Speech Assessments in Hypoplastic Cleft Maxilla Treated by Anterior Segmental Distraction Osteogenesis

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KEYWORDS

Cleft lip and palate, Distraction osteogenesis, Maxillary hypoplasia, Speech articulation test

ABSTRACT

Introduction: Cleft lip and palate (CLP) affect the craniofacial region, with significant implications for speech, hearing, feeding, and facial aesthetics. This study aimed to evaluate the impact of anterior maxillary segmental distraction osteogenesis (AMSDO) using an internal distractor on speech outcomes in CLP patients, assessed using the Pittsburgh Weighted Speech Scale (PWSS).

Methodology: A single-center prospective study was conducted between March 2022 and April 2024. Twelve CLP patients with maxillary hypoplasia, aged 17.8 to 32.6 years, were included. Purposive sampling was done to include patients with complete unilateral or bilateral CLP who exhibited Class III skeletal relationships with reverse overjet greater than 7 mm. Speech assessments were conducted preoperatively (1-3 months before surgery) and postoperatively (5-7 months after distraction). Speech samples were recorded and evaluated by a licensed speech-language pathologist using the PWSS. Statistical analysis was performed using paired t-tests and Chi-square tests.

Results: Significant improvements were observed in the total PWSS score ($p = 0.03$), nasality ($p < 0.001$), phonation ($p < 0.001$), and articulation errors ($p = 0.004$) postoperatively. No significant changes were seen in nasal emission and facial grimace. No correlation was found between the amount of distraction and speech outcomes. Unilateral cleft patients showed better outcomes than bilateral cleft patients, though not statistically significant.

Conclusion: AMSDO significantly improves speech outcomes in CLP patients with maxillary hypoplasia. The PWSS is a reliable tool for assessing these changes. Further research with larger samples and longer follow-ups is warranted to confirm these findings.

1. Introduction

The most frequent primary Cleft lip and palate (CLP) is among the most prevalent congenital disorders affecting the craniofacial region, with significant implications for speech, hearing, feeding, and overall facial aesthetics [1,2]. Speech disorders associated with CLP are particularly concerning as they can hinder social integration and overall quality of life [3,4]. CLP patients often experience a range of speech disorders, including excessive nasality, audible air coming through the nose, and incorrect articulation [5]. The cause of these speech problems is velopharyngeal insufficiency (VPI), a condition in which air escapes through the nose during speech due to insufficient closure of the soft palate against the posterior pharyngeal wall [6].

Speech difficulties significantly impact the quality of life for both young and adult CLP patients [6]. Young patients often face academic challenges, social isolation, bullying, and emotional distress due to poor speech intelligibility, affecting their self-esteem and mental health [7,8]. Adults with CLP experience persistent speech problems that limit professional opportunities, cause social anxiety, and lead to isolation, affecting personal relationships and contributing to long-term psychological disorders like depression and anxiety [9].

Primary surgical treatments aim to restore anatomical integrity and functionality, yet secondary interventions are often necessary to address residual deformities and functional impairments [10,11]. Anterior maxillary segmental distraction osteogenesis (AMSDO) is one such intervention that corrects maxillary hypoplasia, a frequent complication in CLP patients [12]. AMSDO not only enhances facial aesthetics by improving midface projection but also positively affects dental occlusion, which can have a secondary benefit for speech function by providing a more favourable oral environment for articulation [13]. However, the direct impact of AMSDO on speech outcomes remains a subject of ongoing research.

AMSDO can be carried out by either internal or external distractors. The skull serves as an external stabilization point for distraction in the rigid external distraction system. Despite being widely used due to its simplicity of use, removal, and adjustment, the device can be difficult to use, causing external scars, infections, injuries to nerves or tooth buds, and being socially and psychologically unacceptable to the mature patient [14]. Internal distractors are designed in a subtle way, which increases their social convenience and acceptance [15].

The evaluation of speech disorders in CLP patients is complex and multifaceted, involving perceptual, instrumental, and sometimes, acoustic analyses [16]. Several rating scales such as Great Ormond Street Speech Assessment, Cleft Audit Protocol for Speech-Augmented (CAPS-A), Americleft Speech Protocol, Universal Parameters for Reporting Speech Outcomes, and Pittsburg Weighted Speech Scale (PWSS) are used to evaluate speech in CLP [17,18,19,20,21]. PWSS is a commonly used perceptual tool that assesses speech characteristics, including resonance, nasal air emission, and articulation. The PWSS assigns weights to different speech parameters, providing a comprehensive measure of speech proficiency in CLP patients [21].

Previous literature has compared speech outcomes between Le Fort l osteotomy and Le Fort l distraction osteogenesis by several methods such as nasoendoscopy, nasometry, and perceptual speech assessments by sleep language pathologists [22,23,24]. The perceptual speech assessments in a study by Chung et al. used PWSS, a study by Rao Janardhanan et al. employed Universal Parameters for Reporting Speech Outcomes while most other studies developed their own assessment scale for rating speech outcomes [23,25,26,27]. However, literature evaluating speech outcomes following AMSDO using an internal distractor is limited with no standardised protocol followed [12,14,25]. The present article aimed to delve into the effects of AMSDO using an internal distractor on speech outcomes in CLP patients, evaluated using the PWSS.

2. Methodology

2.1 Study Setting

This is a single-centered prospective study conducted at ********** between March 2022 to April 2024. An institutional review board ethical clearance was obtained before starting this study (IHEC/SDC/ORTHO-2105/24/158). The study was performed in compliance with the institutional guidelines and written informed consent from all the participants was obtained.

2.2 Study Sample

Calculation of the sample size was performed using the G Power software (version 3.0.10). Sample size calculation was done based on a study by Lin et al. [12]. This study included 12 patients who were admitted at Saveetha Dental College, Saveetha Institute of Medical and Technical Sciences, from March 2022 to April 2024 and had maxillary hypoplasia related to cleft lip and palate. The participants consisted of 8 male cases and 4 female cases aged between 17.8 to 32.6 years. There were two subjects with a complete bilateral cleft and ten with a complete unilateral cleft. All patients had been previously treated with cheiloplasty between the ages of 3 to 6 months, followed by palate repair surgery between 12-18 months of age and secondary alveolar bone grafting in adolescence. This prevents bias in pre operative speech quality of the selected participants. Based on cephalometric analysis, the patients were found to have a hypoplastic maxilla and a Class III skeletal relationship with a reverse overjet greater than 7 mm. Purposive sampling which was non-probabilistic in nature was utilized to select participants who met the inclusion criteria.

2.3 Perceptual speech assessments

Speech samples of the patients were recorded in a soundproof room using a Sennheiser Profile USB Microphone (Sennheiser Electronics SE & Co. KG, Germany) at a standard distance of 15 cm from the mouth. Every patient repeated the same standardised text samples. A thorough evaluation of nasal and non-nasal phonemes, as well as variations in resonance at different speech production levels, was made possible by the recordings, which also included syllable repetition, phrase repetition, counting, and conversational speech samples. The preoperative speech recordings were performed 1 to 3 months prior to the surgical procedure, and the post operative samples were procured between 5-7 months following the end of the active distraction phase. The postoperative samples were collected 3 months after removal of the distractor device so as to not influence the speech outcomes. A single licensed speech-language pathologist with more than 5 years of specialised experience with cleft patients carried out the PWSS perceptual speech assessments of all the patients. Video recordings of spontaneous speech samples were taken for evaluation of nasal emission and facial grimace, attributes of the PWSS. The PWSS speech scale rates the following five aspects of speech: phonation, articulation, nasal emission, facial grimace, and nasality. The cumulative score obtained by adding the assigned points can be used to evaluate the perceptual speech related to VPI. The speech evaluation was graded overall in the following manner: 0 as competent; 1-2 as competent to borderline competent; 3-6 as borderline competent to borderline incompetent; and 7 and above as incompetent (Table 1).

Table 1: Pittsburgh Weighted Speech Scale

Nasal Emission (0-3, Highest Value)		
	Right	Left
Not present	0	0
Inconsistent, visible	1	1
Consistent, visible	2	2
Nasal escape or nasals appropriate - Reduced	0	0
Nasal escape or nasals appropriate - Absent	0	0
Audible	3	3
Turbulent	3	3
Facial Grimace (0/2 if Present)		
Nasality (0-4, Highest Value)		
Normal	0	
Mild hypernasality	1	
Moderate hypernasality	2-3	
Severe hypernasality	4	
Hypo-/Hypernasality	2	
Cul de sac	2	
Hyponasality	0	
Phonation (0-3, Highest Value)		
Normal	0	
Mild hoarseness/breathiness	1	
Moderate hoarseness/breathiness	2	
Severe hoarseness/breathiness	3	
Reduced loudness	2	
Tension in system	3	
Articulation		
Normal	0	
Developmental errors	0	
Errors from other causes not related to VPI	0	
Errors related to anterior dentition	0	
Reduced intraoral pressure for sibilants	1	
Reduced intraoral pressure for other fricatives	2	
Reduced intraoral pressure for plosives	3	
Omission of fricatives or plosives	2	
Omission of fricatives or plosives plus hard glottal attack for vowels	3	
Lingual-Palatal sibilants	2	
Pharyngeal fricatives, plosives, backing, snorts, inhalations or exhalation substitutions	3	
Glottal stops	3	
Nasal substitution for pressure sounds	4	

2.4 Customised internal distractor device

Custom-made internal distractors were prefabricated on casts. Either a 11 mm or a 13 mm hyrax screw, depending on the amount of advancement required, was placed in an anteroposterior direction parallel to the sagittal plane. The hyrax screw arms were fused to the premolar and molar bands in such a way that the hyrax screw lies on the plane along which the anterior distraction is to take place. The maxillary or mandibular teeth were given acrylic capping to disocclude the dentition.

2.5 Surgical procedure

Surgery first approach was used in these patients; therefore, no pre-surgical orthodontics was performed. Anterior maxillary osteotomies were performed under general anaesthesia. Based on root angulations assessed from preoperative radiographs, vertical maxillary osteotomy cuts were either between the first molars and second premolars or between the first and second premolars. Pre-surgical orthodontics for root divergence was not required for any of the patients. Bilateral modified high level horizontal anterior maxillary osteotomy cuts were made. The surgical procedure was carried out by the same surgeon for all 12 patients. Adequate mobilization of the anterior segment was ensured and the prefabricated distraction device was cemented onto the teeth using resin modified glass ionomer luting cement (GC FujiCEM™, GC Corporation, Tokyo, Japan). After a 5-days latency period, the frequency of activation of the hyrax screw was 0.5 mm twice a day (1 mm/day). After the required amount of advancement, the distractor was sealed and left in place for a period of three months to allow for consolidation.

2.6 Statistical analysis

Statistical analysis was conducted using IBM SPSS software (version 26.0). Descriptive statistics summarised the patients' characteristics and speech outcomes. Paired t-tests compared pre- and post-operative PWSS scores. The correlation between the magnitude of maxillary distraction and the resultant speech outcomes was examined using the t test. Chi quare test was used to determine the correlation between the cleft defect type and speech changes. Intra examiner reliability was evaluated with kappa statistics. A p-value below 0.05 was deemed to be statistically significant.

3. Results

All 12 patients underwent successful AMSDO and their maxillary hypoplasia improved. There were no complications such as development of oronasal fistulas, haemorrhage, infection or necrosis in any of the patients. The distractors were intact and stable post surgery till the end of the consolidation phase. Post consolidation radiographic analysis revealed mean anterior maxillary advancement was 10.35 ± 1.59 mm as measured between the proximal surfaces of the roots of the teeth adjacent to the distracted bone. On analysis of speech, no patient exhibited worsening of their speech outcomes. The mean duration of consolidation period for the patients was 87 (64 - 96) days. The average duration for speech evaluation post-surgery was 6.04 ± 0.95 months. The intra-rater reliability examined using kappa statistics revealed a value of 0.84 which suggest substantial agreement between the values.

3.1 Total PWSS score

Mean pre operative PWSS score was 12.83 ± 6.89 and it improved to 7.05 ± 5.02 post operatively (Table 2). A statistical difference was seen between the two ($P = 0.03$). A significant decrease in the total score by 5.68 ± 2.46 was seen post operatively. Pre-operative speech assessment revealed that none of the patients showed competent nor competent to borderline velopharyngeal competent speech. Borderline to incompetent speech was seen in 5 of 12 (41.67%) and incompetent in 7 of 12 (58.33%) patients. Speech evaluation post surgery revealed to be competent in none of the patients, competent to borderline in 2 of 12 (16.67%), borderline to incompetent in 7 of 12 (58.33%), and incompetent in 3 of 12 (25%). In general, there was a notable improvement in post-operative speech scores compared to pre-operative speech scores. Speech did not deteriorate in any of the patients.

3.2 Nasal Emission

There was no statistically significant difference between the pre operative (1.76 ± 0.68) and post operative (1.34 ± 0.80) scores ($P = 0.179$). Consistent and visible nasal emission seen in the patients prior to the surgery with no clinically significant difference post surgery. The nasal emission distorted some of the pressure consonants in the cleft patients.

3.3 Facial Grimace

Facial grimace did not show a significant difference in the scores pre (1.26 ± 0.33) and post (0.97 ± 0.75) operatively ($P = 0.233$). Facial grimace observed in the cleft patients were essentially unchanged probably attributed to the habitual muscular functions of adult patients.

3.4 Nasality

The mean score of nasality across all 12 subjects was 2.24 ± 0.69 preoperatively and 0.90 ± 0.40 postoperatively. A statistically significant difference in the nasality was seen between the pre and post operative scores ($P < 0.001$). Eight patients (75%) demonstrated a clinically significant decrease in hypernasality. Speech changed from mild to normal in three subjects, from moderate to mild in four others, and from severe to mild in one other. None of the patients demonstrated hyponasality neither pre nor post operatively.

3.5 Phonation

A statistically significant difference was seen in the phonation scores between pre surgery (1.3 ± 0.280) and post surgery (0.60 ± 0.25) ($P < 0.001$).

3.6 Articulation

The average number of articulation errors for each patient was 7.78 ± 3.96 before surgery and 4.00 ± 1.10 errors after surgery with a mean improvement of 3.78 errors. A statistically significant decrease in articulation errors was seen post operatively ($P = 0.004$). After surgery, eight out of the twelve patients (75%) showed improvement in their articulation (i.e., a decrease of more than two errors). The articulation of four patients (25%) remained primarily unchanged. None of the patients' speech deteriorated. Prior to surgery, lingual palatal sibilants and omissions of plosives and fricatives were the most common articulation errors. The patients' anterior dentition was corrected after surgery, which resulted in lesser oral distortions. Post surgery, the articulation improved to reduced intra oral pressure for sibilants and fricatives for most patients. The articulation errors showing the most improvement after surgery were fricatives.

3.7 Correlation between magnitude of distraction and speech outcomes

Patients were categorised into those with less than 10 mm advancement (4 out of the 12) and those with advancement greater than 10 mm (8 out of 12). Chi square test revealed no significant difference between the amount of advancement and speech outcomes.

3.8 Correlation between cleft defect type and speech outcomes

Patients with unilateral cleft defect showed a better post operative speech scoring compared to patients with bilateral cleft defects. However, this difference was not statistically significant.

Table 2: Results table

Parameters	Pre Mean Score ± SD	Post Mean Score ± SD	Mean Difference ± SD	P value
Nasal Emission	1.76 ± 0.68	1.34 ± 0.80	-0.42 ± 0.30	0.179
Facial Grimace	1.26 ± 0.33	0.97 ± 0.75	-0.29 ± 0.23	0.233
Nasality	2.24 ± 0.69	0.90 ± 0.40	-1.34 ± 0.23	0.000
Phonation	1.3 ± 0.28	0.60 ± 0.25	-0.70 ± 0.10	0.000
Articulation	7.78 ± 3.96	4.00 ± 1.10	-3.78 ± 1.86	0.004
Total Score	12.83 ± 6.89	7.05 ± 5.02	-5.68 ± 2.46	0.03

$p<0.05$ is significant

4. Discussion

Maxillary advancement of a greater magnitude was obtained with AMSDO compared to other techniques [28,29]. Numerous drawbacks of traditional methods were avoided with the midmaxillary interdental osteotomy [30]. Advancing of the entire maxilla sagittally in cleft patients carries a risk of velopharyngeal incompetence and a decrease in the length of the soft palate [31,32]. Given that AMSDO does not impact the velopharyngeal sphincter, it is suggested as a substitute for traditional Le Fort I surgery and rigid external distractors for cleft patients with notable VPI. The patients' facial aesthetic was enhanced by the simultaneous regeneration and advancement of surrounding soft tissue in addition to the advancement of bone. With the increased distraction that the AMSDO provides, facial aesthetics and occlusion can be dramatically improved without interfering with speech mechanisms.

The findings from the present research indicate a significant improvement in the speech outcomes following AMSDO procedure. The PWSS scores showed a significant increase after completion of the distraction. PWSS scores improved from incompetent to borderline - incompetent in 4 patients, from borderline - incompetent to competent - borderline in 2 patients. A significant decrease in the hypernasality and phonation of speech was noted. A decrease in nasal airway resistance due to increased nasal airway volume following AMSDO could be attributed to this. The number of articulation errors decreased post operatively. An improved skeletal relationship between the maxillary and mandibular jaw bases along with correction of the anterior dentition diminished the articulation errors. No difference was seen in nasal emission and facial grimace. A possible explanation for this could be the habitual movements of the facial musculature. These learned neuromotor patterns can be challenging to correct as adults. None of the patients showed a worsening of their speech outcomes.

There was no correlation seen between the amount of distraction achieved and their corresponding speech outcomes. Patients with distraction greater than 10 mm showed no difference from the patients with distraction less than 10 mm. This may be attributed to the fact that anterior segmental distraction does not influence velopharyngeal function. Several studies correlated the amount of maxillary distraction after Le Fort l osteotomy with the speech outcomes. Studies by Ko et al.,[33] and Harada et al.,[34] report velopharyngeal dysfunction when maxillary distraction is greater than 15 mm. However, in this study, the velopharyngeal competency is not subject to change, as only anterior segmental maxillary distraction is performed. Patients with unilateral cleft defects showed better outcomes compared to patients with bilateral cleft but the difference was not significant. Previous literature states that bilateral cleft patients presented with poorer speech intelligibility compared to unilateral cleft patients [35,36,37]. The non-significant difference may be attributed to the small sample size.

Speech difficulties in adulthood are highly damaging, for the likelihood of improving them with speech therapy or surgery is considerably diminished [38]. Enhancement of diction in the treated patients resulted from improved tongue mobility due to the expansion of the oral cavity and corrected occlusal relationships. The refined alignment and incisor relationship may be responsible for the improvement in speech comprehension. AMSDO can potentially alter the airflow pressure in the mouth to enhance the resonance of the nasal cavity.

The results of this study correlate with those of Lin et al., [12] where an improvement in nasal cavity resonance, speech intelligibility, and velopharyngeal closure was seen in a few patients. Perpetual assessments by speech-language pathologists were conducted. Nasal emission remained unchanged, and the speech outcomes did not worsen after AMSDO. A study by Rao Janardhan et al., measured speech outcomes following AMSDO using Universal Parameters scale and found an enhancement in the clarity of speech in two cases and speech acceptability in another two cases [25]. A study by Bevilacqua et al., evaluated speech by fiberoptic video nasoendoscopy and spontaneous speech samples with the help of speech-language pathologists [14]. Significant improvements in articulation were reported and none of the patients exhibited an increase in hypernasal speech.

There is a scarcity of literature presenting speech outcomes following AMSDO in cleft patients using standardised assessment methods. The significant improvement in speech outcomes post-AMSDO highlights the importance of considering this procedure for CLP patients with maxillary hypoplasia. Speech therapy should be integrated into the treatment plan to maximize speech improvements.

Limitations

The limitations of this study include an inadequate sample size that restricts the generalizability of the findings. Moreover, the six-month follow-up period may not be sufficient for measuring long-term speech outcomes. Additional research with bigger sample sizes and extended follow-up durations are needed to confirm these results.

5. Conclusion

Anterior maxillary distraction offers a promising intervention for addressing maxillary hypoplasia in CLP patients, with significant improvements in speech outcomes. The Pittsburgh Weighted Speech Scale is an effective tool for evaluating these changes. While this study provides valuable insights, further research with larger cohorts and longer follow-ups is essential to fully understand the impact of AMSDO on speech in CLP patients.

Author Contribution: Conceptualization - N.R.S; Data curation - N.R.S; Formal analysis- P.T.R; Investigation – N.R.S, P.T.R; Methodology - N.R.S, P.T.R; Project administration – P.T.R; Resources- P.T.R; Supervision – P.T.R; Validation -.P.T.R; Visualization – N.R.S; Roles/Writing - original draft – N.R.S; and Writing - review & editing – N.R.S, P.T.R.

Disclosure of Interests: Authors declare no conflict of interest

Acknowledgment: I would like to thank my dean, Dr. Aravind Kumar, for giving us this opportunity to grow and expand our knowledge in research.

Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

Footnotes

Abbreviations

CLP - Cleft lip and palate

AMSDO - Anterior maxillary segmental distraction osteogenesis

VPI - Velopharnygeal insufficiency

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