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Assessing the Impact of Maxillary Advancement on Speech among Cleft Lip and Palate Patients using Borel-Maisonny Classification

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Abstract

Purpose: The aim of our study is to describe the effect of maxillary advancement on speech in our patients born with unilateral cleft lip and palate or bilateral cleft lip and palate .

Patients and Methods: This retrospective study, perceptually assessed, from January 2013 to December 2023, preoperative and postoperative speech of children born with a cleft, after maxillary advancement through a Le Fort I osteotomy. All children were operated and followed by the same team. Speech was evaluated by a certified speech-therapist using a modified Borel-Maisonny classification, divided into three categories: cat. A for velopharyngeal competent (1), cat. B for velopharyngeal borderline competent (1/2, 2b), cat. C for velopharyngeal incompetent (2, 2m). Preoperative and postoperative lateral cephalometric radiographs were used to measure the amount of maxillary advancement. The Fischer Exact test was used to assess the results.

Results: 43 non-syndromic children (27 males, 16 females) were assessed. The mean advancement of point A was 6.5 mm horizontally. 83.7% (36) of the patients had a good speech -cat A and B- preoperatively. 94.3% (34) of them maintained their phonatory score after the Le Fort I osteotomy, while 5.7% (2) velopharyngeal borderline competent patients -cat B, worsened their speech score resulting in a velopharyngeal incompetency -cat C- postoperatively. In the seven patients who were velopharyngeal incompetent -cat C- before surgery (16.3 %), no change was observed post-operatively. The cleft type and the amount of maxillary advancement did not significantly influence the velopharyngeal function.

Conclusions: Patients with a preoperative Velopharyngeal Competency (VPC) were not at risk of a worsened velopharyngeal function after maxillary advancement, while some of those with borderline velopharyngeal competency before surgery evidenced and unfavorable speech outcome. Consequently, patients exhibiting borderline features of velopharyngeal function should be counseled about the risk of speech disorders following maxillary advancement surgery.

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Introduction

Cleft Lip and Palate (CLP) is one of the most common congenital craniofacial anomalies, resulting from incomplete fusion of facial structures during early embryonic development. It is characterized by a rift in the lip, the palate, or both, which can vary in severity and requires multidisciplinary care. One of the prominent issues in CLP patients is maxillary hypoplasia, which results from factors such as impaired sagittal and transverse growth, as well as the timing and type of primary palate and/or lip surgeries. Consequently, children with CLP often present with maxillary retrusion. According to the literature, maxillary advancement is required in 25 to 40 % of patients with CLP [1]. The most common orthognathic procedure used to achieve maxillary advancement is the Le Fort I osteotomy. While this intervention has been shown to yield favorable outcomes in terms of occlusion and facial aesthetics, its impact on speech remains a subject of debate in the literature.

In 1977, Witzel and Munro [2] reported the case of a 16-year-old boy with unilateral Cleft Lip and Palate (uCLP) who expressed dissatisfaction with his speech after maxillary advancement performed by a Le Fort I osteotomy. Given that speech is a cornerstone of social interactions, it represents one of the most important functional goals in the management of CLP patients. Therefore, this finding raised concerns among cleft care teams. Subsequent studies have aimed to elucidate the impact of maxillary advancement on speech. While some researchers suggest minimal changes in speech and intelligibility postoperatively, others report significant alterations in speech production and resonance. Many authors have found that bringing an operated cleft palate forward may cause a Velopharyngeal Incompetency (VPI), by inducing a lack of closure between oral and nasal cavities, resulting in the difficulty to produce resonated vowels and high-pressure consonants [3]. Developed by a French speech therapist in 1975, the Borel-Maisonny Classification [4] (BMC) offers a perceptual framework for assessing speech outcomes following maxillary advancement and allowing a systematic evaluation of speech and intelligibility. In the University Hospital Center of the Canton of Vaud (CHUV), as also in many French-speaking countries, the BMC (Table 1) is commonly used to evaluate speech outcomes, including nasal air emissions and other indicators of VPI.

The aim of this study is to evaluate the effect of maxillary advancement on speech in patients with uCLP and Bilateral Cleft Lip and Palate (bCLP) by using a score based on the BMC, a perceptual methodology tailored for French-speaking populations. Additionally, the study seeks to improve presurgical counseling by providing personalized prognoses based on patients' scores.

Material and methods

This study protocol was approved by the Ethical Committee of The Canton of Vaud (CER-VD 2024- 00273), in adherence to the principles outlined in the declaration of Helsinki.

We identified all children born with uCLP or bCLP who underwent a Le Fort I osteotomy for maxillary advancement to address maxillary hypoplasia, at the CHUV, from January 2013 to December 2023. Patients with associated malformations, incomplete data, or primary surgery performed elsewhere were excluded. The assessments were carried out retrospectively. All patients were French-speakers and underwent preoperative and postoperative speech evaluation. Standard upper airway assessments were documented on the basis of patient history of nasal obstruction with the presence of snoring, sleep apnea and mouth breathing, as well as nasal airflow during resonated vowels and high-pressure consonants with the Glatzel mirror. Speech was evaluated according to BMC. In addition, voice quality, articulation disorders associated with VPI, and intelligibility were also analyzed. Assessment was performed separately by two qualified speech pathologists experienced in CLP speech disorders. In addition, subjects requiring a pharyngoplasty had instrumental assessment of nasalance using a 622 Kay Electronics nasometer (Kay Elemetrics, Pine Brook, NJ, USA) and nasofibroendoscopy before the operation.

Since, in the literature, VPI is the most usual means to define an altered speech, we correlated the presence and severity of nasal air flow with velopharyngeal competence (VPC), leading to grading speech into three categories (A, B, C) in alignment with the BMC, by associating perceptual evaluation of speech (hypernasality) and nasal airflow (Table 1): Category A for patients with a VPC who had an excellent speech (no nasal airflow and no hypernasality); Category B for borderline velopharyngeal competent patients with a VPI but no hypernasality and non-audible nasal airflow and a good intelligibility. Category C for patients with a VPI with hypernasality and audible airflow and bad intelligibility. The presence of articulation compensations, such as pharyngeal or glottal articulation, were not described in our classification. We named this the Borel-Maisonny's score; it allowed us to compare our results with those in the literature.

All patients had undergone primary lip repair between five and six months of age and primary palate repair between 6 and 12 months of age, by the same pediatric plastic surgeon, following the Malek procedure [5,6]. At the age of three, all children were evaluated by a multidisciplinary team. This team was composed of two pediatric surgeons, two pediatric Ear, Nose and Throat (ENT) specialists, two maxillofacial surgeons, an orthodontist, two speech therapists, a psychologist and a coordinating nurse. The child and their parents were seen according to the child's needs, typically on an annual or biannual basis. Besides, parents were provided with strategies to encourage babbling and early verbal communication from the age of one. Children were routinely monitored by an ENT specialist by otoscopy, tympanometry and hearing tests to exclude chronic otitis media. They were checked every two to three years, if they had no sign or symptoms of chronic ear disease or long-term hearing impairment.

From the age of five, dental disorders were managed by the same orthodontist. The orthodontic treatment was, for most patients, correlated with the planning of the alveolar bone graft, aiming to expand the maxilla and then close the cleft in the alveolar process allowing the emergence of permanent teeth in a favorable periodontal environment.

In early adolescence, the relationship between the upper and lower jaws and the malposition of the teeth were assessed. For our group of patients, a purely orthodontic approach was insufficient to correct dentofacial deformity. The need for orthognathic surgery was based on clinical evaluation and data available from the analysis of the lateral cephalograms. Orthognathic surgery was performed by the same team of maxillofacial surgeons. The maxillary advancement was performed through a Le Fort I osteotomy mostly in one piece. Due to a possible transversal collapse of the maxillary arches on each side of the cleft, caused by the scarring tissue, the maxilla rarely needs to be segmented in two or three pieces before advancement.

In the case of combined orthodontic and surgical approaches, three phases can be described: (1) presurgical coordination of upper and lower dental arches, with braces for several months; (2) surgical correction; and (3) postsurgical orthodontic treatment for a few months.

Preoperative and postoperative lateral cephalometric radiographs were taken, in occlusion and lips at rest. Preoperative radiographs were taken, on average, 1.3 months before surgery (0.25-5). Postoperative images were taken, on average, 9.6 months after the surgery (1-30). Cephalometrics radiographs were traced and analyzed by the same orthodontist with Quick Ceph Systems® and oriented along the Frankfort horizontal plane. Cephalometrics landmarks were established to evaluate sagittal movements of the maxilla. The reference plane used for superimposition of the per- and-post-operative tracings was the Sella-Nasion plane (SN). The length of anterior-posterior advancement was determined by measuring the advancement of the point A (the most concave point of the anterior maxilla) on the superimpositions pre and postoperatively. The BMS before and after surgery were compared in order to assess any changes in speech, resonance, and intelligibility following maxillary advancement surgery.

To assess the impact of maxillary advancement on speech outcome in children with UCLP or BCLP, the Fischer Exact Test was used with $p<.05$ considered as statistically significant.

Results

Between January 2013 and December 2023, 61 patients with uCLP or bCLP in our department underwent a maxillary advancement through Le Fort I osteotomy. Eighteen (18) had been excluded because of associated malformations, neurological anomalies, primary surgery in another center or incomplete data. 43 children, 27 males and 16 females, were included in the study (28 with uCLP- Group 1, 15 with bCLP- Group 2) (Table 2). Preoperative speech evaluation was conducted at a mean of 13 months (2-40), and postoperative evaluation at a mean of 16.9 months (6-47), with three patients evaluated between 6 and 12 months. In our group, one patient had synkinesis before surgery which persisted after maxillary advancement, reported as articulation compensation, which is not part of the BMS. Orthognathic surgery was performed at a mean age of 16.4 years (range 13-21) with five patients having concomitant mandibular setback osteotomies.

In addition, seven patients presented with a palatal fistula, one of which was closed during the surgery. Furthermore, in the 11 patients who had undergone a pharyngoplasty prior to the Le Fort I osteotomy, no significant differences in speech parameters were observed after the procedure.

Postoperative speech evaluation enabled the distinction between resonance and nasal airflow resulting from the fistula and the velopharyngeal sphincter. Among these patients, speech assessment indicated stable post-operative velopharyngeal function.

Group 1 (Table 3): This group consisted of 28 patients (9 females, 19 males) with uCLP. Preoperatively, 12 (43%) demonstrated a VPC with a normal speech (category A). Postoperatively, ten of these patients maintained an excellent velopharyngeal function and speech, while two exhibited good speech with a non-audible nasal airflow but remained in category A since the nasal airflow was due to the presence of a palatal fistula in both cases. Thirteen (13) patients (46.4%) had a borderline VPC with a good speech (category B) preoperatively, 12 had unchanged velopharyngeal function postoperatively, and one experienced worsening of velopharyngeal function and speech with hypernasality and audible nasal airflow (category C). Finally, three (10.8%) exhibited a VPI and a poor speech with lack of intelligibility (category C) preoperatively, and no change was observed postoperatively.

Group 2 (Table 4): This group included 15 patients (7 females, 8 males) with bCLP. Preoperatively, six (40%) demonstrated a VPC with an excellent speech (category A), of whom five maintained excellent speech postoperatively, while one reported non-audible nasal airflow postoperatively due to a palatal fistula, while the velopharyngeal sphincter remained competent after the surgery (category A). Five patients (33%) had velopharyngeal borderline competency (category B) and four of them had a good speech while one had a poor speech because of the presence of a palatal fistula preoperatively: this one improved his speech after the fistula closure combined with the maxillary advancement; one altered his velopharyngeal competency and experienced poor speech outcomes (category C) after surgery. In the four (26.7%) who had exhibited speech preoperatively with lack of intelligibility (category C), no change was observed postoperatively.

Lateral cephalometric analysis

Post-operative radiographs were missing for five patients. The extent of maxillary advancement ranged from 2 to 11 mm (mean, 6.5 mm). In Group 1, the mean advancement was 6.1 mm (range 2-11 mm) and in group 2, 8.2 mm (range 5-10 mm). In only one patient was there more than 10 mm of anteroposterior movement of the upper jaw.

Table 1: Borel-Maisonny classification and the Borel-Maisonny's score.

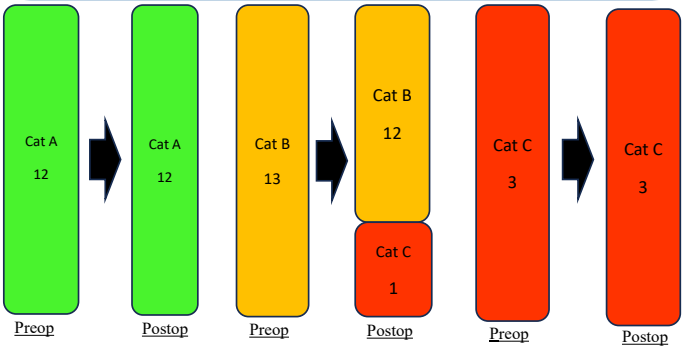
Type O	No phonation
Type 1	Excellent speech: no nasal airflow no hypernasality
Type 1/2	Good speech, intermittent nasal airflow emission, no hypernasality, good intelligibility
Type 2b	Good speech with non-audible continuous nasal airflow, no hypernasality, good intelligibility
Type 2	Poor speech with hypernasalty and continuous nasal airflow, good intelligibility
Type 2m	Poor speech with continuous nasal airflow, hypernasality, bad intelligibility

Category A Category B Category C

Table 2: Demographic and clinical characteristics (N= 43): number (%), unless otherwise stated.

SEX	
Male	27 (62.8)
Female	16 (37.2)
CLEFT TYPE	
Unilateral cleft lip and palate	28 (65.1)
Bilateral cleft lip and palate	15 (34.9)
OTHER SURGERY	
Pharyngoplasty previous to orthognathic surgery	11 (25.6)
Additional mandibular set back during Le Fort I osteotomy	5 (3.5)
MEAN AGE at the orthognathic surgery *(years),* (range) 16.4 (14-21)	
AMOUNT of Maxillary advancement *(mm),* mean (range) uCLP: 6.1 (2-11) bCLP: 8.2 (5-10)	

Table 3: Demographic and clinical characteristics (N= 43): number (%), unless otherwise stated.



Green referring to normal velopharyngeal competence (Cat.A), orange to velopharyngeal borderline competence (Cat B), red to velopharyngeal incompetence (Cat C)

Table 4: Speech results pre and postoperative based on Borel-Maisonny's score after maxillary advancement through Le Fort I osteotomy in 15 children born with bilateral cleft lip and palate.

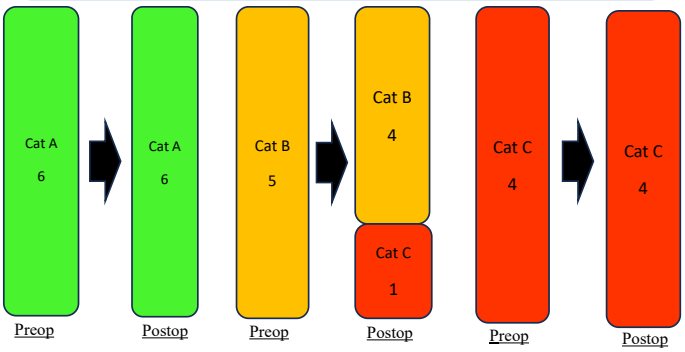


Table 5: Speech results pre and postoperative based on Borel-Maisonny's score after maxillary advancement through Le Fort I osteotomy in 43 children born with uni and bilateral cleft lip and palate.

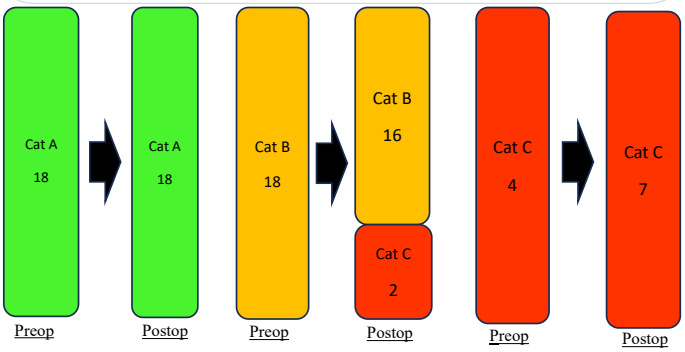


Table 6: Proposition of equivalence between different speech evaluation scales: Pittsburgh Weighted Speech Scale (PWSS), Cleft Audit Protocol for Speech Augmented (CAPS), Borel-Maisonny's score (BMS), 5-point scale velopharyngeal function.

	PWSS	CAPS	BMS	5-point scale VPF
Competent	0	Dark green	Category A (type 1*)	0
Borderline competent	1-2	Light green	Category B (type 1/2*, 2b*)	1
Borderline incompetent	3-6	Yellow-orange	Category C (type 2*)	2-3
Incompetent	≥ 7	Red	Category C(Type 2m*)	4

*Borel-Maisonny classification

There were no perioperative complications.

Comparison of speech outcomes between the two groups showed no significant difference.

Discussion

This study demonstrates that in the combined groups A and B, 94.3% of children with excellent or good speech preoperatively maintained their speech quality following maxillary advancement via Le Fort I osteotomy. Two children with borderline VPC, one with uCLP and one with bCLP developed VPI after the surgery.

By the late 1960s, Obwegeser [7] introduced the concept of maxillary advancement through Le Fort I osteotomy in routine surgical practice. Concurrently, Jabaley and Edgerton [8] hypothesized that moving the maxilla forward could potentially lead to speech impairment due to changes in the pharyngeal airways. However, after conducting a study on a 18-year-old male without a cleft palate, they found no significant alterations in the relationship between the velum and the posterior pharyngeal wall following maxillary advancement.

Later, in 1976, Schwarz et al. [9] evaluated speech in 31 patients with CLP and nine patients without cleft who underwent maxillary advancement. They concluded that maxillary advancement could enhance the quality of speech articulation without causing any speech impairment. However, the methodology used for evaluation was not clearly described.

In 1977, Witzel and Munro reported a case of reduced speech intelligibility following maxillary advancement in a 16-year-old male with CLP [2]. Pre-operative assessments showed normal nasality with minor articulation errors. Post-operatively, the patient showed significant hypernasality during speech, attributed, according to cephalometric analysis, to a lack of contact between the soft palate and the posterior pharyngeal wall. Witzel et al. concluded that patients with CLP were at risk of velopharyngeal dysfunction after maxillary advancement, highlighted by speech disorders.

Subsequent research has explored the impact of maxillary advancement on speech through various assessment methods, using perceptual methodology alone or combined with instrumental one. No consensus emerges regarding the correlation between maxillary advancement and speech disorders resulting from VPI.

Studies have shown that VPI results in distinct speech disorders, different from articulation errors, associated with malocclusion or labial incompetency in CLP patients with dental and skeletal abnormalities [10]. Proper velopharyngeal function is crucial to produce high-pressure consonants and orally resonated vowels, necessitating the closure of the velopharyngeal sphincter for sound resonance. Impaired velopharyngeal function allows airflow and acoustic energy to escape into the nasal cavity, leading to audible nasal airflow and hypernasal resonance, which can profoundly impact communicative intelligibility.

In 2006, Chancharoensook [11] reviewed 39 studies spanning over three decades, identifying post-operative hypernasality predominantly in cleft lip and palate patients with borderline velopharyngeal function pre-operatively. 74% of the studies evaluated speech perceptually, as in our study. Perceptual assessments methods, such as the Cleft Audit Protocol for Speech (CAPS) [12], Pittsburgh Weighted Speech Scale (PWSS) [13] or

its variant and the five-point scale of velopharyngeal function, have been almost exclusively used in categorizing speech in the literature.

Using the CAPS, based on assessment of nasality, articulation, nasal air emission, borderline velopharyngeal function would be reported as "light green", "borderline competent" (1-2), "insignificant borderline" (1) (Table 6) [14]. The PWSS is more subjective, based mainly on nasality. PWSS is more operator-dependent than CAPS, where velopharyngeal function is scored from 0 (competent) to 7 (incompetent). We believe that when investigating French speakers, the BMS should be used, given that there are three main (vowel) formants that vary depending on the linguistic data [15]. In French, the main formant is produced in the oral cavity, requiring more frequent velopharyngeal closure than in English. Therefore, in French-speaking countries, speech therapists mostly use the BMC for perceptual evaluation of speech. This is, to our knowledge, the first article referring to the BMC when investigating the effect of maxillary advancement on velopharyngeal function through speech disorders. Kaldub et al. [14] proposed an interesting equivalent table for a speech evaluation scale, allowing the use of BMC in international literature, enabling us to compare our results with those found in studies using English-speaker speech assessments, through the BMS.

Witzel [16] defined borderline velopharyngeal function as a "small pinhole gap in the velopharyngeal valve at maximum closure" in patients presenting "normal resonance or clinically insignificant hypernasality and inaudible nasal emissions". The lack of closure might be caused by the inability of the pharyngeal scarring soft tissues to adapt to the velopharyngeal space expansion induced three-dimensionally in CLP patients. Indeed, the anterior-posterior translation has an impact on soft palate muscles; however, with structural adaptation, a non-repaired palate could adjust to changes in the pharyngeal depth allowing preservation of previous speech abilities. In a retrospective study, Alaluusua [17] et al. assessed one hundred non-syndromic CLP patients perceptually over 10 years with a 5-point scale of velopharyngeal function, and instrumentally with a nasometer. They demonstrated, consistent with our results, that patients with borderline velopharyngeal function were at risk of experiencing speech problems after maxillary advancement independently of the cleft type. Indeed, according to the equivalent table, patients with speech classified as 1/2, 2b in the BMC are considered patients with borderline velopharyngeal function (Category B) and were those who showed a speech deterioration post-operatively [18-20].

In contrast, Semdberg [21] concluded that maxillary advancement had no significant impact on velopharyngeal function in a retrospective study with 13 CLP patients (7 bCLP, 4 uCLP). Mean amount of advancement was 6.2 mm (2-9) and mean follow-up 13 months. Blind perceptual analysis of speech and velopharyngeal function was performed through a 5-point scale (0-4). Interestingly, from the 11 patients, eight additionally had mandibular set back; one of them had meanwhile received a pharyngeal flap. Another patient underwent maxillary advancement with pharyngeal flap during the same operation. Only one patient had symptoms of preoperative VPI and was considered as borderline. After the surgery, two patients, including the borderline competent, showed a moderately reduced overall impression of speech which was not considered statistically significant. Those results could be explained by the additional surgeries undergone by most of the patients making comparison with maxillary advancement alone difficult. Moreover, since set back mandibular osteotomy could reduce tensions in muscles involved in velopharyngeal gap closure, this combined procedure may allow a better velopharyngeal competency [22].

In addition, other authors [23,24] reported a relevant association between mandibular set back and speech improvement. The borderline velopharyngeal status did not appear to influence postoperative outcomes. Yet, one explanation could be that a lesser advancement was needed to reach satisfactory maxillary projection in the group with additional mandibular set back. Furthermore, as also in our study, no difference in speech parameters was found between patients who had had a pharyngoplasty preoperatively and those with no pharyngoplasty. We reported no speech improvement in patients who underwent bimaxillary osteotomy. However, non-audible nasal airflow was observed in one case, likely due to air escaping through a palatal fistula present before surgery and which may have been enlarged by the forward movement of the maxilla. It is important to note that only five patients underwent additional mandibular setback in our cohort, which represents a sample size too small to draw consistent conclusions. The existing literature on the subject is insufficient to define the best procedures to prevent speech disorders after maxillary advancement in CLP patients, since only a few studies have attempted to compare bimaxillary osteotomy with mandibular set back and Le Fort osteotomy alone. Janulewicz et al. [1] focused on this point, assessing speech through PWSS in 54 CLP patients, of whom 34 had Le Fort I osteotomy and 20 bi-maxillary osteotomies. The speech of patients with borderline velopharyngeal function was altered by the surgery. Improvement in articulation was found in most patients. No difference was found in the incidence or the increase of hypernasality between the two groups. It is important to note that one of the limits of the study was the early post-operative evaluation, starting after three months. In fact, it is now well demonstrated that transient speech impairment could be observed up to three months after the surgery [20,25]. Moreover, stable outcomes are observed after at least one year, taking into account the risk of relapse during the first 12 months following surgery. Therefore, a longer period, of at least 12 months, may be needed for a proper evaluation of the permanent speech improvement, our primary concern. Hence, their conclusions should be taken carefully.

It is hypothesized that the extent of maxillary advancement might negatively impact postoperative velopharyngeal function, particularly if the advancement exceeds 10 mm [18,26,27]. However there is no consensus in the current literature on this issue. Studies by David et al [28]. and Sader et al [26]. have reported impaired velopharyngeal function in patients with advancements exceeding seven mm and ten mm, respectively. Similarly, Chua et al. [29] indicated that even a modest advancement of four mm could impair velopharyngeal function, as evidenced by speech disorders. Conversely, Schultz et al. [30], using the CAPS to assess 18 CLP patients, found no correlation between VPI and advancements averaging 9.8 mm (range 3-12 mm). In alignment with Kim et al. [27], for CLP patients requiring more than ten mm advancement, we frequently perform additional mandibular setback due to the challenge of mobilizing scarred soft tissues. Despite the smaller size of the cohort, our findings align with these results, showing no significant correlation between maxillary advancement amount and postoperative speech impairment.

A review of the literature by Pereira [31] and Vella [32] highlights that most studies are of level III or IV evidence with fewer than 40 patients. A key strength of our study is the larger number of patients, which reduces the likelihood of a type II error. Furthermore, Pereira et al. [31] noted that few studies evaluated speech later than 12 months post-surgery. In our study, only three adolescents were assessed before 12 months. And yet, it is important to consider the changes in pharyngeal morphology due to natural growth as a potential bias.

This study has several limitations. Firstly, it is a retrospective study. Secondly, our evaluations relied solely on perceptual methodology, whereas the gold standard for speech evaluation should be both perceptual and instrumental [21,29,31]. Lastly, maxillary advancement was measured on cephalometric radiographs taken at non-standardized follow-up periods; some radiographs were obtained two years post-surgery in growing adolescents.

Our data suggest that in uCLP or bCLP patients, the preoperative speech score, using the Borel-Maisonny classification, may be directly influenced by maxillary advancement via Le Fort I osteotomy, regardless of the extent of advancement or additional mandibular set-back. This indicates that anteroposterior advancement of the upper jaw should not be considered the unique predictor of a high risk of VPI.

Conclusion

In our study, patients mostly maintained their speech after maxillary advancement surgery through Le Fort I osteotomy. Our results suggest that CLP patients exhibiting borderline features of velopharyngeal function should be counseled about the risk of speech disorders following maxillary advancement surgery. Further prospective randomized controlled multicentric studies with larger sample sizes are needed to validate these findings and improve our clinical practice in the management of CLP patients undergoing maxillary advancement surgery.

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