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A Lingual Ultrasound Study of Speech in Patients With Cleft Lip and Palate Following Orthognathic Surgery

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Abstract

Objectives: This study evaluated the effects of orthognathic surgery on speech of Class III (‘underbite’) patients with repaired cleft lip and palate (CLP) through ultrasound imaging, aerodynamic assessment and articulatory analyses before and after surgery to evaluate how surgery impacts articulation, velopharyngeal (VP) function and hypernasality.

Materials and Methods: Tongue gestures of five Class III patients with repaired CLP, five Class III patients without CLP and five Class I controls were visualised through ultrasound imaging pre- and post-surgery. Tongue Dorsum Excursion Index and Tongue Constraint Position Index (TCPI) were calculated using ultrasound, and VP insufficiency (VPI) indices were measured using pressure flow assessment and compared with qualitative-perceptual ratings by Speech-

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Author Contributions

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Conflicts of Interest

The authors declare no conflicts of interest.

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Language Pathologists (SLPs). Linear mixed-effects models were used to test for significant post-operative changes in Dorsum Excursion Index and TCPI among consonants. Pressure flow measures are expected to estimate VP gap size during speech using PERCI-SARS.

Results: Patients with CLP had significantly lower Dorsum Excursion Index for /k/ before front ($p = 0.001$) and back vowels ($p < 0.001$). TCPI revealed consistently lower values for patients with CLP than controls and Class III patients, with inter- and intra-speaker variation for all consonants. Pressure flow data of two patients with CLP showed evidence of VPI and altered nasality post-surgery.

Conclusion: Patients with CLP present with lower Dorsum Excursion Index and TCPI when compared to controls, suggesting more anterior tongue positioning. Dorsum Excursion Index and TCPI can be used to understand tongue shape and movement changes, potentially elucidating functional effects of jaw surgery on speech. Dorsum Excursion Index and pressure flow data indicate increased compensatory articulations and VPI after orthognathic surgery for patients with CLP, impacting speech.

1 | Introduction

Cleft lip and palate (CLP) is one of the most common congenital malformations, with an incidence of 1 in every 700–1000 births [1]. Most cases are sporadic, with no association with other syndromes or genetic conditions [2, 3]. Nearly half of patients with CLP present with speech distortions during childhood compared to 3.6% among their non-cleft peers [4–6]. Young children with CLP have speech delays that persist after palate repair [7]. The American Cleft Palate Craniofacial Association recommends primary palate repair surgeries to be performed by 18 months of age or earlier when possible [8]. Depending on the time of intervention, cleft size and state of surrounding tissues, different techniques can be used to achieve palatal closure, with common procedures being a two-flap palatoplasty, Furlow double opposing Z-palatoplasty and two-stage palatoplasty in the United States [9].

Following palatal surgeries, patients with CLP may present with speech alterations, including hypernasality and audible nasal air emission (NAE), often due to inadequate velopharyngeal (VP) closure, a condition known as velopharyngeal insufficiency (VPI) [7]. Hypernasality occurs when there is excessive sound energy and airflow change in the nasal cavity during the production of oral sounds. The increased energy affects vowel quality and is also problematic for consonants that depend on increased intraoral pressure, such as stops (/p b t d k g/) and fricatives (/s z ʃ f v/).

Severe malocclusion further exacerbates speech impairment in patients with CLP [10]. Scar tissue caused by early surgical repair restrains maxillary forward growth, leading to a Class III ‘underbite’ malocclusion. As a result, patients with CLP often undergo orthognathic surgery to advance their maxilla once growth is complete [11]. Independent of CLP, Class III ‘underbite’ is associated with a high incidence of structural speech sound disorders due to abnormal oral anatomy [12, 13]. Patients with CLP and Class III malocclusions may be doubly impacted in their speech, with consonant distortions related to their malocclusion and resonance issues from hypernasality due to VPI [7, 14, 15]. Therefore, patients with CLP may suffer from obligatory hypernasality and structurally based ‘lisping’ that affects

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consonant articulation. Furthermore, patients with CLP and underbites often fail to respond to speech therapy and are considered ‘non-stimulable’ because of their structural speech sound disorders; speech-language pathologists (SLPs) often recommend deferring treatment until malocclusion correction [16].

Patients with CLP often produce intelligible but socially marked, negatively evaluated speech, impacting self-confidence and quality of life [17]. Children with CLP often have learning challenges and lower-than-average educational attainment, unrelated to intelligence or genetic predispositions [18]. Speech is proposed as one of the factors impacting childrens’ educational outcomes [18, 19]. Despite multiple surgeries to improve palatal defects and muscle function, nearly half of patients with CLP suffer from persistent speech distortions into adulthood [16].

Studies have investigated articulation through ultrasound imaging to understand the aetiology of speech distortions in patients with CLP and have identified unique articulatory characteristics in children with CLP. Bressman et al. [20] described compensatory articulations of /k/, such as tongue blade elevation, through qualitative analysis. Zharkova et al. [21, 22] developed two indices to quantify variations in tongue posture to evaluate compensatory articulations in CLP speech. The Dorsum Excursion Index evaluates the degree to which the tongue dorsum is involved during the articulation of a particular sound. The Tongue Constraint Position Index (TCPI) evaluates how far forward or backward the highest point of the tongue is relative to the tongue dorsum. Both indices were measured on ultrasound data from adults without speech disorders [22], typically developing children, and children with CLP, with and without VPI [23, 24]. Less is known about Dorsum Excursion Index in young adult patients with CLP, who have nearly concluded jaw development and are years past peak pubertal growth, unlike growing children featured in prior studies (ages 7–14) [23–25]. Furthermore, little is known about speech changes and articulatory adaptation mechanisms when older patients with CLP undergo orthognathic surgery to correct their Class III malocclusion.

This study aimed to evaluate the effects of orthognathic surgery on the speech of patients with CLP through ultrasound imaging, aerodynamic assessment and articulatory analyses before and after jaw surgery, to evaluate how surgery impacts articulation, VP function and hypernasality. We hypothesise that patients with CLP will exhibit different lingual strategies for consonants post-surgery, while also presenting with hypernasal speech with moderate to inadequate VP closure post-surgery due to the enlargement of the VP gap from maxillary advancement. To test our hypotheses, tongue gestures were visualised through ultrasound imaging before and after surgery. Dorsum Excursion Index and TCPI were calculated using ultrasound, and VP indices were obtained using pressure flow for quantitative measures and compared with qualitative perceptual ratings by SLPs.

2 | Material and Methods

2.1 | Study Setting and Participants

This pilot longitudinal observational cohort study included five participants with repaired CLP (two females and three males), aged 16–21 years old (Table 1). All patients

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were referred to our dentofacial disharmony (DFD) clinics in orthodontics and oral and maxillofacial surgery (OMFS) with a history of repaired non-syndromic CLP (referred to as 'CLP'). At the time of their initial visit, patients were orthodontically decompensated and ready for orthognathic jaw surgery with a Le Fort I maxillary advancement, with or without a mandibular bilateral sagittal split osteotomy setback. All enrolled patients were diagnosed by a boarded orthodontist and oral surgeon as having concluded a majority of growth (CVM 5–6) and having Class III DFD malocclusion, with a negative overjet (OJ < 0 mm) due to maxillary deficiency and/or mandibular prognathism with a Class III dental and skeletal presentation (Table S1). Additionally, five Class III patients without CLP who underwent orthognathic surgery (referred to as 'Class III') and five Class I control patients who received non-surgical orthodontics were selected to compare with the CLP cohort, matching for age and gender (five females, five males; ages 15–22) (Table S2). Two of our Class III patients with CLP had a two-flap Bardach palatoplasty procedure at our centre as young children; the other three patients with repaired CLP had palatoplasty procedures performed elsewhere, with an unknown surgical approach. Participants were enrolled from June 2022 to August 2024 from the OMFS and Orthodontics Departments at the Adams School of Dentistry, University of North Carolina (UNC) at Chapel Hill. All adult participants provided written informed consent, and minor participants gave assent while a legal guardian gave written consent. The research protocol was approved by the UNC Institutional Review Board (IRB) (IRB#19–1196).

2.2 | Speech and Clinical Records

Speech and clinical records were collected at one time point for the five Class I control subjects (at least 2 months into fixed orthodontics with braces) and two time points for five Class III, and five Class III patients with CLP (T0: 1 month before orthognathic surgery and T1: 2–4 months after surgery). At each visit, patients were directed to read 20 English phrases containing target words containing the initial consonants /s/, /t/, /ʃ/ ('sh'), /k/ and following vowels /æ/, /ɑ/, /i/, /u/ (/æ/ as in 'cap', /ɑ/ as in 'cop', /i/ as in 'key' and /u/ as in 'coo'). Targets were embedded in a carrier phrase ('Say___again') to elicit connected speech (Table S3). Each phrase was repeated three times and presented to each participant in a random order. Additional speech audio recording procedures were done as described in Tran et al. [26]

2.3 | Ultrasound Imaging

Ultrasound imaging was conducted with a standard medical ultrasound with a microconvex probe (Transducer model number MC4–2R20S-3, frequency range 2.0–4.0 MHz; Telemed, Vilnius, Lithuania, EU) connected to a portable ultrasound system (MicrUs EXT-1H; Telemed, Vilnius, Lithuania, EU). The probe was secured with an UltraFit headset, allowing for consistency and stabilisation of probe placement [27]. Real-time tongue movement imaging was done with the ultrasound system connected to a laptop running Articulate Assistant Advanced software (AAA) version 221.4.1 [28–30]. Settings were chosen for better visualisation of the tongue (100 fps, field of view: 101.2°, depth of 80 mm). Ultrasound data were obtained by placing the long axis of the probe parallel to the sagittal plane, providing a midsagittal view of the tongue. A microphone (AKG, model C-520; Harman International Industries, USA) attached to the headset allowed simultaneous audio

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and ultrasound recording. Patients were recorded swallowing water to image the palate and were recorded biting down and pressing their tongue against the bottom of a tongue depressor to visualise the occlusal plane. Examples of ultrasound frames during speech are shown in Figure 1A.

2.4 | Ultrasound Evaluation

The tongue surface was automatically traced using DeepLabCut [31] models developed by Wrench and Balch-Tomes [30] in AAA and then hand-corrected. Tongue Dorsum Excursion Index was measured in accordance with Zharkova and Cleland et al. [21–23] Dorsum Excursion Index directly measures the extent of tongue dorsum activity during articulation. A straight line (*n*) connects both ends of the tongue spline, and a perpendicular line (*d*) is located at the midpoint of *n* (Figure 1B). Dorsum Excursion Index is a ratio (*d/n*) and therefore is not influenced by variation in ultrasound probe angle or tongue size. The higher the Dorsum Excursion Index value, the more involved the dorsum is for that particular production. TCPI uses the lines *d* and *n*, as well as the longest perpendicular line from *n* (*l*) to locate the highest point of the tongue. TCPI is calculated by dividing the distance between *d* and *l* and dividing that by half of *n* [22]. According to Zharkova, TCPI values are more positive or negative based on how far forward or backward the, 'part of the tongue closest to the palate is' [22].

Fricatives (/s/, /ʃ/) were visualised using the centre frame of each production, whereas stops (/t/, /k/) were visualised just before their release bursts (i.e., brief burst of air when tongue closure for /t/ and /k/ is released) to show where the tongue makes a closure. Dorsum Excursion Index and TCPI values were automatically extracted using the analysis values feature in AAA and then manually checked for accuracy and to ensure the selected frames for stops /t/ and /k/ were indeed before the release burst.

A linear mixed-effects model was performed to test the significance of Dorsum Excursion Index and TCPI change post-operatively for each consonant using the lme4 [32] and sjPlot [33] packages in R. Dorsum Excursion Index and TCPI were the response variables, and an interaction between patient group, consonant and following vowel (front vs. back) was tested, with the speaker as a random effect. Bootstrapping was performed in order to obtain estimates and significance of each model using the lmeresampler [34] package in R. Alpha parameters were set at $p < 0.05$.

2.5 | Pressure Flow

To assess VP gap size during speech, pressure flow was measured using the PERCI-SARS approach, as described (Microtronics Inc., Chapel Hill, NC) [35]. Three patients with CLP were recorded saying the syllables /pʌ/, /pi/, /bʌ/, /bi/ and /mi/ and the word 'hamper'. They were repeated in sets of three (e.g., /pi pi pi/), and each set was produced four to six times. Area (mm^2) of the VP gap was calculated using the orifice equation (Table S4) [36, 37]. Orifice area values < 5 mm^2 are considered adequate, values between 5 and 19 mm^2 are considered marginal, and values ≥ 20 mm^2 are considered inadequate. The Palatal Closure Efficiency Index (PaCE) was also calculated for repetitions of /pi/ and /mi/ using the formula: PaCE = 100 − ((/p/ area ÷ /m/ area)*100) [35]. It is hypothesised that

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patients with VP gaps ≥ 5 mm^2 preoperatively will be more likely to exhibit VP dysfunction post-operatively due to enlargement of the VP gap from maxillary surgery.

2.6 | Perceptual Evaluation

Perceptual speech analyses were adapted from Vallino and Tompson [38]. Patients were evaluated by two SLP for auditory and visual distortions of the sounds /sɑ/, /si/, /sɪsi/ (‘sissy’), /tʌ/, /ti/, /ʃɑp/ (‘shop’), and /ʃuz/ (‘shoes’) as described in Lathrop-Marshall et al. [12] (Table 2). The evaluations were then combined into a single column for each sound. Each sound was then classified as either ‘normal’ or ‘distorted’ based on the agreement between the two raters. Points of difference were resolved with a consensus discussion between the SLPs.

2.7 | Self-Reported Outcomes

Patient-reported outcomes are important for evaluating whether treatments address patients’ motivations for seeking care [39]. Thus, we asked Class III patients with and without CLP about their concerns before orthognathic surgery and their perceptions of their speech following the procedure using questionnaires. Survey questions were sourced from existing UNC DFD patient forms and subsequently refined by the study team. A survey expert from the UNC Odum Institute for Research in Social Science evaluated the questions, which were subsequently pretested for clarity and revised (pretesters: two orthodontic residents, four undergraduates) before use during study visits (Figure 2, Table S5). All surgical patients were asked, ‘Do you perceive a change in these factors following your jaw surgery? If so, was the change positive, negative, or no change?’ Patients were asked about their speech, dental aesthetics, joint function, facial aesthetics, ease of eating and chewing, self-confidence and overall happiness.

3 | Results

This study included an examination of five Class III patients with CLP, five Class III patients without CLP and five Class I controls. Demographics and occlusal findings are in Table 1 and Table S1, respectively. All patients with CLP had a history of lip and palate repair in childhood, as well as previous speech therapy.

Dorsum Excursion Index values for all cohorts indicate a systematic and expected ranking of consonants, where /k/ has the highest Dorsum Excursion Index, followed by /ʃ/ (‘sh’), and then /s/ and /t/ (Tables S6 and S7). This is expected based on the different places of lingual articulation of /k/, /ʃ/, and /s/. Predicted Dorsum Excursion Index values for each consonant and each group are shown in Figure 3A,B along with an example of /k/ articulations before front vowels by CLP1 with normal velar closure (red) and with a common compensatory articulatory movement (blue, elevated tongue blade with a low Dorsum Excursion Index value) in Figure 4. Dorsum Excursion Index for consonants /s/, /t/, and /ʃ/ was all significantly different from the reference level, defined as /k/ before back vowels in controls (Table S7). This reference level was selected because we anticipate a large difference in /k/ articulation for the CLP cohort relative to controls, particularly before front vowels. For controls, the difference in Dorsum Excursion Index for consonants before

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front vowels (/æ/ as in ‘cap’, /i/ as in ‘key’) was not significant compared to consonants before back vowels (/ɑ/ as in ‘cop’, /u/ as in ‘coo’).

Class I control speakers had higher Dorsum Excursion Index values for /k/ than the CLP cohort before and after surgery. Patients with CLP had significantly lower Dorsum Excursion Index for /k/ before front vowels ($p = 0.0099$) relative to controls before and after surgery (Table S7). However, Class III patients without CLP did not show lower Dorsum Excursion Index values for /k/ before front vowels before or after surgery, unlike patients with CLP, indicating that this lowering of Dorsum Excursion Index values is not a feature of Class III malocclusion but is related to repaired CLP. Results from pairwise comparisons confirm that /k/ is significantly different before front vowels than back vowels for the CLP cohort before and after surgery ($p < 0.001$), unlike controls and Class III patients without CLP. Although this distinction is expected in non-disordered English speakers, the elevation of the tongue blade during /k/ productions is unique to the CLP cohort. The difference in Dorsum Excursion Index between speakers with CLP and controls for /k/ was greater before front vowels than back vowels, which was not the case for Class III patients without CLP who remained in line with controls before and after surgery. Dorsum Excursion Index values for /k/ before back vowels appeared to decrease post-operatively for the CLP cohort, whereas /k/ before front vowels appeared unchanged after surgery, yet lower than controls.

Differences in Dorsum Excursion Index were also observed for /s/. As with /k/, pre-op CLP productions of /s/ before back vowels were close to controls but significantly different and decreased overall post-operatively, whereas /s/ before front vowels remained lower compared to controls. Differences in /s/ Dorsum Excursion Index for CLP and control speakers are less extreme compared to differences in /k/. Class III speakers without CLP do not show this same trend, having similar Dorsum Excursion Index values for /s/ at pre-op and post-op time points relative to Class I controls. Meanwhile, Dorsum Excursion Index for /ʃ/ for the CLP cohort was in line with Dorsum Excursion Index for /s/ for Class I controls, before both front and back vowels. Class III patients without CLP did not show this pattern, having a stronger distinction between /s/ and /ʃ/ in both vowel contexts.

Figure 3C,D shows the average TCPI across groups, consonants, and phonological contexts. Few significant differences regarding TCPI were found using the mixed-effects linear regression model, but several interesting trends were noted in this small, likely underpowered cohort (Tables S6 and S8). Overall, TCPI was lower for all consonants and phonological contexts for the CLP cohort than Class I controls. All consonants except /ʃ/ were significantly different from one another among controls, which is expected because these sounds have different places of articulation in the oral cavity. Class III pre-op TCPI data were in line with the CLP cohort for /k/ and /s/ then increased to be in line with Class I controls after surgery. TCPI for /ʃ/ and /t/ before back vowels was significantly different from controls ($p = 0.019$) for both time points in patients with CLP. TCPI for /k/ before /i/ was lower for the CLP cohort compared to controls but increased in the direction of controls after surgery. There was notable variation in the TCPI results for other consonants, which was anticipated based on prior reports [22, 24]. The effect of following vowel type on TCPI across consonants was not significant. It is possible that measuring the middle frame of the production of fricatives eliminated any coarticulatory effect on TCPI.

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Pressure flow data were available for three patients with CLP (CLP2, CLP4, and CLP5) (Table S4). Before surgery, all three patients had adequate orifice areas for /pi/. For 'hamper,' one patient (CLP4) had adequate closure, and the other two patients had marginal levels of VP closure preoperatively. Orifice area for /pi/ before surgery was lower overall compared to 'hamper', which is expected due to the preceding nasal consonant before the /p/. After surgery, all three patients showed adverse closure for 'hamper,' and two out of three exhibited inadequate closure for 'pi' (CLP2, CLP4). Interestingly, the two patients who had the most marked increase in /pi/ orifice area (CLP2: pre-op 1.9 mm^2 to post-op 39 mm^2, CLP4: pre-op 0.3 mm^2 to post-op 27 mm^2) were also evaluated as hypernasal by SLPs and had self-reported negative speech outcomes after surgery (Table 2; Tables S4 and S5). PaCE data for CLP2 and CLP4 also suggest increased VPI, with values changing from 93% pre-op to 0% post-op in CLP2 and 99% pre-op to 21% post-op in CLP4 for /pi/, respectively.

Self-reported data (Figure 2; Table S5) for the five patients with CLP and five Class III without CLP indicate heterogeneity in speech outcomes consistent with SLP evaluations. Although some patients with CLP reported speech improvement (40%), others indicated no change (20%) or negative changes in their speech (40%). Class III patients without CLP reported similar results in improvement (40% improved), whereas reporting less negative changes in their speech (20%). All surgical patients reported improvements in their dental aesthetics, and most indicated positive change in their facial appearance (80%) and happiness (60% CLP, 80% Class III) post-operatively. Class III patients without CLP reported more positive changes across more functional domains than patients with CLP (Figure 2). Some patients showed improvement in one functional domain but a decline in another after surgery (Table S5).

Regarding blinded SLP perceptual evaluations, particular sounds and nasality either remained the same or worsened after surgery in the five patients with CLP (Figure 5, Table 2). In contrast, Class III patients without CLP generally remained the same or improved after surgery. There were no changes in /sɑ/, /si/ or /sisi/ ('sissy') for any patient with or without CLP. SLP evaluation found three out of five patients with CLP demonstrated an increase in hyper- or hypo-nasality, consistent with pressure flow measures; two of these patients also self-reported a negative change in speech (Table 2; Tables S4 and S5). There was an increase in distortions in /tɑ/ for three patients (CLP2, 4, 5), in /ti/ for two patients (CLP2, 4), which is possibly related to insufficient intraoral pressure during the production of /t/ due to increased VP gap size after surgery. Class III patients without CLP presented normal product ions of /t/. One patient with CLP also showed increased distortion for /ʃ/ (CLP3). Perceptual evaluations indicate speech either remains the same or worsens after orthognathic surgery in Class III patients with CLP, likely related to worsening nasality, whereas speech remains the same or improves after surgery for Class III patients without CLP.

4 | Discussion

This study presented longitudinal calculations of two tongue shape indices in patients with CLP who underwent orthognathic surgery to correct their Class III malocclusions in comparison to Class I well-proportioned individuals, as well as Class III patients without CLP who underwent similar orthognathic procedures. Dorsum Excursion Index

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and pressure flow data suggest increased compensatory articulations and VPI, respectively, after surgery for patients with CLP. Dorsum Excursion Index for /k/ before back vowels decreased post-op, whereas Dorsum Excursion Index for /k/ before front vowels remained significantly lower than Class I controls and Class III without CLP even before surgery, indicating a unique feature exclusive to patients with CLP. Furthermore, decreases in Dorsum Excursion Index for /s/ post-op were found before both front and back vowels. Lower Dorsum Excursion Index values for /s/ compared to controls aligned with the blinded SLP evaluations—the only patient who was not evaluated as having /s/ distortions (CLP3) had higher Dorsum Excursion Index values, which is more aligned with control subjects. When /s/ is articulated too far forward in the vocal tract (i.e., dental, interdental), Dorsum Excursion Index will be lower compared to the typical alveolar constriction associated with /s/. The decrease in /t/ Dorsum Excursion Index after surgery aligns with blinded SLP evaluations, which found increased distortion in the /t/ sound, potentially related to inadequate oral pressure and VPI. Orifice area and PaCE index data both suggest increased VPI for all three patients with CLP measured, corroborated by SLP perceptual evaluation. In contrast, Class III patients without CLP showed a reverse trend in /t/. Patients who present with adequate oral pressure and VP function had higher pre-op Dorsum Excursion Index values than patients with CLP, and post-op values increased approaching the Class I controls. This aligns with SLP evaluations which rated all /t/ instances as normal for Class III patients without CLP.

Class I controls and Class III speakers without CLP showed the same Dorsum Excursion Index patterns as found in Zharkova [22] and Cleland et al. [23] where /k/ shows higher values than /ʃ/, and /ʃ/ shows higher values than /s/ and /t/. This indicates that Dorsum Excursion Index effectively measures articulatory variations in different consonants, regardless of occlusion type or the presence and type of speech distortions. The CLP cohort showed consistently lower Dorsum Excursion Index values and decreased away from control and Class III without CLP averages after surgery. Although this does not align with the expectations set by Zharkova [22], it does align with Cleland's [23] argument that excessive dorsum raising is not as common in CLP populations as originally thought, and that Dorsum Excursion Index can investigate differences in articulation across groups in other ways. For example, lower Dorsum Excursion Index values for /s/ can be interpreted as more anterior constrictions in the vocal tract, leading to dental or interdental realisations of the sound. Dorsum Excursion Index for /ʃ/ among patients with CLP was within the range of Dorsum Excursion Index values for /s/ for Class I controls and Class III patients without CLP. This is supported by the lower TCPI averages and the consistent /ʃ/ distortion noted in the blinded SLP evaluations.

Dorsum Excursion Index results for /k/ align with the findings in Cleland et al. [23] with controls having consistently higher Dorsum Excursion Index than patients with CLP. However, there was variation between speakers within both the CLP and control cohorts. For example, CLP3 was evaluated as sounding normal for both time points and showed minimal change in Dorsum Excursion Index after surgery, with mean Dorsum Excursion Index values for each phone aligning with controls. Meanwhile, CLP1 was evaluated as having /s/ distortions and hyponasality before and after surgery. Although CLP3 had /k/ Dorsum Excursion Index values in line with the control cohort, CLP1 exhibited extremely

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low Dorsum Excursion Index values for /k/ before front vowels. The articulation for /k/ before front vowels for CLP1 involved contact between the tongue surface and the palate as well as tongue blade elevation, which led to productions of the velar stop /k/ that have a distinct, high frequency noise (hissing sound) after the release burst (as shown in Figure 4), a type of compensatory articulation described by Bressman et al. [20] Although CLP1's mean Dorsum Excursion Index for /k/ before back vowels was in line with controls (T0: 0.582; T1: 0.653), /k/ before front vowels consistently showed the pattern of palatal constriction and tongue blade elevation, with a mean Dorsum Excursion Index of 0.385 and 0.473 for each timepoint.

Trends involving TCPI were less distinct than Dorsum Excursion Index, with some inter- and intra-speaker variation for all consonants. However, TCPI was consistently lower for the CLP cohort compared to controls. Class III patients without CLP before surgery also had consistently lower TCPI than controls, but after surgery their TCPI increased to be in line with controls. According to Zharkova et al. [22], TCPI will be lower for speakers with CLP compared to non-disordered speech due to a 'retracted tongue position'. More data were needed to understand the relationship between Dorsum Excursion Index and TCPI across consonants and phonological contexts, both for typically developing speakers and speakers with CLP. Notably, pre-op Class III TCPI data were consistent with the CLP cohort for /k/ and /s/ but then increased to be in line with Class I controls after surgery. This suggests one mechanism by which speech changes after surgery in Class III patients without CLP. Additionally, comparisons in articulatory changes after orthognathic surgery between Class III patients with and without CLP with larger cohorts will shed further light on what changes in articulation after surgery are unique to patients with CLP.

VP orifice area and PaCE index measurements from pressure flow data are indicative of increased VPI and align with the blinded SLP evaluations surrounding nasality (Figure 5A,B). Increased orifice area and decreased PaCE after surgery were expected due to VP gap enlargement after maxillary advancement. With the increase in VPI comes an apparent increase in audible NAE during the production of certain consonants (/k/, /ʃ/, /t/). Although audible NAE was not quantitatively evaluated, an overall increase in symptoms (flutter and/or turbulence) was noticed post-operatively and should be examined with more detail in future research. Overall, it appears that speech perceptually stays the same or worsens after maxillary advancement in Class III patients with repaired CLP, likely related to changes in VPI and nasality. As a result, if a Class III patient with repaired CLP is pursuing orthognathic surgery for speech improvement, they may be better suited to a different surgical or therapeutic approach. Alternatively, if patients are primarily interested in facial and dental enhancements and are less concerned with speech and hypernasality, then orthognathic surgery may be the preferred treatment route. Class III patients' (without CLP) speech perceptually stayed the same or improved after surgery and had no change in nasality and VPI, suggesting surgery may improve their speech.

Although the imaging protocol for patients with CLP was the same as for controls and Class III without CLP, imaging of the palate was more challenging. During swallowing, the tongue approximates the surface of the palate as it pushes the bolus being swallowed backward. In ultrasound imaging of swallowing, it is possible to mistake the air immediately above the

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floor of the nasal cavity as the surface of the palate in the oral cavity. This was a consistent issue with the examined CLP data. However, articulatory information in ultrasound imaging for consonants /k/ and /t/ allows one to see the point of closure at the velum and alveolar ridge, respectively. This information can be combined to achieve a more accurate palatal trace.

Study limitations include a small, underpowered sample size, which could include variable results that should be interpreted cautiously. Hence, results should be interpreted individually as opposed to generalised conclusions. Additionally, similar linguistics studies have had comparable sample sizes [22, 24]. There were also racial and ethnic distribution differences between the control, Class III and CLP groups, with the Class I control group represented by a homogenous white cohort compared to a heterogeneous Asian/Hispanic/Latino CLP group and Caucasian/Hispanic/Latino Class III group. The Class III without CLP group had greater ethnic diversity than the Class I controls. A future study will include a larger, adequately powered cohort with more balanced racial and ethnic distributions to reduce sample variations. We also calculated Dorsum Excursion Index and TCPI for one ultrasound frame for each token; future research should investigate trajectories of Dorsum Excursion Index and TCPI throughout the entire production of consonants. Additionally, we could not account for different palatoplasty techniques employed during palate repair for patients with CLP, as those procedures were conducted over a decade prior to patients' enrolment in our study; palatal procedures commonly completed at our centre include two-flap Bardach palatoplasty (for two of the patients with CLP). There was likely variability in which procedures were performed as three patients transferred care to our team after their palatoplasty. Furthermore, post-operative data for this study was collected 2 months after surgery, when some post-operative swelling can still be present [40]. Perhaps, this is resulting in hyponasality in some patients. Future follow-ups will collect data 1 year after surgery, eliminating any influence of post-surgical swelling. Additional data were needed to understand the relationship between Dorsum Excursion Index, TCPI, and cephalometric changes after orthognathic surgery. Future studies should compare Dorsum Excursion Index and TCPI for tokens with and without audible NAE to explore the correlation between VPI and NAE in patients who underwent orthognathic surgery to correct skeletal malocclusions.

As long-term care providers of patients with CLP, orthodontists would benefit from familiarity with speech sciences and the mechanisms by which speech is affected in this high-need patient cohort. This will assist orthodontists in appropriately diagnosing and referring patients for speech evaluations and understanding relevant literature for interdisciplinary management of patients with CLP who present with severe malocclusions and speech disorders.

5 | Conclusion

- Dorsum Excursion Index and TCPI are useful indices that can be used to understand how articulatory tongue shape and movement change across different participants and time points, providing helpful information about the effects of orthognathic surgery on speech function.

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- Patients with CLP present a pattern of more anterior tongue gestures after surgery (i.e., lower Dorsum Excursion Index and TCPI) when compared to Class I controls and Class III without CLP.
- Dorsum Excursion Index and pressure flow data suggest increased compensatory articulations and VPI following orthognathic surgery for Class III patients with CLP.
- In Class III patients with repaired CLP, speech perceptually stays the same or worsens after maxillary advancement likely due to increased VPI and altered nasality.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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Data Availability Statement

The data that supports the findings of this study are available in the Supporting Information of this article.

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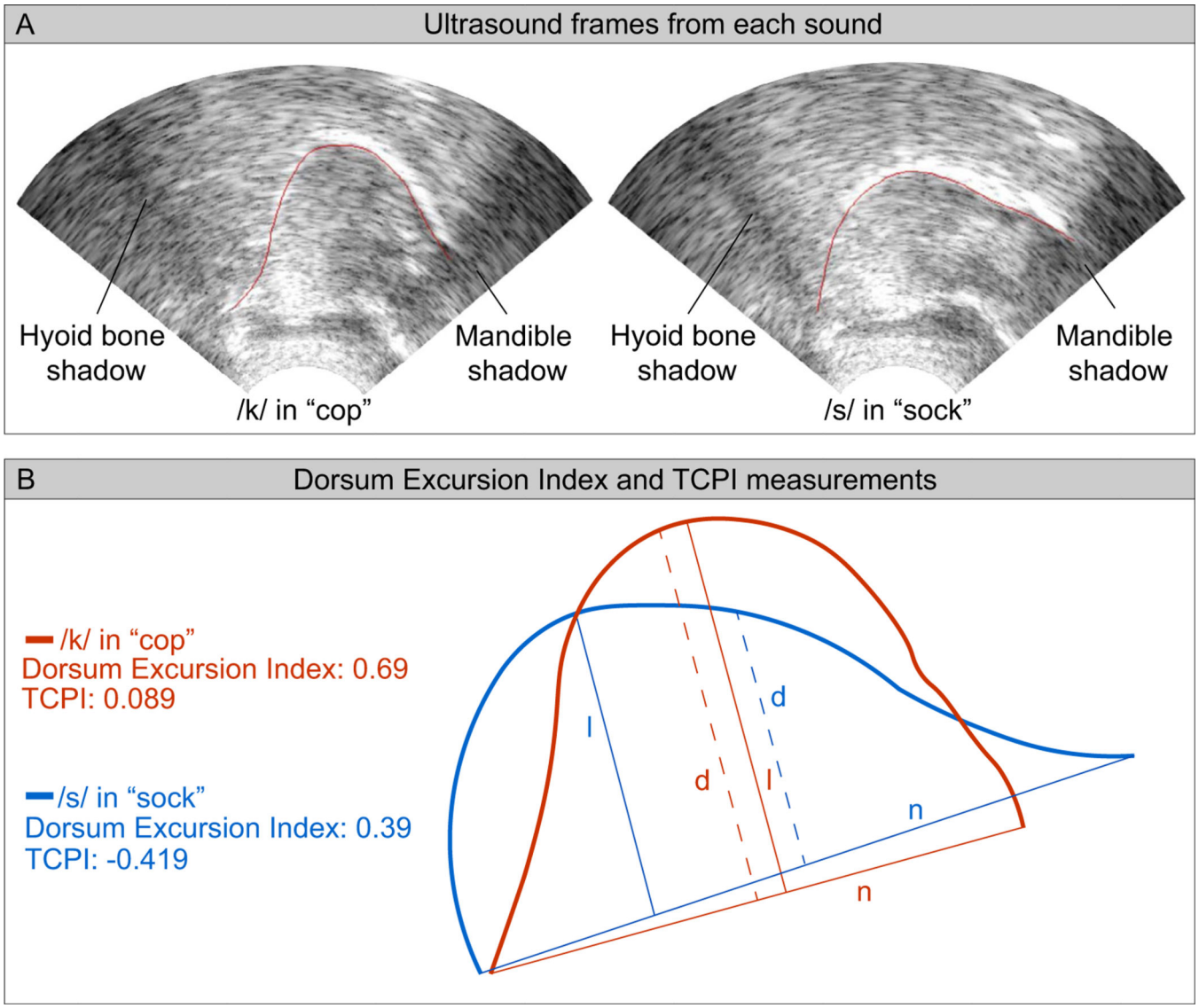


FIGURE 1 |.

(A) Dorsum Excursion Index and TCPI Measurements. Example ultrasound frames of /k/ in ‘cop’ (left) and /s/ in ‘sock’ (right) spoken by Control3. Red lines represent traces of the tongue surface, spanning from the tongue tip (right) to the tongue root (left), traced by DeepLabCut. Dark regions on the left and right sides of the ultrasound image are the hyoid bone shadow and mandible shadow, respectively. (B) Dorsum Excursion Index for /k/ in ‘cop’ (red) and /s/ in ‘sock’ (blue) for CLP3. Dorsum Excursion Index is a ratio calculated by dividing the length of line *d* by the length of line *n*. Higher Dorsum Excursion Index values are associated with greater involvement of the tongue dorsum. TCPI is calculated by dividing the distance between *d* and *l* by half of *n*.

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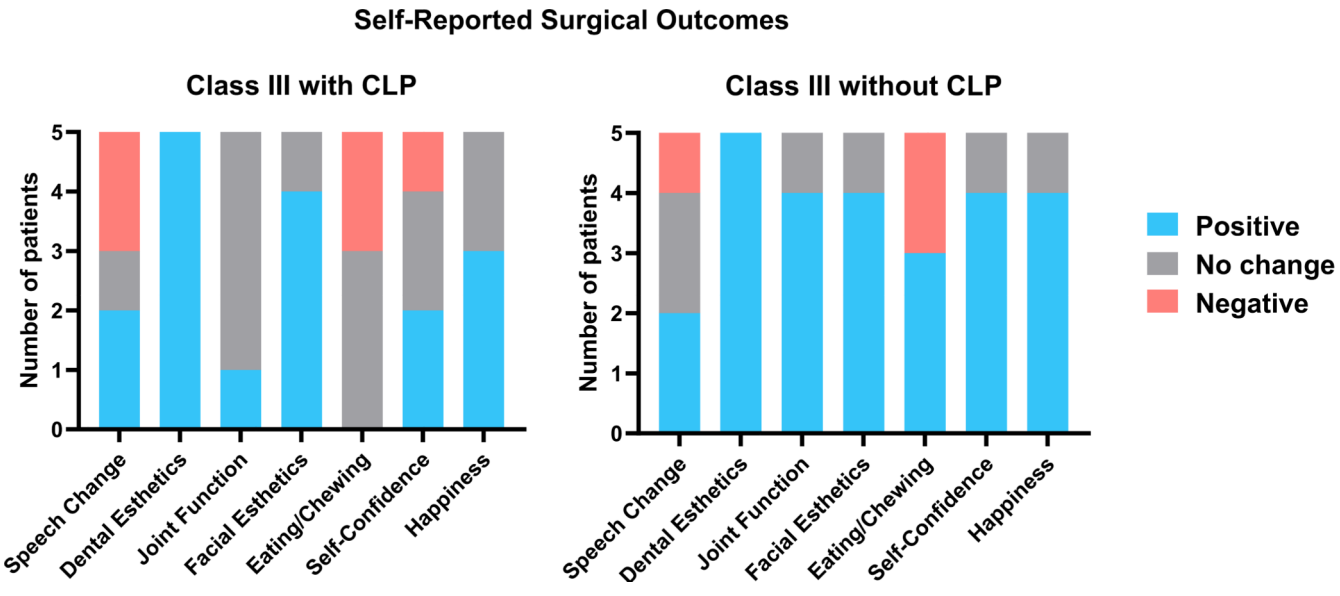


FIGURE 2 |.

Self-reported surgical outcomes from all surgical patients (Class III with and without cleft lip and palate [CLP]). Patients reported changes as positive, negative, or as unchanged post-orthognathic surgery. Patients reporting positive change are shown in blue, no change in grey, and negative change in salmon. Data found in Table S5.

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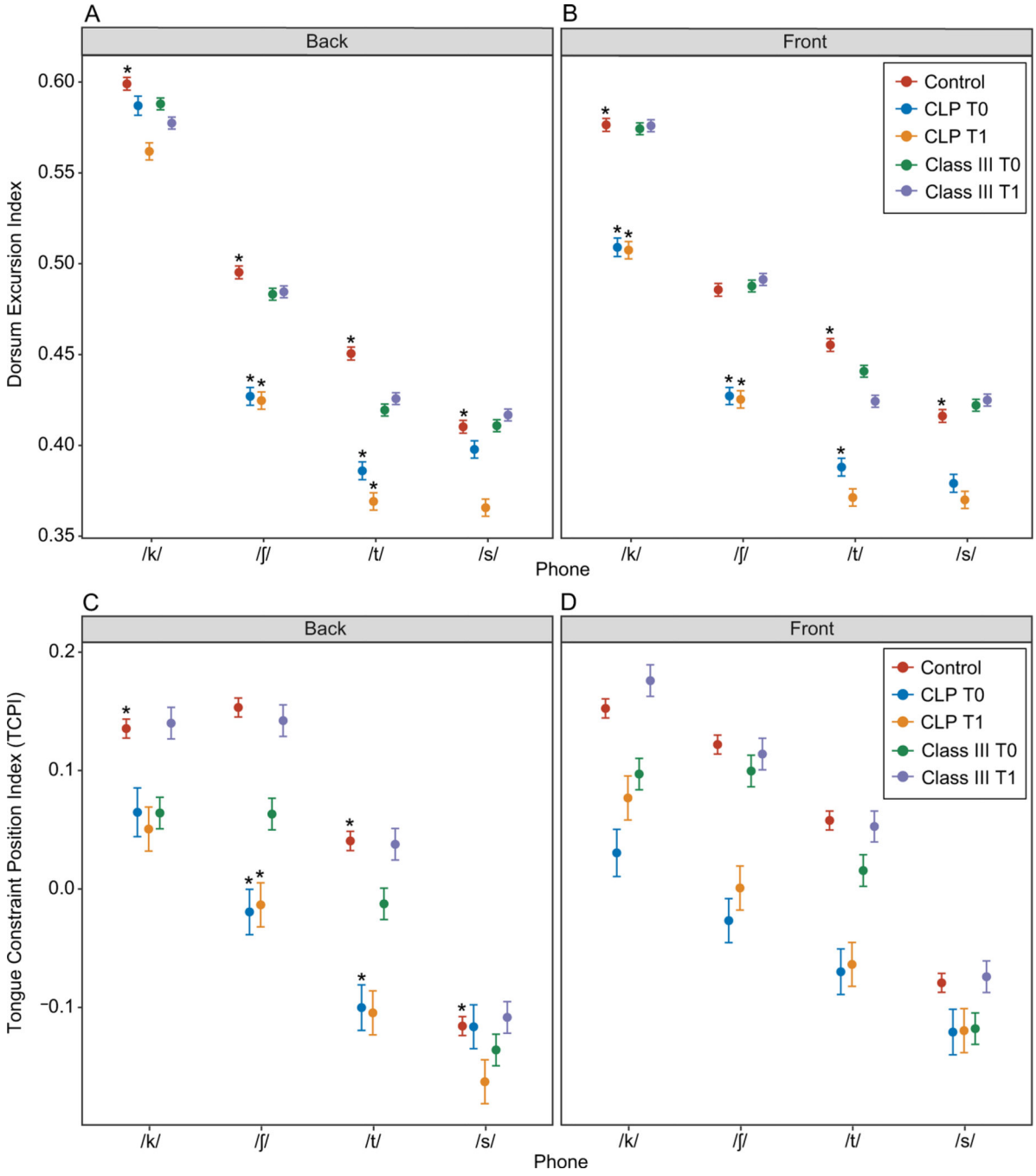


FIGURE 3 |.

(A and B) Predicted Dorsum Excursion Index values for consonants before back vowels /ɑ/ and /u/ (upper left) and front vowels /i/ and /æ/ (upper right) based on the linear mixed-effects model. Each dot represents the fitted values and standard error for each group (red: Class I control; blue: Pre-op CLP; yellow: Post-op CLP; green: Class III without CLP Pre-op; purple: Class III without CLP Post-op). (C and D) Predicted TCPI values for consonants before back vowels (bottom left) and front vowels (bottom right). Asterisks represent significant differences relative to reference levels ($p < 0.05$). Reference levels

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were defined as Dorsum Excursion Index for /k/ before back vowels. This reference level was selected because we anticipate a large difference in /k/ articulation for the CLP cohort relative to controls, particularly before front vowels. Bars represent standard error. Data found in Tables S6–S8.

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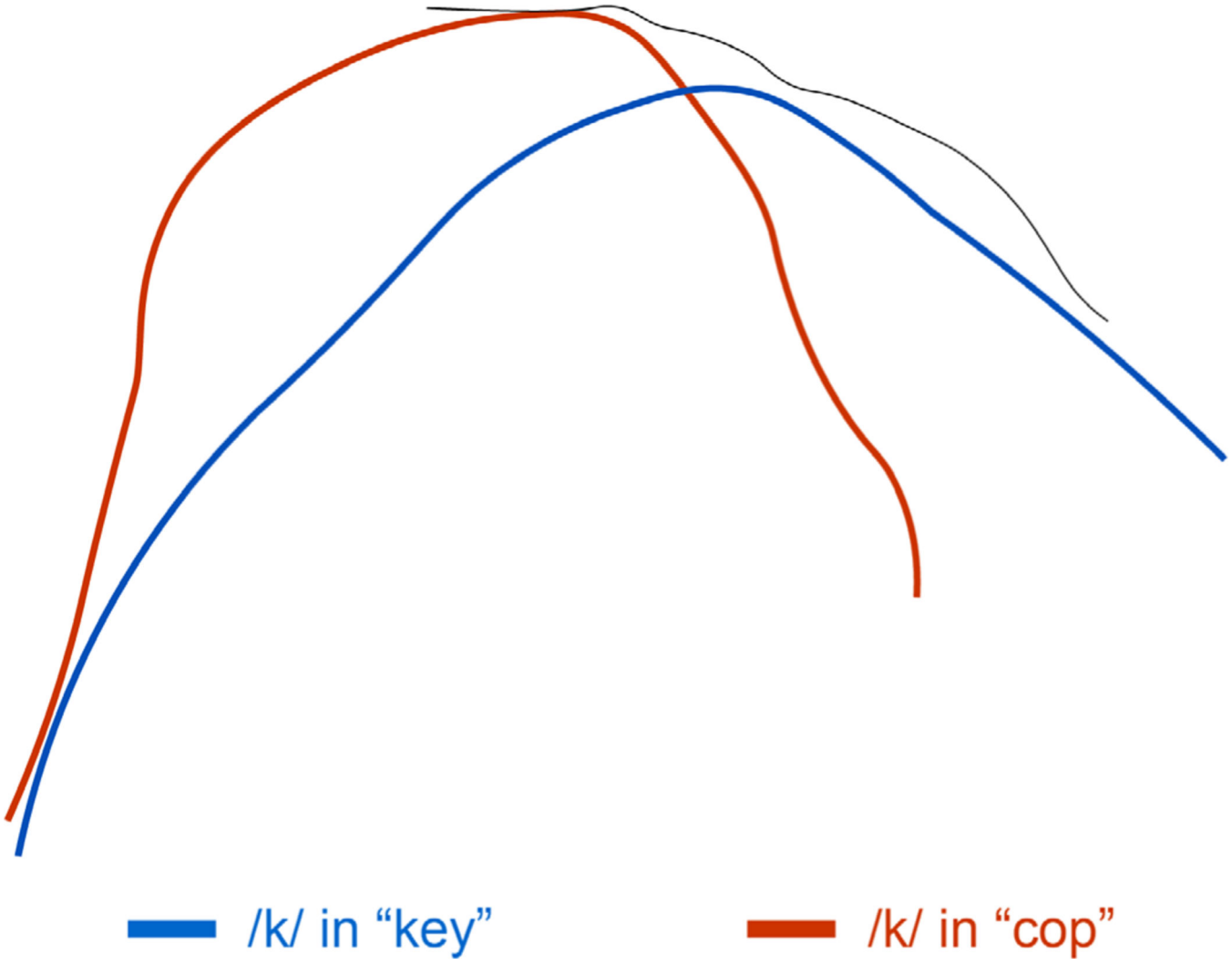


FIGURE 4 |.

CLP1 produces /kɑ/ (red) with a typical velar closure (Dorsum Excursion Index = 0.681, TCPI = 0.266), but produces /ki/ (blue) with an elevated tongue blade (Dorsum Excursion Index = 0.382, TCPI = 0.102), a common compensatory articulation within CLP speech. Articulations of /k/ with a raised tongue blade such as the blue spline above tend to be accompanied by atypical, high-frequency noise (hissing sound). The black spline represents the palate trace. CLP, cleft lip and palate.

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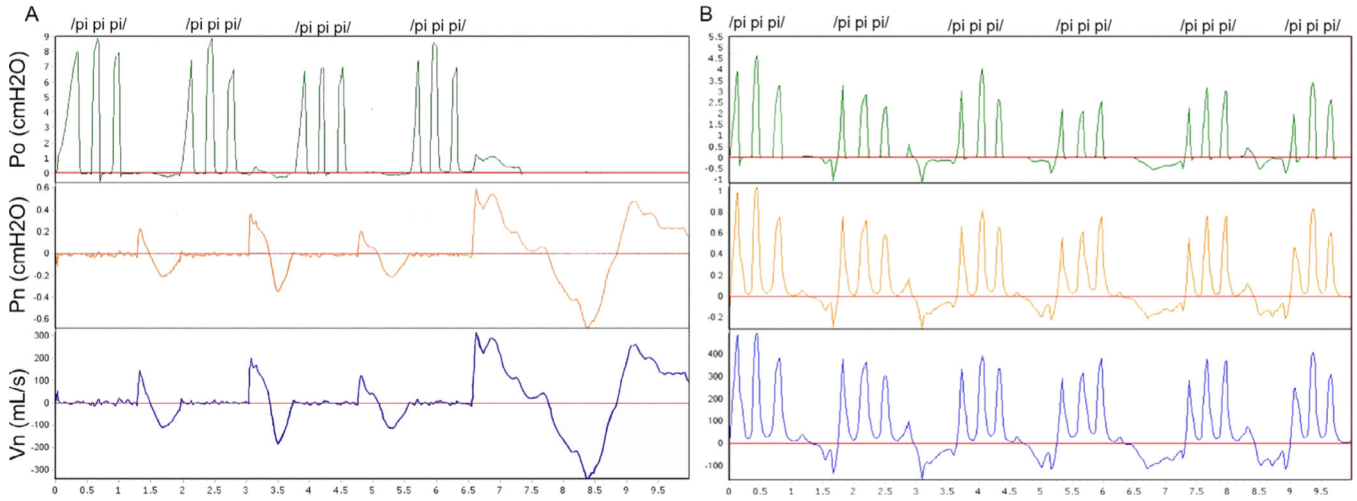


FIGURE 5 |.

(A) Pre-surgery pressure flow data for a participant with adequate velopharyngeal closure during four repetitions of /pi pi pi/. There is a lack of nasal airflow during the syllables, indicating essentially complete velopharyngeal closure; estimated area was < 1 mm^2. (B) Post-surgery pressure flow data for the same participant during six repetitions of /pi pi pi/. There is consistent nasal airflow during each syllable indicating inadequate velopharyngeal closure or velopharyngeal insufficiency; estimated area was 27 mm^2. Pn, nasal air pressure; Po, oral air pressure; Vn, nasal airflow.

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TABLE 1 |

Participant demographics.

	Patients with CLP (*n* = 5)	Controls (*n* = 5)	Class III without CLP (*n* = 5)
Age	Mean: 19 years ± 2 years Range: 16–21 years	Mean: 18 years ± 3 years Range: 15–22 years	Mean: 20 ± 3 years Range: 18–25 years
Sex	2 females; 3 males	2 females; 3 males	3 females; 2 males
Race	3 Asian; 2 Caucasian	5 Caucasian	3 Caucasian; 2 Hispanic/Latino
Ethnicity	1 Hispanic/Latino	0 Hispanic/Latino	2 Hispanic/Latino
Occlusion	5 Class III; 1 Anterior Open Bite (CLP2)	5 Class I	5 Class III
Surgeries	CLP1 & CLP2 received Lefort I 3-piece maxillary advancement surgeries, CLP3 & CLP5 received Lefort I 1-piece maxillary advancement surgeries and CLP4 received a Lefort I 1-piece advancement with a mandibular BSSO setback	Non-surgical	2 patients received Lefort I 1-piece advancement, 3 patients received Lefort I 1-piece advancement with a mandibular BSSO setback

Abbreviations: BSSO, Bilateral Sagittal Split Osteotomy; CLP, cleft lip and palate.

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TABLE 2 |

Perceptual evaluation by speech language pathologists.

Patient	/sɑ/	/si/	'sissy'	/ʃ/	/tɑ/	/ti/	Nasality
CLP1							
T0	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Hypo (mild)
T1	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Hypo
CLP2							
T0	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Normal
T1	Distorted	Distorted	Distorted	Distorted	Distorted	Distorted	Hyper
CLP3							
T0	Normal	Normal	Normal	Normal	Normal	Normal	Normal
T1	Normal	Normal	Normal	Distorted	Normal	Normal	Normal
CLP4							
T0	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Hypo
T1	Distorted	Distorted	Distorted	Distorted	Distorted	Distorted	Hyper
CLP5							
T0	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Hypo
T1	Distorted	Distorted	Distorted	Distorted	Distorted	Normal	Hypo (mild)
Class III 1							
T0	Distorted	Distorted	Distorted	Normal	Normal	Normal	Normal
T1	Normal	Normal	Normal	Normal	Normal	Normal	Normal
Class III 2							
T0	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Normal
T1	Distorted	Normal	Distorted	Normal	Normal	Normal	Normal
Class III 3							
T0	Normal	Normal	Normal	Distorted	Normal	Normal	Normal
T1	Distorted	Distorted	Distorted	Normal	Normal	Normal	Normal
Class III 4							
T0	Distorted	Distorted	Distorted	Normal	Normal	Normal	Normal
T1	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Normal
Class III 5							
T0	Distorted	Distorted	Distorted	Distorted	Normal	Normal	Normal

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Patient	/sɑ/	/si/	‘sissy’	/ʃ/	/tɑ/	/ti/	Nasality
T1	Distorted	Distorted	Distorted	Normal	Normal	Normal	Normal

Note: Sounds were evaluated as ‘normal’ or ‘distorted’. Distortions could be any of the following: Interdental, dental, backed, lateralized, or whistle. Overall nasality (normal, hypernasal (hyper), hyponasal (hypo)) was evaluated as well.

Abbreviation: CLP, cleft lip and palate.

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