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Case Study
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Use of Ultrasound Biofeedback in Assessment and Treatment of Speech Disorders in a 22q 11.2 Deletion Syndrome with Submucous Cleft Palate- A Case Report

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Abstract

Background: The purpose of this study is to present a case that uses ultrasound biofeedback (UBF) to establish the correct production of /s/ sound in a 7-year-old child with 22q11.2 deletion syndrome and submucous cleft palate with a persistent speech sound disorder to illustrate how UBF can be used in a clinical setting.

Methods: The patient underwent sixteen treatment sessions using UBF. Percent consonants correct measurements taken across five data points in cluding twice before treatment (week 1 and week 4), mid-treatment (week 8), post-treatment (week 16), and after a month follow-up.

Results: After employing UBF intervention, percent correct consonants reached 80% accuracy. Despite the child's previous history of unsuccessful speech therapy, the use of UBF resulted in observable changes in tongue shape during the production of the /s/ sound following the treatment.

Conclusion: Ultrasound can be beneficial in showing new tongue movement and placement for improved intelligibility. Ultrasound stands to make a significant contribution to the treatment and diagnosis of speech sound errors, but more research is needed to understand how ultrasound is used with this clinically-diverse population.

Keywords: Ultrasound; Cleft palate; 22q deletion; Ultrasound therapy; Articulation; Speech sound disorders; Biofeedback

Introduction

More than 50% of children with cleft lip and palate (CLP) achieve typical speech by the age of 5 [1,2]. There are several factors that may contribute to the persistence of speech difficulties beyond the age of 5. One of the most important ones is the co-occurrence of cleft palate with syndromic conditions affecting the vocal tract and articulators. There are more than 500 defined Mendelian syndromes include cleft palate as a feature [3] accounting for about 50-55% of cases in cleft palate only patients and 30% in CLP patients [4,5]. 22q11.2 deletion syndrome also known as velocardofacial (VCF) or DiGeorge (DGR) syndrome is the most common cause of syndromic palatal anomalies and velopharyngeal dysfunction (VPD) [6]. As documented, more than 90% of children with 22q11 present with a speech-language disorder which could involve velopharyngeal closure [7,8].Velopharyngeal dysfunction (VPD) and submucous cleft palte have been reported in 67% of children with 22q11. Speech sound production skills are worse in children with 22q11 than individuals with isolated cleft palate. Those misarticulations could occur due to dysarthria, apraxia of speech, VPD, or phonological-developmental delay. Obligatory errors such as weak pressure consonants, nasalization of phonemes, audible nasal air emission as well as compensatory misarticulations such as glottal stops, pharyngeal fricatives/stops, nasal fricatives, and laryngeal fricatives, have been reported to persist into late childhood and adolsenece [9,10]. Therfore. it is crucial for SLPs in schools and other settings to be aware of available intervention approaches for this group of childen. One of which is ultrasound biofeedback (UBF).

Ultrasound visual biofeedback (UBF) is becoming increasingly popular as a biofeedback technique for children with diverse speech sound disorders. Therapy using UBF has been mainly focused on children with hearing impairment, apraxia of speech, and individuals with residual sound errors with unknown etiology [11].The instrumentation has been used to characterize characteristics of cleft palate speech in several studies [12-15].However, it has only been used in intervention in one small study involving two participants with CLP [16].

Ultrasound biofeedback therapy uses the real-time imaging capabilities of ultrasound to augment speech-sound therapy sessions with a visual representation of the tongue as it moves during speech. What the monitor displays from the ultrasound is actually the tongue-air interface, a bright white line created by the large amplitude of ultrasound echoes due to the acoustic impedance mismatch between the tongue structure and the air. The ultrasound transducer, or wand, is placed submentally (below the chin) and is typically oriented either to capture a sagittal image or a coronal image of the tongue surface (Figure 1). Ultrasound can show the tongue-air interface of the root, dorsum, blade, and near-tip regions (but typically not the tip, especially for alveolar consonants). In addition, it cannot reveal articulatory gestures outside the oral space, such as lip constriction or jaw excursion, palate, and velum without special measures. As such, it is a method of identifying tongue movement or tongue placement during speech but without reference to other articulatory structures.

This case study aimed to describe an evidence-based treatment approach using UBF provided to a child with 22q11.2 deletion

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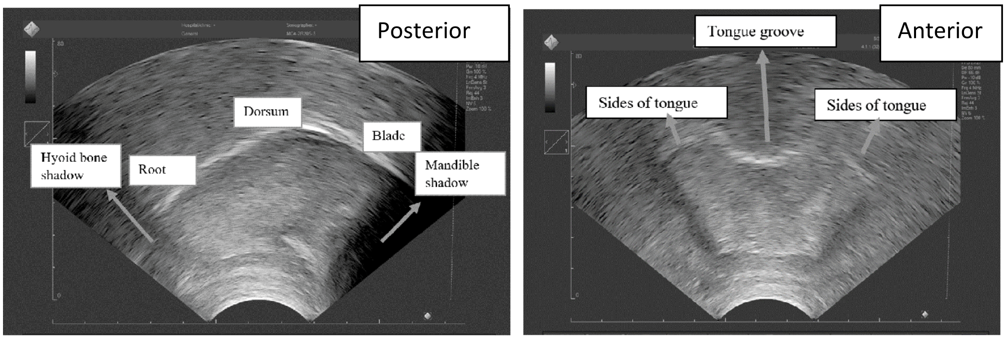

Figure 1: Ultrasound images of the tongue in sagittal (left) and coronal (right) planes.

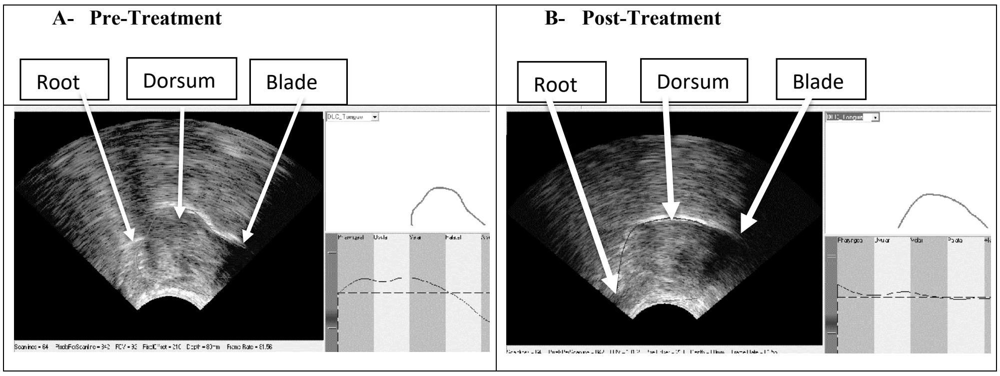

Figure 2: The Visual Display of Ultrasound Data from the Articulate Assistance Advanced (AAA) Software During the Production of /s/ in the Word /sɛvɪn/, 3A: Data Captured Pre-treatment, 3B: Data Captured Post-treatment

syndrome and submucous cleft palate with persistent speech sound disorders associated with it. Additionally, we documented the effectiveness of intervention through UBF in improving the child's articulation accuracy, tongue configuration, and intelligibility. The study is unique because it takes an exploratory approach to different steps of assessment and treatment with this client using UBF. It also helps readers to understand how UBF can assist in better identifying the type of errors and treatment planning. The treatment goal was to establish a correct motor plan for the sound disordered and generalizing it to variable speech contexts.

Case presentation: The Client is a 7-year-old girl diagnosed with 22q11 deletion syndrome. She was enrolled in speech therapy at 3 years old when her preschool teacher noted that other children had trouble understanding her and she seems "muffled". Medical history included a diagnosis of an autoimmune disorder (Age 4), tonsillectomy and adenoidectomy (Age 5), submucouse cleft palate (Age 6), and sensorineural mild-moderate hearing impairment in both ears (Age 6). The client's speech evaluation from the craniafacial team report showed moderate-severe hypernasality, inaudible nasal emission, and articulation errors.

Per mom's report, the client had made some progress in her speech goals at school but recently plateaued and seemed unmotivated to continue to work with her school SLP. It was the parents decision at the moment not to purse surgical intervention for her VPD due to several medical and behavioural diagnoses that could impact the outcome of surgery. Therefore, the client's school SLP recommended that she try ultrasound speech therapy as a part of a grant study affiliated with the craniofacial team.

Procedures: assessments

Americleft speech assessment was conducted as an initial assessment, the SLP noted that the client's speech was hypernasal (score=3), and in addition to some developmental errors, all three cleft speech charactristics (CSC) groups of errors were noted (Posterior oral CSCs, score=1; non-oral CSCs, score=1; Passive CSCs, score=2; Noncleft speech immaturities/errors, score=1, and Anterior errors "dentalized productions", score=2).

The client indicated errors such as devoicing of almost all fricatives and affricatives, distortions in pre-vocalic /r/, inconsistent use of glottal fricatives for the /s/ and /z/ sounds, and dentalized distortions in /s/ and /ʒ/. The devoicing errors could not be addressed using ultrasound, so the SLP selected the /s/ sound to be targeted in therapy. The SLP asked the client to keep the ultrasound wand under her chin and say "See", "Saw", "Say", and "So" (along with other s-initial and s-final simple syllable words). In most s-initial contexts, the dorsum of the client's tongue raised up and back towards the uvula while simultaneously making an inappropriate elevation near the soft palate, suggesting an undifferentiated tongue gesture (Figure 2A) for pre-treatment tongue posture for the /s/ in the word /sɛvɪn/). Despite the error initially seemed to resemble a palatal fricative due to raised dorsum in mid-palatal area, it was accompanied with a backward movement of the tongue root and constriction in the glottal area, resulting in its perceptual resemblance to a glottal fricative. Various factors could contribute to the development of persistent fricative distortions in individuals with cleft palate. These include hearing impairment, presence of VPD, and dental crowdings resulting from malocclusions (Zajac et al., 2021). It appears that the client had some of these factors that could have impacted the production of fricatives.

Treatment plan: After her ultrasound session, the SLP explained that our tongue is made up of different muscles that work together to make certain shapes for speech sounds. In child-centered language, she explained to the client , "The tongue is like a hand: you can move part of your hand (some fingers) without moving the other part (the other fingers). You can move part of your tongue without moving the other part, too, but it can be difficult to learn that if you don't see it. Now that we can see it, we might learn new ways of moving our tongue." In order to set realistic expectations, SLP also told the client and her mother that the ultrasound might help, but it might take a while for noticeable progress to happen.

Intervention included 2x a week therapy sessions for 8 weeks (45 min duration) with two follow up sessions scheduled 4 weeks apart at the end. The SLP chose to target the /s/ in isolation to establish a motor plan for the sound. The SLP focused on modeling (using ultrasound on herself) the tongue movements for the /s/ sound and showing pictures of what the tongue shape looked like for the consonant. When the client was able to identify the "right" tongue shape vs. an "old tongue shape" (consistent with her original method of producing /s/), the SLP encouraged her to explore ways of making this shape, targeting both the tongue tip up and tongue tip down positions for the /s/ sound. Per the ultrasound images and what SLP observed from her attempts, the tongue tip-up position seemed to suit the client's pronunciation needs better. She was able to keep the tip towards the alveolar ridge during /s/ attempts in 8 out of 10 tries. The initial syllabic structure introduced to the client was /si/ to encourage anterior tongue movement (due to client's pattern of glottal articulation). The SLP later moved to /s/ syllables with the vowel /a/, as the client had some dentalized errors, a wider jaw opening in this vowel inhibited tongue protrusion. The target sound /s/ was practiced across all vowel contexts with an accuracy goal of 80% in all contexts. The SLP praised the client with knowledge of performance (KP) feedback: "Good work, you were able to raise your tongue blade by itself-I did not see the tongue is raising in the back!" The SLP emphasized what the different parts of her tongue were doing when the client moved from one phoneme to the next.

Consistent with work on the implementation of ultrasound for other articulation errors in published protocols, the SLP focused on making sure that the client could produce the target sound in a given context with an accuracy rate of around 8 out of 10 tries before moving on to another context. Client ended the first session with an overall /s/ accuracy rate in most CV contexts at 80%. Two exceptions were the /su/ and /so/ context (65% accuracy). The client was also able to name parts of the tongue (tip, blade, dorsum, root, and sides) and explain what they had to do for "her" (tip up) /s/.

Follow-up and outcomes: Over subsequent sessions, the client progressed with other vowel contexts in word-initial and final positions, phrases, sentences, /s/ blends, multi-syllabic words, and complex sentences. The percentage of correct target consonants was calculated as the key outcome measure using fifty untreated words four times throughout the duration of the stud (Figure 3). displays the scores of percent consonants correct measurements taken twice before treatment (week 1 and week 4), mid-treatment (week 8), post-treatment (week 16), and after a month follow-up. Two listeners who were blind to the research design and the data point timeline scored the probes' responses using audio/video ultrasonic recordings. The reliability of these correct/incorrect judgments on a point-by-point basis was calculated using Cohen's kappa and was $\kappa = .65$.

Discussion

As the client made progress with target sounds in various contexts, the SLP would trial a "no ultrasound" condition, where the client would put the ultrasound wand down and try to reproduce her successful production without visual support. If she struggled with this task, the SLP would ask her to try it again with the ultrasound wand, and remember what her tongue had to do, and once success was established, they would then repeat the "no ultrasound" condition. The purpose of this was to help the client be able to practice at home where there would be no ultrasound feedback.

After seeing the client for 16 sessions, the SLP noted that client was able to correct her errors without the ultrasound (or other cues) about 80% of the time. When, needed oral cues such as "Remember your tip high in the screen!" helped the client to correct herself around 85% of the time. The client's level of accuracy without ultrasound (and with the support of oral cues) helped the SLP decide that she could return to her school SLP for continued treatment, with the opportunity to return for further ultrasound speech therapy if the family desired.

Conclusion

Ultrasound biofeedback is a non-invasive, and real-time method of

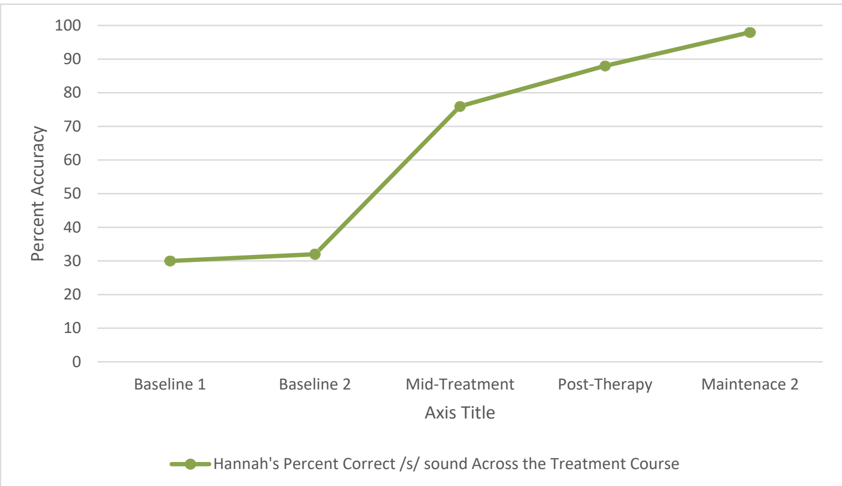

Figure 3: Client's percent consonants correct (PCC) in untreated probe lists across the course of treatment.

visual feedback that, like other motor-based visual feedback techniques, may improve accuracy of diagnostics and therapy cues for persistent speech errors in children with syndromic cleft palate. Enrollment in ultrasound studies is often restricted to individuals with typical craniofacial anatomy, making it difficult to know how craniofacial disorders (i.e., pierre robin sequence, macroglossia, and submental scarring secondary to surgery) will present when using ultrasound. Such restricted participant eligibility criteria are common in early-stage, explanatory research studies, where researchers seek to know if the intervention can work under highly-controlled conditions (Thorpe et al., 2009). However, case studies of participants with craniofacial anomalies enrolled in ultrasound speech treatment programs remain a promising research area. More research is needed to understand best practices for ultrasound biofeedback implementation with this clinically-diverse population.

Consent

We obtained institutional review board approval for the case study, and the parent needed to sign an informed consent form at the start of treatment. To protect the participant's identity, she will simply be referred to as "the client" and her mother as "the mother."

Conflict of interest statement

Author has no relevant conflict of interest to report.

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