Associations Between Severity of Clefting and Maxillary Growth in Patients With Unilateral Cleft Lip and Palate Treated With Infant Orthopedics

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Objective: **The purpose of this study was to examine possible associations between severity of clefting in infants and maxillary growth in children with complete unilateral cleft lip and palate.**

Design: **This was a retrospective study of measurements made on infant maxillary study casts and maxillary cephalometric variables obtained at 5 to 6 years of follow-up.**

Setting: **The study was performed at the Institute of Reconstructive Plastic Surgery of New York University Medical Center, New York, New York.**

Patients: **Twenty-four consecutive nonsyndromic unilateral complete cleft lip and palate patients treated during the years 1987 to 1994.**

Interventions: **All the patients received uniform treatment (i.e., presurgical orthopedics followed by gingivoperiosteoplasty to close the alveolar cleft combined with repair of the lip and nose in a single stage at the age of 3 to 4 months). Closure of the palate was performed at the age of 12 to 14 months.**

Results: **Infant maxillary study cast measurements correlated in a statistically significant manner with maxillary cephalometric measurements at age 5 to 6 years.**

Conclusions: **The results demonstrate the large variation in the severity of unilateral cleft lip and palate deformity at birth. Patients with large clefts and small arch circumference, arch length, or both demonstrated less favorable maxillary growth than those with small clefts and large arch circumference or arch length at birth.**

KEY WORDS: *infant orthopedics, maxillary growth, severity of clefting, unilateral cleft lip and palate*

Despite the general opinion that deficiency of tissue, probably seen as width of the alveolar cleft and position of the maxillary segments, is an essential variable affecting the growth of the maxilla following lip, palate, and nose repair (Robertson and Fish, 1975; Ross, 1987; Millard and Latham, 1990; Bardach and Kelly, 1991), there are only a few studies aimed at examining this possible association. More often, the effect of treatment, particularly surgical technique, timing, and the expertise of the surgeon, has been studied and considered to have a great impact on the growth and development of the craniofacial complex in children with cleft (Ross, 1987; Roberts et al., 1991; Shaw et al., 1992). Other factors, such as presurgical orthopedics and orthodontic treatment, are also considered to influence the final growth outcome.

Most previous studies concerning associations between severity of clefting and facial growth used presurgical study cast measurements as initial values and study cast and cephalometric analysis at a later stage. Schwarz et al. (1984) concluded that no single measurement of the casts could reliably predict the occurrence of crossbites at the early mixed dentition stage. However, when more variables were pooled, approximately 90% of crossbite or no-crossbite cases could be predicted. Friede et al. (1988) reported that the preoperative study cast and frontal cephalometric measurements used to predict maxillary growth and occlusal relationships explained only

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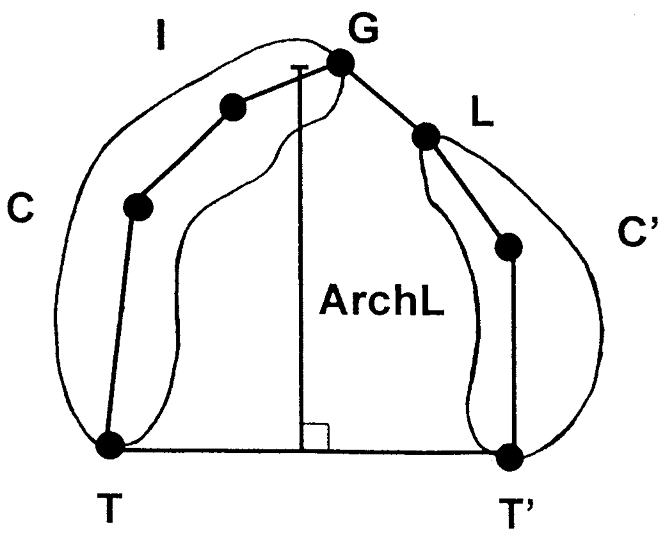

FIGURE 1 **Reference points marked on the study casts. G = midpoint of the margin of the alveolar process medial to the cleft; L = midpoint of the margin of the alveolar process lateral to the cleft; I = point of intersection between the alveolar ridge and groove of the median labial frenum; C, C′ = point of intersection between the alveolar ridge and groove of the lateral labial frenum; T, T′ = tuberosity points, junction of the alveolar ridge with the outline of the tuberosity. Width of the cleft (G-L), arch circumference (T-C-I-G + L-C′-T′), anterior (C-C′) and posterior (T-T′) arch widths, and arch length (G perpendicular to T-T′ line) were measured on the study models.**

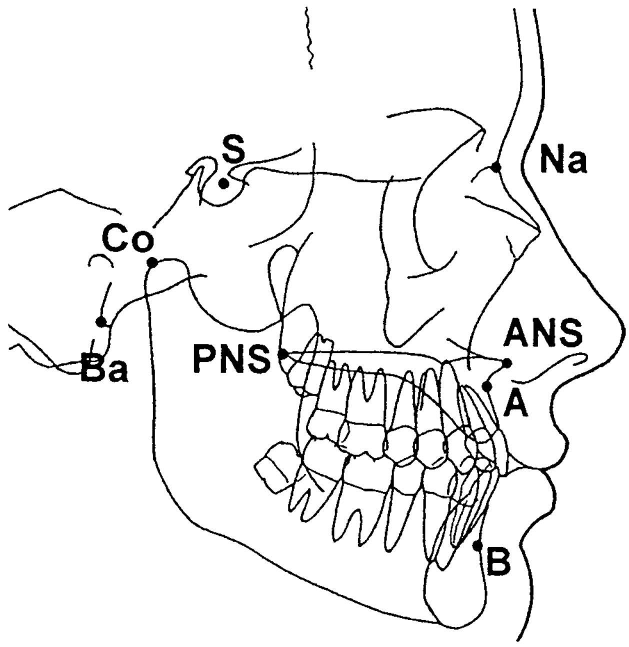

FIGURE 2 **The length of the maxilla (ANS-PNS, Co-A) and the relationship of the maxilla and mandible to the cranial base (SNA, BaNA, SNB) were measured on the lateral cephalograms.**

half of the variation found later. The width of the palatal cleft measured directly at palatoplasty was not found to correlate with the severity of the malocclusion at the age of 4 years (Suzuki et al., 1993). In a recent study by Johnson et al. (2000), no correlation was found between the initial defect and occlusal score at the age of 6 years. All these studies aimed at exploring the relationship between initial deformity and occlusion rather than midface skeletal development.

The present study was initiated on the basis of the observation that there is large variation in the severity of unilateral cleft lip and palate deformity at birth, which may at least partially be due to varying amount of tissue deficiency. If an association exists between initial deformity and maxillary skeletal growth, one can hypothesize that the midface growth problem associated with tissue deficiency will be an issue throughout the postnatal growth period and may be less related to the given treatment. One cannot, however, rule out the possibility of treatment exacerbating intrinsic growth problems (Ross, 1987).

The specific aim of this study was to examine whether there are associations between the initial extent of clefting and maxillary growth.

Materials and Methods

This retrospective study involved 24 consecutive patients with nonsyndromic unilateral complete cleft lip and palate (UCLP) treated at the Institute of Reconstructive Plastic Surgery of New York University Medical Center during the years 1987 to 1994 with good-quality study casts and lateral cephalograms. All the patients received uniform treatment, presurgical orthopedics, earlier with a pin-retained appliance (n = 9) or currently with a nasoalveolar molding appliance (n = 15) as described by Grayson et al. (1999). The aim of infant orthopedics was to bring the alveolar segments together before primary surgery. This made the use of gingivoperiosteoplasty possible without undermining and stretching of soft tissues (Millard and Latham, 1990) to close the alveolar cleft, combined with repair of the lip and nose in a single stage at the age of 3 to 4 months (Cutting et al., 1998). Closure of the palate was performed at the age of 12 to 14 months (Cutting, 1999). All the operations were performed by the same plastic surgeon.

Maxillary study casts were obtained at the initiation of molding therapy (mean 25.8 days, range 6 to 75 days). Conventionally used landmarks (Friede et al., 1988; Seckel et al., 1995) were lightly marked on the casts with a 0.5-mm pencil. Cleft width (G-L), arch circumference (T-C-I-G + L-C′-T′), anterior (C-C′) and posterior (T-T′) arch width, and arch length (G perpendicular to T-T′) were measured with a digital sliding caliper (Fig. 1). In addition, ratios between cleft width and arch circumference (G-L/T-C-I-G + L-C′-T′), and cleft width and arch length (G-L/G perpendicular to T-T′) were calculated. Once the measurements were made, the points were immediately erased. Repeating landmark identification tested reproducibility of the measurements and measurements on 10 randomly selected study casts after at least 1 month by the same investigator. Errors in landmark position and measurements for each variable were calculated from the formula

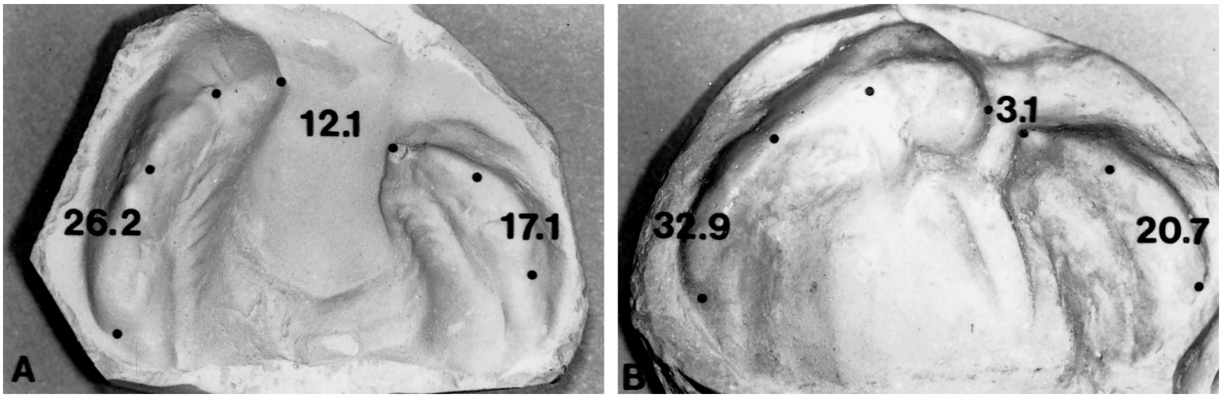

FIGURE 3 Study casts of two 6-day-old infants illustrate variation in the severity of clefting found in patients with complete unilateral cleft lip and palate. The present results indicate that because of a large cleft and short arch circumference (A), less favorable maxillary growth can be anticipated, compared with the child with a small cleft and large arch circumference (B). Measurements are in millimeters.

$\sqrt{\Sigma\ d^2/2n}$, where d is the difference between the first and the second measurement.

Lateral cephalograms of the same patients were obtained at 5- to 6-year follow-up (mean 5.4 years, range 4.3 to 7.1 years) before any orthodontic or orthopedic treatment was started. The cephalograms were traced, landmarks identified, and measurements performed to assess the length of the maxilla (ANS-PNS, Co-A) and the relationship of the maxilla and mandible to the cranial base (SNA, BaNA, SNB; Fig. 2). With the exception of point A, all the other cephalometric landmarks and lines used are well known in cephalometrics. Since point A is difficult to define in young children with UCLP, a constructed maxillary point was used (Tindlund, 1989): nasal line (through ANS and PNS) was drawn, and the point was located on the most anterior maxillary surface 7 mm below the line. Two orthodontists examined the tracings and location of the landmarks. Discrepancies among examiners were resolved by arbitration.

Pearson's correlation coefficients were calculated between the study cast measurements and cephalometric variables.

Results

Reproducibility testing of the study cast analysis showed the range of measurement errors to be from 0.17 to 1.35 mm (Table 1) (i.e., all the errors were less than 4.1% of the mean of the corresponding measurement). The errors were well within the same range and magnitude than in a previous study particularly aimed at examining the reliability of maxillary study cast measurements of infants with cleft lip and palate (Seckel et al., 1995). Therefore, it can be concluded that the present measurement errors are insignificant as far as the reliability of the study cast results is concerned.

Several statistically significant correlations were found between the initial study cast measurements and the cephalometric variables at the mean age of 5.4 years (Tables 2 and 3). Cleft width as a single measurement showed statistically significant correlations, at $p < .01$ level, with the length of the maxilla (Co-A, ANS-PNS) and with the relationship of the maxilla to the cranial base (SNA, BaNA). When the cleft width ratio, either with arch circumference or arch length, was examined, the correlations were more significant. On the other hand, no statistically significant correlations were found among any of the cast measurements and mandibular position in relation to cranial base (SNB).

Discussion

In the present study, the effect of phenotypic variability, particularly the severity of clefting on maxillary growth following cleft repair, was investigated. Other factors, such as the timing and technique of surgery and the individual performing the operations, were kept constant. In addition, to decrease

TABLE 1 Combined Error in Landmark Positioning and Measuring (in Millimeters) as Calculated for Each Study Cast Measurement

Variable	*Error*
G-L	0.17
C-C′	0.56
T-T′	0.65
Arch length	0.79
Arch circumference	1.35

TABLE 2 Means, Standard Deviations, and Ranges of the Infant Study Cast Measurements and Cephalometric Variables at the Age of 5.4 Years

	n	*Mean*	*SD*	*Range*
G-L (mm)	24	7.9	3.71	0.9–16.4
Arch C (mm)	24	51.0	5.47	42.0–60.4
Arch L (mm)	24	19.6	2.13	15.1–24.7
A Width (mm)	24	26.8	2.57	20.7–30.5
P Width (mm)	23	34.8	2.93	28.4–40.2
SNA (deg)	24	78.8	3.72	71.5–85.5
BaNA (deg)	24	60.6	3.95	54.0–68.5
SNB (deg)	24	76.1	3.78	68.0–83.0
CoA (mm)	24	78.3	5.07	67.5–88.0
ANS-PNS (mm)	24	45.8	3.55	38.5–52.5

TABLE 3 Pearson's Correlation Coefficients Between Infant Study Cast Measurements and Cephalometric Variables at the Age of 5.4 Years

	G-L	*Arch C*	*Arch L*	*A Width*	*P Width*	*G-L/Arch C*	*G-L/Arch L*
SNA	−.655*	.493**	.015	−.223	.387	−.657*	−.616*
BaNA	−.649*	.532**	.167	−.242	.398	−.652*	−.637*
SNB	.332	.296	.011	−.032	.024	−.296	−.276
CoA	−6.83*	.509**	.189	−.406**	.359	−.704*	−.696*
ANS-PNS	−.520*	.447**	.217	−.279	.420**	−.559*	−.556*

* $p < .01$.
** $p < .05$.

other sources of variability, only nonsyndromic patients with UCLP were included in this study.

Two different infant orthopedic appliances, pin-retained or nasoalveolar molding appliance, were used in the treatment. Our results show, however, that there is no statistically significant difference in the craniofacial skeletons of children treated with these two modes of infant orthopedics (Vendittelli et al., 2000). This justified pooling of the groups for the study.

In contrast to previous studies (Friede et al., 1988; Schwarz et al., 1984; Suzuki et al., 1993; Johnson et al., 2000) with the aim of examining associations between the initial condition and total facial growth, we correlated infant maxillary study cast measurements to growth of the maxilla only. No attempt was made to determine correlations to maxillomandibular relations or occlusion. This may be the reason we found statistically significant correlations between the severity of cleft deformity in infancy and maxillary cephalometric variables at age 5 to 6 years.

The present study was a correlation study, and since a proof of correlation does not indicate a cause-and-effect relationship but only co-occurrence, these findings have to be interpreted with caution. The various aspects of surgery being evidently of prime importance regarding facial growth in children with UCLP (Ross, 1987; Roberts et al., 1991; Shaw et al., 1992) were not evaluated here. However, in the present study, every patient received presurgical molding therapy to close the alveolar cleft and approximate lip segments. This procedure reduced cleft width variation by the time of the primary surgery. The reduced variation in cleft width was found to reduce variability of surgical technique (extent of undermining), which reportedly results in more uniform midface growth within this population (Wood et al., 1997). Our findings show that the patients with UCLP with large clefts and small arch circumference or arch length had less favorable maxillary projection than those with small clefts and large arch circumference or arch length by the age of 5.4 years. Despite differences in maxillary growth, overall facial appearance could still be acceptable, depending on mandibular growth and extent of dentoalveolar compensations.

The finding that all patients with UCLP are not the same at birth and remain to be characterized by their severity of initial deformity has important clinical implications. Treatment outcomes, and favorable or unfavorable maxillary growth, may be anticipated according to initial severity of the cleft deformity and less related to the given treatment. More importantly, treatment protocol could vary according to severity of the initial deformity. Yet, in the case of a child with a large cleft and small arch circumference, we might still propose closure of the cleft with presurgical orthopedics and gingivoperiosteoplasty while being aware of the higher risk for maxillary growth impairment associated with the initial tissue deficiency. Thus, we would take advantage of the benefits of the gingivoperiosteoplasty (i.e., reduced need for bone grafting at the time of canine eruption, reduced occurrence of soft tissue fistulae, and improved prognosis of bone grafting if required [Santiago et al., 1998]).

Our findings may also have implications for the design of research protocols in cleft lip and palate growth studies. Patients with cleft have traditionally been grossly grouped for clinical and research purposes (e.g., bilateral, unilateral, and cleft palate only). However, because there is a demonstrated variation in severity of cleft deformity, children with UCLP could be sorted by severity before correlating specific treatment variables with outcomes. According to our findings, a child with UCLP with small cleft and large arch circumference would probably have a different outcome with regard to maxillary growth from a child with a large cleft and short arch circumference. If the effect of clinical variables, such as surgery, on facial growth is based on findings of children with large clefts and short arch circumference, different results and interpretation would probably be attained than if patients with UCLP with small clefts and large arch circumference were evaluated (Fig. 3). Thus, a revision of the traditional pooling of patients would appear recommendable.

In conclusion, our findings confirm previous opinions that not all children with UCLP are the same and that the severity of clefting is an essential variable affecting maxillary growth in patients with UCLP (Robertson and Fish, 1975; Ross, 1987; Millard and Latham, 1990; Bardach and Kelly, 1991). Even more variation in outcomes can be anticipated when variability as a consequence of clefting is superimposed on normal biologic variation of the craniofacial complex (Vig, 1990; Molsted, 1999). The present findings are naturally applicable to the used treatment protocol. Previous studies suggest that prediction accuracy may vary between groups of children with cleft according to different treatment methods (Schwarz et al., 1984; Friede et al., 1988). Therefore, future studies are warranted to examine whether the association between initial cleft severity and maxillary growth can be found in children with UCLP treated with different protocols.

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