ANATOMICAL AND PHYSIOLOGICAL CHANGES FOLLOWING PRIMARY PALATOPLASTY USING THE BUCCAL FLAP APPROACH

by

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Individuals born with cleft palate typically undergo primary palatoplasty between 6-12 months of age to repair their palate. Traditional primary palatoplasty surgical techniques include the V-Y pushback, the intravelar veloplasty, the von Langenbeck approach, and the Furlow Z-plasty. However, approximately 5-20% of patients present with velopharyngeal dysfunction post-surgery and require a secondary surgery for speech. The buccal flap approach, another surgical technique, has been used at the time of primary palatoplasty, during secondary surgery for velopharyngeal dysfunction (VPD) and for fistula repair. Study outcomes related to the buccal flap approach have failed to fully document the resulting anatomic changes in the velopharyngeal (VP) anatomy but have reported promising perceptual speech outcomes.

The study was designed to use magnetic resonance imaging (MRI) to evaluate how the use of the buccal flap approach, used during primary palatoplasty, alters VP anatomy and physiology. Specifically, VP anatomy was evaluated for anatomical differences and function in comparison to non-cleft individuals to determine if the use of a buccal flap creates a more normal VP system for adequate VP closure. Significant differences for effective velar length, effective

VP ratio, sella-nasion-subspinale (SNA) angle, sella-nasion-supramentale (SNB) angle, and velar stretch were noted between the two groups. There were no significant differences for velar thickness, velar length, VP portal depth, nasion-sella-basion (NSBa) angle or subspinale-nasion-supramentale (ANB) angle.

This study defined anatomic changes resulting from primary palate repair using the buccal flap approach. Results from the study add valuable information to the literature on VP and craniofacial changes following the use of this specific surgical technique. As a secondary contribution, this study highlights the utility of using MRI to quantify the changes that occur to the VP anatomy following the buccal flap surgical approach and may help improve diagnostic and treatment approaches for individuals with cleft palate.

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LIST OF ABBREVIATIONS

VP	Velopharyngeal	1
SLP	Speech language pathologist	1
VPI	Velopharyngeal insufficiency	2
PRISMA	Preferred Reporting Items for Systematic Review and Meta-Analysis	3
EMBASE	Excerpta Medica Database	3
CINAHL	Cumulative Index to Nursing and Allied Health Literature	3
CAPS-A	Cleft Audit Protocol for Speech-Augmented	11
VPD	Velopharyngeal dysfunction	17
MRI	Magnetic resonance imaging	29
3D	Three-dimensional	35
2D	Two-dimensional	35
DICOM	Digital Imaging and Communication in Medicine	35
PPW	Posterior pharyngeal wall	36
SNA	Sella-nasion-subspinale	36
SNB	Sella-nasion-supramentale	36
ANB	Subspinale-nasion-supramentale	36
NSBa	Nasion-sella-basion	36
ANCOVA	Analysis of Covariance	41
ICC	Intra-class correlation coefficient	41

CHAPTER 1

LITERATURE REVIEW

This chapter provides a systematic review of the buccal flap surgical approach for cleft palate repair[1]. The impact of the buccal flap approach on speech and surgical outcomes pertaining to velopharyngeal (VP) anatomy and function is summarized. Limitations in the current research and areas of future research interests are discussed.

INTRODUCTION

Orofacial clefting is one of the most common birth defects. A cleft palate occurs when the roof of the mouth does not fuse together properly during fetal development. This results in an opening between the nasal and oral cavity which can affect feeding, speech, and resonance. Individuals born with cleft palate typically undergo a primary palatoplasty between 6-12 months of age to reconstruct the palate and establish a normal VP mechanism. The importance of the primary palatoplasty is to ensure normal speech and resonance post-surgery. There are various surgical procedures designed to repair a cleft palate which report different speech and surgical outcomes. In cleft palate care, the speech-language pathologist (SLP) is involved in the evaluation of the patient along his/her lifespan, particularly when primary palatoplasty fails to result in normal VP function. Given speech is the primary outcome measure for success of primary palatoplasty, examining surgical techniques from this perspective is critically important.

The goal of primary palatoplasty is to reconstruct the palate and achieve normal speech and resonance while not inhibiting maxillofacial growth. Primary palatoplasties aim to create a

[1] **Haenssler, A.E.**, & Perry, J.L (2020). The effectiveness of the buccal myomucosal flap on speech and surgical outcomes in cleft palate: A systematic review. *International Journal of Speech & Language Pathology and Audiology, 8*, 50-58.

separation between the oral and nasal cavity and create proper elevation and retraction of the velum necessary for adequate VP closure during speech and swallowing. The success of a repair is based on normal resonance, assessed when speech develops, and normal maxillofacial growth, assessed after maxillofacial growth is complete. Although multiple techniques exist, the most commonly used methods for primary palatoplasty include the Furlow double opposing Z-Plasty, straight-line intravelarveloplasty, or V-Y pushback [1]. The effectiveness of these traditional surgical techniques resulting in normalized speech is reported to range from 80% to 95% [2, 3]. Secondary surgeries for velopharyngeal insufficiency (VPI) are performed to correct postoperative VPI and aim to achieve adequate VP closure necessary for speech. The goal of secondary surgery for VPI is to create a seal between the oral and nasal cavity while avoiding airway obstruction. Common surgical techniques include a pharyngeal flap, sphincter pharyngoplasty, palatal lengthening procedure, palate re-repair, or posterior pharyngeal wall augmentation. The effectiveness of these surgical techniques results in 70.7% of patients achieving normal resonance and approximately 8.7% requiring further surgery for speech [4].

In recent years, favorable post-operative speech and maxillofacial growth results have been reported with the use of the buccal myomucosal flap for primary palatoplasty, secondary surgery for VPI, and fistula repairs [5, 6, 7, 8, 9]. The buccal myomucosal flap approach consists of dividing the hard and soft palate junction and creating a defect that will be reconstructed using the buccal myomucosal flaps, which are raised from the inner aspects of the cheek. In the use of a bilateral buccal flap, the first flap is interposed in the opening between the hard and soft palate and sutured in the nasal layer with the mucosal side facing the nasal lumen. The second flap is then sutured in the oral layer, with the mucosal side facing the oral lumen [1, 10, 11]. The reported benefits of the buccal myomucosal flap are to close the palate without tension, lengthen the palate, reconstruct the levator veli palatini sling, achieve normal maxillofacial growth, and achieve normal resonance [5, 6, 7, 8]. The buccal myomucosal flap approach is different from more traditional surgeries in that it adds muscle to the defect between the hard and soft palate in an effort to decrease tension and lengthen the palate, while other surgical approaches rely on the existing palatal tissue to close the cleft.

Given the emerging number of publications on improved outcomes following the buccal myomucosal flap approach, the authors performed a systematic review of the current literature on the impact of the buccal myomucosal flap approach. The purpose of the study was to assess the effectiveness of the buccal myomucosal flap approach using the primary outcome measure of speech and secondary outcome measure of surgical complications. Outcomes following the use of buccal myomucosal flap approach used for primary palatoplasty and secondary surgery for VPI are considered separately.

METHOD

Literature Search

This systematic review followed the checklist and guidelines outlined by the Preferred Reporting Items for Systematic Review and Meta-Analysis (PRISMA) [12]. Specifically, PRISMA uses an evidence-based method including completion of a 27-item checklist and following a four-phase flow diagram to ensure transparent and comprehensive reporting methods. A meta-analysis and assessment of risk bias were not performed due to the heterogeneity in assessment and outcome measures. Electronic searches of the following databases were used to identify studies: Excerpta Medica Database (EMBASE), Pubmed, Scopus, Cumulative Index to Nursing and Allied Health Literature (CINAHL), and Cochrane on

June 21, 2019. The search strategy included synonyms for the surgical procedure ((buccal flap OR buccinator flap OR buccal myomucosal flap) AND (cleft palate OR submucous cleft palate)). No limits were placed in the search criteria. Reference lists of all relevant publications were searched for additional papers.

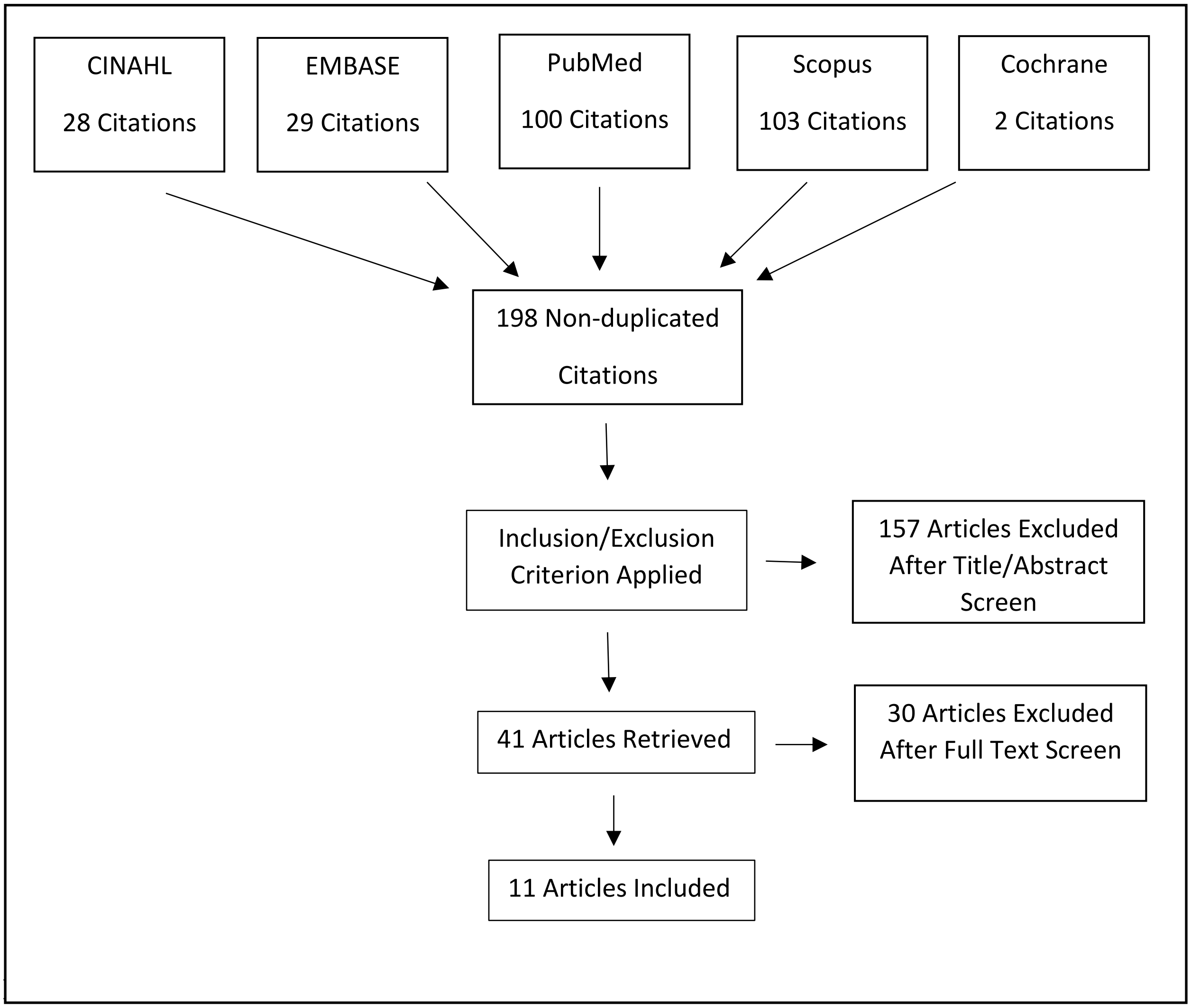
Inclusion/Exclusion Criteria

Studies were screened for inclusion using the following inclusion criteria: human study of patients undergoing the surgical technique for primary cleft palate repair or secondary surgery for VPI. Studies were also included if they contained one of the following inclusion criteria: (1) postoperative perceptual speech assessment reported and/or (2) visualization of postoperative VP anatomy. Studies were screened for exclusion using the following exclusion criteria: articles not published in the English language, had an inadequate description of methods used to analyze speech and surgical outcomes, as well as letters, presentations, case reports, technical notes, and cadaver studies.

Study Selection

A 3-stage screening process was used. In the first stage of screening, article titles were reviewed and excluded based on their lack of relevance to the review. Articles that were remaining were then reviewed at the abstract level and removed if found to be irrelevant to the review. At the third stage, the full-text articles were reviewed. To avoid overlapping patient populations, authorship and date of recruitment were compared. Articles with patient populations that were found to overlap were compared, and the article with the most comprehensive data was included. The PRISMA flow diagram is presented in Figure 1.

Figure 1. Study selection PRISMA flow diagram



The following data were extracted from each article: author(s), year of publication, procedure name, reason for procedure, number of patients, syndrome diagnosis, mean age at surgery, mean length of follow-up for speech assessment, methods of speech assessment, resonance pre- and postoperatively, nasal air emission pre- and postoperatively, methods for VP assessment, VP function pre- and postoperatively, postoperative complications, and need for further surgery. The data were further divided and reviewed based on the surgery type, i.e., primary palatoplasty or secondary surgery for VPI.

Outcomes of Interest

The primary outcome variable evaluated was speech. Speech outcomes were subdivided into nasal air emission, resonance, and intelligibility. The methodology used for assessing speech outcomes was extracted and included speech assessment carried out by a SLP, blinded speech assessment, speech assessment by more than one SLP, and use of a validated perceptual speech assessment. Outcomes of interest pertaining to the VP function include the methodology used for imaging, the assessment scale used to rate VP gap and competency, and the post-operative VP gap size using the specified scale in the study. The secondary outcome measure was surgical complications.

Data Analysis

A formal meta-analysis of the study data was not possible due to the variability in methods of speech and VP assessment and reporting of results among the studies. Tables were constructed to summarize and explain the findings of the included studies. For each study, the number of patients reported in each outcome of interest was recorded. Individual percentages were calculated using the number of patients in each study as the denominator. Average percentages were calculated manually by adding the total patients reported in each outcome of interest and using the total number of patients from all of the studies as the denominator.

RESULTS

The literature search identified 262 potential articles (Figure 1). After removing duplicated articles and the application of the inclusion and exclusion criteria to titles and abstracts, 41 articles progressed to the next level of screening. The remaining articles were reviewed in their entirety and a total of 11 studies met the inclusion criteria. The studies included were published between 2003 and 2018 and were all retrospective reviews published in the Cleft Palate-Craniofacial Journal (n=4), Plastic and Reconstructive Surgery (n=3), Journal of Cranio-Maxillo-Facial Surgery (n=1), the Journal of Plastic, Reconstructive and Aesthetic Surgery (n=1), Ceyon Medical Journal (n=1) and the Journal of Oral Maxillofacial Surgery (n=1). Studies originated from the United States (n=4), Europe (n=4), Asia (n=2), and South America (n=1). Comparison of the study characteristics revealed differences in the study populations (including the inclusion or exclusion of patients with syndromes), pre- and post-operative assessments for speech and VP function, and recorded outcomes. Details for each study are presented in Table 1.

Table 1. Study Characteristics and Speech Outcomes, Sorted Alphabetically by Procedure

Study	Procedure	N; Mean age at surgery (range)	Syndrome (%)	Post-surgery follow-up (range)	Normal resonance	Normal nasal emission	Intelligibility/ normal speech improved	Postoperative complications	Need further surgery for VPD	VP Assessment & Criteria	VP Gap Pre-op	VP Gap Post-op
Primary Palatoplasty												
Chen (2003)	Bilateral musculomucosal buccal flap	26; 6.4 years (3-12)	0	2-8 years	-	-	-	-	-	Nasopharyngoscopy; Good VP closure: <5mm; Poor VP closure: <5 and <10mm	-	20/24 (83%) had good VP closure; 4/24 (17%) had poor VP closure
Mann (2017)	Double opposing Z-plasty ± buccal flap	505; -	69 (14.1%)	7.76 years	I	-	I	8.78%	7.1%	-	-	-
Jackson (2004)	Buccal myomucosal flap	156; 6.3 ± 2 months	0	36 months	91.10%	-	89% normal speech	3.60%	10 (6.4%)	Videofluoroscopy; Competent: no gap; Minimally competent: 0-5mm; Moderately competent: 6 to 10 mm; Grossly incompetent: >10mm	-	91.1% Competent; 6.2% Marginal; 2.7% Incompetent
Secondary Surgery for VPI												
Ahl (2016)	Buccinator mucomuscular flap	103; 5.5 years	19 (18.4%)	14 months (5-23)	I	I	I	10 (9.7%)	14 (13.6%)	-	-	-
Denadai (2017)	Bilateral buccinator myomucosal flaps	53; 19.3 years (5-50)	0	3,6,12,15 months	72%	83%	85% normal speech	11 (21%)	8 (15%)	Nasopharyngoscopy; Complete; Pinhole (bubbling); Small (<80, >100% closure); moderate (50-80% closure); large (<50% closure)	Moderate: 19/53 (36%); Large: 34/53 (64%)	Complete: 41/53 (77%); Pinhole: 7/53 (13%); Small: 5/53 (10%)
Dias (2016)	Buccinator myomucosal flap	34; -	0	6 months-1 year	24 (70.6%)	29(85%)	I	-	-	-	-	-
Hens (2013)	Bilateral buccinator myomucosal flaps	32; 8 years (3-28)	16 (50%)	9.2 months (5.7-17.2)	21 (81%)	No notable difference	-	4 (12.5%)	6 (19%)	Lateral videofluroscopies; Presence or absence	27/30 patients with a gap	20/30 (66.7%) no gap; 7/30 (23.3%) patients with a gap

Hill (2004)	Bilateral buccinator musculomucosal flap	16; 8.5 years (4-19)	0	3, 6, 12 months	87%	I	93%	4 (25%)	Not noted	-	-	-
Logjes (2017)	Unilateral myomucosal buccinator flap	42; 4.9 years (2.6-17.6)	8 (19%)	1.2 years (.5-2.1 years)	I	-	32 (80%)	1 (.02%)	7 (16.7%)	Nasopharyngoscopy; Adequate VP closure; Subadequate VP closure; Inadequate VP closure	-	Improvement in 24/29 (83%)
Mann (2011)	Buccinator flaps	24; 10.3 years (3-16)	6 (25%)	58 months	I	I	I	2 (7%)	1 (.04%)	-	-	-
Robertson (2008)	Unilateral buccal myomucosal flap	22; 8.5 years (1-23)	4 (18%)	-	14 (47%)	13 (65%)	I	-	5 (23%)	Videofluoroscopy; Competent: no gap; Minimally competent: 0-5mm; Moderately competent: 6 to 10 mm; Grossly incompetent: >10mm	17/20 had VPI	16/20 no VPI

Abbreviations: I, improved; VP, velopharyngeal; VPI, velopharyngeal insufficiency

Study Participants

Study size ranged from 16 to 505 participants across research studies, with a mean of 92 patients and median of 34. Not all patients were included in the speech analysis in three studies due to age of the patient or absence of pre-and post-operative speech samples. The included studies were separated into two groups based on if the patients received a primary palatoplasty or secondary surgery for VPI. Three studies dealt with patients undergoing primary palate repair and eight studies examined secondary surgical findings. A total of 687 patients underwent the buccal myomucosal flap for primary palatoplasty and 326 patients underwent secondary surgery for VPI.

In all studies, cleft types were presented either descriptively or using the Veau classification system [13]. Participants with syndromes were included in six studies, for a total of 122 participants [1, 7, 8, 11, 14, 15].

Speech Assessments and Outcomes

Primary Palatoplasty Speech Outcomes

Resonance and speech intelligibility improved in the two studies that reported the variables following primary palatoplasty. Jackson et al. [6] reported normal resonance was achieved in 91.1% of patients and reported 89% of patients achieved normal speech with the use of the buccal myomucosal flap technique. Data related to normal nasal emission was not reported in either of the two studies.

Secondary Surgery for VPI Speech Outcomes

Following secondary surgery for VPI, 74% of patients achieved normal resonance with the use of the buccal myomucosal flap technique. Nasal emission data were collected in seven of

11 studies. The studies reported an improvement in nasal air emission, with 79% of patients presenting with no nasal air emission post-surgery. Normal speech was reported in 85% of patients and 80% of patients achieved normal intelligibility.

Speech Assessments

Perceptual speech assessment scales varied widely between studies and are detailed in Table 2. Five studies reported the perceptual speech assessment using an in-house scale. The other five studies used previously published perceptual speech assessment scales, consisting of the Bzoch [16], the GOS.SP.ASS.'98 [17], and the Cleft Audit Protocol for Speech-Augmented (CAPS-A), which was derived from the GOS.SP.ASS.'98 [17]. Nasal air emission was rated using scales ranging from 3-point to 5-point scales. Resonance was rated using scales ranging from 3-point to 6-point scales. The speech assessments were carried out by SLPs in all the studies, however, not all studies included ratings by multiple SLPs to ensure reliability. The SLP was blinded to the study in six of the eight studies. Information about the experience level or training of the SLP for cleft palate speech errors was provided in six out of 11 studies.

Table 2. Perceptual Speech Assessment Details

Study	Perceptual assessment tool	Assessment by SLP	Assessment by more than one SLP	Blinded speech assessment
Jackson (2004)	Bzoch, 1977	Yes	No	No
Mann (2017)	In-house scale	Yes	No	No
Ahl (2016)	Cleft Audit Protocol for Speech-Augmented (CAPS-A)	Yes	Yes	Yes
Denadai (2017)	In house-scale	Yes	Yes	Yes
Dias (2016)	GOS.SP.ASS.'98	Yes	No	Yes
Hens (2013)	Cleft Audit Protocol for Speech-Augmented (CAPS-A)	Yes	No	Yes
Hill (2004)	In house-scale	Yes	Yes	Yes
Logjes (2017)	In-house scale, Nasometry	Yes	Yes	No
Mann (2011)	In house-scale	Yes	No	No
Robertson (2008)	Bzoch, 1977	Yes	No	Yes

Velopharyngeal Function Assessment and Outcomes

Overall, patients receiving the buccal flap approach for both primary palatoplasty and secondary surgery for VPI demonstrated an improvement in VP closure. "Good" VP closure was achieved in 87% of patients following primary palatoplasty, and 73% of patients following secondary surgery for VPI, across 2 studies. Post-operative imaging was included in six out of the 11 studies and is detailed in Table 1. Imaging included nasopharyngoscopy in three out of the six studies [5, 15,18] and videofluoroscopy in three out of the six studies [6, 7, 11]. Assessment of visual VP function scoring varied between studies with some using a 2-point scale to define the function of the VP mechanism, while other studies used a 4-point scale.

Surgical Outcomes

In terms of negative surgical outcomes, 7.5% of patients were reported to have complications and 7.7% required further surgery following primary palate repair. The only surgical complications reported following primary palatoplasty were fistulas. Following secondary surgery for VPI, 12.6% of patients were reported to have complications and 14.5% required further surgery. The most common surgical complications reported following secondary surgery for VPI were fistulas, dehiscence, and partial flap necrosis.

DISCUSSION

The purpose of this systematic review was to determine the effectiveness of the buccal myomucosal flap in primary and secondary cleft palate repairs, as measured by speech outcomes (primary outcome) and rate of surgical complication (secondary outcome). The buccal myomucosal flap approach aims to close the palate without tension, lengthen the palate, reconstruct the levator muscular sling, not inhibit craniofacial growth and achieve proper oral-nasal resonance for speech [5, 6, 7, 8]. However, these claims have not been rigorously investigated, as more traditional primary and secondary techniques have previously been. The success of primary palate repairs and secondary surgery for VPI is measured by speech, the development of fistulas, the need for additional surgery, and proper craniofacial growth and development [19]. This review has noted wide heterogeneity across studies in the reported sample sizes, patient criteria, and methodologies used for measuring outcomes for both speech and VP competence.

Systematic reviews have previously been conducted on surgical interventions for primary palatoplasties, specifically on the Furlow double-opposing Z-plasty and straight line intravelarveloplasty [20]. Timbang et al. [20] reported hypernasality for 11.1-20% of individuals with an isolated cleft palate and 29.1-33.3% of individuals with a unilateral cleft lip-cleft palate following the straight line intravelarveloplasty. For individuals treated with the Furlow double opposing Z-plasty, 13-14.3% of individuals with an isolated cleft palate and 8.9-18.5% of individuals with a unilateral cleft lip-cleft palate presented with hypernasality post-surgery [20]. The percentage of individuals with hypernasality following the buccal myomucosal flap (8.9%) is lower than the percentage of individuals with hypernasality from the Timbang et al. [20] review.

Surgical success is typically measured by the patient's speech and resonance. In the current review, improvements in resonance, nasal emission, and intelligibility were reported in all studies with the use of buccal myomucosal flap approach when used as a secondary speech surgery to treat VPI. A systematic review by de Blacam et al. [4] reported normal resonance in 70.7% of individuals who received a posterior pharyngeal wall augmentation, palatoplasty, a pharyngeal flap, or a sphincter pharyngoplasty, while the current review reported 74% of patients achieved normal resonance following the buccal myomucosal flap approach. Normal nasal emission was reported in 65.3% of individuals and improved intelligibility was reported in 86.5% of individuals following traditional surgical techniques for VPI [4]. The current review reported 79% of patients presented with no nasal air emission, 85% with normal speech and 80% achieved normal intelligibility post-surgery following the buccal myomucosal flap repair. Based on these comparison literature reviews, the buccal myomucosal approach reported more successful speech outcomes for resonance and nasal emission than the traditional surgical techniques for VPI. de Blacam et al. [4] reported an improvement in intelligibility while the current review reported normal speech and intelligibility. The results between the two reviews cannot be compared accurately due to the variability in the definition used for assessing overall intelligibility and normal speech.

It was observed that there is substantial variation in the reporting of speech outcomes across the study reported as part of this systematic review, and therefore results should be interpreted with caution. Half of the articles included in the review used non-standardized in-house scales. The remaining five articles included published scales but varied in the scale used for the assessment. It is important for future studies to incorporate evidence-based metrices to assess speech outcomes. Evidence based-metrices are used to establish evidence-based practices which can be incorporated into clinical practice [21]. Not only does this allow for clinicians to properly assess speech and resonance for individuals with cleft palate, but also allows to an accurate comparison of surgical outcome across studies. Henningsson et al. [22] reported the need of consistent speech parameters and speech-sampling procedures across centers in order to accurately compare results across patient groups and languages. Henningsson and colleagues

[22] provided a framework within which speech results can be reported in a consistent manner. To report speech outcomes, a set of five universal speech parameters are recommended including: hypernasality, hyponasality, audible nasal air emission and/or nasal turbulence, consonant production errors, and voice disorder. Two global parameters, speech understandability and speech acceptability, are also recommended. Allori et al. [23] also reported a standard set of outcome measures for comprehensive cleft care. Speech and communication were included as an outcome domain and included intelligibility, articulation, and VP competence as specific aspects for consideration. Validated instruments including the Intelligibility-in-Context scale, Percent Consonants Correct Scale, and VPC graded rating scale, are recommended to assess the speech and communication outcomes. It is important to note that seven articles were excluded during the search criteria because they reported an "improvement in speech" but included an inadequate description of the speech assessment used to reach that conclusion. In many cases, these appeared to be anecdotal statements that were not based on a formal method of review. The inadequate description of the speech assessment does not allow for an accurate comparison of the results to other studies. Data reporting methods varied across studies, with some only listing an improvement in speech, others reported the percentage of patients with an improvement following a statistical comparison between pre-and post-operative speech samples. In future studies, there is a need for additional statistical analysis of the results, including statistics between cleft groups and syndromes, to accurately compare results between institutions. Power analysis should also be included to ensure sample sizes contain enough power to detect differences among groups.

Four studies performed inter- or intra-reliability on the speech assessments and only six studies used speech language pathologists who were blinded to the study. The inconsistency in speech assessment methods makes it difficult to draw meaningful comparisons between studies. Intra- and inter-rater reliability should be used with a standard protocol to ensure proper speech ratings. The studies originated in multiple countries which further limits comparisons as sounds vary between languages as well as the assessments typical used in each country.

Six studies assessed VP closure using videofluoroscopies or nasopharyngoscopy. This assessment was used to determine if the surgery resulted in a competent VP mechanism. The scales used to assess the VP gap varied from a 2-point scale to a 4-point scale, therefore, making it difficult to make meaningful comparisons between studies. Although the patients were all assessed using a point scale, there was variability in the way the studies reported the outcomes. Some studies presented the data as an improvement in VP closure while others reported descriptive statistics for the closure for each point on the scale. All studies reported an increase in VP closure post-surgery, suggesting that the surgery is successful in altering the musculature necessary for the VP sphincter to function appropriately. However, the levator veli palatini muscle, the muscle that is altered in this surgical technique and is the primary muscle for velar elevation has not been assessed directly through use of imaging.

Surgical complications were reported in eight out of 11 studies, while the need for additional speech surgery was reported in nine out of 11 studies. Systematic reviews have previously been conducted on surgical interventions for primary palatoplasties, specifically on the Furlow double-opposing Z-plasty and straight line intravelarveloplasty [20] and velopharyngeal dysfunction (VPD) [4]. Timbang et al. [20] reported mean failure rates for individuals with an isolated cleft palate were 9.7% for the Furlow double opposing Z-plasty and 16.5% for the straight line intravelarveloplasty. In the unilateral cleft palate group, the authors reported mean failure rates of 11.1% for the Furlow double opposing Z-plasty and 17.1% for straight line intravelarveloplasty. The oronasal fistula rate was 7.87% in the Furlow repair group and 9.81% in the straight line intravelarveloplasty group in the Timbang et al. [20] review. Negative surgical outcomes for the buccal myomucosal flap for primary palate repair was 4.86% in the current review, suggesting the surgical technique may be more successful for primary palatoplasties. The most common surgical complications reported were fistulas, dehiscence and partial flap necrosis following the buccal myomucosal flap repair. Postoperative complications were noted in 3% of all patients undergoing a posterior pharyngeal wall augmentation, palatoplasty, pharyngeal flap or sphincter pharyngoplasty for secondary surgery for VPD [4]. However, the percentage of postoperative complications ranged from 0-6.4% between studies. The average percentage of surgical complications for the buccal myomucosal flap (12.75%) is higher than the mean (3%) for traditional secondary surgical methods for VPD from the de Blacam et al. [4] review. The interpretation of the surgical comparisons should be interpreted with caution. The systematic review by de Blacam et al. [4] included four categories of surgery for VPD including the pharyngeal flap, sphincter pharyngoplasty, palatoplasty and posterior pharyngeal wall augmentation. The systematic review consisted of 83 studies that reported perceptual speech assessment or obstructive sleep apnea while the current systematic review included only 11 studies. Both the buccal myomucosal flap studies and the studies included in the de Blacam et al. [4] review did not all include postoperative complications and also varied in the inclusion of syndromes, which would alter the overall percentage of surgical success in both reviews. de Blacam et al. [4] also reported that some studies included patients that had previously undergone surgery for speech. The differences between studies make it difficult to draw comparisons between the surgical techniques.

Timbang et al. [20] reported the need for secondary surgery in the straight line intravelarveloplasty group to be 9.1-29.2% for the isolated cleft palate group and 6.7-19.4% for the unilateral cleft-lip-cleft palate group. The need for secondary surgery in the Furlow double opposing Z-plasty was reported to be 0-11.4% in the isolated cleft palate group and 0-6.7% in the unilateral cleft-lip-cleft palate group. In the current review, 6.5% of patients required secondary surgery following the buccal myomucosal flap for primary palate repair. This is similar to the percentage of patients requiring surgery following the Furlow double opposing Z-plasty repair. In the systematic review of four categories for surgery for VPD (pharyngeal flap, sphincter pharyngoplasty, palatoplasty, and posterior pharyngeal wall augmentation), de Blacam et al. [4] reported 8.7% of patients required further surgery for speech. This is below the percentage of patients that needed further surgery following the buccal myomucosal flap for secondary surgery for VPD (14.5%). Results for the surgical comparisons should be interpreted with caution, as speech assessments varied greatly between studies in the systematic reviews, which alters the decision for secondary speech surgery.

There are limitations to the prior conclusions. There is a wide variability between the studies in the sample size, patient population, and assessment methodologies for speech and VP competence. The inconsistent use of published speech scales and VP closure ratings, as well as how data were reported may have impacted the outcome of the review. The exclusion criteria were used to control for variability between studies, but all variability cannot be eliminated. The surgeon's experience and slight differences in the buccal flap surgical technique are factors that could not be eliminated in this systematic review. The studies included in the review contained cohorts including individuals with syndromes and individuals who were nonsyndromic, as well as individuals with different cleft types. With a wide range of features in the patient population, it was not possible to extract data separately.

CONCLUSION

This systematic review suggests that the buccal flap surgical approach for both primary palate repair and secondary repair for VPI may improve patients' resonance and nasal air emission as well as improve VP closure. The buccal flap surgery used to correct VPI has previously been reported to effectively improve or correct hypernasal speech and minimize the risk of obstructive sleep apnea and hyponasality [24]. The current systematic review and a systematic review for secondary surgery to correct VPI [24] both report improvements in speech outcomes and VP function, suggesting the buccal flap can be beneficial for both primary palatoplasty and secondary surgery for VPI. Future studies should consist of randomized clinical trials to determine the efficacy of the buccal flap surgical approach for primary palatoplasty. There is a need for standardized methods to assess speech and VP competence to allow for precise comparisons between results at different institutions.

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CHAPTER 2

INTRODUCTION

Individuals born with cleft palate present with various degrees of anatomical differences in the VP muscles: Anatomical differences can result in abnormalities in the function of the VP muscles and their ability to achieve VP closure required for normal speech (Ha et al., 2007; Tian et al., 2010a; Tian et al., 2010b; Perry et al., 2011; Kotlarek et al., 2017; Perry et al., 2018). The VP sphincter consists of the velum and pharyngeal walls, which close during speech to separate the oral and nasal cavity. This process is necessary for proper oral-nasal resonance in speech. VP dysfunction occurs when the VP sphincteric mechanism fails to create a seal between the oral and nasal cavity. Predictors of VP dysfunction include short soft palate length and an increased pharyngeal depth. These anatomical differences can result in a VP gap and hypernasal speech (D'Antonio et al., 2000). Velar stretch, the ability for the velum to increase in length from rest to elevation during phonation, has been reported to be a predictor for adequate VP closure necessary for proper resonance (Pruzansky & Mason, 1969).

The goal of primary palatoplasty, performed around the child's first birthday, is to create a separation between the oral and nasal cavities and to create proper elevation and retraction of the velum against the posterior pharyngeal wall during speech and swallowing. Primary palatoplasty aims to reconstruct the palate and release and reorientate the levator veli palatini muscles. The levator veli palatini muscle is the primary muscle of velar elevation, therefore, the release and reorientation of the muscle facilitate mobility of the velum necessary to achieve VP closure (Smith & Kuehn, 2007). In non-cleft anatomy, the levator veli palatini muscle forms a cohesive sling through the velum. For individuals with cleft palate, at birth the levator veli palatini muscles are abnormally attached to the palatine bones, which prevents adequate mobility of the muscle. The success of primary palatoplasties is based on: (1) normal resonance (assessed when speech develops) and (2) normal maxillofacial growth (assessed after maxillofacial growth). It is reported that as many as 20% of patients will present with VP dysfunction post-surgery and require a secondary surgery for speech (Fisher & Sommerlad, 2011; Lithovious et al., 2014).

Patient-specific surgical planning is proposed to improve these poor outcome rates: Primary palatoplasties are typically performed between 6-12 months of age. Kollara et al. (2014) and Perry et al. (2018) have demonstrated significant anatomical differences between race, gender, and cleft anatomy. The authors suggest that surgery for cleft palate may be optimized by tailoring surgery to be patient-specific based on the pre-surgical anatomy. Tailoring the surgery to the specific patient allows for the establishment of a VP system that is a mechanistically advantageous system for speech. Perry et al. (2018) further proposed that the distance from the posterior palate to the location of the levator veli palatini muscle in the velum is a critically important anatomic arrangement for normal speech. Specifically, among individuals with repaired cleft palate and normal speech the VP anatomy varied significantly among the study group and compared to a control non-cleft group. However, this effective velar length distance was the single variable that remained constant among all participants with normal resonance.

Computational modeling has been used to study the cause-effect relationship between variations in VP anatomy and VP function. Inouye et al. (2015a) examined different surgical reconstructive procedures utilizing varying degrees of levator veli palatini overlap. The results suggest that there is an optimal level of overlap of the levator veli palatini muscle to maximize the ability of the levator to generate proper closure force and therefore normal speech. This is of importance because there is variability in the range of overlap of the levator veli palatini muscle in different surgical techniques. Inouye et al. (2015b) assessed how anatomic variability impacts the VP mechanism. It was reported that some anatomical parameters have a greater influence on VP mechanics. Velar length was reported to be influential to closure force. Effective interventions to improve VP function include decreasing VP distance and increasing the levator veli palatini cross-sectional area. Velum stiffness, possibly due to scar tissue, was also reported to play a role in VP closure force. If the velum is stiffer, the VP distance, velum-levator veli palatini angle and then velar length may be even more important to achieve proper VP closure. Decreasing the VP distance can involve autologous fat transplants, surgery with levator veli palatini overlap, or retropositioning (Inouye et al., 2015b). The musculus uvulae is also reported to be involved in VP closure, however, this muscle is dysmorphic or absent in individuals with cleft palate. Inouye et al. (2016) found that the musculus uvulae acts as a space-occupying structure and velar extensor. It is suggested that for individuals with cleft palate, the implantation of autologous or engineered tissues at the velar midline may assist in VP closure (Inouye et al., 2016). In summary, results from these computational modeling studies further emphasize that patient-specific surgical planning is important for individuals with disadvantageous anatomy, such as cleft palate, to ensure adequate VP closure necessary for normal speech.

The literature lacks documentation of the anatomic outcomes and the effects of different surgical techniques: Before understanding patient anatomies, it is critical to document and define how different types of primary palatoplasty techniques affect or change the VP anatomy. Knowledge of anatomical outcomes is important for surgeons to decide which technique would be effective based on the patient's pre-surgical anatomy. Studies on common surgical interventions report speech outcomes to assess post-surgical outcomes. Perceptual speech assessments of resonance are conducted following primary palatoplasty at 3-7 years of age, to assess for hypernasality, weak consonant production, and nasal air emission and to determine if the primary palatoplasty was successful. Timbang et al. (2014) performed a systematic review to compare the success of the Furlow Double Opposing Z-Plasty and the Straight-Line Intravelar Veloplasty methods for cleft palate repair. The studies included in the review reported fistula rates and speech outcomes. <u>The review failed to describe the alterations to the anatomy that resulted in the reported speech outcomes</u>. The success of primary palatoplasties is also based on normal maxillofacial growth. Individuals born with cleft palate frequently have facial growth abnormalities including Class III malocclusion, maxillary hypoplasia, and retrusion and a concave appearance to their facial profile (Oberoi et al., 2008). Maxillary deficiency following primary palatoplasty is thought to be related to an intrinsic primary defect and caused by scarring from surgical reconstruction (i.e., primary palatoplasty). As a result, approximately 27% of patients with repaired unilateral cleft lip and palate undergo orthognathic surgery when midface growth is complete (Ross, 1987; Oberoi et al., 2012). <u>Therefore, studies should assess alterations to both VP and craniofacial variables following primary palatoplasty to determine the effects of different surgical techniques</u>.

Evaluating the surgical claims and outcomes by quantifying the anatomical changes can validate or refute the claims and allow for more patient-specific surgical selection. Different surgical techniques have different claims on the effect of the anatomy (Table 3). Literature on surgical techniques, such as the Furlow Z-plasty, originally reported the surgery lengthened the palate. This claim has since then been confirmed using quantitative measurements on the anatomy post-surgery, using visualization. Kotlarek (2019) examined the use of the buccal fat pad graft in primary palatoplasties, which involves the placement of the pedicled buccal fat pad graft at the palatine aponeurosis. At five years post-operatively, persistence of the fat pad was observed and an increase in velar length and thickness was reported. The current study examined a surgical approach that similarly functions to provide increased velar tissue. However, unlike the buccal fat pad which uses adipose tissue, a buccal flap approach involves the use of a muscular band of tissue from the buccinator muscle. *We hypothesized the use of a muscular tissue within the velum would provide an advantageous VP system for VP closure.* The addition of the muscle and the muscle's blood supply claims to add tension free closure and the repositioning of the levator veli palatini muscles in a more anatomical position (Mann et al., 2017).

Table 3. Primary palatoplasty surgical techniques and surgical outcome claims.

Surgical Technique	**Surgical Claims**
Furlow Double Opposing Z-Plasty	Lengthens the palate and tightens the muscular sling
Intravelar Veloplasty	Release and reorientation of the levator
Buccal Fat Pad Graft	Lengthens the palate, thickens the velum
Buccal Flap	Lengthens the palate

Study outcomes related to the buccal flap approach have failed to document the resulting anatomic changes in the VP anatomy: The buccal flap surgery is a technique that falls within the anatomic cleft restoration philosophy. Specifically, the technique is unique in that it involves the use of a buccal flap consisting of buccal muscle tissue, raised from the inner aspects of the cheeks, which is retropositioned in various positions depending on the cleft type to restore the hard and soft palate defect(s). These techniques have been outlined by Robertson et al. (2008)

and Mann et al. (2017). For simplicity, the term buccal flap approach will be used throughout the paper. The surgical approach aims to close the palate without tension, lengthen the palate by allowing the velum to move posteriorly, reconstruct the levator muscular sling, not inhibit craniofacial growth, and achieve proper oral-nasal resonance for speech (Chen & Zhong, 2003; Jackson et al., 2004; Robertson et al., 2008; Ahl et al., 2016). Studies have reported an improvement in resonance, nasal air emission, and overall speech intelligibility following the buccal flap approach. The literature on the buccal flap approach has presented promising perceptual results but fail to evaluate the anatomical outcomes to achieve VP closure. This study evaluated the surgical claims and outcomes by visualizing and quantifying the anatomical changes post-surgery following craniofacial maturation. Results from the study aimed to validate or refute the surgical claims which can influence patient-specific surgical choices based on the patient's anatomy.

The overarching aim of this study was to characterize the anatomic and physiologic changes as a result of the buccal flap surgical approach. Specifically, we used magnetic resonance imaging (MRI) to analyze VP and craniofacial anatomic changes following the buccal flap approach. We also analyzed the function of the musculature during phonation. The anatomic parameters were then characterized by comparing individuals with the buccal flap approach to individuals with non-cleft anatomy.

CHAPTER 3

METHODS

Participant Demographics

Participant distribution is presented in Table 4. Participants included a total of 20 adult males between the ages of 19-30 years of age. Participants in the two groups were aged matched up to 6 months of age. Specifically, groups included:

-10 adults born with cleft palate who received the buccal flap surgical approach at the time of primary palatoplasty

-10 adults without a history of cleft palate

The age of 19-30 years was selected to ensure the primary palatoplasty was successful for speech and did not inhibit craniofacial growth. To control for the impact of additional craniofacial surgeries on the outcomes, the participants in the cleft palate group who received the buccal flap surgical approach at the time of primary palatoplasty did not receive a secondary speech surgery or a maxillary advancement surgery. This age range was selected because it is beyond the peak of maxillofacial growth. The maxilla reaches maturity at age 14 for females and age 15 for males. The mandible reaches maturity at age 14 for females and age 16 for males (Waitzman et al., 1992a, Waitzman et al., 1992b). Due to the known racial impact on velopharyngeal variables (Kollara et al., 2016), only one racial group was used in this study.

Speech ratings were made by a primary SLP rater with extensive experience in the perceptual assessment of speech in individuals with cleft palate. A secondary SLP rated the speech recordings to allow for inter-rater reliability. Each SLP rated the audio recorded speech sample using the Cleft Audit Protocol for Speech-Augmented-Americleft Modification (CAPS-

A-AM) ratings paradigm, with hypernasality rated on a 5-point scale (0=absent, 1=borderline/minimal, 2=mild, 3=moderate, 4=severe).

Table 4. Participant distribution among groups.

N=30	**Group**	**Description**
10	Cleft with buccal flap	Primary palatoplasty with buccal flap approach
10	Non-cleft	Normative control group

Inclusion criteria. To be enrolled in either group, all participants must have been adult males between 19-30 years of age. Participants in the buccal flap group had a Veau 3 or 4 cleft palate and received a primary palatoplasty with the buccal flap approach by the same surgeon (Dr. Robert Mann).

Exclusion criteria. Participants were excluded if they presented with syndromes or musculoskeletal disorders. Individuals who received a secondary surgery for VPI or received orthognathic surgery were excluded. Adults who were unable to consent for themselves or did not speak English were also excluded.

Justification for age and race selection. Adult participants between the ages of 19-30 years of age were selected because maxillofacial growth is complete by this age (Waitzman et al., 1992a, Waitzman et al., 1992b). This age range was used to ensure the primary palatoplasty was successful for speech and did not inhibit craniofacial growth. Due to the known racial impact on velopharyngeal variables (Kollara et al., 2016), only one racial group was used in this study.

Recruitment. Participants in the buccal flap group (n = 10) were identified via chart review and as they presented at the craniofacial clinic. Potential participants were contacted via phone, mail, or during a clinic appointment and were recruited to participate in the study. Those contacted by phone were provided a predetermined script that outlined the study purpose and discussed what their participation would involve as well as the risks/benefits associated with the study. An additional 9 Caucasian male adults and 1 Hispanic male adult with non-cleft anatomy were identified through the use of a flyer that described the study goals and details regarding participant involvement. Potential participants who contacted the researchers and meet all inclusion and exclusion criteria were provided a predetermined script that outlined the purpose of the study and what their participation would involve. They were also informed of the risks/benefits associated with the study. The non-cleft control participants were age-matched, within six months of age, to the participants within the buccal flap group.

Experimental procedure. This study utilized a prospective study design for the assessment of anatomical and physiological variables. The post-operative MRI data were collected at a single time point.

Surgical methods of the buccal flap approach. All primary palatoplasties using the buccal flap approach were completed by the same craniofacial surgeon (Dr. Robert Mann) of Spectrum Health/Helen DeVos Children's Hospital. The participants received a Z-plasty with the buccal flap approach.

The Furlow double-opposing Z-plasty is a popular technique for primary palatoplasties and is commonly performed among surgeons. The Furlow palatoplasty involves the reconstruction of the levator veli palatini sling and lengthens the velum. This technique lengthens the velum by borrowing tissue from the width of the velum. The palate is closed using opposing Z-plasties, one placed in the oral mucosa and one placed within the nasal mucosa, and results in a scar that looks like a “Z”, as seen in Figure 2. The levator muscle is contained within the mucosal flap and freed from the posterior hard palate. The levator muscle bundles overlap in the posterior soft palate when the flaps are rotated and sutured together (Furlow, 1986).

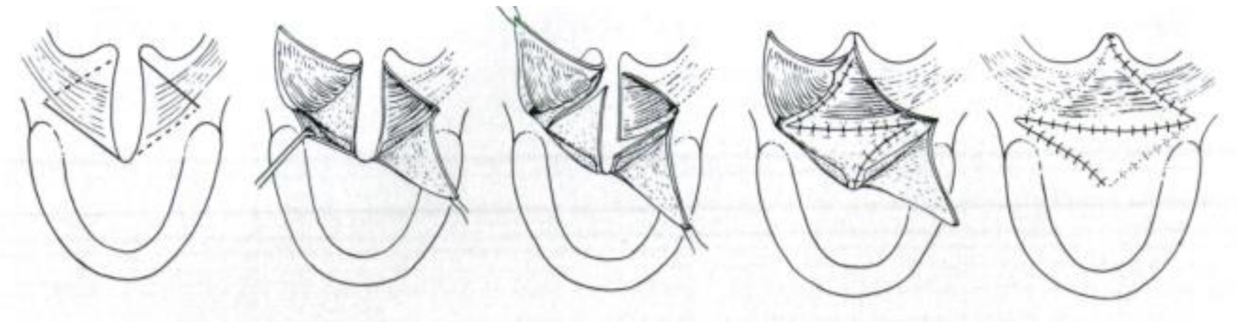


Figure 2. Primary palatoplasty using the Furlow double-opposing Z-plasty. The Z-plasty flaps are elevated and transposed to overlap the palatal muscles.
Source: Furlow, 1986.

The buccal flap surgical approach expands on the Furlow double-opposing Z-plasty and adds buccal flaps from the buccinator muscle to the defect between the hard and soft palate. The cleft margins are incised and the nasal and oral palatal mucoperiosteum is elevated. The Z-plasty flaps are then outlined on the oral surface, similarly to Furlow’s original description. The buccal flaps are then raised and average 3.5 to 5.0 cm in length and 1.2 cm in width. The mucosa is incised along the full length of the flap, revealing the buccinator muscle below. The flap is then elevated from the distal to proximal end, leaving a thin layer of the buccinator muscle over the buccal fat. The flap is rotated so the mucosa faces nasally and bridges over the retromolar trigone and into the defect. The buccal flap is then sutured to the posterior cuff of mucosa along the edge of the hard palate. The Z-flaps are then interdigitated posterior to the buccal flap in a standard Z-plasty fashion. The intravelar sling is completed after the oral myomucosal Z-flap is sutured over the nasal myomucosal flap. A second buccal flap is then raised and rotated to have its mucosa facing orally and sutured into the L-shaped defect, as seen in Figure 3. The buccal flaps may be divided in a second procedure (Mann et al., 2017).

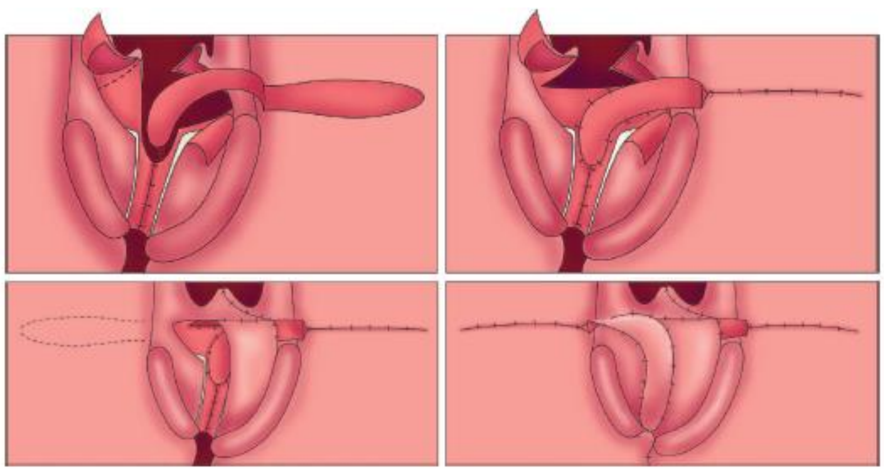


Figure 3. Primary palatoplasty using the buccal flap approach. The buccal flaps are raised and folded into the defect. The Z-flaps are then interdigitated in a standard Z-plasty fashion. Source: Mann et al., 2017

Magnetic resonance imaging. Dr. Perry's previously collected data demonstrate the validity and reliability of using the developed MRI protocols in adults, children, and infants. Spectrum Health/Helen DeVos Children's Hospital, in Grand Rapids, MI, is equipped with a 3 Tesla Siemens MRI machine. Participants who received the buccal flap surgical repair were scanned on the 3 Tesla Siemens MRI machine. The non-cleft participants were scanned at Vidant Medical Center in Greenville, NC. Vidant Medical Center is equipped with a 1.5 Tesla Siemens MRI machine. The scanning protocol outlined below was established on both scanners. Participants were not sedated. Head rotation was minimized using cushions around the head. A high resolution, T2-weighted turbo-spin-echo three-dimensional (3D) anatomical scan called SPACE was used to acquire a large field of view covering the oropharyngeal anatomy (25.6 x 19.2 x 15.5 cm) with 0.8 mm isotropic resolution with an acquisition time of slightly less than 5 minutes (4:52). A midsagittal 2D (two-dimensional) scan with equal resolution was obtained during production of the sounds "S" and "E". These 2D scans took about 7.8 seconds each and allowed the velum to be viewed in an elevated position during phonation. These imaging sequences, described previously (Bae et al., 2011, Perry et al., 2013), provided a 3-D data set of the structures of interest.

Imaging analysis. Image processing was completed using Thermo Scientific™ Amira™ Software (Thermo Fisher Scientific, Waltham, MA, US). Amira has a built-in native Digital Imaging and Communication in Medicine (DICOM) support program which preserves the original anatomical geometry of the data. All measurements were performed by two researchers trained in MRI data analysis using the software. The primary researcher measured all data and was blinded to the participant groups during image analysis. On the midsagittal plane, the following linear velopharyngeal variables were measured: effective velar length, velar thickness, velar length, velar length during phonation, and VP portal depth (measured from the posterior hard palate to the posterior pharyngeal wall (PPW) or adenoid pad). The following craniofacial angles were measured: sella-nasion-subspinale (SNA) angle, sella-nasion-supramentale (SNB) angle, subspinale-nasion-supramentale (ANB) angle, and the anterior cranial base angle (nasion-sella-basion (NSBa) angle). The effective VP ratio was obtained by dividing the effective velar length by the VP portal depth. Velar stretch was calculated by taking the velar length during phonation and subtracting the velar length at rest. Description of variables is presented in Table 5 and visualized in Figures 4-6. Intra- and inter-rater reliability was established by re-measuring all four craniofacial angles for each participant and was completed by two researchers with experience in 3D MRI data analyses.

Table 5. Description of Variables.

Variable	Description	Reference Image
Velar thickness	Distance (mm) from the velar knee to the velar dimple	Figure 4
Velar length	Curvilinear distance (mm) from the posterior hard palate to the tip of the uvula on the midsagittal plane	Figure 4
VP portal depth	Linear distance (mm) from the posterior hard palate to the posterior pharyngeal wall (PPW) or adenoid pad as seen on the midsagittal image	Figure 4
Effective velar length	Linear distance (mm) from the posterior hard palate to the levator muscle bundle as seen on the midsagittal image with the oblique coronal plane overlaid	Figure 5
Effective VP ratio	Calculation obtained from dividing the effective velar length by the VP port depth	
Nasion-sella-basion (NSBa) angle	Anterior cranial base angle (degrees) created from the nasion to sella to basion as seen on the midsagittal image	Figure 6
Sella-nasion-subspinale (SNA) angle	Angle (degrees) created from the sella to nasion to subspinale as seen on the midsagittal image	Figure 6
Sella-nasion-supramentale (SNB) angle	Angle (degrees) created from the sella to nasion to supramentale as seen on the midsagittal image	Figure 6
Subspinale-nasion-supramentale (ANB) angle	Angle (degrees) created from the subspinale to nasion to supramentale as seen on the midsagittal image	Figure 6
Velar stretch	Calculation obtained by the following formula (measured in mm): Velar $\text{length}_{\text{phonation}}$ –velar $\text{length}_{\text{rest}}$	Figure 7

Figure 4. Midsagittal images of linear variables measured. Velopharyngeal variables include the VP portal depth, velar length, and velar thickness.

Figure 5. Midsagittal image with an oblique coronal image plane overlaid. The variable effective velar length is measured from the posterior border of the hard palate to the insertion of the levator veli palatini muscle.

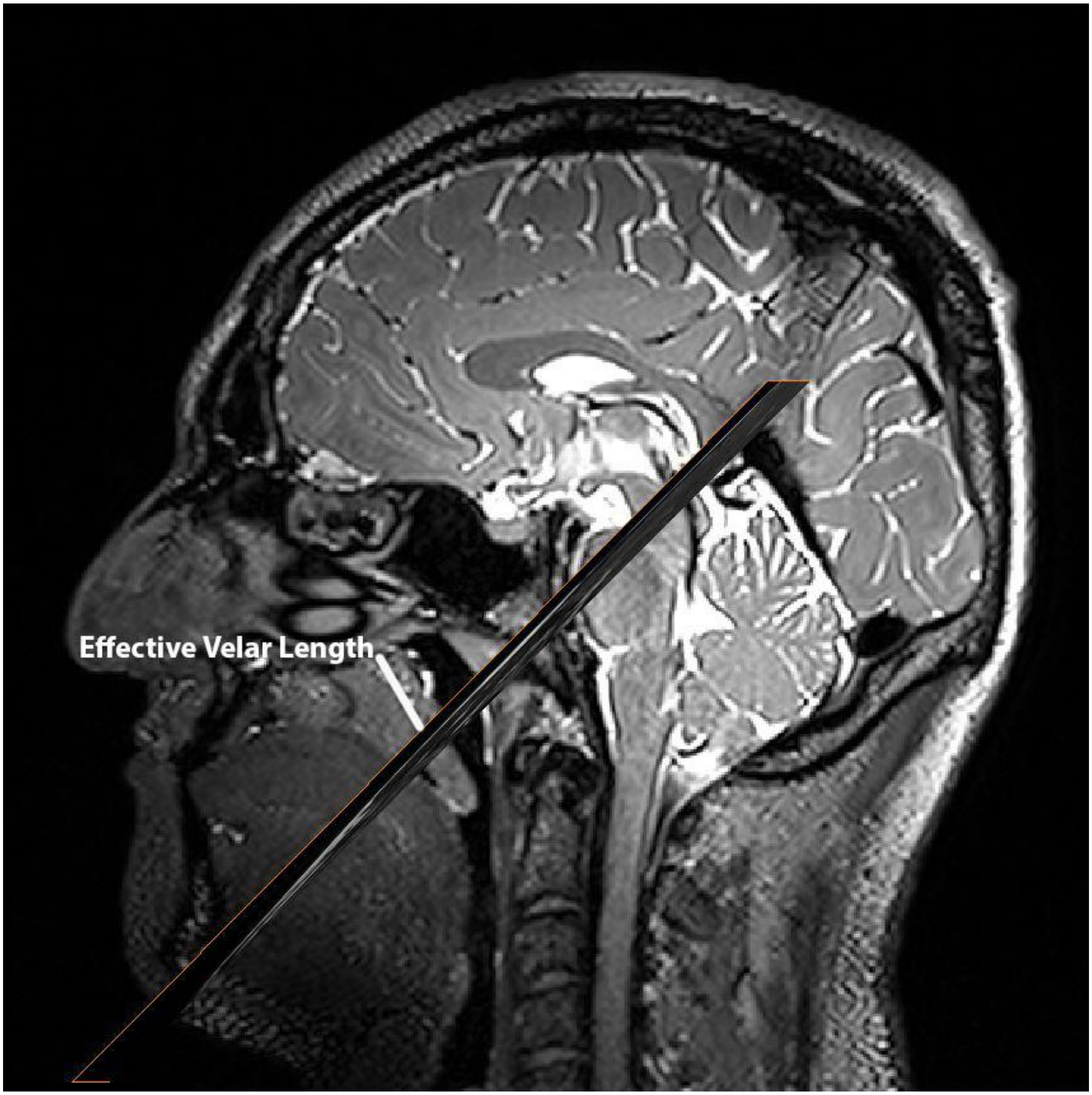
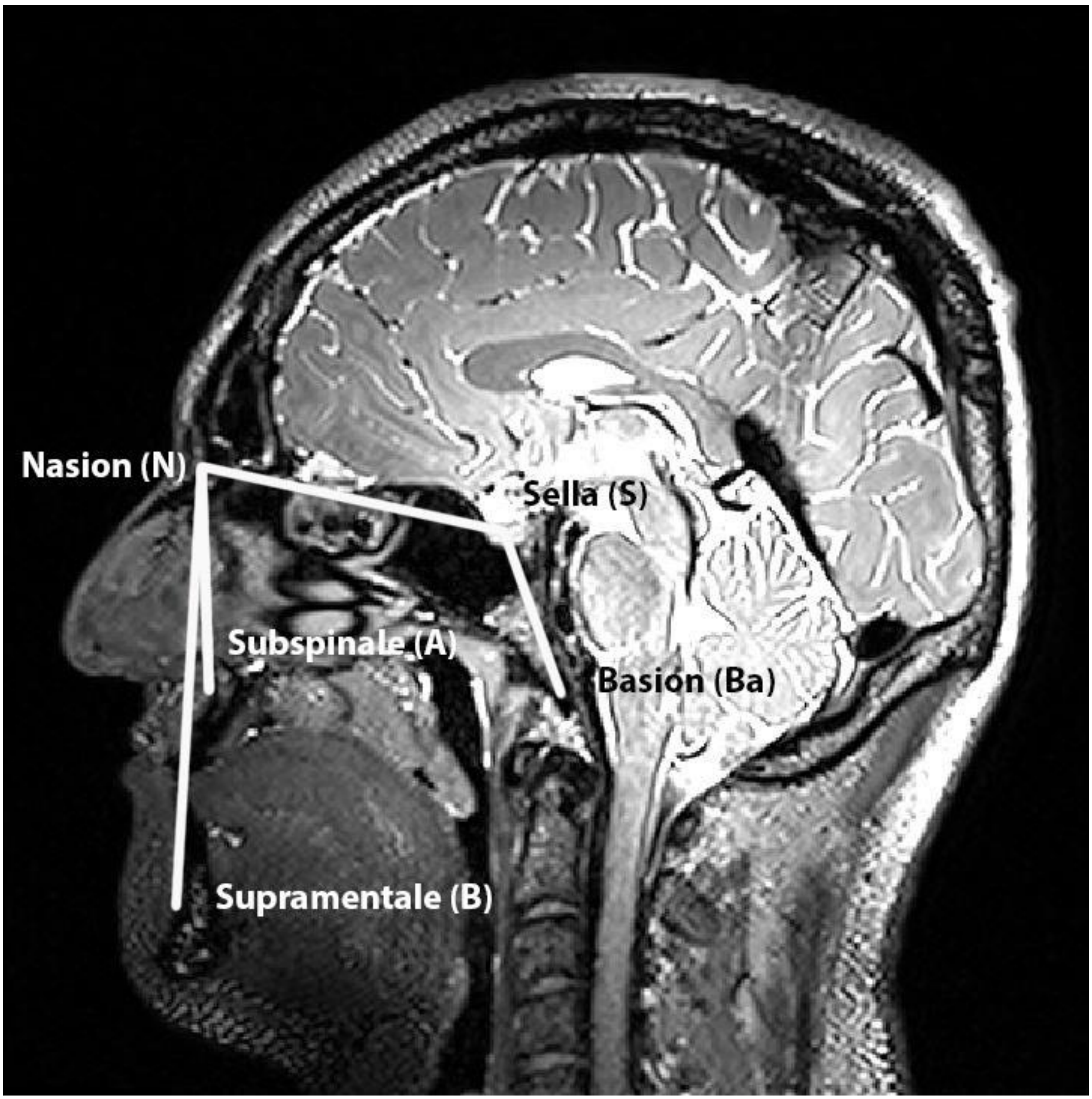


Figure 6. Angular craniofacial measurements taken from the midsagittal image include the sella-nasion-subspinale (SNA) angle, sella-nasion-supramentale (SNB) angle, subspinale-nasion-supramentale (ANB) angle, and nasion-sella-basion (NSBa) angle.



Statistical analysis. Statistical analyses were conducted using IBM SPSS 27.0 (IBM Corp, Aramonk, NY). Descriptive statistics (mean, standard deviation, median, minimum, maximum) for each group were used to compare velopharyngeal and craniofacial measurements. An Analysis of Covariance (ANCOVA) was utilized to compare the values obtained across the two groups, while controlling for height and weight and using robust standard errors to account for heterogeneous variances. Levene's test of equality of variances showed variables with unequal variances ($p < .05$). The assumption of normality was met for all variables, as assessed by Shapiro-Wilk's test ($p > .05$).

Reliability. Intra- and inter-rater reliability was established by re-measuring all cranial base angles due to angles having the lowest reliability in previously reported MR imaging studies of the VP mechanism (Table 6). An intra-class correlation coefficient (ICC) ($\alpha = .05$) was performed to determine intra- and inter-rater reliability. Measurements consisted of NSBa angle, SNA angle, SNB angle, and ANB angle. One participant was excluded from reliability for the SNA angle, SNB angle, and ANB angle due to an image artifact from the participant's dental appliance in the location of the craniofacial measurements. ICC estimates were based on a two-way-random model using a 95% confidence interval. Intra-rater agreement for NSBa angle was .951, indicating excellent reliability. Inter-rater agreement for NSBa angle was .813 indicating good reliability. Intra-rater agreement for SNA angle was .962, indicating excellent reliability. Inter-rater agreement for SNA angle was .889 indicating good reliability. Intra-rater agreement for SNB angle was .901, indicating excellent reliability. Inter-rater agreement for SNB angle was .800 indicating good reliability. Intra-rater agreement for ANB angle was .841, indicating good reliability. Inter-rater agreement for ANB angle was .584 indicating poor reliability.

Differences in the methodology used to measure the ANB angle may be a reason for the poor inter-rater reliability.

Table 6. Intraclass Correlation Results for Reliability Measures

Variable	Inter-rater Reliability		Intra-rater Reliability	
	ICC	95% Confidence Interval	ICC	95% Confidence Interval
NSBa angle (º)	.813	.586-.921	.951	.880-.980
SNA angle (º)	.889	.735-.956	.962	.903-.985
SNB angle (º)	.800	.552-.918	.901	.761-.961
ANB angle (º)	.584	.189-.816	.841	.633-.936

Abbreviations: ICC, intraclass correlation coefficient; SNA, sella-nasion-subspinale; SNB, sella-nasion-supramentale; ANB, subspinale-nasion-supramentale; NSBa, nasion-sella-basion.

CHAPTER 4

RESULTS

Magnetic resonance images were successfully obtained on all subjects using the protocol outlined in the present study. Participants in the study ranged from 19.24 to 30.79 years of age. The median participant age was 23.87 years. The participants in the buccal flap group ranged from 19.24 to 30.79, with a median age of 23.68. The participants in the non-cleft group ranged from 19.92 to 30.61, with a median age of 23.87. Of the participants with a repaired cleft palate, two had a bilateral cleft lip and palate and eight had a unilateral cleft lip and palate. All reported primary palate repairs occurred between the ages of 7-14 months. Hypernasality was rated for all subjects using the 5-point scale. All participants in the control group demonstrated normal resonance. Out of the 10 participants with cleft palate, five were rated as having normal resonance (no hypernasality; rating 0), three were rated as having minimal hypernasality (rating 1), and two had mild hypernasality (rating 2). No participants presented with moderate (rating 3) or severe hypernasality (rating 4).

Within the group with the buccal flap approach, the craniofacial variables SNA angle, SNB angle, and ANB angle were not measured for one participant due to an MR image artifact from the participant’s dental appliance. All results are listed in Table 7.

Table 7. Group Means and Standard Deviations and Results of ANCOVA.

Measures	Mean and Standard Deviations for Non-cleft	Mean and Standard Deviations for Buccal Flap Group	Test for Group Difference[a]	
			F(1, 16)	p-value
Velar thickness	12.02 ± 1.25 mm	11.38 ± 2.48 mm	0.32	.583
Velar length	40.32 ± 3.66 mm	37.80 ± 4.44 mm	1.76	.203
VP portal depth	25.88 ± 1.62 mm	23.90 ± 4.62 mm	1.29	.273
Effective velar length	16.37 ± 1.49 mm	20.01 ± 2.04 mm	18.27	.001*
Effective VP ratio	0.63 ± 0.06	0.87 ± 0.18	13.16	.002*
NSBa angle (º)	124.73 ± 3.95	125.68 ± 6.74	0.16	.696
SNA angle (º)	88.02 ± 3.73	77.08 ± 3.17	40.40	< .001*
SNB angle (º)	83.75 ± 2.48	80.82 ± 2.42	5.64	.031*
ANB angle (º)	4.16 ± 2.12	4.44 ± 1.95	0.06	.938
Velar stretch	5.39 ± 1.92 mm	1.40 ± .99 mm	31.04	< .001*

Abbreviations: VP, velopharyngeal; SNA, sella-nasion-subspinale; SNB, sella-nasion-supramentale; ANB, subspinale-nasion-supramentale; NSBa, nasion-sella-basion.

[a] ANCOVA controlling for height and weight.

*$p < .05$

Aim I: To define anatomic VP and craniofacial changes related to the use of the buccal flap repair to determine if there are any anatomical differences between the individuals with the buccal flap approach group and the non-cleft group.

We hypothesized that the participants with the buccal flap repair would display a similar effective velar length, velar length, velar thickness, VP portal depth, and effective VP ratio post-surgically compared to that of the non-cleft controls. The buccal flap is claimed to lengthen the palate and provide good reconstruction of the levator muscle while not inhibiting maxillofacial growth (Chen & Zhong, 2003; Jackson et al., 2004; Robertson et al., 2008; Ahl et al., 2016). We also hypothesized that the participants with the buccal flap approach repair would demonstrate skeletal alignment similar to the non-cleft controls. The added muscle will in theory not constrict the growth of the maxilla which would decrease the likelihood of a class III malocclusion and therefore demonstrate a similar skeletal alignment to the non-cleft control. The results from the study did not support every hypothesis.

Effective velar length. Mean values were statistically significant between the two groups ($p = .001$). Effective velar length means increased from the non-cleft group (16.37 mm) to the buccal flap group (20.01 mm).

Velar length. Velar length means increased from the buccal flap group (37.80 mm) to the non-cleft group (40.32 mm), but these differences were not statistically significant between groups ($p = .203$).

Velar thickness. Velar thickness means increased from the buccal flap group (11.38 mm) to the non-cleft group (12.02 mm). These differences were not statistically significant between the two groups ($p = .583$).

VP portal depth. VP portal depth means increased from the buccal flap group (23.90 mm) to the non-cleft group (25.88 mm), but these differences were not statistically significant between groups ($p = .273$).

Effective VP ratio. Mean values were statistically significant between the two groups ($p = .002$). The effective VP ratio increased from the non-cleft group (0.63) to the buccal flap group (0.87).

SNA angle. Mean values were statistically significant between the two groups ($p < .001$). The SNA angle was larger for the non-cleft group (88.02°) compared to the buccal flap group (77.08°).

SNB angle. Mean values were statistically significant between the two groups ($p = .031$). The SNB angle was larger for the non-cleft group (83.75°) compared to the buccal flap group (80.82°).

ANB angle. The ANB angle was larger for the buccal flap group (4.44°) compared to the non-cleft group (4.16°), but these differences were not statistically significant between groups ($p = .938$).

NSBa angle. The NSBa angle was larger for the buccal flap group (125.68°) compared to the non-cleft group (124.73°). These differences were not statistically significant between the two groups ($p = .696$).

Aim II: To determine if the use of the buccal flap approach results in similar velar stretch compared to non-cleft adults.

We hypothesized that the individuals with the buccal flap approach would demonstrate a velar stretch similar to the non-cleft adults. Velar stretch, a quantitative measure is used to assess

VP function during speech. Velar stretch is dependent on available muscle mass, and the range and speed of movement during speech (Pruzansky & Mason, 1969). The buccal flap approach places a vascularized and innervated muscle between the hard and soft palate, adding to the available muscle mass. Therefore, we hypothesized that the buccal flaps would aid in velar stretch and allow the individuals to achieve a velar stretch similar to those with non-cleft anatomy. The results of the present study did not support our hypothesis.

Velar stretch. Mean values were statistically significant between the two groups ($p < .001$). Velar stretch means increased from the buccal flap group (1.40 mm) to the non-cleft group (5.39 mm).

CHAPTER 5

DISCUSSION

The buccal flap approach for primary palatoplasty is a well-documented surgical technique. Studies have reported favorable speech and surgical outcomes that suggest the surgical technique creates a favorable VP system by closing the palate without tension, increasing velar length, decreasing the VP gap, reconstructing the levator veli palatini sling, and optimizing maxillary growth (Chen & Zhong, 2003; Jackson et al., 2004; Robertson et al., 2008; Ahl et al., 2016).

Previous studies have not utilized MRI to assess surgical outcomes following the buccal flap approach. Data from this study were collected using MRI to assess anatomical changes to the VP portal and craniofacial changes as a result of the buccal flap approach and were compared to non-cleft participants. Quantitative data for VP and craniofacial variables were obtained to examine the anatomic and physiological impact on VP closure following the buccal flap approach for primary palatoplasty.

Anatomical Changes (Aim I)

Aim I was designed to quantify the VP and craniofacial dimensions of adults surgically treated with the buccal flap approach and compare the anatomical measurements to adults with non-cleft anatomy to determine if there are significant differences between the groups. Significant differences between groups were observed for effective velar length, effective VP ratio, SNA angle, and the SNB angle.

Effective velar length was greater in the buccal flap group in comparison to the non-cleft group. The effective velar length is the portion of the velum that contributes to VP closure during speech. It is the distance between the posterior border of the hard palate and the insertion of the levator veli palatini muscle in the velum. During speech, the rest of the velum hangs below the contact region between the velum and the pharynx and therefore does not assist in VP closure. Results from the study suggest that the participants who received the buccal flap approach have a longer effective velar length; therefore, the levator veli palatini sling is placed more posteriorly during surgery compared to individuals with non-cleft anatomy. This places the levator veli palatini muscle in a more favorable position for VP closure and likely contributes to VP competence. Therefore, the effective velar length may be a significant predictor for a successful palate repair and a predictor for any additional surgical intervention needed. The buccal flap surgical approach creates a muscular structure at the devoid area posterior to the palate. The presence of the muscle mass may decrease the migration of the levator veli palatini muscle toward the posterior palate following surgery; therefore, maintaining a more posterior position compared to individuals with a traditional surgery technique which do not aim to add tissue to the defect. The participants with the buccal flap approach in the current study did not undergo any secondary palate repairs. It is likely that the increase in effective velar length is a contributing factor to the success of the primary palatoplasty using the buccal flap approach and the reduced need for secondary surgical intervention for VPI.

The effective VP ratio was larger in the buccal flap group in comparison to the non-cleft group. The effective VP ratio (0.87) falls within the normative range for adults with non-cleft anatomy (0.43-1.35) (Haenssler, Fang & Perry, 2020). Tian et al. (2010b) reported a correlation between the effective VP ratio at rest and during sustained phonation. Tian et al. (2010b) suggest that the rest effective VP ratio is a predictor for VP competence in the anterior-posterior direction and is a good predictor of maximum velar posterior displacement during speech. This is of clinical importance because the effective VP ratio reported in the current study supports that the levator veli palatini muscle is adequately repositioned during primary palatoplasty to help achieve VP closure in the anterior-posterior direction.

Individuals with cleft palate typically have diminished maxillary growth (Krogman et al., 1975; Hayashi et al., 1976; Ozturk & Cura, 1996). Although there are conflicting results for the cause of the maxillary deficiency, it is generally agreed that it is due to a combination of intrinsic dental, skeletal, and soft tissue deficiencies as well as surgical intervention. The severity of the maxillary deficiency is dependent on many factors including cleft type, surgical repair, and the timing of the repair. In the present study, the SNA angle was significantly smaller in the buccal flap group in comparison to the non-cleft group. This is consistent with previous findings that assessed facial growth for individuals with a repaired cleft palate (Bishara, 1973; Ye et al., 2015). Ye et al. (2015) reported a smaller SNA angle for adults with an operated cleft palate in comparison to non-cleft controls. In the present study, individuals in the buccal flap group also presented with a smaller SNB angle in comparison to the non-cleft adults. A smaller SNB angle was reported for the lip-repaired patients in comparison to non-cleft controls (Ye et al., 2015). Bishara (1973) also reported a statistically smaller SNB angle between the cleft palate and control groups, indicating that the mandible is in a relatively posterior position in relation to the cranial base. The current study supports that the anterior-posterior development of the maxilla is inhibited in individuals with cleft palate repairs. Future research should assess different primary palatoplasty surgical techniques to determine how different surgical techniques alter maxillary growth.

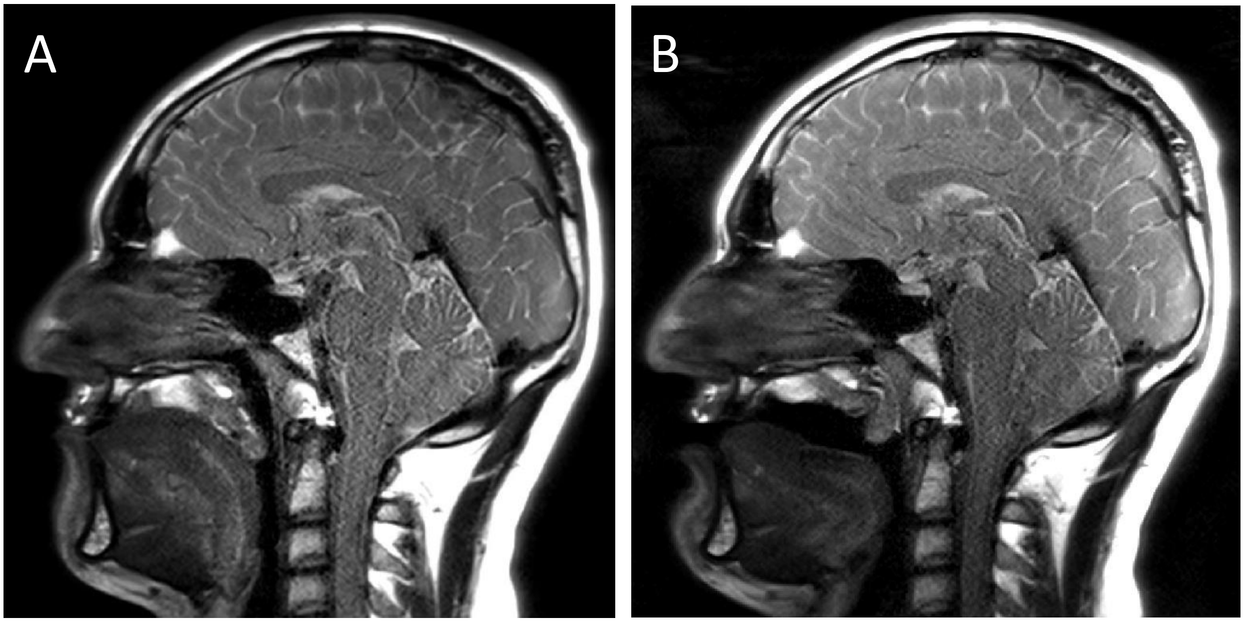
No significant differences between the groups were noted for velar length, velar thickness, VP portal depth, NSBa angle, or ANB angle. Non-significant differences confirm that the anatomical measurements between the two groups were similar. No significant difference between the groups for velar length supports previous surgical claims, although previous claims have not used MRI to assess velar length. Perry et al. (2018) reported statistically significant differences between a cleft group and non-cleft group for velar length, velar thickness, and pharyngeal depth. Previous studies have also reported significantly shorter velar lengths for individuals with repaired cleft palate compared to non-cleft anatomy (Coccaro et al., 1962, Akgüner et al., 1998, Satoh et al., 2002). This is in contrast to the findings in this study. The differences may be due to the surgical technique the adults received during primary palatoplasty. Future studies should assess these variables for individuals with cleft palate who have received different surgical techniques.

Previous literature has stated that a cleft palate does not affect the size or shape of the cranial base in individuals with nonsyndromic cleft palate (Ye et al., 2015). The current study did not find a significant difference between the groups for the NSBa angle, which supports previous literature. Consistent with previous literature (Bishara, 1973), the ANB angle was not significantly different between the individuals with cleft palate and the non-cleft controls. This finding suggests that the maxilla and mandible present with an acceptable relation to each other following surgical intervention. However, Ye et al. (2015) reported a significant difference between the operated cleft palate group and the non-cleft controls. The difference in results may be due to different surgical techniques at the time of primary palatoplasty. Ye et al. (2015) included individuals with a unilateral cleft lip and palate who received two-flap pushback primary palatoplasty, while the current study assessed individuals who received the buccal flap approach at the time of primary palatoplasty.

Physiological Changes (Aim II)

Aim II was designed to assess physiological changes following the buccal flap approach in comparison to the non-cleft group. The velum was viewed at rest and during sustained phonation (Figure 7) to calculate velar stretch, which was used to assess the physiological changes. In the present study, velar stretch was greatest for the non-cleft group. Velar stretch is dependent on available muscle mass, and the range and speed of movement during speech (Pruzansky & Mason, 1969). The buccal flap approach places a vascularized and innervated muscle between the hard and soft palate, adding to the available muscle mass. The muscle placement from the buccal flap approach may assist in velar stretch for individuals with cleft palate. Tian and Redett (2009) reported that the effective VP ratio predicted 75% of the variance for the maximal velar stretch ratio. The authors suggest that less velar stretch is required for VP closure when the individual presents with a longer effective velar length. The individuals in the buccal flap group presented with a longer effective velar length in the current study which may alter the required stretch needed to achieve VP closure. Future research should compare velar stretch between individuals with the buccal flap approach and those who received a traditional surgical repair to determine if the addition of the buccal flaps assists in velar stretch.

Figure 7. Midsagittal images of the velum at rest (A) and sustained phonation (B). The variable velar length is used to calculate velar stretch.



Clinical Implications

Results from the present study add to the literature on documenting qualitative and quantitative surgical outcomes for primary palatoplasty using the buccal flap approach. The findings from the study support many previous surgical claims and provide a novel visualization of the surgical technique post-surgery through the use of MRI. The MR images confirm the levator veli palatini muscle is repositioned in a posterior placement and is cohesive. The surgical technique not only increases the length of the velum but also increases the length of the effective velar length. It is suggested that the effective velar length and the effective VP ratio could be used to predict if an individual is likely to present with VPI (Haenssler, Fang & Perry, 2020).

Therefore, the repositioning and posterior placement of the levator sling in the buccal flap approach is essential in achieving normal VP function.

Limitations

A limitation of this study is the small sample size. The current study was designed to obtain preliminary data to investigate the anatomical and physiological outcomes following the buccal flap surgical approach for primary palatoplasty. Although many studies assessing the VP portal and mechanism using MRI have a total sample size of ten, or groups of ten (Bae et al., 2011, Bae et al., 2016, Kotlarek & Perry, 2017), a larger sample size would allow for better generalization of the surgical outcomes.

An additional limitation to the study was the participant demographics. The participants in the study were predominately Caucasian males. Findings from the current study should not be generalized to individuals with a different race due to known racial differences for VP anatomy. The participants in the buccal flap approach also presented with different cleft types. An increased sample size is also needed to control for the unequal groups for cleft type in the present study. An additional limitation was the use of sustained phonation instead of dynamic speech MRI. MRI sequences using sustained productions simplify the complexity of dynamic speech and the coarticulatory effect of sounds.

Future Directions

Future research should assess how the anatomical changes found in this study impact speech and compare data to traditional cleft palate repairs without the use of the buccal flap repair and to individuals with non-cleft anatomy. An increased sample size would improve generalization of findings and allow for better statistical power. Future research is needed to determine which surgical techniques are the most successful in repairing cleft palate and preventing the need for further surgical interventions. To ensure patients receive the best surgical technique based on their anatomy, additional research is needed to identify patient-specific factors that will predict the best surgical technique for each individual.

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CHAPTER 6

CONCLUSION

This study defined anatomic changes resulting from primary palate repair using the buccal flap approach. This study highlights the utility of using MRI to quantify the changes that occur to the VP anatomy following the buccal flap surgical approach. Significant differences between the two groups were noted for effective velar length, effective VP ratio, SNA angle, SNB angle, and velar stretch. Participants with the buccal flap approach presented with the larger effective velar length and effective VP ratio. The individuals with the buccal flap approach had smaller SNA and SNB angles as well as less velar stretch during phonation. No significant differences between the groups were noted for velar length, velar thickness, VP portal depth, ANB angle, or NSBa angle. Future studies should compare these anatomical changes to traditional cleft palate repairs without the use of buccal flap repair and to determine patient-specific factors that will predict which individuals would benefit most from this surgical technique.

APPENDICES

APPENDIX A. INITIAL IRB NOTIFICATION OF APPROVAL

EAST CAROLINA UNIVERSITY
University & Medical Center Institutional Review Board Office
1L-09 Brody Medical Sciences Building· Mail Stop 682
600 Moye Boulevard · Greenville, NC 27834
Office **252-744-2914** · Fax **252-744-2284** · **www.ecu.edu/irb**

Notification of Initial Approval: Expedited

From:	Biomedical IRB
To:	Lakshmi Kollara Sunil
CC:	
	Jamie Perry
	Jamie Perry
Date:	12/20/2011
Re:	UMCIRB 11-001103
	Variations in VP structure between upright and supine MRI in children

I am pleased to inform you that your Expedited Application was approved. Approval of the study and any consent form(s) is for the period of 12/19/2011 to 12/18/2012. The research study is eligible for review under expedited category #4 and 6. The Chairperson (or designee) deemed this study no more than minimal risk.

Changes to this approved research may not be initiated without UMCIRB review except when necessary to eliminate an apparent immediate hazard to the participant. All unanticipated problems involving risks to participants and others must be promptly reported to the UMCIRB. The investigator must submit a continuing review/closure application to the UMCIRB prior to the date of study expiration. The Investigator must adhere to all reporting requirements for this study.

The approval includes the following items:

Name	Description
assent form | History	Consent Forms
Coloring book for children | History	Additional Items
data coding | History	Data Collection Sheet
Flyer | History	Recruitment Documents/Scripts
Informed Consent Template-No More Than Minimal Risk 5-1-10.doc | History	Consent Forms
letter for parent in the mail prior to study | History	Additional Items
parental permission form | History	Consent Forms
questionnaire | History	Surveys and Questionnaires
Script (email & Telephone) | History	Additional Items
SCRIPT FOR CHILD PARTICIPANT.docx | History	Consent Forms
study protocol | History	Study Protocol or Grant Application

The Chairperson (or designee) does not have a potential for conflict of interest on this study.

IRB00000705 East Carolina U IRB #1 (Biomedical) IORG0000418
IRB00003781 East Carolina U IRB #2 (Behavioral/SS) IORG0000418 IRB00004973
East Carolina U IRB #4 (Behavioral/SS Summer) IORG0000418

APPENDIX B. IRB AMENDMENT APPROVAL LETTER

EAST CAROLINA UNIVERSITY
University & Medical Center Institutional Review Board
4N-64 Brody Medical Sciences Building· Mail Stop 682
600 Moye Boulevard · Greenville, NC 27834
Office **252-744-2914** · Fax **252-744-2284** ·
www.ecu.edu/ORIC/irb

Notification of Amendment Approval

From: Biomedical IRB
To: Jamie Perry
CC:
Jamie Perry
Jamie Perry
Date: 5/14/2018
Re: Ame21_UMCIRB 11-001103
UMCIRB 11-001103
Variations in VP structure between upright and supine MRI in children and adults

Your Amendment has been reviewed and approved using expedited review for the period of 5/13/2018 to 12/4/2018. It was the determination of the UMCIRB Chairperson (or designee) that this revision does not impact the overall risk/benefit ratio of the study and is appropriate for the population and procedures proposed.

Please note that any further changes to this approved research may not be initiated without UMCIRB review except when necessary to eliminate an apparent immediate hazard to the participant. All unanticipated problems involving risks to participants and others must be promptly reported to the UMCIRB. A continuing or final review must be submitted to the UMCIRB prior to the date of study expiration.The investigator must adhere to all reporting requirements for this study.

Approved consent documents with the IRB approval date stamped on the document should be used to consent participants (consent documents with the IRB approval date stamp are found under the Documents tab in the study workspace).

The approval includes the following items:

Document	Description
Child Assent_5_5_2018 (cleaned).docx(0.02)	Consent Forms
Child Assent_5_5_2018 (tracked).docx(0.02)	Consent Forms
Informed Consent(adults)_5_5_2018 (cleaned).docx(0.10)	Consent Forms
Informed Consent(adults)_5_5_2018 (tracked).docx(0.11)	Consent Forms
Informed Consent(children)__5_5_2018 (clean).docx(0.10)	Consent Forms
Informed Consent(children)__5_5_2018 (tracked).docx(0.11)	Consent Forms
letter for MRI (mail)_5_5_2018 (clean).docx(0.04)	Additional Items
letter for MRI (mail)_5_5_2018 (tracked).docx(0.07)	Additional Items
Parental Consent_5_5_2018 (clean).docx(0.02)	Consent Forms
Parental Consent_5_5_2018 (tracked).docx(0.02)	Consent Forms
questionnaire- updated 4_25_2018 (clean).docx(0.02)	Surveys and Questionnaires
questionnaire- updated 4_25_2018 (tracked).docx(0.02)	Surveys and Questionnaires
Script (email & phone) 5_5_2018 (clean).docx(0.02)	Additional Items
Script (email & phone) 5_5_2018 (tracked).docx(0.01)	Additional Items
Survey- Final yes no.docx(0.01)	Surveys and Questionnaires
The Research Protocol_5_5_2018 (clean).docx(0.12)	Study Protocol or Grant Application
The Research Protocol_5_5_2018 (tracked).docx(0.13)	Study Protocol or Grant Application

The Chairperson (or designee) does not have a potential for conflict of interest on this study.

APPENDIX C. SPECTRUM HEALTH IRB APPROVAL LETTER

SPECTRUM HEALTH
Human Research Protection Program
Office of the Institutional Review Board
100 Michigan NE, MC 038
Grand Rapids, MI 49503
616.486.2031
irb@spectrumhealth.org
www.spectrumhealth.org/

APPROVAL OF RESEARCH

September 16, 2019

Robert Mann, MD
Spectrum Health
Plastic Surgery & Craniofacial Serv. PC
426 Michigan NE - Suite 304
Grand Rapids, MI 49503

TYPE OF REVIEW: **Initial, Non-Committee Review**

IRB#: **2019-112** *(please reference this number in all correspondence with the IRB)*

PROTOCOL NAME: **An Anatomic MRI Comparative Analysis between Patients Born with Clefts Treated with the Z-Plasty plus Buccal Flaps Type Repair and Patients Born without Clefts**

Dear Dr. Mann:

The above referenced protocol and associated materials were reviewed and approved by the IRB via expedited review on September 12, 2019 under categories 4, 5, 6 as described in 45 CFR 46.110.

The approval period for this research began on **September 12, 2019**.

The IRB reviewed the following documents related to the approval of the research proposal:

- Initial application signed 05/08/2019
- Intake Form signed 03/07/2019
- Study protocol dated 08/09/2019
- Main Informed Consent dated 08/09/19; Healthy Volunteer Informed Consent dated 08/29/2019
- Data collection sheet dated 01/22/2019
- Phone script dated 01/31/2019
- Eligibility form dated 01/30/2019
- Anatomic MRI study participant sentences (undated)
- Letter template dated 08/09/2019

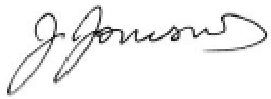
Any changes made to the study following this approval, including informed consent changes, require submission in writing to the IRB and approval by the committee. Changes may not be implemented until approved by the IRB except when necessary to eliminate apparent immediate hazards to the subject. Approval of your research means you are responsible for complying with all applicable policies and procedures of the FDA, OHRP, HIPAA, Spectrum Health, and the Spectrum Health IRB. Also, please be advised that unanticipated problems involving risk to subjects or others must be *promptly* reported to the IRB. You may reference the Investigator Manual for guidance on expectations of the IRB after approval.

Please be advised, this approval letter is limited to IRB review. It is your responsibility to ensure all necessary institutional permissions are obtained prior to beginning this research. This includes, but is not limited to, ensuring all contracts have been executed, any necessary Data Use Agreements and Material Transfer Agreements have been signed, documentation of support from the Department Chief has been obtained, and any other outstanding items are completed (i.e. CMS device coverage approval letters, material shipment arrangements, etc.).

The IRB requires submission of the Study Completion Notification to the committee. If your study has been completed, terminated, or if you do not wish to continue, please submit the Study Completion Notification.

If you have any questions please contact the Spectrum Health IRB office at 616-486-2031, email irbassist@spectrumhealth.org, or visit us on the web at www.spectrumhealth.org.

Sincerely,



Jeffrey Jones MD
Chair, Spectrum Health IRB

cc: Amanda N Rostic, MPH

APPENDIX D. SPECTRUM HEALTH IRB AMENDMENTAPPROVAL LETTER

SPECTRUM HEALTH
Human Research Protection Program
Office of the Institutional Review Board
100 Michigan NE, MC 038
Grand Rapids, MI 49503
616.486.2031
irb@spectrumhealth.org
www.spectrumhealth.org

APPROVAL OF RESEARCH

January 8, 2020

Robert Mann, MD
Plastic Surgery & Craniofacial Serv. PC
426 Michigan NE
Suite 304
Grand Rapids, MI 49503

TYPE OF REVIEW: **Modifications, Non-Committee Review**

IRB#: **2019-112** *(please reference this number in all correspondence with the IRB)*

PROTOCOL NAME: **An Anatomic MRI Comparative Analysis between Patients Born with Clefts Treated with the Z-Plasty plus Buccal Flaps Type Repair and Patients Born without Clefts**

Dear Dr. Mann:

The request for modification of approved human research and associated materials were reviewed and approved on 1/8/2020.

The IRB reviewed the following documents related to the approval of the modification:

- Modification of Approved Research xform signed December 11, 2019, adding Imani Gilbert as a sub-investigator
- Study protocol dated 1/8/2020
- Informed Consent Form Main dated 1/3/2020

This modification also clarifies the following study staff roles as follows: ECU personnel Imani Gilbert and Abby Haenssler will be coming on site to interact with but not consent patients and will have access to PHI and Dr. Perry will be accessing PHI only and will not be on site at Spectrum Health.

The IRB determined re-consenting is not required because no local subjects are enrolled.

If you have any questions please contact the Spectrum Health IRB office at 616-486-2031, email irbassist@spectrumhealth.org, or visit us on the web at www.spectrumhealth.org.

Sincerely,

Jeffrey Jones MD
Chair, Spectrum Health IRB

cc: Emily Beltz, MHA

APPENDIX E. RIGHT TO RESUE PUBLISHED MANUSCRIPT IN DISSERTATION

From: Dr. Ahmed <ahmed@synergypublishers.com>
Sent: Saturday, January 9, 2021 9:18 AM
To: Haenssler, Abigail
Subject: RE: Permission of article use for dissertation

This email originated from outside ECU.

Dear Dr Abigail Haenssler

Thanks for publishing your work in the International Journal of Speech & Language Pathology and Audiology. you are free to include your article in your dissertation.

Kind Regards
Ahmed