

BMJ Open Internet delivery of intensive speech and language therapy for children with cerebral palsy: a pilot randomised controlled trial

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ABSTRACT

Objectives To test the feasibility of recruitment, retention, outcome measures and internet delivery of dysarthria therapy for young people with cerebral palsy in a randomised controlled trial.

Design Mixed methods. Single blind pilot randomised controlled trial, with control offered Skype therapy at end of study. Qualitative study of the acceptability of therapy delivery via Skype.

Setting Nine speech and language therapy departments in northern England recruited participants to the study. Skype therapy was provided in a university setting.

Participants Twenty-two children (14 M, 8 F) with dysarthria and cerebral palsy (mean age 8.8 years (SD 3.2)) agreed to take part. Participants were randomised to dysarthria therapy via Skype (n=11) or treatment as usual (n=11).

Interventions Children received either usual speech therapy from their local therapist for 6 weeks or dysarthria therapy via Skype from a research therapist. Usual therapy sessions varied in frequency, duration and content. Skype dysarthria therapy focused on breath control and phonation to produce clear speech at a steady rate, and comprised three 40 min sessions per week for 6 weeks.

Primary and secondary outcome measures Feasibility and acceptability of the trial design, intervention and outcome measures.

Results Departments recruited two to three participants. All participants agreed to random allocation. None withdrew from the study. Recordings of children's speech were made at all time points and rated by listeners. Families allocated to Skype dysarthria therapy judged internet delivery of the therapy to be acceptable. All families reported that the study design was acceptable. Treatment integrity checks suggested that the phrases practised in one therapy exercise should be reduced in length.

Conclusions A delayed treatment design, in which dysarthria therapy is offered at the end of the study to families allocated to treatment as usual, is acceptable. A randomised controlled trial of internet delivered dysarthria therapy is feasible.

Strengths and limitations of this study

- The study addresses a topic which has been identified as a high priority for research by parents and clinicians.
- Assessors of the primary outcome measure were blind to timing of data collection and participant.
- The study used a mixed methods approach to assess feasibility and acceptability of the intervention and study design.
- Participants needed to have access to the internet and a computer/tablet at home to be eligible to join the trial, which may limit the generalisability of the findings.

INTRODUCTION

Approximately 2 per 1000 infants have cerebral palsy,^{1 2} of whom 20% have motor disorders affecting speech production,³ causing the speech disorder dysarthria. Dysarthria in cerebral palsy can lead to shallow, uncontrolled breathing for speech; breathy, harsh voice; reduced pitch variation/unexpected pitch breaks; hypernasality; poor articulation and difficulties in modulating pitch, loudness and timing to vary intonation.⁴⁻⁷ These characteristics reduce the intelligibility of children's speech, with severe impacts on quality of life,^{8 9} social participation,^{10 11} education and employment prospects.¹²

Therapy to improve speech is limited, but is a priority for families.¹³ Children in the UK receive an average of 20 hours per year to address both communication and speech issues. Intervention often targets word production through the Nuffield Dyspraxia Programme¹⁴ and non-speech oromotor function, which have no supporting evidence in childhood dysarthria.¹⁵ Small-scale studies of therapy to control respiratory effort for speech and coordinating exhalation with phonation to generate a clearer speech signal

and a steady speech rate have shown increases in intelligibility, utterance duration and communicative participation.^{16–19} However, as yet no randomised controlled trials (RCT) of this type of intervention for children with cerebral palsy have been reported.²⁰

To date, therapy focusing on breath support and rate has been provided face to face. But sessions may be difficult to schedule in school given the number required to promote motor learning (18–20 sessions over 4–6 weeks, depending on the therapy programme^{17 18 21}). Attending clinics outside school hours may not be possible because children with cerebral palsy have considerable mobility restrictions, in addition to parents' work and other commitments. A recent systematic review has shown that web-based therapy shows promise as an acceptable, feasible and effective method of delivery for children with speech and language disorders.²² The activities involved in dysarthria therapy (eg, repetition, picture naming, picture description and conversation) are similar to those used in previously tested internet therapy,^{23 24} and thus it may be possible to deliver dysarthria programmes as teletherapies. If internet delivery was found acceptable and feasible to children with cerebral palsy it may increase their access to intensive therapy.²⁵

This study aimed to examine the acceptability of internet delivery of dysarthria therapy and to test the feasibility of conducting an RCT comparing intensive dysarthria therapy targeting respiration, phonation and speech rate delivered via the internet using Skype in increasing the speech intelligibility of children with cerebral palsy and dysarthria with usual therapy. The objectives were: to ascertain recruitment rate, attrition rate, outcome measure completion, data quality and acceptability of the study design to participant children and their families in both randomised arms of the study; to measure adherence to the treatment protocol to those allocated to Skype therapy and gauge families' perceptions of the acceptability of Skype delivery of therapy; and to develop a tool to measure therapy costs. Our previous research sought to capture natural variation and change in speech by recording children on two separate occasions twice before and three points after therapy.^{17 18} This study additionally aimed to determine the minimum amount of data that should be collected in a full trial.

DESIGN

This feasibility study comprised: (1) a single blind, randomised controlled pilot trial to compare intensive dysarthria therapy targeting respiration, phonation and speech rate delivered via the internet using Skype with usual therapy; and (2) a qualitative investigation of the views of children and their parents about the acceptability of the Skype dysarthria therapy and the trial design.

METHODS

Settings

Eight National Health Service (NHS) speech and language therapy departments in Northern England and

one independent school participated in the study. All provided services to children with cerebral palsy and were typical of services which would be invited to take part in a full trial.

Patient involvement

In our initial study of the intervention, therapy was delivered four times per week for 4 weeks. Participants recommended the format be changed to three times per week therapy over 6 weeks.²⁶ Parents of children with speech difficulties associated with cerebral palsy took part in workshops prior to the study to advise on the overall design of the study and select the primary outcome measure of speech intelligibility. A member of the research team (a parent of a child with cerebral palsy who had completed postgraduate training in health research) designed materials for the project and determined strategies for interaction with parents and children, including recruitment. She also created the interview topic guides, and led the dissemination of findings to families through a group workshop and written summaries.

Participants

We aimed to recruit 24 children with cerebral palsy and dysarthria to the study based on the recommendation of Julious.²⁷ These indicate that 12 subjects per group is reasonable to consider feasibility and precision of parameter estimates.

Inclusion criteria comprised: diagnosis of cerebral palsy made by paediatrician or paediatric neurologist; diagnosis of dysarthria made by speech and language therapist (SLT); parents rate children's speech as 'imprecise but usually understandable to unfamiliar listeners in context' or 'unclear and not usually understandable to unfamiliar listeners out of context'²⁸; aged 5–17 years; attending schools in North of England; internet access at home; and parents agree to be randomised to either dysarthria therapy delivered via Skype (herein Skype dysarthria therapy) or usual local therapy with the offer of Skype dysarthria therapy at the end of the trial.

Exclusion criteria for the study were bilateral hearing loss >50 dB (from prior audiological testing); severe visual impairments not correctable with glasses (from prior vision testing); and unable to understand grammatically simple instructions (tested by SLTs).

Intervention

Families used their own personal computer (PC) or tablet and webcams (internal or external) for the therapy. No additional hardware (eg, external microphones) was provided. Smartphones were not used as images of pictures used in therapy would be too small to see clearly. Prior to Skype therapy parents accepted the therapist's Skype contact request and taught their child to recognise the therapist's Skype picture on their PC or tablet. Parents were with the child when the therapist called to start each session. Skype dysarthria therapy was delivered three times per week for 6 weeks following the Speech

Systems Approach.^{17 18} Therapy sessions were delivered by one of three therapists employed for the project (HK, NP, KS), depending on availability. The therapists were generalist paediatric clinicians who were 6 months to 4 years postcertification and who had no prior experience of providing motor speech interventions or teletherapy. Prior to the research, they were trained to conduct the programme according to a written protocol by the lead author through demonstration, discussion and self-study. Each therapy session lasted approximately 40 min. Children were taught to control exhalation and phonation to produce a clear voice in open vowels by copying the therapist's model. Children were given external targets relating to how voice sounds, individualised to children's vocal characteristics, for example, 'say it in a loud/big/strong/steady voice', until the most helpful prompt was found. Children then practised using their (loud, steady, strong) target voice in a hierarchy of speech tasks involving increasingly longer utterances, from short single words to multisyllabic words and phrases in speaking tasks involving progressively greater cognitive processing (eg, conversation, question and answers, guess who games). Children were required to produce their target voice in 8/10 productions in a task to move to the next level of the hierarchy. In phrases, children were encouraged to limit the number of syllables they spoke on a breath and to maintain a speaking rate which sustains the target voice and speech clarity. In each session, children practised using their target voice in at least 120 utterances in four exercises: (1) 10 open vowels; (2) three productions of 10 self-selected phrases that they often use in daily life (herein familiar phrases); (3) 70–80 words and phrases from the speech task hierarchy (eg, 70 single syllable words); and (4) random presentation of 10–20 utterances from the three preceding tasks. Feedback was provided frequently to help children acquire the new voice at each level and then faded to aid retention. Children were also encouraged to monitor their own productions and to use internal feedback (eg, 'How did that sound/feel?'). Parents were free to sit with their child during the therapy. During the first two or three sessions all parents sat near their child, watched how therapists elicited speech behaviours and commented on how their child sounded. Therapists also asked parents to remind children to use their target voice in daily interaction, but did not train them to use other therapy techniques. Once they were familiar with the therapy routine, some parents chose to attend to household tasks out of view of the webcam but within calling distance of the child if any difficulties arose, others continued to sit nearby.

Usual therapy comprised the therapy children were provided by their local therapy services, at usual dose and intensity for 6 weeks.

Recruitment

Children were recruited via SLTs working in participating NHS Trusts and adverts placed in newsletters from parent support agencies (eg, Contact, local authority Disabled

Children's Networks), an independent school for children with disabilities and parent-carer forums. Children who appeared to fit the inclusion criteria were visited at home with their parents. Assessments of speech (Diagnostic Evaluation of Articulation and Phonology,²⁹ Verbal Motor Production Assessment for Children³⁰), receptive language (Test of Reception of Grammar Second Edition³¹) and non-verbal cognition (Raven's Coloured Progressive Matrices³²) were undertaken to confirm inclusion. Adolescents aged 16 years or over and parents provided written consent to participate. Younger children provided verbal or written assent. All recruitment activities, the time taken and any reasons given for declining the study were logged.

Outcome measures

Primary outcome

The aim of dysarthria therapy is to increase children's speech intelligibility. We measured the intelligibility of children's speech in single words and connected speech. As in our previous studies, we elicited single words using the Children's Speech Intelligibility Measure (CSIM),³³ which comprises 200 lists of 50 single words matched in length and complexity. In this assessment, the child repeats words spoken by the researcher. We developed random allocation schedules to ensure that each list was allocated a maximum of twice in the study and no list was allocated to the same child twice. We elicited connected speech using picture description. Children described pictures and the researcher transcribed their speech verbatim, and then repeated the utterances to check their understanding with the child. The research therapists recorded children's single word and connected speech at home on two separate days at 6 weeks and 1 week prior to therapy and at 1, 6 and 12 weeks after therapy completion. Recordings were made using an Edirol 9 digital recorder, with a table-mounted microphone (Audio-Technica Pro 24) placed on a table 80 cm from the child (measured at 180°).

To calculate average percentage intelligibility we played each recording to three unfamiliar adults who had no experience of conversing with people who have speech disorders. We recruited listeners via adverts in newsletters for personnel in regional businesses. Each recording was heard by three different listeners. Each listener was allocated two to three different recordings via a secure web platform with the proviso that they heard the same child only once. Listeners were blind to the time of recording (ie, before or after therapy). They were instructed not to turn up the volume of the recordings (although this could not be enforced). In the single word task listeners heard a word and selected the word from a written list of 12 phonetically similar foils. In the connected speech task listeners orthographically transcribed each phrase they heard. The system constrained playing of recordings so that each word/phrase could be played only once. We provided no information about the participants other than that they had cerebral palsy and were taking part in a

study about their speech. We calculated percentage intelligibility scores for each single word recording and each connected speech recording heard by each listener using the word lists and gold standard transcriptions created by the researchers when making the connected speech recordings as the denominators.

Secondary outcomes

We measured the wider impacts of intervention using the Focus on the Outcomes of Communication Under Six (FOCUS),³⁴ a validated measure of communicative performance and participation. Parents and children's class or form teachers (depending on the child's school) completed the Parent form and Clinician form respectively at 1 week before therapy and 12 weeks after its completion.

Children and parents independently rated the improvements in children's speech using a 7-point scale: 1 indicates speech intelligibility is almost the same, hardly any better at all; 7 indicates speech is a very great deal better.³⁵ Text in the children's scale was accompanied by smiley faces, graduating in size, and numbers to indicate improvements (see online supplementary material 2).

Randomisation

Children were randomly allocated to Skype dysarthria therapy or usual treatment with the offer of Skype dysarthria therapy at the end of the study in a 1:1 allocation ratio, after the first visit to collect data (6 weeks before therapy). Randomisation was undertaken using an independent web-based allocation system hosted by Newcastle Clinical Trials Unit.

Adherence to therapy protocol

The Skype dysarthria therapy requires that children produce at least 120 speech behaviours in four exercises per session. At the start of each exercise the therapist gains eye contact, checks the child's sitting posture, describes the exercise, provides a model and instructs the child to produce the behaviour in their target voice (eg, 'Tell me what these are. Use your strong voice.' We developed a Treatment Integrity Checklist to code the presence of these therapy actions and the number of behaviours elicited in each exercise (see online supplementary material 1). We randomly selected two sessions of each child's Skype dysarthria therapy to be video recorded. A researcher who was not involved in delivering therapy to the recorded child completed the treatment integrity checklist from the video recording. Five of the video recordings were also independently rated by a second rater.

Acceptability of Skype-delivered dysarthria therapy and trial design

Parents and children allocated to the Skype dysarthria therapy group were interviewed three times (1 week before, 3 weeks into and 6 weeks after the intervention) about their experiences of receiving therapy through Skype by a researcher who was not involved in therapy

provision (JS). In the third interview they were also asked about their experiences of randomisation and taking part in the study. Parents and children allocated to usual therapy were interviewed about the study design at the end of the study. Interviews were semistructured and followed a topic guide developed for the study. Questions about the therapy covered perceived benefits and disadvantages of receiving therapy via Skype, any memorable experiences of the Skype therapy, impact of therapy on children's speech and communication and parents' encouragement of children to use their new voice (the latter two topics are unrelated to feasibility and are reported elsewhere³⁶). Questions about the study design asked what it was like to be involved in the research, suggestions for making it easier for families to take part and views about the measures used. All but two children were interviewed separately to their parent/carer to ensure each participant's voice was heard. Initial interviews took place in children's homes. Subsequent interviews were taken at home or via telephone/Skype at the families' convenience. Two children were interviewed at school with their teaching assistant present. All interviews were audio recorded and transcribed orthographically. Interviews with children took approximately 35 min (range 15–45 min) and 45–50 min with parents.

Development of costing tool

A questionnaire to measure time required to provide therapy to children allocated to both Skype therapy and treatment as usual. Therapists recorded: the number of sessions provided to a child; the time taken to prepare, conduct and follow-up each therapy session and the aim and content of sessions for children allocated to treatment as usual; and travel time duration (in 10 min intervals); and the NHS salary band of the person conducting these activities was also collected. We also conducted a literature review of tools to measure benefits suitable for use in an economic evaluation. PubMed, Embase, Medline, PsycINFO, NHS EED and Evidence-Based Medicine Reviews (Ovid) databases were searched to identify literatures on tools to measure benefits for children with speech, language and communication disabilities aged 3–18 years. Full-text articles were included with no year limitation but language restricted to English. The review focused on the relevance of the tool to the client group and the availability of a validated method to convert responses into health state utilities which in turn would be used to estimate quality-adjusted life-years—a common metric used in economic evaluations (further details available from the authors).

Analysis

To determine the feasibility of the study design we assessed the recruitment rate, attrition rate, outcome measure completion and data quality in both randomised arms and adherence to the dysarthria therapy protocol in children allocated to Skype therapy. We used descriptive statistics to assess feasibility and set the following criteria:

- ▶ Seventy-five per cent identified children agree to be randomised to dysarthria therapy or usual treatment.
- ▶ Seventy-five per cent children allocated to the usual treatment group are retained for the duration of the study.
- ▶ Eighty-five per cent recorded sessions reach criterion of 120 behaviours produced across four exercises.
- ▶ Seventy-five per cent children recruited to the treatment group and their parents rate therapy via Skype as acceptable and no insurmountable problems are reported by families.
- ▶ Sixty-seven per cent in the Skype therapy condition and their parents rate therapy as at least somewhat effective at 12 weeks after therapy.

Agreement between raters on the adherence to the therapy protocol was assessed using kappa for the presence of therapist actions at the start of each exercise (eg, describing the exercise, providing a model) and Pearson correlation coefficient for the number of behaviours elicited in each exercise. As the aim of the study was to determine the feasibility of the study design, we did not examine change using our clinical outcome measures.³⁷

To assess acceptability of delivery of the therapy via Skype and acceptability of the study design we analysed transcribed interview data using an inductive thematic analysis.³⁸ One researcher (JS) repeatedly read and compared interview transcripts to develop codes to describe meaning within interviews, and grouped codes into overarching themes and subthemes. Verification of the codes and themes took place through discussion of code examples and potential themes with two other researchers (KB and LP). Disagreements were resolved through further review of the data set and explanation of thinking behind particular coding or themes. Interview transcripts were coded and stored using NVivo V.10.

In the development of a tool to assess therapy costs we used descriptive statistics (median, range) to summarise data relating to therapists' time spent on activities relating to each participant allocated to treatment as usual and completion of each section of the data collection form.

To determine if it is possible to reduce the amount of data collected, we examined the agreement between intelligibility measures taken on day 1 and day 2 of the paired recordings and between pretherapy recordings using mixed effects regression models, with recording pair nested within data collection point, nested within child to obtain intraclass correlation coefficients.³⁹ We also used regression models to examine the effect of word list length on estimates of percentage intelligibility. Lists comprising the first 15, 20, 25 and 30 words were compared with the full 50 words. In the examination of the effect of word list length, we fitted child as a random effect and list length as a fixed effect. We used intraclass correlation coefficients to measure agreement between the reduced lists and the full 50-word list for the full data set, and for pretherapy and post-therapy data separately.

RESULTS

Feasibility

Recruitment

Thirty-five children were identified and contacted about the study, of whom 22 were recruited (8 female, 14 male; mean age 8.8 years (SD 3.2)) between October 2014 and October 2015 (figure 1). As dysarthria in cerebral palsy is a chronic condition and children can remain on SLT case loads throughout childhood, we examined identification of participants by services who had recruited children to previous studies separately from those with whom we had not worked before. Therapists from three Trusts who had identified participants in our previous research identified one, three and four eligible children each, of whom one, two and two respectively were recruited. Therapists from five Trusts that had not collaborated in previous studies identified three to five (median=4) potential participants each, of whom two to four participated (median=3).

All families who fitted the study criteria agreed to be randomised to Skype therapy or treatment as usual with the offer of Skype therapy at the end of the study, surpassing our feasibility criterion of 75%. Identification of the first participant in each of the first wave Trusts took 4–64 days and their recruitment took 19–45 days from identification.

Characteristics of the study population

Table 1 shows the characteristics of the participants in the two groups. There were more boys in the treatment as usual group, and overall this group had slightly lower receptive language and non-verbal understanding than the children allocated to the Skype therapy.

Attrition

None of the participants withdrew from the study. All children allocated to Skype dysarthria completed 6 weeks of intervention. All children remained in the arm to which they were originally allocated, surpassing our feasibility criterion of 75%. However, due to the deadline set by our funding body, four families were unable to complete measures at 12 weeks after therapy.

Outcome measure completion

We completed 206 of 220 planned recordings (94%) of children's speech. Seven recordings, from the last four children recruited, could not be completed by the cut-off for recordings which was imposed by the research time frame. A further seven recordings (from four children) could not take place due to child illness (4) or holidays (3). We recruited 213 listeners over 12 weeks, who rated a mean of three recordings each (SD 0.15). Parent and teacher compliance with questionnaire (FOCUS) completion was low: 29/44 parent questionnaires (66%); 26/44 teacher (59%); 34 pretherapy (77%); 22 post-therapy (50%).

Table 2 provides a breakdown of measures collected for participants in the Skype dysarthria and usual therapy groups.

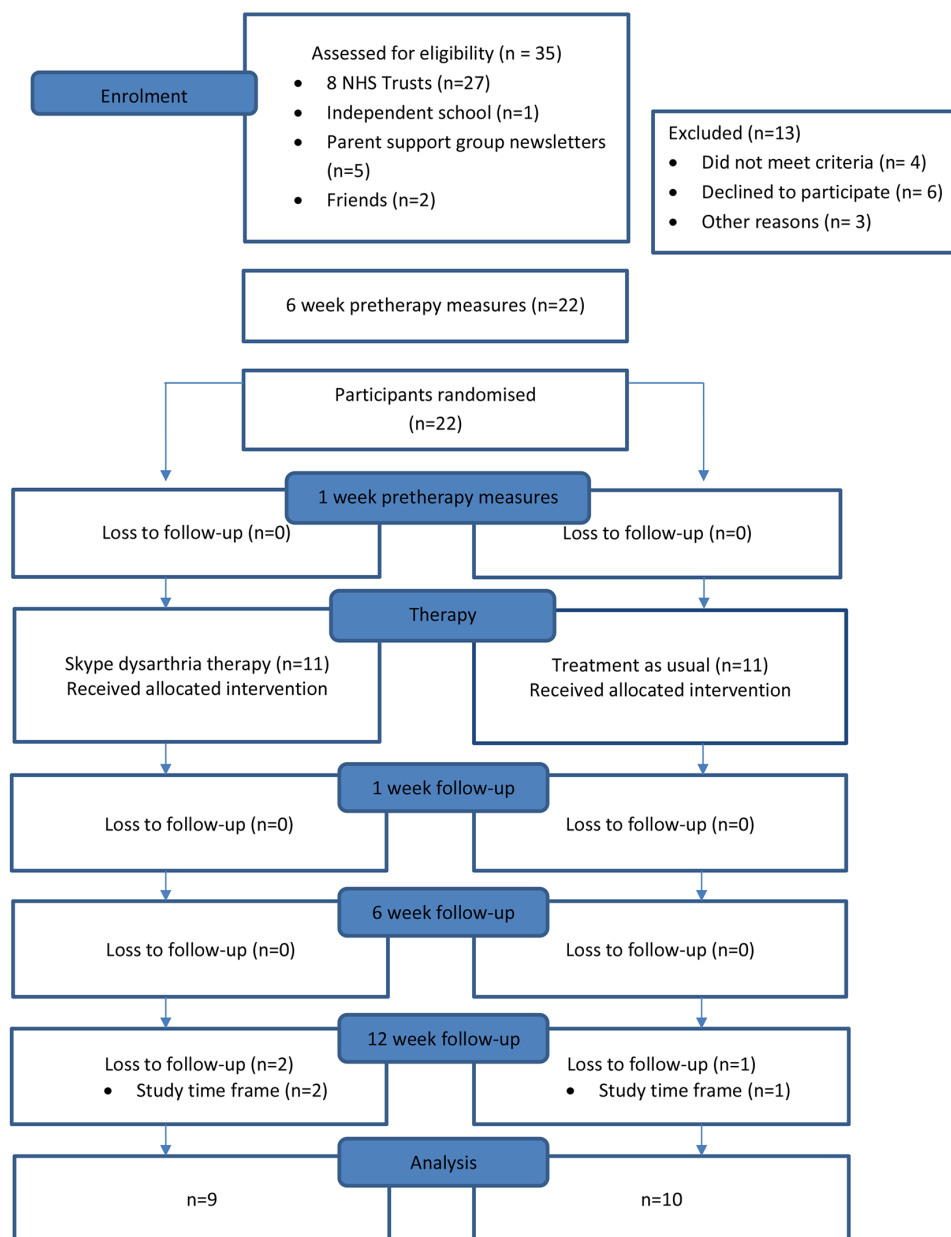


Figure 1 Consolidated Standards of Reporting Trials (CONSORT) flow diagram. NHS, National Health Service.

Adherence to therapy protocol

Children received a mean of 15 sessions (SD 2.7, median 16, range 11–18). Children missed sessions because of illness and competing family commitments. Internet connection issues were encountered in 32/147 sessions (median 2, range 0–10 per child). Therapy time lost to connection problems ranged from 2 to 30 min per child over their programme of therapy (median 15; range 0–38). Longer connection issues were encountered when new devices were used and one session was abandoned due to Skype issues that affected users globally.

Video recordings of 31 Skype dysarthria therapy sessions (19%) were checked for treatment fidelity. Agreement between the two raters on the presence of therapy activities at the start of each exercise was substantial⁴⁰ ($K=0.72$) and high for the numbers of behaviours produced in each exercise ($r=0.99$, $p<0.001$). Twenty sessions (65%)

met the criterion of 120 behaviours (mean 121, range 74–140), thus failing to meet the feasibility criterion of 85% of sessions containing 120 speech behaviours. Failure to elicit 120 speech behaviours arose because the functional phrases selected by five participants for practice in exercise 2 were long and could not be spoken on one breath. This curtailed the time left for rapid elicitation of novel words and phrases and random practice in exercises 3 and 4. In two sessions, Skype connection difficulties reduced time available to complete the exercises.

Effectiveness of therapy

All eight children and eight parents who rated effectiveness of therapy at 12 weeks after intervention deemed it at least moderately effective (median rating by children 5, range 5–7; median rating by parents 6, range 4–7). Should the three children and their parents who were

Table 1 Participant characteristics

	Skype dysarthria therapy group (n=11)	Usual therapy group (n=11)
Sex	5 M; 6 F	9 M; 2 F
Mean age in years (SD)	8.8 (2.1)	8.8 (4.1)
Type of cerebral palsy	Spastic 6 Dyskinetic 5	Spastic 6 Dyskinetic 3 Ataxic 2
GMFCS median (IQR)	III (I, IV)	I (II, IV)
MACS median (IQR)	II (I, III)	II (II, III)
CFCS median (IQR)	III (I, IV)	II (II, III)
Viking Speech Scale median (IQR)	III (II, III)	III (II, III)
Mean language comprehension age (TROG2 age equivalent score)	7.8 (3.3)	5.6 (2.9)
Mean non-verbal understanding age (Raven's Progressive Matrices age equivalent score)	6.6 (2.8)	4.9 (2.0)
Mean length of utterance in words	5.2 (1.9)	5.3 (2.8)
Phonemes articulated (from DEAP, /24)	19.9 (4.5)	18.1 (5.6)

CFCS Communication Function Classification System⁵⁹; DEAP, Diagnostic Examination of Articulation and Phonology; GMFCS, Gross Motor Function Classification Scale⁶⁰; MACS, Manual Ability Classification Scale⁶¹; TROG2, Test of Reception of Grammar second edition³¹.

unable to complete ratings due to time limitations have judged it less than somewhat effective (ie, 8/11 (72.7%) judging therapy as effective), the criterion of 67% rating the therapy as at least somewhat effective would have been reached.

Acceptability

Skype delivery of therapy

Discussion about the delivery of the intervention via Skype centred around four key themes: (1) familiarity with technology; (2) remote connections; (3) fit with family life; and (4) unforeseen benefits.

Familiarity with technology

During the interviews, none of the parents raised concerns about using Skype for therapy. Those allocated to the Skype therapy described themselves as being familiar with using technology for communication; some had used Skype before to keep in touch with relatives and others had used alternative video communication software such as FaceTime. Parents were happy for the children to be contacted by the research therapists via Skype for prearranged sessions once they had accepted the study Skype name as a contact.

Skype I haven't used in probably about five or six years. But yes, we've used FaceTime a lot. (Father A)

Table 2 Outcome measures completed for participants in the Skype dysarthria usual therapy groups

Measure	Skype dysarthria therapy group	Usual therapy group
Number of recordings made/total possible (%)		
6 weeks pretherapy	22/22 (100)	22/22 (100)
1 week pretherapy	21/22 (95.5)	21/22 (95.5)
1 week post-therapy	20/22 (90.9)	20/22 (90.9)
6 weeks post-therapy	22/22 (100)	22/22 (100)
12 weeks post-therapy	17/18 (94.4)	19/19 (94.4)
Number of listeners hearing recordings/total possible (%)		
6 weeks pretherapy	57/66 (86.4)	58/66 (87.9)
1 week pretherapy	62/66 (93.9)	61/66 (92.4)
1 week post-therapy	59/66 (89.4)	60/66 (90.9)
6 weeks post-therapy	58/66 (87.9)	60/66 (90.9)
12 weeks post-therapy	48/54 (88.9)	53/57 (93.0)
Number of FOCUS questionnaires completed/total possible (%)		
Parents pretherapy	8/11 (72.7)	10/11 (90.9)
Parents post-therapy	6/11 (54.5)	5/11 (45.5)
Teachers pre therapy	8/11 (72.7)	8/11 (72.7)
Teachers post-therapy	6/11 (54.5)	4/11 (36.4)
Number of parents rating therapy effectiveness/total possible (%)		
1 week post-therapy	11/11 (100)	NA
6 weeks post-therapy	10/10 (100)	NA
12 weeks post-therapy	9/9 (100)	NA

FOCUS, Focus on the Outcomes of Communication Under Six; NA, not applicable.

I'll show you how to get Skype on and then you'll get that ready. When I tell him, once he's done it, he'll know how to set it up because he's good with computers, so it won't be a problem. (Mother E)

Children were also positive about therapy via their tablet/computer, seeing it as 'cool' (Child G). Most appeared confident about using the technology from the start, although one child who had not used Skype or FaceTime had lots of questions about how to work the software and appeared a little anxious about being able to connect. However, in post-therapy interviews this child reported that she quickly 'got used' to Skype after the first couple of sessions.

I know Skype. Will it ring? (Child Q)

There is a button you have to press, you know the 'enter' button on the computer? When you've got the little arrow, and you can move the arrow around with the mouse? (Interviewer)

I don't know, yes I do. And you just click the button. What if we don't have a mouse? I think it's going to

be better if someone like peeked through the door. If I need help with a hard question and I peek through the door and say I am struggling, and they come and help me. (Child Q)

Remote connections

Parents reported that internet connections were generally good and that when connections were lost they were usually retrievable within a session.

There was one day I think [research therapist] was struggling to connect. She just Skyped a typed message saying, 'I'm just having a few problems.' ... But on the whole our Broadband has been fine because it does stick a little bit sometimes where we are. But we don't seem to have had any issues really. (Mother E)

We never even had any issues with Wi-Fi connection and our Wi-Fi's rubbish. We never got a lost signal through once, I don't think that's happened once. If there is any, it's just like me being the idiot and not turning up the volume up. (Father A)

Parents and children reported enjoying therapy and feeling comfortable with the therapist.

Basically the same. Just on an iPad instead of face to face. (Child R)

I think it's great in the fact that it is over Skype. You've not got people coming in all the time, in and out of the house, which is not a problem. But it makes it a bit more fun for the youngsters, I think, by having Skype. [Child] was sitting there with his iPad and he was really enjoying it. (Mother R)

However, one child reported that sitting in the same position for the duration of the Skype sessions was difficult.

It's difficult to do sitting. On Skype it's different. (Child V)

On Skype it's difficult to sit still for that long. (Mother V)

Overall, although shorter sessions might be better for some children, it seemed that the families did not view the technology as a barrier to therapy.

Fit with family life

Parents and children reported that a key benefit of Skype therapy delivery was being able to have therapy at home; families did not have to rush to attend appointments or make additional car journeys, which are difficult for families of children with disabilities, making clinic appointments stressful for children, parents and siblings. Children liked doing therapy in familiar surroundings.

It's nice that he can do it in his own home, just relaxing...it's not like 'Right come on, we have to go out' and drive there, in and out of his wheelchair...it's just another trip out which we would rather avoid if you don't have to do it. (Mother I)

However, the three times per week schedule was difficult to manage across the full 6 weeks for some families.

Yes it is manageable.... I couldn't say we could do a fourth one. Three is the limit. (Mother V)

Some parents thought that having some of the therapy sessions via the internet at school may facilitate greater engagement of education staff in children's therapy and understanding of it.

I think that would be quite good because then, at least, you'd be able to get a bit more feedback from the school as to how they think that it's improved on. (Mother R)

Unforeseen benefits

Parents reported that therapy delivery via Skype had an unforeseen benefit of promoting children's independence. They described how children used their own computer or tablet, chose where to do therapy and some older participants logged on independently once parents were sure they recognised the Skype contact used by the researchers for the study. Parents described how they could remain nearby, see what was happening in therapy and be on hand to help if was required, but could also attend to other household tasks and spend time with their other children. This meant that parents could be involved but also let their children take control and ownership of their therapy.

I think she feels, it gives her a bit more independence. It's hers. ...We'll pop our head in now and again and make sure she's okay. She doesn't need us at all. She can just do it herself which is great. (Father A)

Yes, kind of, you're listening to how well she's doing, but equally taking in what the girls are doing with her, so that you can kind of apply some of it at home. (Mother G)

Research design

Discussion around the study design centred on the random allocation to Skype therapy or usual therapy and the outcome measures used in study. One of the motivating factors in taking part, even if allocated to usual care, was the offer of therapy via Skype at the end, and this seemed crucial for successful engagement with participants:

I think if it was just for a study [without offer of Skype therapy], I think we would have been less likely to (participate). (Father D)

That was the great thing, it didn't matter when, you were going to get it regardless, so it was worth it. (Mother O)

The visit schedule for the collection of data was time consuming, but deemed to be manageable for the duration of the study.

It is a big commitment, yes. If you've got the time, they do make you aware of what time you're going to need prior to the therapy. So it's not like you're not aware of it. (Mother R)

Just the fact that everything has to be quiet, that's quite tricky with having a three year old who's quite noisy; that was quite hard. And the fact that it had to be all in one week as well. That was quite tricky. But certainly I wouldn't say that it was a negative thing, it just was more difficult to fit in really. (Mother V)

All families allocated to treatment as usual accepted the offer of Skype dysarthria therapy at the end of the study. Having therapy immediately after the end of the study period was seen as preferable for both parents and children allocated to treatment as usual.

It's going to work out that we've had continuous visits, regular visits, and then the therapy will start for them. It's actually maybe worked out quite well. It's not like it's been forgotten and then, 'Oh,' all of a sudden here's the therapy from something you did six months ago. (Mothers F and J)

All parents and all participants reported that having multiple speech recordings made for the study was acceptable, but that the task did become boring. One child reported that she did not like being recorded but quickly got used to it.

I'm used to it now. It's not hard for me, it's easy now. It used to be hard when I first started. Now it's getting easier and easier. (Child C)

Neither participants nor parents reported any concerns about the recordings being heard by strangers in order to estimate children's intelligibility.

You've got to measure the improvement.... There'd be no point in doing it if you weren't going to see a difference... So it has to be somebody who doesn't know the children or who have never heard them before. (Mother E)

Most parents also reported that the FOCUS questionnaire was relevant and easy to complete, and none reported reasons for not returning the data. One mother reported that she had to check the meaning of some items and was concerned that she could not recall previous ratings and wanted to report that she thought her child had improved.

(remember thinking) I'm misinterpreting this. I can't just fathom out what they're asking me. ... I should have taken a photocopy actually of my answers. Just to remember what I'd put last time. (Mother E)

Harms

No adverse events were notified during the study.

Economic measures of therapy costs

Therapists of all children allocated to treatment as usual and the research therapists completed all sections

of the resources questionnaire. Results suggested that some sections could be simplified (therapists' travelling time to see children could be left open for therapists to complete).

Six children allocated to the treatment as usual group received at least one session (median 1; range 0–7) of speech and language therapy from NHS services in the 6 weeks therapy period assessed for the study. Of the six children who did receive input, two were seen for single review sessions only, two received regular therapy focused on expressive language and two received therapy targeting articulation. Therapy sessions were usually 31–40 min in duration. Staff salary bands ranged from 4 (therapy assistant) to 7 (specialist).

The literature review identified 13 articles reporting the use of tools previously used in this client group to estimate health state utilities. Apart from generic tools developed for use in children, for example, HUI2[®], EQ-5D-Y, no other suitable tools were identified. A further tool, the CHU-9D, while not previously used, was also considered potentially suitable.

Reducing the data set

There were strong intraclass correlations between percentage intelligibility across pairs of recordings within the same subject at each of the five time points, for single words this was 0.87 (95% CI 0.79 to 0.91) and connected speech 0.82 (95% CI 0.73 to 0.89). Intraclass correlations were also strong between recordings taken at 1 and 6 weeks before therapy for single words (0.83, 95% CI 0.7 to 0.90) and connected speech (0.73, 95% CI 0.57 to 0.86).

Mixed effects regression models showed very little impact of list length on intelligibility after adjusting for the random effect of child when the full 50-item lists were compared with 30, 25 or 20 items. However, when the first 15 items were compared with the full 50-item list a significant difference was observed (95% CI -3.87 to -0.36). There were strong intraclass correlations between the 20, 25, 30 and 50 words of the CSIM lists, for both the full data set and when pretherapy and post-therapy data were examined separately (table 3).

DISCUSSION

Summary of findings

This study aimed to test the feasibility of conducting an RCT of the clinical and cost-effectiveness of intensive dysarthria therapy versus usual treatment for children with cerebral palsy. Results suggest that an RCT is feasible and acceptable but that the study processes and data collection can be simplified to reduce research costs and burden to participants.

The study design was acceptable to families. Families joined the research because they were seeking therapy to improve children's speech intelligibility and communicative independence, which is highly prized.⁴¹ An important component of feasibility of recruitment was the offer of

Table 3 Mean and SD of percentage of words understood in the first 15, 20, 30 and 40 words and full 50-word lists from the Children's Speech Intelligibility Measure

Words in CSIM lists	Percentage of words perceived correctly by listeners		Comparison with full 50-word list					
			All data		Pretherapy		Post-therapy	
	Mean (SD)	95% CI	ICC	95% CI	ICC	95% CI	ICC	95% CI
Full 50-word list	50.27 (18.51)	48.82 to 51.72	–	–	–	–	–	–
First 30 items	49.78 (19.41)	48.26 to 51.30	0.97	0.96 to 0.97	0.97	0.95 to 0.98	0.97	0.96 to 0.98
First 25 items	49.67 (19.84)	48.11 to 51.22	0.96	0.95 to 0.96	0.96	0.94 to 0.97	0.96	0.95 to 0.97
First 20 items	49.55 (20.25)	47.96 to 51.13	0.93	0.93 to 0.94	0.92	0.90 to 0.94	0.94	0.92 to 0.95
First 15 items	48.45 (21.14)	46.80 to 50.11	0.89	0.87 to 91	0.87	0.83 to 0.90	0.90	0.87 to 0.92

CSIM, Children's Speech Intelligibility Measure; ICC, intraclass correlation coefficient.

the experimental therapy to families allocated to treatment as usual at the end of data collection, even though the treatment may turn out not to be effective. Our findings here concur with the results of previous trials of interventions for children with other types of neurodisability, such as autism,^{42–44} and support the conclusion that parents of children with neurodisability often feel the need to 'do more' for their children continue to seek additional input that could aid their child's development.^{45 46} There was no loss of precision in estimates of variation of intelligibility when we reduced the number of words assessed from 50 to 20, which is similar to other tests of intelligibility.⁴⁷ Future trials could limit data collected to single recordings of 20 single words and picture description at 1 week before therapy, and 1, 6 and 12 weeks after therapy. Return of the FOCUS questionnaire was low (approximately 60%) and could possibly be improved if usual mechanisms of communication (eg, home-school diaries and bags) were used.⁴⁸

Similar to other studies of teletherapy for children with speech disorders and adults with dysarthria,^{22 24 49} interviews with parents and participants suggest that provision of therapy via Skype is feasible and acceptable to families. Therapy was delivered according to the therapy protocol and children and parents reported no difficulties in maintaining rapport with the therapist, supporting other recent research.^{22 24 50} One possible advantage of the study design was that the research therapists were familiar to families by the time therapy was due to commence, having carried out the assessments and speech recordings. Shaw and colleagues, in their study of general practice consultations, found that sessions via videoconferencing flowed more easily when patients and practitioners knew each other and had time to build a relationship.⁵⁰ Internet connection issues affected one in five sessions in our study, but were generally short and resolvable as previous UK research.⁵⁰ However, connection difficulties affected some children more than others, possibly due to broadband speed. Test sessions to establish Skype connection with families and to confirm computer settings would be advisable in future research and if implementing Skype in therapy.⁵⁰ None of the parents reported any concerns about internet safety and were happy

for therapists to contact the family via Skype to start therapy sessions. Individualised actions to maximise internet safety would be required in a trial, as recommended for all children,⁵¹ and Trusts would need to assess information governance issues individually.⁵⁰ However, Skype is now being piloted by NHS Trusts as a medium to conduct consultations and advice exists on its use in clinical settings.^{52–54}

For most children it was possible to complete all therapy exercises within 40 min. Problems that occurred were due to a selecting phrases that were too long to say in one breath. A similar finding has been noted for adults with dysarthria caused by Parkinson's disease.⁵⁵ This could be readily avoided in future trials. Shortening the phrases may also help reduce the overall session length, making them more accessible to children who find it difficult to maintain their concentration.

Therapy was conducted by three generalist clinicians who had no experience of childhood motor speech disorders or teletherapy. As such, they were similar to many NHS therapists who provide services to children in mainstream schools in the UK, but less experienced than therapists who provide specialist services to children with motor disorders.¹⁵ This study has shown that the protocol can be followed by junior therapists and by extension it should be suitable for therapists with greater experience. Future trials should test the effectiveness of the therapy when delivered via local therapists, and include a process evaluation to evaluate barriers to and facilitators of implementation which have not been identified in our research to date.⁵⁶ If a study were clustered at the unit of Trust, to avoid contamination between therapists providing intervention and participants interacting with each other, mean cluster size can be estimated as 3, based on recruitment from Trusts who had not participated in previous studies. We recruited a small number of participants who were known to NHS speech and language therapy services via parent networks and support organisations. Advertisement of trials via parental support networks should be encouraged to reduce gatekeeping.

Limitations

This study recruited participants from eight Trusts within one region. Recruitment may not be representative of Trusts in other parts of the UK. However, to our knowledge no research is being conducted on children's speech elsewhere in the UK and there is sparse research internationally.²⁰ Families participating in this study used their own laptops/tablets for the remote therapy and may represent a 'tech-savvy' group. Teletherapy in NHS is only feasible to families with a computer and internet access; consideration would be needed as to whether this approach might widen socioeconomic health inequalities.

Intelligibility measures were conducted by the research therapists, which could have prompted children to use their new voice during measurement. Future research should ensure that measurements are conducted by personnel who have not been involved in therapy but who have built up a rapport with children so that they feel comfortable during the assessments.

All parents rated speech to have improved at least moderately by 12 weeks. Such lack of variation precludes the calculation of a minimally important difference in speech intelligibility using anchor-based methods and suggests change in percentage intelligibility should be used to inform sample size calculation.⁵⁷ Only one child judged their speech to have improved moderately (level 4); all others judged their speech to be a (very) great deal better (6 or 7) at all time points. High ratings may be due to response bias and social desirability. However, children may have experienced change that led them to report their speech being a (very) great deal better. Furthermore, they may also have developed confidence in or from their ability to be understood, as reported in the interviews. Future research must measure the impact of therapy on children's participation and well-being as well as their intelligibility.¹³

The aim of usual treatment and a very brief description of the intervention was provided by local therapists in our costing tool. Although the tool showed that none of the children in the treatment as usual group received intervention focusing on breath support, phonation and rate to increase the clarity of children's speech, it did not enable us to describe the intervention that the children received. For example, we do not know if motor learning principles that underpinned the Skype dysarthria therapy were included in articulation therapy. Future research should include the aim and focus of treatment as usual and the active ingredients. The Template for Intervention Description and Replication checklist⁵⁸ could be used as a basis for expanding the tool.

CONCLUSIONS

The study suggests that an RCT of dysarthria therapy is feasible, that therapy can be provided via Skype to children with cerebral palsy and that families would participate in a trial if all arms of the study included provision of Skype dysarthria therapy at some point. Future trials

could use a smaller data set to measure intelligibility than previously tested, which would reduce the time and costs required to conduct a trial and reduce the burden of participating in the study for families.

Contributors LP, ES, LV and KB designed the study. LP coordinated the study and took overall responsibility for the research. MO developed the website for conducting listening tasks. HK, NP, JS, KS and PA had responsibility for data collection. HK, NP and KS delivered the therapy. LP, ES, PA, LV, KB and JS analysed the data. LP drafted the paper. All authors have contributed to and approved the final version of the paper.

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