Long-Term Effects of Premaxillary Setback on Facial Skeletal Profile in Complete Bilateral Cleft Lip and Palate

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The purpose of this study was to analyze the long-term effects of premaxillary setback in the surgical treatment of complete bilateral cleft lip and palate (C-BCLP). Roentgencephalograms at adolescence or early adulthood were available for 13 of our longitudinal C-BCLP patients who were treated with premaxillary setback and followed up with serial films since infancy. Six individuals had been subjected to early premaxillary setback to facilitate lip repair. Seven of the patients had had their premaxillae setback at a later age to correct a severe midfacial protrusion in the preschool years. Fourteen other C-BCLP patients, followed up and treated similarly except for the premaxillary setback, served as controls. The average long-term outcome of the setback surgery in both the early and late subgroups was retrusion of the midface and a slightly concave facial skeletal profile.

Premaxillary protrusion, a characteristic of infants with complete bilateral cleft lip and palate (C-BCLP), has always constituted a major problem in the surgical rehabilitation of this type of cleft. Millard's (1977) historical review of treatment of the projecting premaxilla is an interesting account of the efforts to achieve ideal results as early as possible, at a time when so little was understood about the spectrum of severity of the defect and its natural history. Among the surgical procedures that were advocated was premaxillary setback, designed to facilitate lip repair and to fit the premaxilla into the maxillary arch to achieve an idealized arch form at an early age. Although premaxillary setback had been advocated and abandoned by others earlier (Millard, 1977), it was repopularized in the 1950s mainly by Browne (1949) and Cronin (1957).

The question of how premaxillary setback might influence future facial growth was a matter of concern to earlier writers such as Veau, as cited by Millard (1977). Bishara and Olin (1972) concluded that early surgical manipulation of the premaxilla affects the ultimate horizontal growth of the premaxilla. Vargervik (1983) reported similar experience as all of her patients that were treated with premaxillary surgery demonstrated "rather severe midfacial retrusion" in early adulthood. In contrast, Monroe and coworkers published a number of case reports (1959, 1965, 1970) and concluded that long-term growth results were satisfactory. However, they stressed that the osteotomy had to be performed behind the "epiphyseal line" believed to join the vomer to the premaxilla. Similarly, others have maintained that

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it is the execution of the surgery itself, rather than the surgical method, that causes the growth disturbance that sometimes occurs (Glass, 1970).

In 1972, we reported the results of premaxillary setback, both in infants with C-BCLP (early setback) and in preschool children (late setback) (Friede and Pruzansky, 1972). Although ingroup variation was noted, the extreme facial convexity produced by the projecting premaxilla decreased considerably as a result of surgical setback. In some cases, it was suggested that the resultant straight profile, with increasing age, would give a concave profile. However, almost none of the patients studied had reached puberty at the time of our 1972 report, and we could only estimate the final effect on the facial profile. The aim of this communication is to follow up the previously studied subjects who were treated with premaxillary setback. At this point all of the patients that we analyzed have passed the pubertal growth spurt, and a few subjects have reached adulthood. Thus, a more definitive evaluation can be made of the effect of premaxillary setback on facial growth.

Materials and Methods

As in our earlier study, the setback patients were grouped according to age at the premaxillary operation (Table 1). In the early sample, the setback was performed before or at the time of lip repair. This group consisted of 6 patients (3 male, 3 female) with an average age at setback of 4.2 months (range: 1 to 10 months). In the second group, seven patients (3 male, 4 female) had a late premaxillary setback at a mean age of 5 years, 7 months (range: 3 years, 7 months to 7 years, 11 months). The setback was performed after lip closure and generally also after palatal repair. The amount of bone resected at the setback could not be established in this retrospective study.

In 60 percent of the patients (3 early and 5 late cases) the premaxilla was stabilized after the setback with a Kirschner pin for variable periods of time (mean: 9 years, 7 months; range: 1 to 18 years). In one case of late setback, a fixed orthodontic appliance provided fixation in addition to the pin. For the remaining patients, no information could be obtained regarding premaxillary stabilization after the setback surgery.

The ages for lip closure and palatal repair corresponded relatively well between the two subgroups. Lip repair was performed in one or two stages between 3 and 7 months and the palate was closed in one or two sessions (starting with the velum) between 1 and 5 years of age (Table 1).

A group of 14 C-BCLP patients (9 male, 5 female) with similar lip and palatal surgery except that premaxillary setback was not performed (Table 1), also taken from our earlier investigation, was chosen to serve as a control. The criterion for selection of these patients, as well as the setback cases, was the availability of follow-up records beyond puberty. The mean ages of the patients of the 3 subsamples studied in this report were: for the early setback group, 17 years (range: 15 years, 6 months to 18 years, 8 months); for the late setback group, 17 years, 3 months (range: 15 years, 11 months to 19 years, 8 months); and for the nonsetback group, 17 years, 6 months

TABLE 1. Mean Ages (months) for the Early Surgical Procedures of the 3 Groups Studied*

	Premaxillary Setback	*Lip Closure*		*Palatal Closure*	
		One Stage	*Two Stages*	*One Stage*	*Two Stages*
Early setback	4 (N=6)	—	4 and 7 (N=6)	26 (N=4)	—
Late setback	67 (N=7)	4 (N=4)	3 and 6 (N=3)	37 (N=5)	10 and 60 (N=2)
No setback	—	4 (N=7)	4 and 8 (N=7)	40 (N=7)	36 and 53 (N=5)

*No information could be obtained concerning palatal surgery in 4 patients.

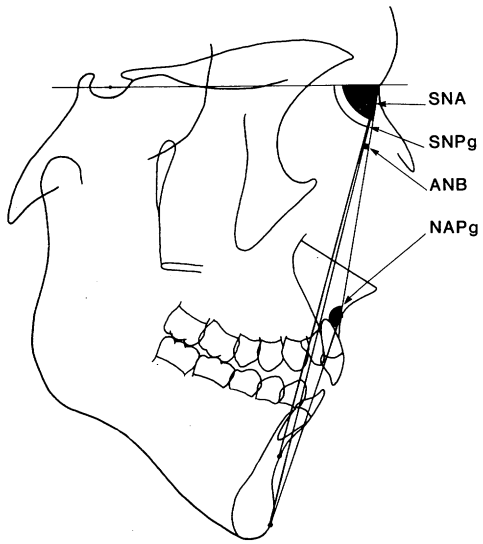


FIGURE 1 Cephalometric variables: measurements employed in the analysis of the patients' lateral roentgencephalograms

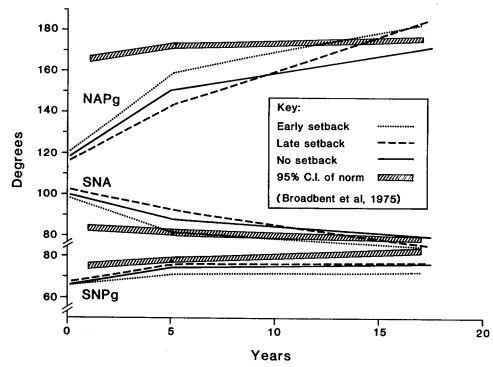


FIGURE 2 Diagram illustrating the long-term development of the means for three of the cephalometric variables in the two setback groups and the control sample treated without premaxillary setback. The 95% confidence intervals of the means for similar variables in individuals without cleft are computed from Bolton Standards (Broadbent et al, 1975) at age 1, 5, and 17 years.

(range: 15 years, 10 months to 19 years, 9 months).

Orthodontic treatment was completed for more than half of the patients of the total sample, and one-fourth of them still wore active appliances at follow-up. The rest had not yet started their final orthodontic or combined orthodontic-surgical treatment.

Tracings of lateral roentgencephalograms were analyzed with concentration on variables related to the skeletal profile (Fig. 1). The following angles, measured between conventional cephalometric reference points, were selected for this study: SNA, SNPg, ANB and NAPg. The first two variables describe the position of the premaxilla and mandible relative to the anterior cranial base, while the latter two measurements represent the relation between the jaws and the skeletal profile. Statistical differences between the means of the three groups were tested nonparametrically with the Mann-Whitney U Test (Siegel, 1956).

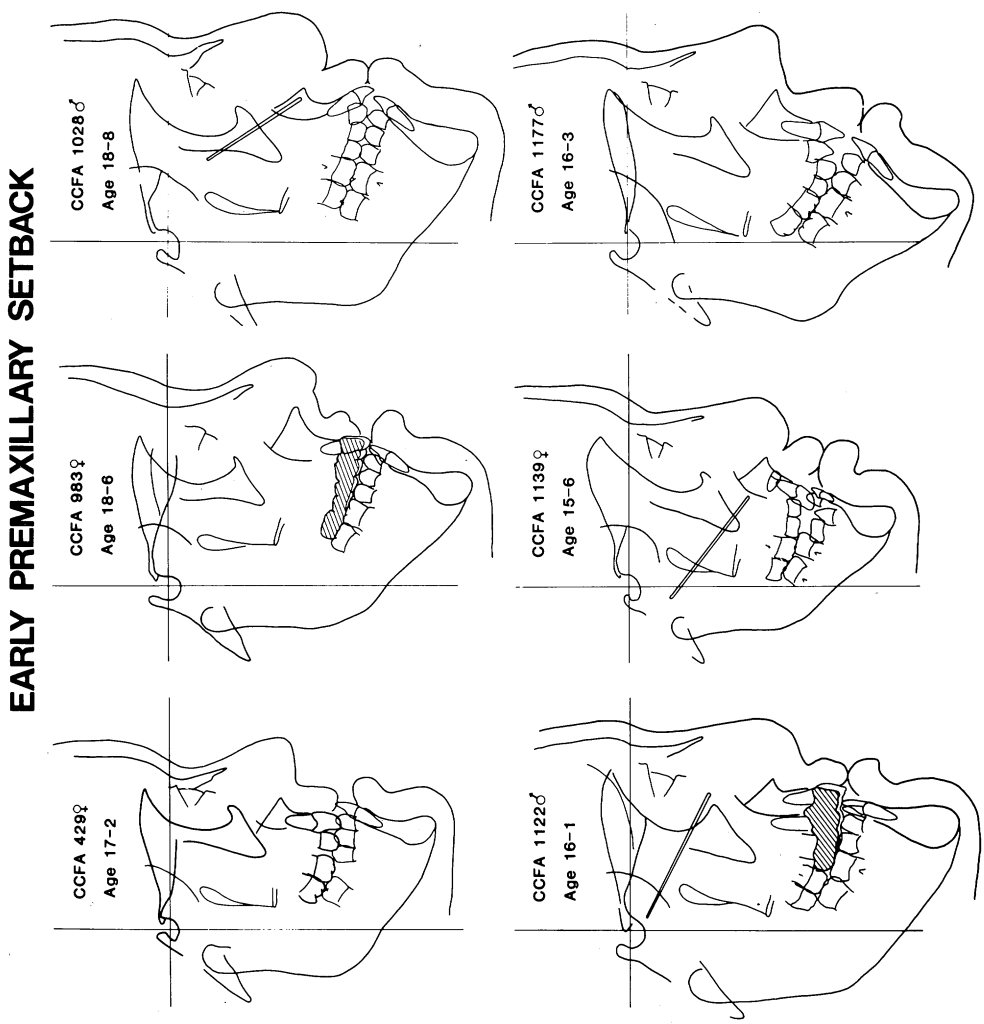
Findings

Although the intent of this report was to focus on the long-term results at adolescence, we felt that it was important to consider how the profile characteristics of the three subgroups had developed since infancy. Comparisons between groups were

TABLE 2. Comparison of Cephalometric Variables at Adolescence Between the Two Setback Subgroups and the Control Bilateral Sample with Cleft Treated Without Premaxillary Setback

	Premaxillary Setback				*No Premaxillary Setback (N=14)*		*Comparisons*		
	Early (N=6)		*Late (N=7)*						
	$\bar{x}$	*SD*	$\bar{x}$	*SD*	$\bar{x}$	*SD*	*A vs B*	*A vs C*	*B vs C*
Age (yrs)	17		17-5		17-6				
SNA	74.5°	2.6	75.4°	5.5	80.3°	3.6	NS*	$p<0.01$	$p<0.05$
SNPg	76.2°	4.2	77.9°	3.1	75.8°	3.7	NS	NS	NS
ANB	0.3°	4.7	−0.2°	5.7	6.4°	3.0	NS	$p<0.01$	$p<0.001$
NAPg	183.2°	9.1	184.9°	12.1	171.0°	6.8	NS	$p<0.01$	$p<0.001$

*NS = no significant difference in mean value between the groups compared.


EARLY PREMAXILLARY SETBACK
CCFA 429♀
Age 17-2
CCFA 983♀
Age 18-6
CCFA 1028♂
Age 18-8
CCFA 1122♂
Age 16-1
CCFA 1139♀
Age 15-6
CCFA 1177♂
Age 16-3

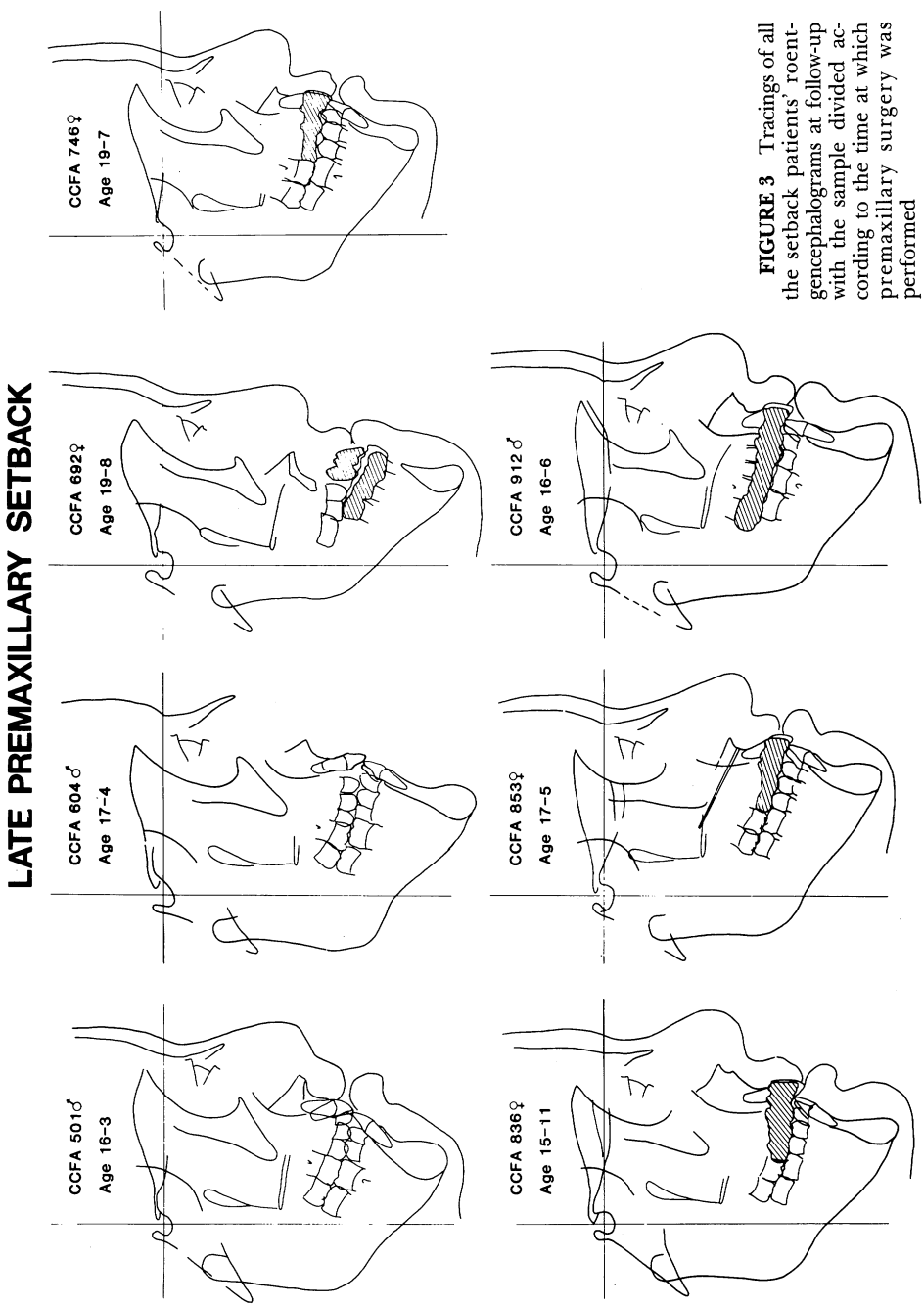


FIGURE 3 Tracings of all the setback patients' roentgencephalograms at follow-up with the sample divided according to the time at which premaxillary surgery was performed

made at a mean age of 2 months (prior to surgery), at 5 years (before the late premaxillary setback procedure), and at the follow-up at 17 years of age. At the preoperative stage, there were no statistically significant differences in the skeletal profile measurements between the three groups (Fig. 2). At the preschool age, the early setback group displayed significantly ($p < 0.01$) less midfacial protrusion than the control C-BCLP patients who were treated without premaxillary setback. The patients selected to undergo late setback surgery showed significantly ($p < 0.01$) more protrusive midfaces than the controls at this stage.

The cephalometric results at follow-up at adolescence indicated that surgical setback of the projecting premaxilla, whether in infancy or later, resulted, on the average, in a slightly concave skeletal profile (NAPg > 180°) (Table 2). In contrast, the control patients without this treatment displayed some facial convexity at a similar age. The difference was caused by more retrusion of the midface in the setback groups than in the patients who had not undergone premaxillary surgery. The average position of the premaxilla at a mean age of 17 years did not differ significantly between the early and late setback groups. This held true for midfacial development, relative to both the anterior cranial base and the mandible. Mandibular position did not differ statistically between any of the subgroups studied.

Lumping of the data by presenting mean values is less than ideal because it obscures individual patterns of facial development. Therefore, in addition to the tabulated values, tracings of the cephalograms of each setback patient at adolescence or early adulthood are presented to supply the reader with a more complete view of the great variation in long-term treatment results (Fig. 3).

There existed differences among patients, not only in end result but also in how the profile characteristics had developed over time. In some patients, the midfacial protrusion was extreme in infancy and was eliminated through the premaxillary setback operation. Continuation of the forward and downward growth of the premaxilla after the operation was a prerequisite for a satisfactory result in early adulthood (Fig. 4). Other infants displayed less than average profile convexity before surgery. In these patients, the growth pattern of the premaxilla was directed mostly downwards after the setback, which had a deleterious long-term effect on the profile and on the relation of the midface to the cranial base and mandible (Fig. 5).

In case 692, the concavity of the profile was extreme at follow-up. The growth pattern of the midface had changed from a forward and downward direction prior to setback to a downward and even backward course after the surgery (Fig. 6). This had been caused by inadequate premaxillary stabilization after the setback surgery, which resulted in pseudoarthrosis between the premaxilla and the bony nasal septum.

Discussion

We recognize that the findings of this study are based on comparatively few C-

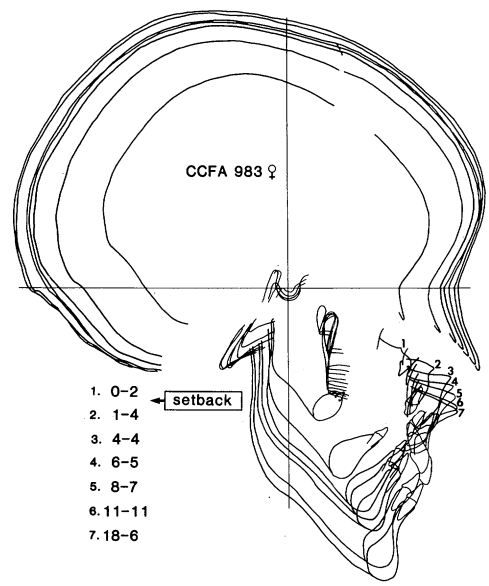


FIGURE 4 Superimposed serial tracings of a patient treated with early premaxillary setback. The extreme preoperative facial convexity was reduced through surgery. Note the continuation of the forward and downward growth of the premaxilla after setback.

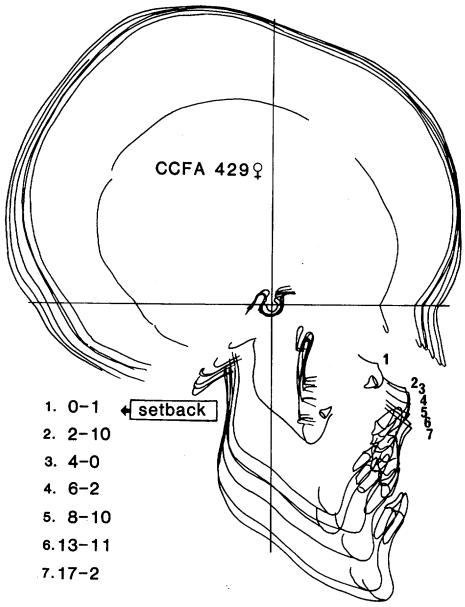


FIGURE 5 Superimposed serial tracings of an early setback patient with less than average midfacial protrusion before surgery. Notice the vertical growth pattern of the premaxilla after setback. Both factors contributed to the development of the concave profile in early adulthood.

BCLP patients, although it can be argued that our sample probably is the largest hitherto reported with longitudinal cephalometric records from infancy to adolescence. We are also aware of the difficulty in selecting a limited number of cephalometric parameters to describe the overall outcome of a particular method of treatment. However, our analysis focused on the most obvious characteristics of a C-BCLP patient, namely, the development of the midface and its influence on the facial profile.

The extreme facial convexity of infants with bilateral clefts decreases considerably with age, as a result of both treatment and growth. The average profile convexity of C-BCLP patients not subjected to premaxillary setback surgery is considered to approximate the norm for individuals without cleft at early adolescence (Narula and Ross, 1970; Friede and Pruzansky, 1972). The straightening of the profile of our control group of patients with bilateral cleft treated without setback seemed to occur somewhat more slowly than in previous reports as the profile had not reached the norm for individuals without cleft at the age of 17 years (Fig. 2). On the other hand, the profile convexity of the two setback groups had decreased too much with a resultant concavity when the patients approached adulthood. Thus, the suspicion expressed in our 1972 paper is confirmed in this study. When examining reasons for such a development, the two setback subgroups have to be considered separately.

Without entering into a discussion about indications for early setback or no setback of the premaxilla in infancy, it should be remembered that the infants of the early setback group did not differ significantly from the other groups with cleft in any of the profile characteristics, including the position of the premaxilla relative to the

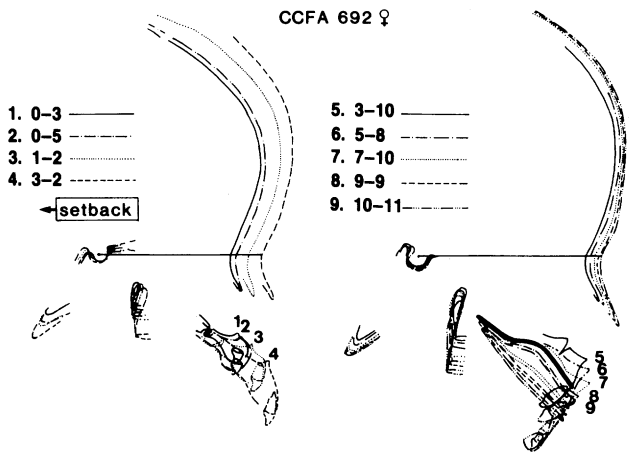


FIGURE 6 Selected serial tracings of case 692 displaying mainly forward premaxillary growth before the setback at age 3 years and 7 months. Pseudoarthrosis developed due to poor premaxillary stabilization with a resultant downward and even backward growth pattern of the premaxilla after the surgery.

anterior cranial base. As the initial convexity, "normal" for C-BCLP babies, was reduced instantly through the early setback of the premaxilla, it should be expected that the mean profile later on would become "abnormally" straight or concave. Variations between patients might be explained on the basis of different genotypes, various amounts of setback, and possible damage to the growth of the vomeropremaxillary suture. In C-BCLP patients (Pruzansky, 1971; Friede and Morgan, 1976) as well as in individuals without cleft (Melsen, 1967), this growth site allows for forward and downward displacement of the (pre)maxilla relative to the vomer.

As the midface of the patients in the late setback group at the preschool age was significantly more protrusive than in the controls treated without setback, the premaxillary surgery seemed to be indicated as the cause at that stage of development. However, many premaxillae were evidently set back more than optimally and the average result was therefore a concave skeletal profile at follow-up. To obtain a good end result, the premaxilla should not be set back to a complete fit in the maxillary arch, as the midface then risks being outgrown by the mandible (Monroe, 1965; Glass, 1970). It has been found that the growth rate of the premaxilla in children with bilateral clefts equals only half of that in children without cleft (Vargervik, 1983). The position of the premaxilla immediately after surgery should correspond to that of other C-BCLP patients of similar age treated without setback. The average profile would then reach the norm value for subjects without cleft in early adulthood.

If case 692, with the rare complication of pseudoarthrosis, was excluded from the late setback group, the mean value for SNA increased from 75.4° to 77.4° and NAPg decreased from 184.9° to 180.7°. From a statistical point of view, this provided a difference in SNA at the 5 percent level of confidence if the late setback patients were compared to the early setback patients. Also, the previously found difference shown in Table 2 when comparing SNA between the late setback patients and patients treated without setback became insignificant. The exclusion of case 692 did not influence the statistical interpretation regarding the angle NAPg. However, comparison between the methods of early and late setback used in this investigation should be made cautiously because of the selection bias in the assignment of patients to the two groups as discussed above. The early setback sample in this study contained a randomized mix of different severities of premaxillary protrusion and profile convexity. The late setback group, on the other hand, was preselected because the patients displayed severe midface protrusion and facial convexity in the preschool years.

Conclusions

Setback of the premaxilla in C-BCLP patients should be performed only in patients with a propensity for extreme profile convexity; otherwise there is a strong risk of profile concavity in adulthood. However, since the degree of midfacial protrusion in infancy appears to be difficult to assess, less definitive methods than early setback, e.g., presurgical orthopaedics or lip adhesion, seem preferable as the treatment of choice. On the other hand, if a C-BCLP patient has been followed up with roentgencephalometry for several years and the normal resolution of the midfacial protrusion has not occurred, a late premaxillary setback is indicated in the preschool years. This study has shown, however, that patients treated with this method may also develop too straight or concave facial skeletal profiles in early adulthood.

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References

Bishara SE, Olin WH. Surgical repositioning of the premaxilla in complete bilateral cleft lip and palate. Angle Orthod 1972; 42:139.

Broadbent BH Sr, Broadbent BH Jr, Golden WH.

Bolton Standards of Dentofacial Growth. Saint Louis: CV Mosby, 1975.

BROWNE D. Hare-lip. Ann R Coll Surg Eng 1949; 5:169.

CRONIN TD. Surgery of the double cleft lip and protruding premaxilla. Plast Reconstr Surg 1957; 19:389.

FRIEDE H, PRUZANSKY S. Longitudinal study of growth in bilateral cleft lip and palate, from infancy to adolescence. Plast Reconstr Surg 1972; 49:392.

FRIEDE H, MORGAN P. Growth of the vomero-premaxillary suture in children with bilateral cleft lip and palate. Scand J Plast Reconstr Surg 1976; 10:45.

GLASS D. The early management of bilateral cleft of lip and palate. Br J Plast Surg 1970; 23:130.

MELSEN B. A radiographic craniometric study of dimensional changes in the nasal septum from infancy to maturity. Acta Odontol Scand 1967; 25:541.

MILLARD DR Jr. Cleft Craft: The Evolution of Its Surgery. II. Bilateral and Rare Deformities. Boston: Little, Brown & Co, 1977.

MONROE CW. The surgical factors influencing bone growth in the middle third of the upper jaw in cleft palate. Plast Reconsr Surg 1959; 24:481.

MONROE CW. Recession of the premaxilla in bilateral cleft lip and palate. Plast Reconstr Surg 1965; 35:512.

MONROE CW, GRIFFITH BH, MCKINNEY P, ROSENTEIN SW, JACOBSON BN. Surgical recession of the premaxilla and its effect on maxillary growth in patients with bilateral clefts. Cleft Palate J 1970; 7:784.

NARULA JK, ROSS RB. Facial growth in children with complete bilateral cleft lip and palate. Cleft Palate J 1970; 7:239.

PRUZANSKY S. The growth of the premaxillary-vomerine complex in complete bilateral cleft lip and palate. Tandlaegebladet 1971; 75:1157.

SIEGEL S. Nonparametric Statistics for the Behavioral Sciences. Tokyo: McGraw-Hill Kogakusha, Ltd, 1956.

VARGERVIK K. Growth characteristics of the premaxilla and orthodontic treatment principles in bilateral cleft lip and palate. Cleft Palate J 1983; 20:289.