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Long-Term Incisal Relationships after Palatoplasty in Patients with Isolated Cleft Palate

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Abstract

Purpose—Various palatoplasty techniques have limited incisions in the hard palate due to concerns that these incisions may limit maxillary growth. There is little convincing long-term evidence to support this. Our purpose is to determine incisal relationships, an indicator for future orthognathic procedure, in patients after repair of an isolated cleft of the secondary palate.

Methods—Our craniofacial database was used to identify patients aged ten years or greater with an isolated cleft of the secondary palate who underwent palatoplasty between 1985 and 2002. Data collected included age at palatoplasty and follow-up, cleft type, associated syndrome, Robin sequence, surgeon, repair technique, number of operations, and occlusion. Incisal relationship was determined through clinical observation by a pediatric dentist and orthodontist.

Results—Seventy eligible patients operated on by 9 surgeons were identified. Class III incisal relationship was seen in 5 patients (7.1%). Palatoplasty techniques over the hard palate (63 of 70 patients) included two-flap palatoplasty, VY-pushback, and Von Langenbeck repair. There was an association between class III incisal relationship and syndromic diagnosis ($p < 0.001$). Other study variables were not associated with class III incisal relationships.

Conclusion—In patients with an isolated cleft of the secondary palate, there was no association between class III incisal relationship and surgeon, age at repair, cleft type, palatoplasty technique, or number of operations. Increased likelihood of class III incisal relationship was associated primarily with syndromic diagnosis.

Keywords

orthognathic surgery; palatoplasty; syndrome; cleft palate; reverse overjet

Background

In the era of modern cleft palate repair, there has been an effort to define the effect of operative intervention on maxillary growth and incisal relationships. However, no consensus based on long-term data has been reached. There is little data describing the effect of these

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incisions in patients with an isolated cleft of the secondary palate, with most studies referring to patients with a cleft lip and palate. Growth disturbance of the maxilla after palatoplasty has been implicated in the ensuing maxillary retrusion^{1,2}. Studies have indicated that patients with unrepaired clefts have normal maxillary growth^{3,4}, while patients who do undergo palatoplasty are at a higher risk for maxillary deficiency^{4,5,6,7,8}. These observations have led some to hypothesize that palatoplasty affects maxillary growth secondary to disturbance of the periosteum, restrictive effects of the scar, and devascularization of the bone^{9,10}. However, there is evidence that directly contradicts the studies referenced above and the true impact of palatoplasty and the exact mechanism of maxillary deficiency remains unclear^{11,12,13}.

Mars and colleagues characterized malocclusion in children with a cleft lip and palate during the early permanent dentition phase using the Goslon Yardstick¹⁴. The anteroposterior arch relationship, in particular the incisal relations, was considered to be the most important aspect of the scoring system to determine outcomes and need for orthognathic intervention. Patients with positive overjet fell into Groups 1 or 2 on the Goslon Yardstick and generally have good to excellent outcomes. Those with edge-to-edge occlusion or reverse overjet fell into Goslon groups 3-5 and have poor outcomes with the need for orthognathic surgery.

In this study, we explore the incisal relationships as a marker for future orthognathic procedure at age 10 years or greater after palatoplasty in patients with isolated cleft of the secondary palate and attempt to delineate the effects of different palatoplasty methods, syndromic diagnosis, and cleft palate type (Veau I or Veau II). It is hypothesized that different surgical techniques of cleft palate repair have no influence on the need for orthognathic surgery based on incisal relationships. This study differs from those previously published reports that focused on patients with nonsyndromic cleft palate and single surgeon outcomes, as we take into account various palatoplasty techniques by multiple surgeons and include patients with a syndromic diagnosis.

Methods

Data for this retrospective study were gathered by reviewing charts of pediatric patients who were seen at a single institution for cleft palate repair between 1985 and 2002, thereby allowing a minimum of ten years for follow-up evaluation. The patient database was used to identify patients with isolated cleft of the secondary palate. Patients with submucous cleft palate were excluded, as were patients with incomplete records or who were lost to follow-up prior to 10 years of age.

Upon approval by the Institutional Review Board, data were collected, including date of birth, age of palate repair, type of cleft (Veau I or II), syndromic diagnosis, Robin sequence, surgeon, repair type, number of operations involving mucoperiosteum over the hard palate, and incisal relationships. Patients with Robin sequence, defined as the clinical triad of micrognathia, glossoptosis, and upper airway obstruction¹⁶, were also identified and evaluated separately.

Incisal relationships were used as a clinical marker for anteroposterior maxillary growth, as used in the Goslon Yardstick evaluation. Patients with edge-to-edge or reverse overjet were defined as having class III incisal relationships for purposes of this study, and would fall into groups 3-5 of the Goslon Yardstick. Two independent and experienced practitioners in the fields of pediatric dentistry and orthodontics determined incisal relationships by clinical observation.

Pearson's correlations between focal covariates were ascertained. Associations with the incidence of class III incisal relationship were determined using the Fisher's Exact¹⁷ and Mann-Whitney U tests. When comparing the incidence of class III relationship to surgeon and to repair technique, the Fisher's Exact test was performed using a script run in MatLab R2011b (Mathworks, Natick, MA USA). MatLab was also used to calculate the Wilson score interval (confidence interval). All other statistical analysis were performed using IBM SPSS Statistics 20. The Helsinki Declaration was read and all guidelines were followed throughout this investigation.

Results

Through our craniofacial database, 87 eligible patients with an isolated cleft palate were identified. Patients with Robin sequence were then excluded from analysis and the remaining 70 patients were further investigated. The overall incidence of patients with edge-to-edge occlusion or reverse overjet was 7.1% (5/70). A syndrome was identified in 21.4% (15/70). Correlations between the main covariates were weak and insignificant (Pearson's $r^2 < 0.03$, $p > 0.15$ for all comparisons, *Table 2*). Veau type ($p = 0.202$), gender ($p = 0.647$), age at repair after seven months of age (Mann-Whitney U = 191.5, $p = 0.802$), and number of procedures (Mann-Whitney U = 189.5, $p = 0.624$) were not associated with a class III incisal relationship. Detailed demographics are included in *Table 1*.

In regard to the palatoplasty technique used, 6 patients underwent a double-opposing Z-palatoplasty, 8 VY-pushback palatoplasty, 49 two-flap palatoplasty, 6 Von Langenbeck procedure, and 1 soft palate repair with intravelar veloplasty as described by Kreins. These were performed between the ages of 7 and 56 months by 9 different surgeons. Class III incisal relationship was found in 1/6 patients (17%) who had undergone a double-opposing Z palatoplasty, 2/8 patients (25%) VY-pushback palatoplasty, and 2/49 patients (4.1%) 2-flap palatoplasty. The VY-pushback technique did have a high incidence of a class III incisal relationship at 25%. However, we can only definitively conclude that repair using two-flap palatoplasty was not related to class III incisal relationship ($p = 0.264$) due to limited numbers for the other techniques.

Of the 5 patients who developed class III, their primary palatal repair was performed by 4 different surgeons. There was no association between class III incisal relationship and any particular surgeon ($p = 0.890$).

Syndromic diagnosis was a significant predictor of class III incisal relationship. In our study population, 15 of 70 (21.4%) patients had an associated syndrome (*Table 3*). All of the children (5/5) with class III incisal relationship at the time of data collection had a

syndromic diagnosis: two with 22q11.2 deletion syndrome, one with Ehlers-Danlos syndrome, and 2 Stickler syndrome. Incidence of class III incisal relationship after palatoplasty in patients with an isolated cleft palate and a syndromic diagnosis was 33.3%. No patients with nonsyndromic isolated cleft of the secondary palate developed class III incisal relationships ($p < 0.001$) (*Figure 1*).

There were eleven patients with a diagnosis of nonsyndromic Robin sequence and all continued to have positive overjet at the time of data collection. In addition, six patients were categorized as syndromic Robin sequence and these individuals were also found to have positive overjet. These patients are not included in the final calculations and numbers presented.

Discussion

Current teaching has suggested that palatoplasty involving the bony portion of the palate during early stages of development may be the basis for maxillary retrusion in patients with a cleft palate. In this study, the total occurrence of edge-to-edge bite or reverse overjet was 7.1% in all patients with an isolated cleft palate. This is comparable to values reported in the literature for class III occlusion in the normal population and children in mixed dentition 4-13%^{18,19,20,21}.

Good and colleagues presented 217 nonsyndromic patients with cleft lip and/or cleft palate who had undergone successful palatoplasty. They reported an overall frequency of LeFort I maxillary advancement of 20.9%. In patients with both cleft lip and palate, 47.7% required maxillary advancement, but in patients with isolated cleft palate, none required an orthognathic procedure¹³. Oberoi and colleagues identified a Lefort I advancement rate of 3.3% for patients with isolated cleft of the secondary palate, while the LeFort I advancement rate was 12.2% and 13.3% for patients with unilateral and bilateral cleft lip and palate respectively²². Both reports support our low incidence of Class III incisal relationships after palatoplasty in nonsyndromic patients with an isolated cleft palate.

Studies using a canine model correlate with our clinical findings. Bardach reported a study of 62 beagle puppies that underwent palatoplasty using his 2-flap palatoplasty technique. He found animals that underwent elevation of two mucoperiosteal flaps with narrow strips of denuded bone on both sides had less severe craniofacial growth aberrations than those in which the defect was left unrepaired²³. Wijdeveld and colleagues also found no effect on vertical or antero-posterior maxillary growth after cleft palate repair in beagle puppies²⁴.

In our patient population, 15 of 70 patients (21.4%) were diagnosed with a syndrome. This number is lower than the traditionally reported 40-60% of patients with a cleft palate²⁵. This may be due in part to our lack of blanket testing for chromosomal abnormalities, such as Stickler or 22q11 deletion syndrome, common in patients with an isolated cleft palate. Furthermore, patients with an isolated submucous cleft palate, a defect known to have a high association with a syndromic diagnosis²⁶ were excluded. Lastly, because mandibular hypoplasia or retrognathia is the hallmark sign of Robin sequence¹⁶, none of these patients

(syndromic and nonsyndromic) developed class III incisal relationships and these patients were not included in final analysis.

Children with a syndromic diagnosis were more likely ($p < 0.001$) to have an edge-to-edge bite or reverse overjet than their nonsyndromic counterparts after primary palatoplasty for an isolated cleft palate. This leads us to question the true impact of primary palatoplasty on the risk for future orthognathic surgery. These data indicate that abnormalities attributed to the described syndromes may have a more significant influence on eventual incisal relationships than repair of the cleft palate.

Some authors have recommended limited periosteal undermining, thereby reducing the area of the palate that is left exposed²⁷. They also suggest avoiding a lateral relaxing incision to reduce scarring and to potentially prevent maxillary retrusion¹⁰. The Von Langenbeck palatoplasty technique has been described to have less impact on midfacial growth¹⁰, and of the 6 patients undergoing this repair in our study, none developed Class III incisal relationship. Two-flap palatoplasty, with wide undermining of the periosteum, was the most commonly performed primary repair in our patient population. Only 2 of 49 patients (4.1%) who underwent this operation developed class III relationships. Anteroposterior maxillary deficiency developed in 1 of 6 patients who underwent a double-opposing Z-palatoplasty and 2 of 8 patients who underwent a VY-pushback palatoplasty. Because of the small sample sizes, we cannot infer whether any repair type is associated with maxillary growth restriction. However, wide undermining of the palate with significant areas of exposed periosteum, such as with the 2-flap palatoplasty technique, did not discernibly increase the risk of class III incisal relationships.

This study presents primary palatoplasties performed by 9 different surgeons. Many prior studies have focused on the results of a single surgeon. Of the 5 patients with class III incisal relationships, primary repairs were performed by 4 different surgeons. It is unlikely based on this information that the development of a class III incisal relationship is associated with any particular surgeon.

Two-stage repair of the cleft palate, with delay in closure of the hard palate has been shown to have a less adverse effect on growth of the maxilla than single stage repair²⁸. However multiple studies have shown that delayed closure of the hard palate leads to speech problems and developmental delay^{29,30,31,32}, and for these reasons our data is encouraging. All of our patients underwent palatoplasty prior to the age of 18 months, with exception of six patients (3 adopted from China and three children with other medical issues that precluded early surgical intervention). Our data suggest that palatoplasty in patients with an isolated cleft palate does not have a deleterious effect on antero-posterior maxillary growth that would lead to an orthognathic procedure. Standard timing and techniques for palatal repair are indeed beneficial for these children in terms of development and speech, and these factors are not associated with class III incisal relationships in this patient population.

We recognize that there are limitations in this study. For one, there were 343 patients with an isolated cleft of the secondary palate who underwent palatoplasty. However, after excluding patients with incomplete records or lost to follow-up in our system and those with a

submucous cleft, 87 patients remained. In addition, 17 patients had Robin sequence and were removed from the main analysis. This is a large portion of our study group and this has an unknown impact on our results. Secondly, overjet is a subjective finding without numeric measurements. While our data were collected by experienced practitioners, we realize that quantitative assessment using lateral cephalograms would have improved the strength of our study and this may be necessary in future analysis. The retrospective nature of this study allows for only the assessment of anteroposterior incisal relationships, which is recognized as the factor of greatest clinical importance in determination of outcomes using the Goslon Yardstick¹⁴.

Conclusion

In patients with isolated cleft of the secondary palate, the diagnosis of a syndrome is the only statistically significant factor that increases the risk of developing class III incisal relationships. Age at primary repair (after seven months of age), number of repairs, cleft type, gender, surgeon, and type of repair do not increase the risk of future orthognathic procedure.

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Figure 1.
Rate of class III incisal relationships in nonsyndromic versus syndromic patients with isolated cleft palate after palatoplasty

Table 1

Demographics of study population

	All patients (%)	Class I & II Incisal Relationships (%)	Class III Incisal Relationships (%)	P=
Number of patients	70	65	5	
Mean age at palatoplasty, mo	14.1	14.3	15.2	0.802
Mean age at follow-up, yr	14.1	13.9	16.4	0.102
Mean number of repairs	1.5	1.45	1.20	0.624
Male:female ratio	29:41 (41:59)	26:39 (40:60)	3:2 (60:40)	0.647
Veau cleft type				0.202
I	34 (49)	31 (48)	4 (80)	
II	36 (51)	34 (52)	1 (20)	
Syndromic diagnosis	15 (21.4)	10 (66.6)	5 (33.3)	<0.001

Table 2

Correlations between variables evaluated. Values are shown as Pearson's r^2 (p -value)

	Number of Procedures	Veau Type	Syndromic
Age at repair	0.001 (0.747)	0.022 (0.166)	0.021 (0.180)
Number of Procedures		0.001 (0.784)	0.005 (0.525)
Veau Type			<0.001 (0.851)

Table 3

Syndromic patients and Class III incisal Relationships

Syndrome	No. of Patients	Class III Incisal Relationships
Hemifacial microsomia	2	0
Van der Woude	1	0
Ehlers Danlos	1	1
Myotonia congenita	1	0
Asperger	1	0
Stickler	4	2
22q11.2 deletion	3	2
Gordon Syndrome	1	0
Klinefelter syndrome	1	0