Original Article

Retrognathic maxilla in individuals born with oral clefts is due to intrinsic factors and not only due to early surgical treatment

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ABSTRACT

Objectives: To determine if the skeletal form of individuals born with oral clefts was associated with maxillary position.

Materials and Methods: Lateral cephalometric radiographs of 90 individuals 8 to 12 years old born with or without cleft lip and palate paired by age and sex were used. Skull base length, cranial base angle, cranial deflection angle, and maxillary skeletal length and position were studied. Also, mandibular skeletal length and position, lower anterior facial height, and dental position were defined. Individuals were divided into three groups: 30 individuals born with cleft lip and palate with Class III malocclusion (UCLP Class III), 30 individuals born with cleft lip and palate with Class I malocclusion (UCLP Class I), and 30 individuals born without cleft lip and palate with Class III malocclusion (non-cleft Class III).

Results: When comparing the UCLP Class III group with the UCLP Class I group, there were differences in maxillary position ($P < .001$) and mandibular position ($P = .004$) found. No differences were found when comparing the UCLP Class III group with the non-cleft Class III group.

Conclusions: There are intrinsic factors that affect craniofacial morphology of individuals born with cleft lip and palate. (*Angle Orthod.* 2021;91:243–247.)

KEY WORDS: Cleft lip; Cleft palate; Class III malocclusion; Facial growth

INTRODUCTION

The craniofacial configuration may involve different combinations of morphological abnormalities of the cranial base, maxilla, and mandible, as well as vertical facial dimensions[1] and may be affected by genetic and environmental factors.[2,3] These factors include congenital anomalies that can affect maxillofacial growth. The most prevalent congenital anomalies in the human face are clefts of the lip and palate.[4] In individuals affected with cleft lip and palate, a high frequency of Class III phenotype is found compared to the general population,[3] and this difference may be caused by the surgical procedures done to repair the defect[5,6] and innate growth deficiency.[7–10] Studies that investigated intrinsic maxillary deficiency in cleft lip and palate were performed in unoperated cleft lip and palate patients and there are controversies in the literature. Some studies demonstrated that the maxillary length in individuals born with cleft lip and palate was no different from the population born without the defect.[11,12] That result motivated the suggestion that just the iatrogenic surgery may affect maxillary growth. This was in contrast to studies that showed a shorter maxillary length in unoperated individuals born with cleft lip and palate,[7,8] which suggested that there is intrinsic growth impairment that affected facial morphology later in life.[9]

Cranial variations affect the craniofacial configuration of sagittal malocclusions. In the case of Class III malocclusion, reduced cranial base length and cranial base angle were reported.[13] Studies of individuals born with cleft lip and palate analyzing the cranial base did not find a difference between patients born with or without the defect independent of the malocclusion[1]

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and studies analyzing the effect of palate repair on the cranial base concluded that the surgical procedures did not affect the cranial base configuration.[14,15] The study of the craniofacial form in individuals born with cleft lip and palate has been typically designed to compare them with individuals that have Class I profiles.[7,8] Fewer studies compared individuals born with cleft lip and palate that were Class III with individuals born without clefts that were also Class III.[1,4]

Despite many studies showing the high-frequency of the Class III phenotype in individuals born with cleft lip and palate, the craniofacial form of these subjects has not been analyzed more in depth. The goal of this study was to determine if the skeletal cranial form of individuals born with cleft lip and palate was associated with the prognathism profile.

MATERIALS AND METHODS

The protocol of this study was approved by the University Hospital Lauro Wanderley research ethics committee Universidade Federal da Paraíba (CAAE number-1345081999995185) and signed written consent was obtained from all participants.

The sample was comprised of 90 lateral cephalograms, 30 from individuals born with cleft lip and palate with Class III malocclusion (19 boys, 11 girls; mean age: 9.4 years), 30 from individuals born with cleft lip and palate with Class I malocclusion (19 boys, 11 girls; mean age: 9.5 years), and 30 from individuals born without clefts with Class III malocclusion (14 boys and 16 girls; mean age: 9.0 years). The inclusion criteria were: good quality radiographs with clear visualization, before any orthodontic treatment was performed, early or late mixed dentition (8–12 years), and absence of syndromes. In both Class III groups, pre-treatment Wits appraisal of −2.0 mm or less was required for the individual to be included in the study.

The individuals born with clefts underwent surgical correction for cleft lip/palate at an early age (lip surgery at a mean age of 6 months and palate surgery at a mean age of 18 months). All surgeries were done by the same surgeon, with the same technique (Millard for unilateral lip repair, Spina for bilateral lip repair, and von Langenbeck for palatoplasty).[16,17]

Cephalometric analysis was carried out by the same operator tracking landmarks to define the size and position of the maxilla (Condylion to point A [Co-A] and Nasion perpendicular to point A [Nperp-A]), size and position of the mandible (Condylion to Gn [Co-Gn] and Nasion perpendicular to Pogonion [Nperp-pog]), cranial base ([S-N]; Saddle angle [N-S-Ar]; cranial deflection [Ba-Na x Or-Po]), vertical measurements (Lower anterior facial height [ANS-Me]), and dental positions (1.palatine plane and IMPA). Measurements were made in millimeters or degrees and interpretation was done using reference values and standard deviations established by McNamara,[18] Jarabak,[19] and Ricketts[20] considering age and sex.

The individuals born with cleft lip and palate with Class III malocclusion (unilateral cleft of the lip or palate [UCLP] Class III group) were compared to the other groups.

Statistical Analysis

Twelve cephalometric images were retraced to assess intraobserver reliability. Logistic regression was used to determine if any of the assessments of craniofacial form predicted the occurrence of Class III. P values .05 or lower were considered statistically significant.

RESULTS

Intraclass correlation (ICC) of 0.80 to 0.86 were found for the cephalometric measurements.

Vertical measures were analyzed using standards of McNamara and were scored as normal in 40 individuals (10 individuals born with clefts that were Class III, 13 born with clefts that were Class I, and 17 born without clefts), as reduced in 32 (10 individuals born with clefts that were Class III, 12 born with clefts that were Class I, and 10 born without clefts), and as increased in 18 (10 born with clefts that were Class III, five born with clefts that were Class I, and 3 born without clefts).

Upper and lower incisor positions were classified as retroclined, proclined, or well-positioned. Among the individuals born with clefts that were Class III, 12 had retroclined upper incisors, 13 had well-positioned upper incisors, and five proclined. For the lower incisors, nine individuals born with clefts that were Class III were retroclined, 16 well-positioned, and five proclined. Among the individuals born with clefts that were Class I, 12 had retroclined upper incisors, 11 well-positioned, and seven proclined. For the lower incisors, seven individuals that were born with clefts that were Class I were retroclined, 16 well-positioned, and seven proclined. Finally, among the individuals born without clefts, six had retroclined upper incisors, nine were well-positioned, and 15 proclined. For the lower incisors, seven were retroclined, 16 were well-positioned, and seven proclined.

When comparing the UCLP Class III group with the UCLP Class I group, there were differences in the maxillary position (Nperp-A: $P < .001$) and in the mandibular position (Nperp-Pog: $P = .004$). Differences were not found in the cranial base, dental position, and vertical measurements between these groups (Table 1).

Table 1. Multinomial Logistic Regression of the Craniofacial Components Among Individuals Born With Clefts That Were Class III With Individuals Born With Clefts That Were Class I and Unaffected Individuals That Were Class III

	Cleft Class III × Non-Cleft Class III				Cleft Class III × Cleft Class I			
		95% Confidence Interval				95% Confidence Interval		
Predictor	Odds Ratios	Lower	Upper	*P*	Odds Ratios	Lower	Upper	*P*
Maxillary length	1.1	0.91	1.32	.31	1.03	0.8	1.32	.84
Mandibular length	1.02	0.88	1.19	.8	0.99	0.81	1.22	.93
Cranial base	0.94	0.76	1.16	.55	1.08	0.84	1.37	.55
Sadle angle	0.91	0.82	1.01	.07	1.0	0.91	1.11	.91
Maxillary position	1.01	0.71	1.43	.97	2.24	1.46	3.43	<.001*
Mandibular position	1.1	0.92	1.31	.29	0.72	0.58	0.9	.004*
Upper incisor position	1.05	0.98	1.12	.14	1.04	0.96	1.12	.27
Lower incisor position	1.04	0.93	1.17	.46	0.99	0.87	1.13	.88

When the UCLP Class III group was compared with the noncleft Class III group, there were no differences in any cephalometric measurements (Table 1).

DISCUSSION

Craniofacial deformities vary in severity and specific dental and skeletal combinations may predict better or worse treatment outcomes. In Class III, maxillary retrusion, mandibular prognathism, protrusive upper incisors, and retrusive lower incisors are the combinations that indicate the need for orthodontic surgical treatment, considering the severity of these deformities.[21]

To study the Class III configuration of individuals born with cleft lip and palate more in detail, the Class III configuration in UCLP was compared to Class III individuals born without clefts and individuals born with UCLP with Class I malocclusion. If deficiency of facial growth only resulted from iatrogenic primary surgery repairs,[22–25] it was expected that the Class III pattern in individuals born with clefts would have a purely maxillary contribution with the other craniofacial characteristics showing normal values. These values should differ from those in individuals born without clefts or from a Class III configuration in an individual that had never been subjected to surgical repair.

Regarding the sagittal skeletal relationship, the combinations of Class III malocclusion were retrusive maxilla with mandibular prognathism, maxillary retrognathism only, and mandibular prognathism only. In this study, there were differences similar to other studies.[1,4] In the UCLP Class III group, there were three cases with maxillomandibular Class III, eight cases with only mandibular involvement, and 19 maxillary Class III cases. On the other hand, in the non-cleft Class III group, there were 12 cases with maxillomandibular involvement, four cases with only mandibular prognathism, and 14 cases with maxillary retrognathism only.

The maxillary length in the UCLP Class III group was the shortest among the three groups, as reported previously.[1] Similarly, there was a more retropositioned maxilla in this group compared to the UCLP Class I group (Table 1). On the other hand, there was no difference in maxillary position between cleft and noncleft Class III individuals. These data suggested that the lip and palate repair was not the only factor responsible for a Class III profile and maxillary growth restriction. These results were in agreement with Liao and Mars (2005),[9] which reported that there was a more retrusive maxilla in unoperated individuals born with cleft lip and palate compared to an unaffected population, and more severe retrusion in operated UCLP patients compared to unoperated UCLP patients.

In a previous study of individuals born with UCLP, which evaluated maxillary growth using Goslon scores, different maxillary growth responses were found: 43% had Goslon 1 and 2 (good maxillary growth), 32% Goslon 3 (regular maxillary growth), and 25% Goslon 4 and 5 (severe maxillary deficiency). All of these patients were treated by the same surgeon who used the same technique, which suggested that these differences were explained by individual variation that affected maxillary growth and were not only the consequence of the surgical procedure.[26]

The sagittal position of the mandible differed among the groups studied (Figure 1). Comparing the UCLP Class III to the UCLP Class I only group, mandibular position showed a difference ($P = .004$) in agreement with studies that found normal mandibular growth and a retropositioned mandible in individuals born with cleft lip and palate compared to individuals born without clefts.[27] These findings suggested different Class III etiopathogenesis. In individuals born without clefts, Class III was related to genetic variants associated with mandibular growth[28,29] and, in individuals born with clefts, the Class III may have been due to other genetic variants[18] and iatrogenic factors.[22,25]

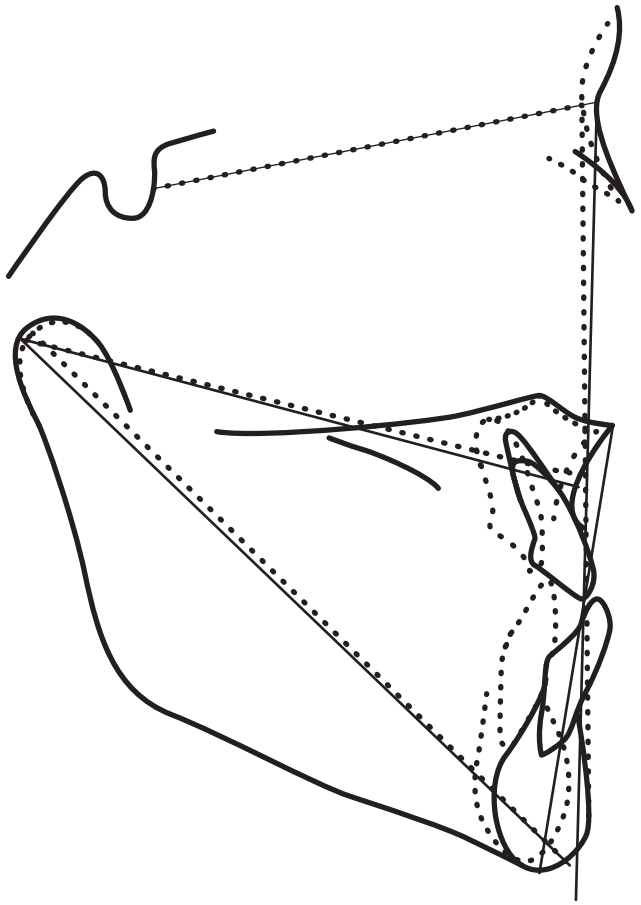

Figure 1. Superimposition of cephalometric measurements included in the study showing that, on average, individuals born with clefts that are Class III (dashed line) have shorter maxillary and mandibular lengths, apparent shorter cranial base, shorter height of the lower anterior portion of the face, and retroinclined upper incisors compared to individuals born without clefts that are Class III, who also appear to have upper incisors projected forward (continuous lines).

Although not significantly different, a shorter cranial base length was seen in 17 individuals born with UCLP and Class III compared to 10 individuals born with UCLP and Class I, which suggested that some UCLP cleft cases may have a Class III craniofacial configuration due mostly to a shortened cranial base.[30,31] No difference in the cranial base length between the UCLP Class III group and the non-cleft Class III group was found.

Analyzing the angular measurements, there was no difference in the cranial deflection and in the saddle angle. Unaffected Class III individuals showed smaller angles in agreement with previous reports of non-cleft cranial configurations.[1,31] Aware that the surgical procedures did not affect the cranial base[14,15] but affected the maxillomandibular relationship,[30,32] these findings suggested that intrinsic jaw factors in the individuals born with cleft lip and palate contributed to the Class III in addition to the surgical procedures.[9,32]

Vertical pattern was analyzed in the linear dimensions and classified according to McNamara scores as normal, reduced, and increased; no differences were found among the groups studied.

More protrusive upper incisors in the group of individuals born without clefts and more retrusive upper incisors in individuals born with clefts ($P = .02$) were found, which is probably related to muscle pressure created by the surgical repair. This difference was not found when comparing the UCLP Class III and UCLP Class I groups ($P = .43$). Lower incisor inclination showed no difference among the groups studied.

It is a consensus in the literature that surgical repair may affect maxillary growth and that maxillary position is the outcome to analyze and compare surgical outcomes. However, the findings presented here showed that, beyond iatrogenesis, maxillary growth may cause a high frequency of Class III in individuals born with cleft lip and palate and it is important to consider this aspect when making decisions regarding the management of patients. A limitation of the study was that the patients analyzed were young and had not yet been through their growth spurt. It is not possible to affirm this would not have altered the results, at least in part.

CONCLUSIONS

- There is a difference in the craniofacial configuration of the Class III phenotype depending on whether an individual was born with or without cleft lip and palate.
- Beyond surgical procedures, there may be intrinsic factors related to the cranial base that affect the craniofacial morphology of individuals born with cleft lip and palate.

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