted to have a broad range of views. We distributed the information sheet through the Down syndrome Research Forum, professional networks, through social media and SaLT managers and personal contacts. We aimed to recruit a diversity of SaLTs specifically in relation to gender, ethnicity and geographical area. In the last 3 months of the study, SaLTs were interviewed to explore their views on a parent-delivered intervention for young children with Down syndrome and their views and willingness to participate in future RCTs.

A topic guide was developed by the researchers addressing the study aims and used flexibly following the lead of the interviewees (Appendix 5). Data were collected via one-to-one interviews conducted via MS Teams. Researchers reviewed and edited the interview transcripts auto produced by MS Teams.

Analysis

Descriptive statistics (socio-demographic, language and cognitive abilities and health status) were collated and summarised. Parent/carers' satisfaction with the intervention was also summarised.

Quantitative analyses

All statistical analyses were pre-specified in a statistical analysis plan (SAP) which was agreed and signed off by the trial statistician and chief investigator prior to commencement of any analyses. As this study only aimed to address feasibility objectives, no formal hypothesis testing was undertaken to make between-group comparisons, but rather summary statistics were calculated by allocated group and overall at each time point. For each outcome, point estimates of SDs as well as associated 60%, 80% and 95% Confidence Intervals (CIs) are presented in line with Browne's recommendation to use the limit of the one-sided 80% CI for an SD obtained from a pilot study to inform subsequent sample size calculation [47].

Qualitative analyses

The data from the parental responses (n=11) regarding their reasons for not taking part in the study were summarised.

The data from the interviews with the SaLTs on their views of a parent-delivered social communication intervention were analysed using Reflexive Thematic Analysis [40], an approach to data analysis that enables patterns of meanings across a dataset to be developed. Data were coded inductively using an essentialist approach to report the experiences, meanings and reality of participants [41]. Author (X) familiarised herself with the data and generated codes using NVivo (Version 20.6.1.1137). Codes were then discussed with the first author and refined. Where disagreements arose, agreement was reached by consensus. Author (X) then grouped the codes into themes. These themes were discussed and defined by the first author.

Results

Participant recruitment and flow

The SaLTs from the three participating sites approached 38 eligible participants. Of these, 18 declined to participate. Of those who declined, 11 parents provided reasons for declining which are the following: illness in family and not the right time, child was too ill to participate, family expecting a new baby so no time for intervention, other children in the family were home schooled (due to lockdown) so parents felt unable to spend extra time with their child with Down syndrome, preference for an SaLT to work with their child directly and one of the parents/carers was not keen on taking part.

Twenty parents completed the consent forms, and 19 families were randomised (one family did not complete the baseline assessments and hence was not randomised). We did not recruit our target of 25 participants due to the restrictions arising from the COVID-19 pandemic which delayed recruitment and reduced the recruitment window by 6 months. The recruitment period varied between sites. Recruitment started between 9th September and 5th December 2020 and

closed on 30th June 2021 with duration lasting between 7 and 10 months. Of the 19 participants, 9 were randomised to the intervention group and 10 to the control group. There were no exclusions after randomisation. There was a loss of 4 participants at the post-intervention follow-up (2 participants from the intervention group and 2 from the control group) and we only have a reason from one participants from the control group. There was a loss of 3 participants at the 6-month follow-up and no reasons were provided by the participants. See Figure 1 below for Participant flow.

16 SaLTs approached from 3 NHS sites to
invite eligible families

Families invited to participate (n=38)

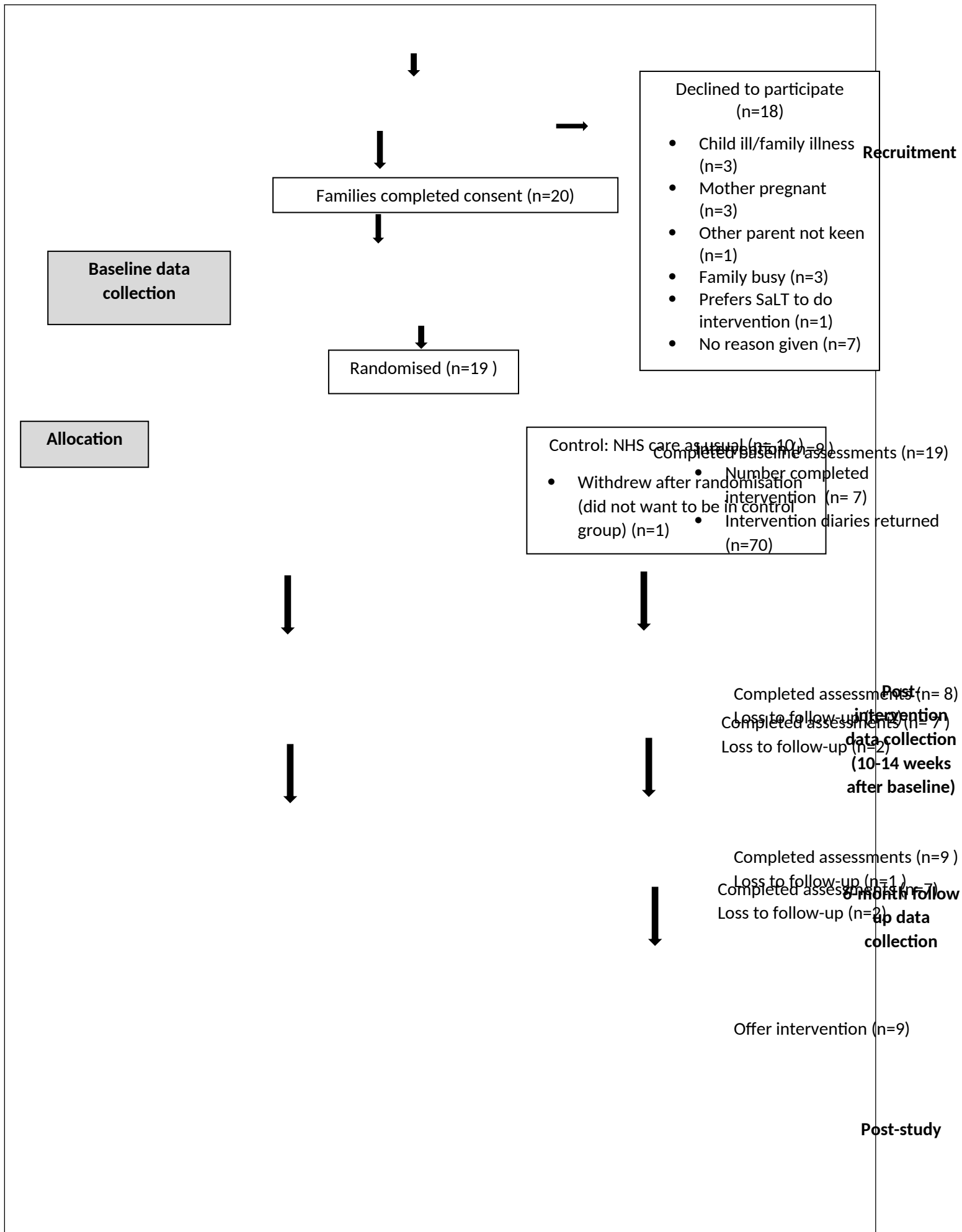


Figure 1: Study flow diagram

Note: NHS – National Health Service; SaLTs – Speech and Language Therapists

Although SaLTs were the obvious professionals to help recruit children with Down syndrome, not all young children with Down syndrome receive support from NHS SaLT services. We therefore explored other recruitment routes, including paediatricians, General Practitioners, NHS networks of health professionals and charities which focus on supporting children with Down syndrome using clinical NHS networks and regional charities. Seventeen out of the 20 recruited families came through NHS SaLT services. Three families were recruited through charities and were advised to register with their local NHS SaLT service so that they could get support, or alternatively would be supported by the principal investigator of the main NHS site (BHFT).

Participants

The children had a mean age of 20.3 months and 6 (32%) were female. Having completed the consent form and the baseline assessment, the parent of one child decided not to proceed with the study once they were informed that their child was randomised to the control group. However, given that baseline data were already collected and the participant randomised, their data are included. The sample's baseline characteristics including age, gender, adaptive functioning, and childcare are presented in Table 1. Characteristics of parents participating are presented in Table 2.

Table 1: Baseline demographic and clinical characteristics of the child participants in the intervention and control groups

n (%) unless otherwise stated	Intervention (N = 9)	Control (N = 10)	Overall (N = 19)
Mean (SD) [Range] Child's age (months)	n = 9 20.6 (9.2) [8, 32]	n = 10 20.1 (9.2) [11, 36]	n = 19 20.3 (9.0) [8, 36]
n (%) Child's Gender (Female)	5 (56%)	1 (10%)	6 (32%)
n (%) >3 weeks premature	3 (33%)	3 (30%)	6 (32%)
n (%) Concerns about vision	1 (11%)	4 (40%)	5 (26%)
n (%) Diagnosed mental, physical or emotional disability	8 (89%)	8 (80%)	16 (84%)
n (%) Concerns about hearing	4 (44%)	8 (80%)	12 (63%)
n (%) history of ear infections	0 (0%)	0 (0%)	0 (0%)
n (%) hearing test	9 (100%)	10 (100%)	19 (100%)
Mean (SD) [Range] hours in childcare per week	n = 9 12 (10.8) [0, 27]	n = 10 14.1 (16.8) [0, 50]	n = 19 13.1 (13.9) [0, 50]
n (%) Type of Childcare			
• Family member	2 (22%)	3 (30%)	5 (26%)
• Child minder	3 (33%)	0 (0%)	3 (16%)
• Nursery	2 (22%)	4 (40%)	6 (32%)
• Nanny/Au pair	0 (0%)	0 (0%)	0 (0%)
• Other	0 (0%)	0 (0%)	0 (0%)
• N/A	2 (22%)	3 (30%)	5 (26%)
n (%) receive Portage	3 (33%)	6 (60%)	9 (47%)
n (%) receiving support from speech and language services	7 (78%)	10 (100%)	17 (89%)
Mean (SD) [Range] Vineland Adaptive Behaviour Scale			
• Communication sub-domain	n = 9 39.8 (11.8) [24, 57]	n = 10 25.3 (11.3) [7, 39]	n = 19 32.2 (13.5) [7, 57]
• Daily living skills sub-domain	n = 9 17.2 (8.7) [12, 38]	n = 10 9.0 (2.8) [5, 14]	n = 19 12.9 (7.4) [5, 38]
• Socialisation sub-domain	n = 9 42.7 (6.8) [32, 55]	n = 10 33.1 (10.0) [13, 43]	n = 19 37.6 (9.7) [13, 55]
• Overall score	n = 9 99.7 (21.7) [68, 137]	n = 10 67.4 (21.4) [29, 92]	n = 19 82.7 (26.7) [29, 137]

Table 2: Baseline demographic characteristics of the parents

n (%) unless otherwise stated	Intervention (N = 9)	Control (N = 10)	Overall (N = 19)
Mean (SD) Parent age (years)	n = 9 38.4 (3.6) [33, 44]	n = 10 37.3 (4.8) [30, 45]	n = 19 37.8 (4.2) [30, 45]
n (%) Parent Gender (Female)	9 (100%)	10 (100%)	19 (100%)
n (%) parent occupation status			
• <i>Employed full time</i>	1 (11%)	4 (40%)	5 (26%)
• <i>Employed part time</i>	5 (56%)	2 (20%)	7 (37%)
• <i>Self-employed</i>	1 (11%)	0 (0%)	1 (5%)
• <i>Unemployed</i>	1 (11%)	3 (30%)	4 (21%)
• <i>Employed (on parental leave)</i>	1 (11%)	1 (10%)	2 (11%)
n (%) Highest level of parent education			
• <i>None</i>	0 (0%)	0 (0%)	0 (0%)
• <i>GCSEs/O-levels</i>	0 (0%)	0 (0%)	0 (0%)
• <i>A-levels</i>	2 (22%)	2 (20%)	4 (21%)
• <i>NVQ/HND</i>	0 (0%)	3 (30%)	3 (16%)
• <i>Degree</i>	5 (56%)	2 (20%)	7 (37%)
• <i>Postgraduate degree</i>	1 (11%)	2 (20%)	3 (16%)
• <i>Other</i>	1 (11%)	1 (10%)	2 (11%)
n (%) born in the UK	4 (44%)	7 (70%)	11 (58%)
Mean (SD) [Range] If not born in the UK, number of years in the UK	n = 5 16.3 (5.5) [8.5, 23]	n = 3 21.0 (7.9) [15, 30]	n = 8 18.1 (6.4) [8.5, 30]
Partner (other parent) characteristics			
Mean (SD) [Range] Partner age	n = 9 37.8 (3.8) [33, 43]	n = 10 37.5 (6.4) [29, 47]	n = 19 37.6 (5.2) [29, 47]
n (%) Partner Gender (Female)	0 (0%)	0 (0%)	0 (0%)
n (%) partner occupation status			
• <i>Employed full time</i>	6 (67%)	8 (80%)	14 (74%)
• <i>Employed part time</i>	1 (11%)	1 (10%)	2 (11%)
• <i>Self-employed</i>	0 (0%)	1 (10%)	1 (5%)
• <i>Unemployed</i>	1 (11%)	0 (0%)	1 (5%)
• <i>Employed (on parental leave)</i>	0 (0%)	0 (0%)	0 (0%)
n (%) Highest level of partner education			
• <i>None</i>	0 (0%)	1 (10%)	1 (5.3%)
• <i>GCSEs/O-levels</i>	0 (0%)	2 (20%)	2 (11%)
• <i>A-levels</i>	0 (0%)	0 (0%)	0 (0%)
• <i>NVQ/HND</i>	0 (0%)	2 (20%)	2 (11%)
• <i>Degree</i>	4 (44%)	3 (30%)	7 (37%)
• <i>Postgraduate degree</i>	4 (44%)	0 (0%)	4 (21%)
• <i>Other</i>	0 (0%)	0 (0%)	0 (0%)

Note: GCSE – General certificate of secondary education; NVQ – National vocational qualification;
A-levels – Advanced level qualifications

The results of the proposed outcome measures are presented in Table 3 below. Data are presented at baseline, immediately post-intervention (3 months after the baseline) and follow-up (6 months following the end of the intervention).

Outcomes

The aim of this feasibility trial was to obtain descriptive statistics that can be used to calculate the sample size needed for a subsequent definitive trial. A summary of the descriptive statistics for the primary and secondary outcome measures is presented in Table 3 below.

Table 3: Summary statistics for proposed outcome measures

Mean (SD) [Range] Outcome	Baseline			Post-Intervention			Follow-up		
	Intervention (N = 9)	Control (N = 10)	Overall (N = 19)	Intervention (N = 7)	Control (N = 8)	Overall (N = 15)	Intervention (N = 7)	Control (N = 9)	Overall (N = 16)
RCDI									
<i>Receptive Language (RCDI-U)</i>	n = 9 127.3 (178.0) [1, 567]	n = 10 69.5 (59.7) [0, 181]	n = 19 96.9 (129.4) [0, 567]	n = 7 217.6 (146.5) [52, 372]	n = 8 55.6 (38.4) [9, 128]	n = 15 131.2 (130.1) [9, 372]	n = 7 229.7 (151.1) [71, 468]	n = 9 91.0 (62.8) [0, 173]	n = 16 151.7 (127.6) [0, 468]
<i>Expressive Language (RCDI-E)</i>	n = 9 53.7 (84.0) [0, 268]	n = 10 11.1 (11.2) [0, 32]	n = 19 31.3 (60.7) [0, 268]	n = 7 51.4 (60.2) [0, 148]	n = 8 12.5 (14.2) [0, 34]	n = 15 30.7 (45.4) [0, 148]	n = 7 99.7 (75.1) [10, 217]	n = 9 26.4 (27.4) [0, 86]	n = 16 58.5 (63.8) [0, 217]
<i>RCDI-Total</i>	n = 9 131.2 (125.1) [4, 384]	n = 10 74.6 (57.1) [0, 156]	n = 19 101.4 (97.1) [0, 384]	n = 7 269.0 (167.1) [65, 474]	n = 8 68.1 (48.0) [9, 162]	n = 15 161.9 (154.5) [9, 474]	n = 7 323.3 (199.6) [65, 618]	n = 9 117.4 (78.9) [30, 259]	n = 16 207.5 (174.3) [30, 618]
CSBS									
<i>Social Composite</i>	n = 9 18.0 (3.4) [11, 22]	n = 10 11.9 (2.8) [8, 17]	n = 19 14.8 (4.4) [8, 22]	n = 7 21.4 (1.6) [19, 23]	n = 8 13.9 (3.4) [10, 18]	n = 15 17.4 (4.7) [10, 23]	n = 7 21.1 (1.6) [19, 24]	n = 9 14.3 (3.9) [9, 22]	n = 16 17.3 (4.6) [9, 24]
<i>Speech Composite</i>	n = 9 7.4 (2.1) [4, 10]	n = 10 4.7 (2.8) [0, 9]	n = 19 6.0 (2.8) [0, 10]	n = 7 8.6 (2.6) [5, 12]	n = 8 7.0 (1.8) [5, 9]	n = 15 7.7 (2.3) [5, 12]	n = 7 10.4 (2.2) [7, 13]	n = 9 6.7 (3.4) [1, 11]	n = 16 8.3 (3.4) [1, 13]
<i>Symbolic Composite</i>	n = 9 10.8 (4.0) [4, 16]	n = 10 7.7 (3.2) [2, 13]	n = 19 9.2 (3.8) [2, 16]	n = 7 13.6 (2.9) [10, 17]	n = 8 9.8 (3.0) [6, 14]	n = 15 11.5 (3.5) [6, 17]	n = 7 14.1 (2.8) [9, 17]	n = 9 11.4 (3.6) [5, 16]	n = 16 12.6 (3.5) [5, 17]
<i>Total</i>	n = 9 36.2 (8.0) [21, 48]	n = 10 24.3 (7.4) [12, 33]	n = 19 29.9 (9.7) [12, 48]	n = 7 43.6 (5.5) [36, 49]	n = 8 30.6 (7.0) [21, 38]	n = 15 36.7 (9.1) [21, 49]	n = 7 45.7 (5.1) [38, 53]	n = 9 32.4 (7.5) [19, 42]	n = 16 38.3 (9.3) [19, 53]

Note: RCDI-Reading Communicative Development Inventory; CSBS -Communication and Symbolic Behaviour Scale

Acceptability of the intervention to SaLTs

The final sample included 12 paediatric SaLTs (2 males, 10 females) with a range of experience and from a range of different NHS trusts in England. 6 SaLTs had been part of the intervention study and 6 had not. They all either currently worked for the NHS, or had done so in the past 2 years. The interview data indicate that 11 out of the 12 SaLTs were supportive of a parent-delivered intervention focusing on early social communication skills for young children with Down syndrome. All SaLTs taking part agreed that parent-delivered interventions should be offered by SaLT services and that such interventions were in alignment with existing provisions:

"so that's the way our service is now developing. It's more focused on. You need to do these things at home and then contact us if you've still got concerns" (SaLT X01)

"so I work in early years in the NHS so all our interventions are, primarily, are, parent led". (SaLT Y03)

Some SaLTs felt that alternative/additional interventions were necessary in addition to this intervention:

"I don't think that parent led intervention, working on play, etc., is enough for children with Down syndrome [.....] so I think that the evidence and my own experience shows that direct therapy, regular direct therapy for children with Down syndrome does work..." (SaLTZ05)

All SaLTs either have or would continue to recommend parent-delivered interventions to parents and were most likely to offer them to parents of children who were under five. When asked about how the interventions they offer may be similar or different from our intervention, some similarities were identified (for example: use of parent diaries, instructions given). The main difference identified was that our proposed intervention was much more structured.

"it's not something that we've used as in such a structured way" (SaLT Y03)

Acceptability of the intervention to parents

This was assessed using a questionnaire which was sent to the parents in the intervention group only, once they had completed the study. 90% parents reported that they were very satisfied with the intervention, one parent was fairly satisfied, and one parent was neutral. All parents reported

that they thought their child's responding to shared attention had improved following the intervention. Most parents reported that their child's language and communication had improved as a result of improvement in shared attention.

"We found that he is trying to communicate more and getting our attention until he was able to get what he was asking for or wanting to do" (Parent 7)

"Definitely. For example she can keep eye contact better and can express her needs on a more understandable way" (Parent 3)

Most parents reported seeing improvement in other areas of development in their child such as visual awareness, visual tracking, eye contact, copying of actions, gross motor skills etc.

"Gross motor skills improved a lot: nearly running and jumping" (Parent 1)

"...Visual tracking and eye contact" (Parent 5)

Most parents also reported that they had changed the way they communicate with their child after the intervention by using more descriptive language, providing more language input, encouraging their child to use different toys, using techniques learnt from the intervention in everyday situations.

"...Trying to input more verbal language than Makaton signs" (Parent 1)

"I am giving more description and talking to him more since we started the intervention" (Parent 4).

Parents were also asked for any other general comments and feedback. There was a mixture of comments with some helpful suggestions of what we may need to adjust in future, which included more instructions on what to do after the child had passed the final level and also the suitability of the intervention for children who were at the older end of our age range.

"It was very beneficial intervention for both of us, thank you for giving us this valuable opportunity.

We had a great support from the study team. They helped/guided us immediately when needed."

(Parent 4)

"The exercises were generally well explained in the manual, and the videos were helpful. Could potentially have more instructions about what to do if you complete the final level, unless it's ok to just practise whichever aspects of the previous levels you/your child wants to (which is what we did)." (Parent 2)

"It was a good and fun programme to see the way in which X understands and responds." (Parent 6)

"I think X was too young at the beginning and found the repeats boring but it was not a problem after a few weeks. The programme should have been longer a bit because we needed to repeat certain weeks and could not reach level 10. We really enjoyed the sessions and the quality time we spent with each other while doing it." (Parent 3)

Adherence to the intervention and contamination

Adherence phone-calls (week 4 and week 8) were completed for 7 (of the 9 families) in the intervention arm. These 7 families completed all follow up assessments as well. There was acceptable evidence from the data collected in the telephone contacts that the parents were following the instructions in the manual and completing the different stages of the intervention as suggested by the manual. 7 out of 9 families who were randomised to the intervention arm completed intervention diaries (78%), and all parents whose children completed all three assessments completed all diaries (100%) and all adherence calls. The mean length of session was 17 minutes (range 5 to 30 mins) and parents spent 60 mins per week on average with a range of 58min to 64 minutes. Parents carried out on average 4 sessions per week, with a range of 2 to 6 sessions per week. This is in line with the instructions in the manual. Inspection of the parent completed diaries showed that over 90% of the parents completed the diaries correctly. We did not collect diaries from the participants in the control arm.

Estimate of the sample size of a future trial to evaluate the effectiveness of the early social communication intervention

The proposed primary outcome in a subsequent definitive trial is the total number of words (expressive, receptive and signed, across multiple languages if the participant is multilingual) as measured using the R-CDI [21], assessed at the primary endpoint of 6 months following the end of the intervention. In order to achieve 90% power to detect a between-group difference of 100 words at the 5% two-sided significance level, a total of 228 participants (114 per arm) will need to be followed up and provide valid outcome data at the primary endpoint. This assumes a standard deviation of the outcome of 231 words, which conservatively represents the upper limit of the 80% confidence interval of the standard deviation estimated from the feasibility study. In consultation with our PPI group and the currently available literature on how children with Down syndrome acquire language [42,43] as well as the relatively simple nature of the intervention, it was agreed that 100 words would be a clinically important difference. An increase of 100 words in a child's repertoire will bring a child with Down syndrome closer to the point of starting to combine words into sentences (the average 24-month-old child has 297 words and is combining words into sentences [44]). A change of 100 words, on average, represents a minimum clinically important change in the primary outcome, and characterises a meaningful improvement in language development compared to typical development in this population. Assuming a loss to follow-up of 20% (similar to the feasibility study), the final indicative recruitment target in a definitive trial is therefore a total of 290 participants. Assuming an average of 20 children per site over a recruitment period of 24 months, we would need 15 sites.

Several scenarios were considered (see table 4 below).

Table 4: Sample Size Scenarios

Standard Deviation	Target Difference	Standardised Effect Size	Required Participants followed up	Target sample size (assuming 20% loss to follow-up)
Base Case				
231	100	0.43	228	286
Vary SD				
129	100	0.78	72	90

143		0.70	88	110
154		0.65	102	128
174		0.57	130	164
210		0.48	188	236
270		0.37	310	388
Vary Target Difference				
	80	0.35	354	444
	90	0.39	280	350
231	110	0.48	188	236
	120	0.52	158	198

Note: SD – standard deviation

Health economics/Health outcomes

Three measures were employed to look at parent/carer and infant quality of life. These measures were chosen and agreed jointly by parents of children with Down syndrome during a focus group interview and these parents did not take part in the intervention (as they had older children with Down syndrome). The following measures were used: Adult Carer Quality of Life Questionnaire (ACQOL) [37]; the Infant and Toddler Quality of Life Questionnaire (ITQOLSF47) [36] and the Hospital Anxiety and Depression Scale (HADS) [38]. Table 5 below shows the summary statistics of the health outcomes measures.

Table 5: Summary statistics of health outcome measures

Mean (SD) [Range] Outcome	Baseline			Post-Intervention			Follow-up		
	Intervention (N = 9)	Control (N = 10)	Overall (N = 19)	Intervention (N = 7)	Control (N = 8)	Overall (N = 15)	Intervention (N = 7)	Control (N = 9)	Overall (N = 16)
Adult Carer Quality of Life									
Total Quality Sum Score (raw score)	n=9 79.9 (18.9) [49,105]	n=10 79.3 (19) [43, 108]	n=19 79.6 (17.9) [43,108]	n=7 87.7 (16.1) [39,109]	n=8 74.3 (23.3) [40,112]	n =15 77.5 (20.7) [39,112]	n=7 79.3 (19.7) [51,108]	n = 9 73.4 (18) [42,96]	n = 16 76 (18.2) [42,108]
Infant-Toddler Quality of life (standard score)									
	n=9 63.3 (23.8) [30,100]	n=10 67 (18) [30,85]	n=19 65.3 (20.4) [30,100]	n=7 72.1 (23.6) [30,100]	n=8 62.2 (16.2) [30,85]	n=15 66.6 (19.7) [30,100]	n=6 67.5 (22.1) [30,85]	n=9 67.8 (18.9) [30,85]	n=15 67.7 (19.4) [30,85]
HADS									
Depression overall score	n=9 3.7 (3.2) [0,9]	n=10 6.8 (4.5) [1, 12]	n=19 5.3 (4.2) [0,12]	n=7 3.3 (2.4) [0,6]	n=9 7.4 (4.9) [2,18]	n=16 5.6 (4.4) [0,18]	n=7 5.8(5) [0,15]	n=9 7.2 (2) [5,11]	n=16 6.3 (4) [0,15]

HADS	n=9	n=10	n=19	n=7	n=9	n=16	n=7	n=9	n=16
Anxiety	6.3 (3.9)	6.8 (4.5)	8.5 (5.5)	4.3 (2)	9.8 (6.4)	7.4 (5.6)	7.7 (5.7)	12 (4)	9.3 (5.4)
overall score	[3,14]	[1, 12]	[1,14]	[1,7]	[4,20]	[1,20]	[2,18]	[7,17]	[2,18]

Note: HADS – Hospital Anxiety and Depression Scale

During the feasibility study, we estimated that the intervention cost is around £142-£174 per child. This includes 1 hour per child of SaLT time (the cost can vary between £82-£114 per hour, PSSRU 2021 [45] and the material used is estimated to cost approximately £60 per child (this includes a canvas bag, colour printed manual on special non-tear paper, 7 different toys and postage and packing). We estimate that to run a full trial with 228 children the cost of the intervention would be between £32,376 – £39,672. This does not include the cost for the families. The cost to families will be calculated in a full trial.

Adverse events and safety

No adverse events were reported.

Discussion

The aim of this study was to explore the feasibility of running a full-scale RCT to evaluate the effectiveness and cost-effectiveness of an early parent-delivered social communication intervention for young children with Down syndrome in addition to standard SaLT care compared to standard SaLT care only. To this end, in this feasibility trial we investigated if we could recruit enough children with Down syndrome and parents/carers, the most relevant recruitment pathways and the retention rate, how acceptable a parent-delivered intervention would be for parents/carers and also for SaLTs, adherence to protocol, ways to assess cost effectiveness and participant numbers for a full-scale RCT.

Recruitment and retention

We originally planned to recruit 25 children with Down syndrome over a period of 12 months. However, due to the COVID-19 pandemic and the interruption and delays this caused to the trial, our three sites were only open for recruitment for between 7 and 10 months (rather than the

planned 12 months) and we recruited 20 participants. We are reasonably confident that if we had had the three sites recruiting for the full 12 months, we would have reached our target of 25 participants. Our sample size calculation suggests that we would need to recruit 290 participants with a view to having 228 participants with a full data set for a full RCT. This means 15 sites are needed which will recruit on average 20 participants each over a period of 24 months, and this we believe is feasible. We also established during the feasibility study that the most effective recruitment route was via SaLT services and charities supporting families with children with Down syndrome.

Retention was monitored by the clinical trial manager who followed up families and who was responsible for letting the research assistant know when families were due to be sent assessments. Of the original 19 families who were randomised and who completed baseline assessments (Time point 0), 15 families completed the second assessment (Time Point 1), which is retention of 79% and 16 families completed the third assessment (Time point 2), which means retention was 84% overall. Retention of 70% is the minimum specified if a study is going to be included in a Cochrane Review [46].

Acceptability to parents and SaLTs

Both parents of children with Down syndrome and SaLTs were generally very positive about the acceptability of the intervention. For the SaLT services, barriers identified included mainly time and resource. The positive perspective of the intervention by parents is likely a reflection of the positive active engagement of our PPI group, which resulted in the information given to parents being clear, provided in 'parent-friendly' format, accessible and easy to follow. The high acceptability of the intervention for SaLTs was likely facilitated by the ongoing conversations and engagement with the NHS sites about the intervention delivery and how well it would fit within current models of delivery of early interventions prior to the study commencing. For the parents, barriers identified were mainly the child's age as some parents felt that their children were either too young or too old to fully benefit from the programme. Also, some parents felt that we could have suggested more

activities for children who managed to go through all levels of the intervention before the end of the 10 weeks.

Treatment fidelity and adherence

Based on the adherence phone calls in weeks 4 and 8, and submitted parent diaries, we are satisfied that the intervention was delivered by the parents with high fidelity and as described in the manual and videos. This is based on the fact that 100% of the parent diaries (for the parents who provided complete data sets for all time points) were returned, over 90% of the diaries were completed correctly and the adherence phone calls were all completed and did not identify any issues. It is very important that the materials are self-explanatory and that parents can use them autonomously, with minimal support from SaLT services. All parents who responded to the post-intervention questionnaire stated that the materials were clear and easy to follow.

The SaLTs who participated in the interviews also all commented that the materials provided were clear and straightforward. Although this did not come up during the study, we are aware that our current materials need to be updated so that they are inclusive and representative of the population as a whole in terms of diversity and we will be updating these before we run a full-scale trial.

Health economic measures and cost effectiveness

The economic analysis results show that the intervention can be delivered at a reasonable cost. We recommend that a detailed cost data collection is conducted in a full trial to make sure all NHS resources are included. We also recommend collecting data on the private costs for the families (e.g. private cost for other health services, adaptations, special toys, time off work, transport costs). In this study we have identified three health outcome measures of effectiveness and we recommend that these can be translated in utility values to be used in a future cost-utility analysis.

Conclusion

This feasibility study is the first-step in the development of an evidence-based theoretically-driven early social communication intervention for young children with Down syndrome, bridging a gap in the current evidence-base for intervention for very young children. Based on the outcomes, and specifically given the rates of recruitment, retention and data completeness and based on the finding that the study procedures were acceptable for parents and SaLTs a full scale trial appears feasible and warranted.

Abbreviations

ACQOL - Adult Carer Quality of Life

A-Levels – Advanced level qualifications

BHFT - Berkshire Health NHS Foundation Trust

CDI- Communicative Development Inventory

CIs -Confidence Intervals

CSBS - Communication and Symbolic Behaviour Scale

GCSE – General Certificate of Secondary Education

HADS – Hospital Anxiety and Depression Scale

ITQOL - Infant Toddler Quality of Life

NELFT - North-East London NHS Foundation Trust

NHS - National Health Service

NVQ – National Vocational Qualification

OHFT - Oxford Health NHS Foundation Trust

PPI - Public and Patient Involvement

R-CDI - Reading Communicative Development Inventory

RCT- randomised controlled trial

SaLTs – Speech and Language Therapists

SaLT - Speech and Language Therapy/Therapist

SAP - statistical analysis plan

SD - standard deviation

Trial Status

Ethical approval was granted on 4th August 2020 for the amended protocol: (South Central - Berkshire Research Ethics Committee ref: 19/SC/0572, IRAS Project ID: 252332). Recruitment opened on 9th September 2020 for BHFT, 10th November for NELFT and 9th December for OHFT. Recruitment closed on 30th June 2021 for all sites.

Declarations

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Ethical approval and consent to participate

The study has been given ethical approval by the Berkshire Research Ethics Committee (reference number IRAS 252332). All participants gave informed written consent (parents provided consent on behalf of the children). The trial was conducted in accordance with the principles of the Declaration of Helsinki (1996) and the Good Clinical Practice guidelines.

Consent for publication

This was granted in writing by Berkshire Healthcare Foundation Trust on 29th March 2023.

Availability of data and materials

The dataset resulting from the study are currently being stored on a secure non-publicly available server at the University of Reading. Anonymized data will be made available through the University of Reading Data Repository.

Competing interests

The authors declare that there are no competing interests

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Authors' contributions

VS led on the design of the study and writing of this paper. All other authors contributed to the design and/or data analysis and/or implementation of the study.

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Appendix 1: Weekly diary

HOW TO COMPLETE THE WEEKLY DIARY

Dear parents,

Here are some pointers on how to complete your weekly intervention diary.

1. As you can see, we have allowed for up to 6 sessions per week as some of you may choose to work with you child 3 times a week, 20 mins each time, or 4 times per week, 15 mins each time, or 6 times per week, 10mins each time. You could also do a combination of these. For example, your first session could be 20 mins, your next 4 sessions could be 10 minutes each. It's ok as long as the total time adds up to 1 hour.
2. It is also possible that your child is ready to move from one level to the next within the week. That's ok, please indicate in the relevant box which level you are working at.
3. Please also indicate if your child needed to prompt them to respond (out of 5 attempts)
4. When the child is able to respond to 4 out of 5 attempts unprompted, put a tick next to that toy and use different toys (as the child needs to be able to respond unprompted on 4 out of 5 attempts to 5 different toys).
5. Please remember that your child is ready to move to the next level when they are able **to respond without prompts** on 4 out of 5 attempts with 5 different toys. If your child responded on 4 out of 5 attempts with prompts, they need more practice.
6. Also please write which toys you have used.
7. We have also included a 'Comments Box' for each week, so that you can put in anything that you have observed. For example: a child may have got really obsessed with one specific toy, or your child spontaneously said/signed a new word during the session, or you struggled to understand what you were supposed to do.

See below for an example completed diary for ONE week and parent's comments.

Child's name: Jane				Week of intervention (1-10): 1		
-	Session 1 Date (dd/mm/yyyy) 20/01/2020	Session 2 Date (dd/mm/yyyy) 22/01/2020	Session 3 Date (dd/mm/yyyy) 23/01/2020	Session 4 Date (dd/mm/yyyy) 26/01/2020	Session 5 Date (dd/mm/yyyy)	Session 6 Date (dd/mm/yyyy)
Level (1-7)	1	1	1	1		
Session length (min)	15	10	20	15		
Number of toys used (up to 5)	2	1	3	2		
Toy 1	Car	Car ü	Ball ü	Bus ü		
Child responded unprompted	0	4	4	4		
Child responded when prompted	1	1	1	1		
Child did not respond when prompted	2	0	0	0		
Toy 2	Ball		Bus	Train		
Child responded unprompted	0		3	3		
Child responded when	1		2	2		

prompted						
Child did not respond when prompted	1		0	0		
Toy 3			Train			
Child responded unprompted			2			
Child responded when prompted			3			
Child did not respond when prompted			0			
Toy 4						
Child responded unprompted						
Child responded when prompted						
Child did not respond when prompted						
Toy 5						
Child responded unprompted						
Child responded when prompted						
Child did not respond						

when prompted						
Summary of week:						
Move to next level: YES NO ü						
Any other comments:						

As you can see, in the first session the parent used 2 different toys (car, ball), in the second session 1 toy (ball) as it was a short session, in session 3 the parent used 3 different toys (ball, bus, train) and in the last session, the parent used 2 different toys (bus, train).

In session 1, the parent used prompts 3 times with the first toy (car) to get a response from the child and 2 prompts for the second toy (ball), however the child only responded to one of these.

In session 2, the parent used prompts once to get a response from the child, whereas on 4 out of 5 attempts, the child responded without prompts. Hence the parent has put a tick next to the word 'car'

In session 3, the parent used one prompt for the first toy (ball) to get a response from the child and the child responded unprompted to the other 4 attempts. Hence the parent has put a tick next to the word 'ball'. For the second toy (bus) the parent used a prompt on 2 out of 5 occasions and for the third toy (train), the parent used a prompt on 3 out of 5 occasions.

In session 4, the parent used one prompt for the first toy (bus) to get a response from the child and the child responded unprompted to the other 4 attempts. Hence the parent has put a tick next to the word 'bus'. For the second toy (train), the parent used a prompt on 2 out of 5 occasions.

The child is not able to move from level 1 to level 2 yet because the child was able to respond unprompted on 4 out of 5 requests for 3 different toys. So in week 2, the parent will continue to work on level 1.

Appendix 2: Adherence to intervention

Adherence and compliance with manual- telephone call

Date of call	
Participant ID	
Name of person making the call	
Intervention week	<ul style="list-style-type: none"> • Week 4 • Week 8
Q1: How many sessions have you done with your child during the past week?	<ul style="list-style-type: none"> • 1 • 2 • 3 • 4 • 5 • 6 • 7 • More than 7
Q2: How long was each session approximately?	<ul style="list-style-type: none"> • 1-5 minutes • 6-10 minutes • 11-15 minutes • 16- 20 minutes • More than 20 minutes
Q3: Which level have you focussed on during the last week?	<ul style="list-style-type: none"> • 1 • 2 • 3 • 4 • 5 • 6 • 7
Q4: Tell me about the last session - what did you do and which toys did you use	
Q5: How did your last session go?	<ul style="list-style-type: none"> • My child engaged with the toys and we completed all the steps • My child engaged with the toys and we completed most of the steps • My child engaged with the toys and we completed some of the steps • My child engaged briefly with the toys • My child did not engage with the toys
Q6: How has your child responded to the sessions	<ul style="list-style-type: none"> • Very engaged • Mostly engaged

during the past week?	<ul style="list-style-type: none"> • Sometimes engaged • Rarely engaged • Not engaged
Q7: What do you usually do if your child doesn't engage?	
Q8: Have you completed your weekly diary for the past week?	<ul style="list-style-type: none"> • Yes • No
Q9: Have you got any other comments?	<ul style="list-style-type: none"> • Yes • No <p>If yes, then elaborate here</p>

Appendix 3: Contamination questionnaire

CON01_Assessment of contamination

Date (DD/MM/YYYY):	Participant ID:

Have you heard about the social communication intervention? Yes / No

If so how?

.....

Have you seen the intervention manual? Yes / No

Have you seen the intervention being done? Yes / No

Appendix 4: Parent satisfaction questionnaire

Evaluating an early social communication intervention for young children with Down syndrome (ASCEND): a feasibility study.

Parent questionnaire – satisfaction of intervention

1. How satisfied are you generally with the intervention programme? (please circle)

Very satisfied Fairly satisfied Neutral Fairly dissatisfied Very dissatisfied

2. Do you think your child's responding to shared attention has improved since starting the intervention?

Yes

No

Comments: _____

3. Do you think any improvement in shared attention has influenced your child's speech/language/communication skills?

Yes

No

Comments: _____

4. Have you noticed any improvement in other areas of your child's development?

Yes

No

Comments: _____

5. Have you changed anything about your communication with your child since the start of the intervention?

Yes

No

Comments: _____

6. Any general feedback would be greatly appreciated – what worked well, what could be improved, why are you satisfied/dissatisfied with the programme?

Appendix 5: Topic guide for interviews with SaLTs about intervention acceptability

TOPIC GUIDE FOR INTERVIEWS WITH SPEECH AND LANGUAGE THERAPISTS

One of the objectives of the Feasibility study is to assess the acceptability of the intervention to speech and language therapists and effectiveness of recruitment of children with Down syndrome by SaLTs. To achieve this objective, all speech and language therapists who had facilitated recruitment and/or the delivery of the intervention during the feasibility study, as well as other SaLTs from Oxfordshire, Berkshire and other counties with paediatric caseloads are invited to participate in an interview with a member of the research team. From this pool of participants, we will purposively sample so that all SaLTs who had supported the delivery of our intervention during the feasibility study (10-12) take part, and SaLTs who were not involved in the feasibility study with a range of specialisms are also represented.

The interview will be conducted either over the phone or face to face. It will be audio-recorded for later transcription and analysis.

The following topic guide will be used during the interviews.

1. The Research Assistant introduces themselves and thanks the participant for agreeing to take part. The interview will last about 30 mins
2. Ask if the SaLT took part in the Feasibility Trial

YES

NO

If they answered YES, ask the following questions:

- What did you think of the Parent manual provided (in terms of usability and quality of information)?
 - Do you think that parents *could* use the information provided in the manual? (probe why could/couldn't use the information)
 - Do you think parents used the information in the manual? (probe why would/wouldn't use the information)
 - What do you think worked well?
 - What did not work well?
- What did you think about the training the research team provided?
 - Was it useful, and if so how?
 - Was it necessary, and if so why and how? (probe if there were bits that were more or less necessary)
 - What do you think worked well?
 - What do you think didn't work well?
 - Would you have been able to understand the information in the Parent Manual without the training provided?
- Do you think that this type of parent-led intervention should be offered and supported by SaLT services? Probe:
 - Can you tell me more about why you think that
 - Are there other types of intervention that SaLT services could offer that would be better?
 - Are other services (not SaLT) better placed to offer this type of intervention?
 - What do you think parents think about SaLT services offering this kind of intervention?

- What do you think about Clinical Trials?
 - Do you think they are a good way of evaluating interventions?
(probe why/why not)
 - Do you think they are practical? (probe why/why not)
 - Do you think they are a good thing for patients and/or practitioners?
(probe why/why not)
- Would you be interested in being part of a clinical trial which is looking at implementing a parent-led intervention for young children with Down syndrome?
 - Would you find the prospect of being involved in a clinical trial appealing (probe why/why not)
 - Is there anything that would make you reluctant to be involved in a clinical trial?

If the SaLT answered NO for question 2, then ask the following:

- What do “parent-led interventions” mean to you? Here, we mean an intervention which is entirely delivered by a parent following a manual, with occasional support by an SaLT), even it is confirming SaLTs understanding (*aim is to ensure a shared understanding of parent led interventions*)
- What do you think about this kind of parent-led intervention?
- Have you ever recommended to a parent a parent-led intervention in your paediatric practice? Probe:
 - What made you recommend it?

- Was it a *parent led intervention* like the one in this study? (probe how was it similar/different)
- Do you think it was a success?
- Would you recommend a parent-led intervention in future? (probe why/why not, if yes to whom/in what circumstances)
- What do you think about Clinical Trials?
 - Do you think they are a good way of evaluating interventions? (probe why/why not)
 - Do you think they are practical? (probe why/why not)
 - Do you think they are a good thing for patients and/or practitioners? (probe why/why not)
- Would you be interested in being part of a clinical trial which is looking at implementing a parent-led intervention for young children with Down syndrome?
 - Would you find the prospect of being involved in a clinical trial appealing (probe why/why not)
 - Is there anything that would make you reluctant to be involved in a clinical trial?

Appendix 6: Intervention levels (1-7) based on Whalen & Schreibman (2003)

Level 1	Response to hand on object (adult gently puts child's hand on object to draw child's attention to it)
Level 2	Response to tapping object (adult taps object to draw child's attention to it)
Level 3	Response to object activation (adult uses a mechanical toy which moves/makes a noise to draw child's attention to it)
Level 4	Eye contact – the adult encourages child to make eye-contact while also engaging with a toy/object
Level 5	Following points – choice of two toys - the aim is for the child to follow adult's eye-gaze by following adult's pointing
Level 6	Following points in a book - The goal is for the child to follow the adult's eye-gaze (line of regard) by following adult's points in a book
Level 7	Following points around the room - The goal is for the child to follow the adult's eye gaze (line of regard) by following the adult's point to something that is outside of their visual field e.g. behind them or something they have to look up or down for, or to the left and right of the child.