# Protocol for SonoSpeech Cleft Pilot: A pilot randomised control trial of ultrasound visual biofeedback versus standard intervention for children with cleft lip and palate.

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## Abstract

Background: Children with cleft lip and palate can continue to have problems producing clear speech after surgery. This can lead to social, emotional, and educational challenges. Typical treatment involves teaching children the correct tongue movements to produce speech sounds. This is known as articulation intervention. However, this intervention is challenging because the tongue is hidden from view and movements are difficult to see and describe. This pilot randomized control trial will try a new treatment, ultrasound visual biofeedback.

Methods/Design: The Sonospeech project will enroll up to 40 children with cleft lip and palate aged 4;6 to 16 in a single-centre two-arm parallel group pilot randomized controlled trial with blinded assessors. Children will receive either six sessions of U-VBF or articulation intervention. The primary goals of this pilot are to determine recruitment/attrition rates; to measure pre-post follow up complete; and acceptability of the randomization and interventions to families.

Discussion: Larger trials of speech interventions for children with cleft lip and palate are needed. This pilot/feasibility study will determine whether a larger randomized control trial comparing ultrasound and articulation interventions is feasible.


*Keywords*: Cleft lip and palate, articulation intervention, ultrasound visual biofeedback

## Background

Cleft lip and palate (CLP) is the most common congenital craniofacial abnormality, occurring in one in 700 births (Bellis & Wohlgemuth, 1999). Problems producing intelligible speech occur in CLP, even after surgery to repair the palate, and in some children cleft speech characteristics (CSC) persist requiring intervention from a speech and language therapist (Medina, 2019). This unclear speech has adverse social and educational consequences, with the speech of children with CLP rated as more likely to belong to someone who was "unhealthy", "no friends" and "ugly"(Lee et al., 2017). Standard treatment is articulation intervention (AI). This approach involves teaching children correct placement for their articulators (primarily the tongue) through verbal description and demonstration (Bessell et al., 2013). AI is a challenging intervention for both the clinician and the patient because speech movements are both difficult to see and describe, due to the main articulator, the tongue, being largely hidden from view. This problem can be circumvented by using instrumental articulatory techniques which view and measure the articulators directly. Over the last few decades electropalatography (EPG) has dominated the literature as the instrumental technique of choice for people with cleft lip and palate. EPG measures tongue-palate contact using an artificial palate with electrodes embedded in it. This real-time dynamic image of tongue-palate contact can be used as a biofeedback tool to teach patients about correct placement of the tongue. However, a Cochrane review of EPG by Lee et al. (2009) found that only one study met inclusion criteria. Despite a large number of studies using EPG, most were small n or case studies. It is likely that larger studies using EPG are sparse because of practical issues with using this tool. Each patient requires an expensive custom-made palate, moreover, this custom-made palate only fits for a limited time period due to changing dentition and planned surgeries. In contrast, ultrasound visual biofeedback (U-VBF) is

growing in popularity as biofeedback tool for children with speech disorders (Sugden et al., 2019). U-VBF holds several practical advantages over EPG: it is cheaper and does not require individualised equipment. Moreover, while EPG images only tongue-palate contact from the alveolar region to the boundary of the hard and soft palate, U-VBF images from near the tongue tip to the root, with pharyngeal articulations, common in CLP, clearly visible. This makes U-VBF arguably the technique of choice for CLP (Bressmann et al., 2011) yet it is relatively new to the Speech and Language Therapy clinic. This is because it is only now that ultrasound systems for measuring articulation provide fast enough frame rates at an affordable cost. A recent study showed that ultrasound can be used to identify all of the CSC described in the instrumental literature and that ultrasound assessment has better reliability than traditional perceptual approaches (Cleland, Lloyd, et al., 2019). However, U-VBF has to our knowledge only be used in one small study with two participants with CLP (Roxburgh et al., 2016). In children with other types of speech sound disorders U-VBF shows positive outcomes for the majority of children and it is particularly useful for establishing new articulations (Sugden et al., 2019): an area of particular difficulty in CLP (Bessell et al., 2013). U-VBF is therefore potentially a useful tool for establishing new articulations in children with CLP. The aim of this study is therefore to assess the feasibility and inform the design of a full scale RCT of U-VBF for children with cleft speech characteristics.

## Methods/Design

### Aims

The aim of this study is to assess the feasibility and inform the design of a full-scale RCT of U-VBF for children with cleft speech characteristics. The primary aim of U-VBF is to enable learning of new articulatory gestures (new speech sounds), with secondary aims of improving intelligibility and health-related quality of life.

### Design and Setting

Mixed methods will be employed. The study is a single blind pilot randomised controlled trial, with control offered U-VBF therapy at end of study. A qualitative study (focus group) of the acceptability of both interventions and the study design will also be undertaken. All intervention will take place at the Royal Children's Hospital Glasgow in Speech and Language Therapy Rooms. Eligibility screens and pre- and post-intervention assessments will take place either in person in a university clinic room or via telehealth (Zoom or TEAMS).

### Research questions

No definitive comparisons of the interventions will be undertaken. The feasibility of a full scale RCT will be determined by evaluating a number of objectives against set success criteria (bulleted below) taken from a similar pilot RCT of children with speech disorders (Pennington et al., 2019):

**Objectives**:

1. To determine recruitment and attrition rates

- 75% of children and their families identified agree to participate

- 75% of children allocated in each group are retained for the duration of the study

2. To measure pre-post and follow-up outcome measure completion

- 75% of outcome measures are completed

3. To measure within-session outcome measure completion

- Data is reported from 75% of intervention sessions

4. To determine acceptability of randomisation to children and their families

- 75% of children and their families rate randomisation as acceptable in a questionnaire

5. To determine the acceptability of ultrasound visual biofeedback as an assessment tool (both groups) and intervention tool (U-VBF group)

- 75% of children and their families rate ultrasound as an acceptable technique in a questionnaire.
- Focus group analysis contains

6. To measure adherence to the treatment protocol

- 75% of sessions reach the minimum dosage of 100 trials in both treatment arms

**Methods**

A single-centre two-arm parallel group pilot randomised controlled trial with blinded assessors will be carried out. Cases will be stratified by three age groups (4;6 -6;0; 6;1-8;0 and and >8;0 years). Due to the nature of the U-VBF, therapists and patients will not be blinded to treatment allocation but the limitations of this will be mitigated by the use of an assessor blinded to group and evaluators blinded to both group and treatment time point.

**Participants**

Children aged 4;6 to 16 will be recruited from the West of Scotland Cleft Lip and Palate Service. Inclusion criteria are regarding age and cleft-type are broad to reflect current practice, however, inclusion criteria for type of cleft speech characteristic are narrower to ensure children are likely to benefit from either U-VBF or the control intervention. Children are eligible if they have at least one speech error that would normally be a candidate for articulation intervention. We aim to recruit 20 children to each arm of the trial. Children with a bilateral hearing loss of greater than 30dB (from previous reports), planned surgery within the next three months, or severe language deficit (from previous SLT reports and a receptive vocabulary standard score<70 on the BPVS-3, (L. Dunn, 1997) ) will be excluded. Following baseline the children will be randomised by the Glasgow Clinical Trials Unit in a 1:1 ratio, stratified for age. Children randomised to the control arm will be offered U-VBF at the end of the trial if they still present with CSCs which are candidates for U-VBF as previous studies have shown this improves the acceptability of a randomised trial to families (Pennington et al., 2019).

**Eligibility/Baseline Assessments:**

We will screen potential participants from case-notes and invite them to attend an initial screening and baseline assessment. This assessment will be either in person or via video conferencing, with in-person preferred. Screening assessment will comprise the British Picture Vocabulary Test 3 (L. Dunn, 1997) to screen for adequate receptive vocabulary; and a speech assessment protocol to determine whether patients present with at least one cleft speech characteristic which would be amenable to both U-VBF and the control intervention. This assessment protocol comprises the Diagnostic Evaluation of Articulation and Phonology (Dodd

et al., 2002) articulation and phonology subtests and a ultrasound tongue imaging protocol designed in a previous project (Cleland, Lloyd, et al., 2019) to identify covert speech errors from consonants at all places of articulation and sentences from the GoS.SP.aSS.'98/ CAPS-A (Sell et al., 2009) (Appendix 1). Families who opt for the assessment over video-conferencing will complete the same assessments, but the ultrasound tongue imaging protocol will be replaced with a perceptual assessment of the same materials[1].

**Speech Target Selection**

Children with CLP may present with multiple CSCs affecting intelligibility. We intend will select as intervention targets speech sounds which are 1. amenable to treatment with both interventions and 2. likely to have the biggest functional impact on intelligibility. Following the screening assessments we will select wordlists targeting each child's specific lingual errors from a battery. In English, lingual speech sounds (imageable with ultrasound and amenable to treatment with both interventions) are /t,d,n,r,l,s,z,ʃ,ʒ,tʃ,dʒ,j,k,g,ŋ/ and all vowels. Children with CLP are most likely to have difficulty with anterior consonants /t,d,s/. Wordlists containing these speech sounds will also form a key outcome measure (see below). Two wordlists per error type will be selected, firstly an "untreated probe" i.e. the words will not be used in the course of therapy, this allows us to check for generalisation of targets. Secondly a "treated probe", containing high frequency and functionally useful words will be used to train speech targets in the course of therapy. The wordlists contain lingual targets in increasingly complex contexts

[1] The assessment sessions are not part of core clinical care and therefore we will offer these over video-conferencing to comply with local Covid restrictions under which travelling for research assessments may not be an "essential journey".

from single syllable words/pseudowords through to multi-syllabic words and sentences. Where the child has more than one error, multiple wordlists will be used, however only one treatment target (the speech sound with the most errors) will be selected and the other errors will serve as "control segments" (i.e. speech sounds that should not improve during the course of therapy unless maturation is a factor). Wordlists will be analysed for percentage target consonants correct (PTC). Children must score <30PTC at baseline to be eligible for the study.

**Interventions**

Both interventions will be delivered by the cleft palate specialist SLTs in the Children's Hospital. Therapy in both treatment arms will be once per week for six sessions with each session lasting 45mins. The number of sessions is pragmatic in nature, reflecting current practice, and is designed to highlight initial response to both interventions. It is likely that some children in both arms of the trial may require further speech intervention in the future (after the follow-up measures are taken) and this will be provided in line with standard practice, which includes further U-VBF. Both interventions will focus on acquisition of new speech sounds. Previous studies show that a new speech sound can be acquired within one to two sessions (Cleland, Scobbie, et al., 2019) of U-VBF for most children but that some children take four to six sessions. Target articulations will be decided individually. Both interventions begin with a pre-practice phase where the aim is to teach the child to approximate the target articulation before they can begin the practice phase where at least 100 repetitions are required for learning and generalisation. This dosage will be measured in both interventions. In this pilot we will focus on both the pre-practice phase and the first stage of practice: acquisition of a new sound in simple contexts such as "ta, tea, toe" building to short words such as "tap, team, tore" as this is feasible

within six intervention sessions. If children are super-responders (i.e. they quickly retain the new speech sound and are able to produce it in complex contexts) then the protocol will also allow us to measure this.

### Articulation Intervention (AI)

This intervention involves working on a single speech sound at a time. The SLT uses modelling, demonstration, verbal description, and feedback in the pre-practice phase to teach the child the new sound at first in limited contexts and then in words and finally in conversation in the practice phase. To increase parity with U-VBF, and in line with newer theories of motor learning (Ruscello & Vallino, 2014), we will standardised the in-session dosage during the practice phase to at least 100 trials, i.e. each child will be given 100 attempts to articulate their target articulation in each session.

### Ultrasound Visual Biofeedback (U-VBF)

This intervention is grounded in the principles of motor learning. The patient sees a real-time image of their tongue moving and guided by the SLT uses this biofeedback to learn a new articulation, building productions to increasingly complex contexts, as in AI. Again a minimum of 100 trials are required in the intervention. The intervention is set out in an open access manual (Cleland et al., 2018) and involves using the software Sonospeech™. The software has functionality to be used as an assessment and intervention tool, allowing the SLT and patient to record and playback ultrasound video with synchronised audio or to view it live. The clinicians delivering the intervention have completed training and currently use ultrasound in their clinical practice.

**Outcome Measures**

A blinded assessor will collect measures at baseline (-t1: pre-treatment), 2 months post-randomization (t7, to allow for any delays in referral to therapy), and 3 months post-randomization (t8) to see if any benefit is maintained. Assessment will take place in a university clinic or via video-conferencing and will be carried out by a research SLT blinded to group for primary outcome measures and by the treating SLTs for within-session measures. See table 1.

**Within-treatment session outcomes**

Previous research suggests that one of the main benefits of U-VBF may be efficiency rather than overall efficacy of treatment (Sugden et al., 2019). We will therefore measure treatment response during each session, rather than just after the course of treatment. The treating SLTs will therefore audio record short treated, and untreated word lists at every treatment session (t1-t6). From this we will determine how quickly children achieve a new articulation as a measure of response to treatment. These will be rated at the end of the project by SLTs blinded to group.

**Candidate Primary Outcome Measures**

The key primary outcome linear measure for change in speech will be percentage target consonants correct (PTC). We will measure this at single sound level (stimulability and in /aCa/ contexts); single word level; and sentence level in treated and untreated wordlists (see above section on speech target selection). All direct speech measures will be recorded with audio and where possible ultrasound tongue imaging in both groups, allowing us to perform ultrasound analysis of data from both groups.

**Patient Reported Outcome Measures**

We will use the Intelligibility in Context Scale (McLeod et al., 2012) as a carer reported outcome. This short scale asks parents/carers to rate how easy to understand their child is to a variety of listeners ranging from family members to strangers. Quality of life will be measured using the CLEFT-Q speech function and quality of life scales for children aged 8 and over (Klassen et al., 2018). We will also use the Experience of Service Questionnaire (Brown et al., 2014) to measure patient and carer satisfaction with both interventions at the end of the project.

**Intervention Acceptability Measures and Qualitative Evaluation**

Families will complete a questionnaire about the acceptability of both interventions at the end of the study. Parents/carers and children over 12 will be invited to join focus groups to discuss their experiences of taking part in the trial and to contribute to planning a larger trial. Each focus group will include up to 10 participants. Responses to the focus groups will be analysed using thematic analysis.

Table 1 shows the timeline for the project, including the timing of each assessment.

| | STUDY PERIOD | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | | | | | | | | | | | |
| | Enrollment & Elligibility | Allocation | Treatment | | | | | | Post Treatment Measures | Post Treatment Measures 2 | Qualitative Evaluation |
| Time Point | "-t1" | 0 | t1 | t2 | t3 | t4 | t5 | t6 | t7 | t8 | t9 |
| **ENROLLMENT** | | | | | | | | | | | |
| Informed Consent/assent | x | | | | | | | | | | |
| Eligibility Measures | x | | | | | | | | | | |
| Allocation | | x | | | | | | | | | |
| **INTERVENTIONS** | | | | | | | | | | | |
| Articulation Intervention | | | ◆ | — | — | — | — | ◆ | | | |
| Ultrasound Visual Biofeedback | | | ◆ | — | — | — | — | ◆ | | | |
| **ASSESSMENTS** | | | | | | | | | | | |
| Perceptual rating of targeted consonant from treated & untreated wordlists | x | | | | | | | | x | x | |
| Perceptual rating of within-treatment wordlists | | | x | x | x | x | x | x | | | |
| Patient Reported Outcome Measures | x | | | | | | | | x | x | |
| Patient Reported Experience Measures | | | | | | | | | x | x | |
| Intervention Acceptability Questionnaire | | | | | | | | | | x | |
| **Qualitative Evaluation** | | | | | | | | | | | |
| Parent Focus Group | | | | | | | | | | | x |
| Young People Focus Group | | | | | | | | | | | x |

## Analyses and Statistical Power

Definitive comparisons of the interventions will not be undertaken due to the feasibility nature of the study. Details of patient screening, recruitment, retention, withdrawal, follow-up will be summarised (see research questions above). Adherence to U-VBF will be measured according to the number of patients who complete the intervention in accordance with the treatment manual. Adherence to treatment dosage will be recorded using an intervention pro-forma in each session where the SLT records a tick mark for each trial (i.e. each time the patient attempts to produce the speech sound in treatment, this should be around 100 for both interventions). All sessions will be audio-recorded (consent permitting) and 20% of sessions will be fidelity checked for dosage and adherence to protocol.

### Ultrasound/Speech Analysis

All of the speech measures at baseline, and follow-up will be recorded with simultaneous ultrasound in both groups by a research SLT blinded to group. Our previous work showed that the addition of ultrasound to transcription increases inter-rater reliability and allows identification of covert (imperceptible to the ear alone) errors. This will allow us both to calculate PTC (the primary outcome measure) with increased reliability and to perform an error analysis. 25% of the data will also be rated by two specialist cleft SLTs (not involved in the project) trained in ultrasound-aided transcription. These SLTs will also rate the audio recordings from the within-treatment sessions, blinded to group.

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