Craniofacial Morphology in Unilateral Cleft Lip and Palate Prior to Palatoplasty

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Roentgencephalometric studies were carried out in 30 boys with unilateral cleft lip and palate approximately five years of age, none of whom had had a palatoplasty. A comparison with 27 normal individuals matched in age showed that most basic deviations of the craniofacial configuration recorded in adults developed at an early age, often prior to palatoplasty, i.e., reduced height of the upper face, maxillary dento-alveolar retroinclination, displacement of the upper jaw backwards, widening of some components of the maxillary complex, and a shortening of the mandibular body and ramus associated with changes in the mandibular shape. Only the length of the upper jaw was not reduced. The shortening of this dimension occurred postoperatively at a more mature age. The described basic deviations exerted untoward effects on further facial parameters (proportions, retrusion of the jaws, limitation of anterior growth rotation, maxillomandibular and other interrelations, and malocclusion). The height of the upper lip showed a similar shortening as in adults, but lip thickness was not reduced. There was good prominence of the upper lip.

Changes of the craniofacial configuration in adult males with unilateral cleft lip and palate that were characteristic for their final development after the completion of all therapeutic procedures were described in an earlier paper (Šmahel and Brejcha, 1983). The basic deviations observed consisted of a shorter depth of the maxilla and of its displacement backwards, deficient vertical growth of the upper face, retroinclination of the dentoalveolar component of the upper jaw, increase of some width parameters of the zygomaticomaxillary complex (nasal cavity and interorbital distance), and a growth deficiency of the lower jaw associated with changes in its shape. These basic deviations led to numerous other changes in the facial configuration in clefts (including facial proportions, maxillomandibular relations, and profile appearance). Soft-tissue changes consisted mainly of a reduced thickness and height of the upper lip. The purpose of the present study was to ascertain which of these deviations were present prior to the primary surgical repair of the palate and which developed later. This would enable a better insight into the causes and mechanisms of their development.

MATERIAL AND METHODS

The series included 30 boys with unilateral cleft lip and palate. They were examined on their admission for primary palatoplasty. Fifteen of the boys had complete clefts and 15 incomplete clefts; 10 of the latter group had a persisting tissue bridge at the lower margin of the nostril and the other five had both soft tissue and skeletal bridge across the anterior maxilla. The group mean age was 5 years 3 months, and ages ranged from 4.5 to 6 years. All boys still had deciduous incisors present, and none of them had any associated inborn malformations. The lip had been repaired according to the technique of Tennison, utilizing a narrow periosteal flap, in

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most cases at about 8 months of age. Perioplasty was not used in individuals with a bony bridge nor in half of those with a soft-tissue bridge across the cleft. The patients were not subjected to any other surgical procedures. Presurgical infant orthopaedics was not used, but in some individuals limited orthodontic therapy with removable appliances had been applied during the primary dentition. All patients were operated upon at the Clinic for Plastic Surgery in Prague.

The series of controls consisted of 27 normal boys, matched in age and selected at random from kindergartens in Prague. Their mean age was 5 years 2 months. All of the control subjects still had deciduous incisors. Cases with severe orthodontic anomalies were not included. None of these children underwent orthodontic treatment.

Cephalograms were obtained under standard conditions with the patient in centric occlusion. All films were measured by one of the authors with the help of several measuring instruments. The method was described in one of our earlier papers (Šmahel and Brejcha, 1983). The craniometric points used in the present study are marked on Figures 1 and 2, and the reference lines on Figure 3. The middle point between the two sides was marked in double contours. Measurements from craniometric points to a reference plane are calculated as the perpendicular distance from the point to the plane, for example, Cd-NSL. Angles are designated by three points, for example, S-N-Ss, or as the angular relationship of two lines or planes, for example, ML/RL. The thickness of the soft tissue profile observations are recorded as follows; Ss_t' and Pr_t are measured parallel to the palatal plane, Id_t and Sm_t' represent the smallest dimension, Pg_t' is measured perpendicular to the line N′–Pg′. The overjet was measured as the distance between the incisal edges of the maxillary and mandibular central incisors, parallel with the plane of occlusion. Observations were not included in the data when it was not possible to determine accurately the craniometric points, reference lines, or dimensions. The maximum deletion of data was in four cases, where the observation Pr_t could not be adequately determined.

The results were tested with the F-test and the t-test. The interorbital index was calculated according to the formula:

$$100 \times \frac{Mo - Mo}{Ek - Ek}$$

Results

The data are summarized in Tables 1 and 2 and in the schematic illustration, Figure 4. The only characteristic of the neurocranium which differed significantly from values in controls was the smaller height of the neurocranium (Ba-Br). The presellar (N-S) and the postsellar (S-Ba) parts of the cranial base and the length of the foramen magnum (Ba-O) were slightly shortened. However, the curvature of the base was unchanged (N-S-Ba).

More marked deviations were found in facial configuration. The upper face was reduced in anterior (N-Sp) and posterior (Pmp-NSL) heights, accompanied by elongation of nasal bones (N-Rhi). A slight retroinclination of the palatal plane (PL/VL) was related to the marked reduction of the posterior height. The depth of the upper jaw was not reduced (Sp-Pmp, Ss-Pmp, Ss-Pl); however, in relation to the cranial base, the maxilla was displaced backwards (Ptm-VL, Pmp-VL, Pl-VL). This contributed to the slight maxillary retrusion (S-N-Ss) and to the shortening of the bony framework of the nasopharynx (Pmp-Ba). There was a rather marked retroinclination of the alveolar process (ASL/PL) and of the maxillary incisors (ISL/PL).

The lower jaw was smaller, with a shortening of mandibular body (Pgn-Go) and ramus (Cd-Go) lengths. This shortening of the ramus resulted in a steeper slope of the body (ML/VL) and a more obtuse gonial angle (ML/RL, $p < 0.1$), as well as other adaptive changes, i.e., a more acute chin angle (CL/ML), retroinclination of lower incisors (IIL/ML) and an increase in the anterior mandibular height (Ii-Gn, Id-Gn). Because of this adaptation, the inclination of the lower incisors (IIL/NSL) and of the anterior wall of the mandibular symphysis (CL/NSL) toward the cranial base remained unchanged. The shortening of the body contributed to retrogenia (S-N-Id, S-N-Sm, S-N-Pg). The position of the mandibular joint (Cd-NSL, N-S-Cd) and the inclination of the ramus (RL/NSL) were unchanged. There was a posterior rotation of the mandible (N-S-Pgn, ML/VL).

Because of the identical retrusion of both jaws, the convexity of the profile (N-Ss-Pg) and the sagittal maxillomandibular relations (Ss-N-Sm) were not disturbed. The deviation of the vertical maxillomandibular relations (PL/ML, $p<0.1$) and the increased height of the lower

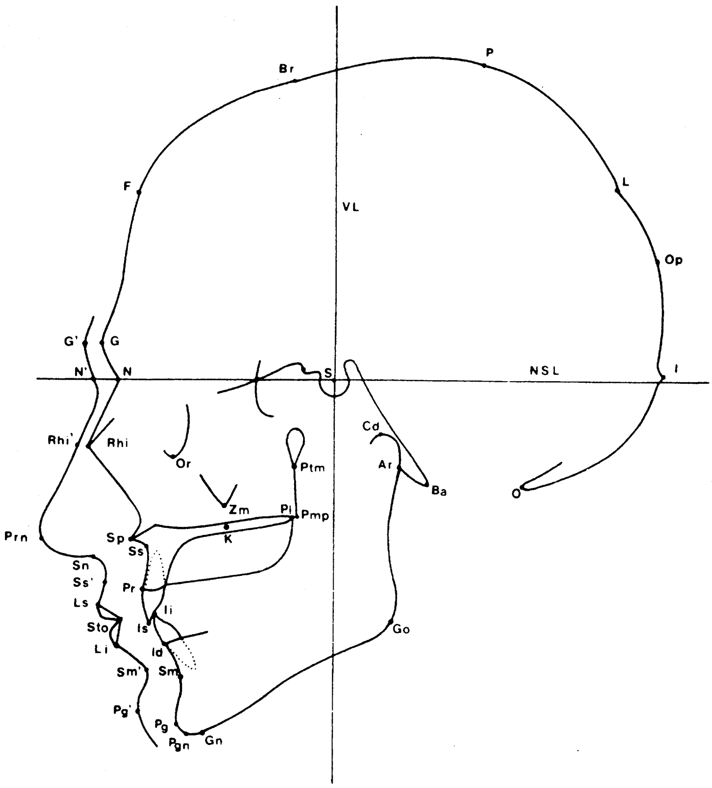

FIGURE 1 Cephalometric points used for the assessment of lateral radiographic films: Ar (articulare) = intersection of inferior contour of the clivus and posterior contour of the ramus; Ba (basion) = most posteroinferior point on the clivus; Br (bregma) = intersection of the coronal suture and lamina externa of the cranial vault; Cd (condylion) = most superior point on the condylar head; F (frontale) = intersection of the perpendicular to the dimension N-Br through its midpoint and lamina externa of the cranial vault; G (glabella) = most prominent point of the supraorbital ridges; G′ (soft glabella) = point on the soft profile contour over G parallel to NSL; Gn (gnathion) = lowest point of the mandibular symphysis; Go (gonion) = point on the angle of the mandible determined by the axis of ML/RL angle; I (inion) = top of the protuberantia occipitalis externa; Id (infradentale) = point of the gingival contact with lower central incisor; Ii (incision inferius) = incisal tip of the lower central incisor; Is (incision superius) = incisal tip of the upper central incisor; L (lambda) = intersection of the lambdoid suture and lamina externa of the cranial vault; Li (labrale inferius) = margin of the vermilion of the lower lip; Ls (labrale superius) = margin of the vermilion of the upper lip; N (nasion) = most anterior point on the frontonasal suture; N′ (soft nasion) = intersection between NSL and soft profile contour; O (opisthion) = most posterior point of the foramen magnum located by the prolongation of the posterior wall of the spinal canal up to the occipital bone; Op (opisthocranion) = point on the surface of the cranial vault farthest from nasion; P (parietale) = intersection of the perpendicular to the dimension Br-L through its midpoint and lamina externa of the cranial vault; Pg (pogonion) = most anterior point on the bony chin; Pg′ (soft pogonion) = most anterior point on the soft tissue chin; Pgn (prognathion) = point on the mandibular symphysis farthest from Cd; Pl (palatale) = most posterior point of the palatal processes; Pmp (pterygomaxillare palatinum) = intersection of palate line with the fissura pterygomaxillaris; Pr (prosthion) = point of gingival contact with upper central incisor; Prn (pronasale) = point on the top of apex nasi; Ptm (pterygomaxillare) = most inferior point of fossa pterygopalatina where fissura pterygomaxillaris begins; Rhi (rhinion) = most inferior point on the nasal bone; Rhi′ (soft rhinion) = point on the soft profile contour over Rhi; S (sella) = centre of sella turcica; Sm (supramentale) = deepest point on the anterior contour of the mandibular symphysis; Sm′ (soft supramentale) = deepest point on the soft contour of the lower jaw; Sn (subnasale) = point at which columella merges with the upper lip; Sp (spinale) = tip of the anterior nasal spine; Ss (subspinale) = deepest point of the subspinal concavity, Ss′ (soft subspinale) = deepest point of the upper lip; Sto (stomion) = point of contact of the upper and lower lip.

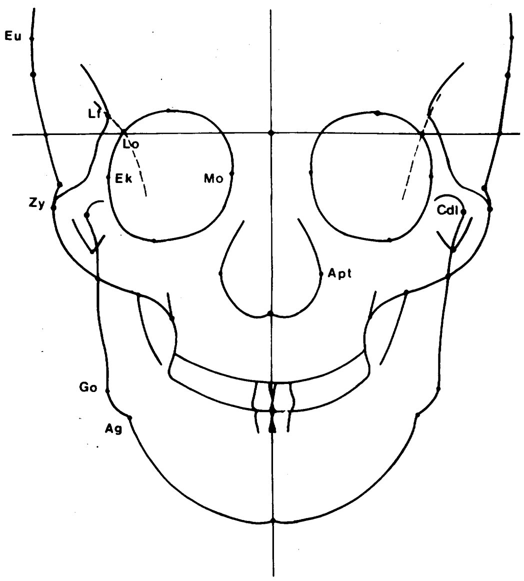

FIGURE 2 Cephalometric points on anteroposterior radiographic films: Ag (antegonion) = highest point in the antegonial notch; Apt (apertion) = most lateral point of the nasal cavity; Cdl (condylion laterale) = most lateral point on the condylar head; Ek (ectoconchion) = most lateral point of the orbital contour; Eu (euryon) = most lateral point of cranial vault; Go (gonion) = most lateral point of the mandibular angle; Lf (laterofrontale) = point of intersection between lateral margin of the ala major and lateral margin of the proc. zygomaticus of the frontal bone; Lo (lateroorbitale) = point of intersection between lateral margin of the ala major and contour of the orbita; Mo (medioorbitale) = most medial point of the orbital orifice; Zy (zygion) = most lateral point on the zygomatic bone.

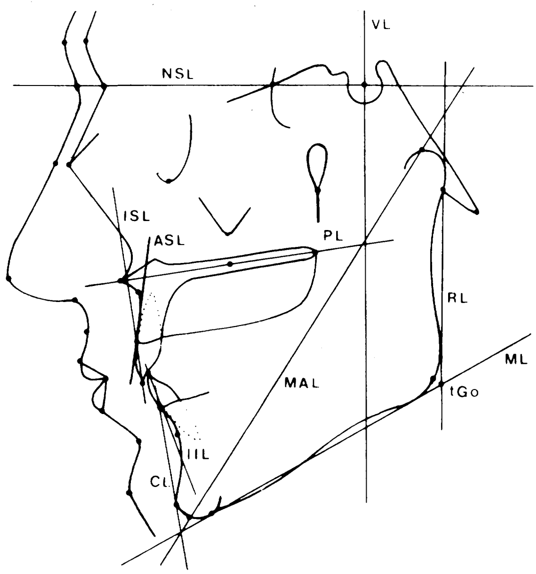

FIGURE 3 Reference lines plotted on lateral radiographic films: NSL = line through N and S; VL = perpendicular to NSL through S; PL = line through Sp and Pl; CL = line through Pg and Id; ML = tangent to the mandibular body through Gn; RL = tangent to the mandibular ramus through Ar; MAL = line through Pgn and Cd; ASL = tangent to the maxillar alveolar process through Pr; ISL = line through Is and Pr; IIL = line through Ii and Id; tGo = tangenta gonion point.

face (Sp-Pg) also were not significant. The relation of the posterior (S-Go) to the anterior (N-Gn) height of the face confirmed the restriction of anterior growth rotation during the development of the face (60.52% versus 62.75% in controls). The dentoalveolar retroinclination of the maxilla resulted, on the average, in an edge-to-edge bite.

Many parameters of the soft tissue profile were related to the configuration of the underlying skeletal framework. This applied to characteristics showing deviations (retrusion of the face: S-N′-Ss′, S-N′-Sm′, S-N′-Pg′), as well as to those which did not differ from the controls (the convexity of the face: N′-Prn-Pg′, N′-Sn-Pg′, Ss′-N′-Sm′). A reduction of the nasal depth (Prn-Sn, Prn-Sp), reflected the flattening of the nasal apex. The height of the upper lip was shortened as well; however, the shortening was significant only in measurements of its dermal portion

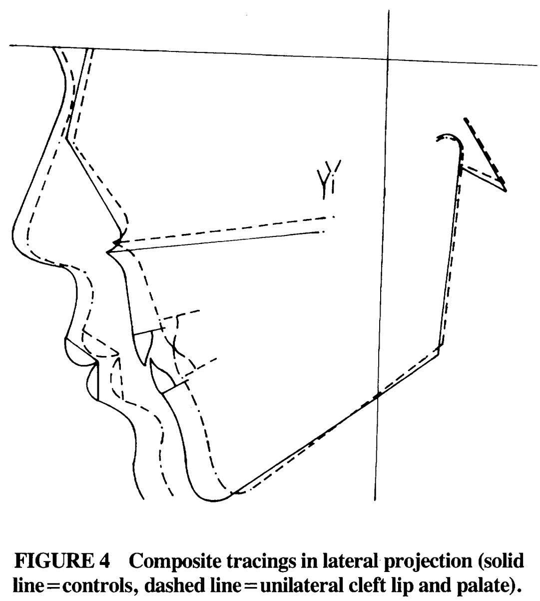

FIGURE 4 Composite tracings in lateral projection (solid line = controls, dashed line = unilateral cleft lip and palate).

TABLE 1 Cephalometric Radiographic Characteristics in Lateral Projection in Boys with Unilateral Cleft Lip and Palate Prior to Palatal Surgery (in mm or degrees)

Variable	$\bar{X}$	*SE*	*Dif**	*Variable*	$\bar{X}$	*SE*	*Dif**
Cranium				*Mandible*			
Ba-Br	139.10	0.87	−3.63†	Pgn-Go	56.27	0.59	−2.29**
Ba-O	35.93	0.45	−1.37**	Cd-Go	43.07	0.53	−3.26‡
N-Ba	94.67	0.78	−2.03**	S-Pgn	99.33	0.88	−3.41**
N-S	64.37	0.56	−1.44**	Ii-Gn	35.20	0.32	+1.09**
S-Ba	36.93	0.48	−1.66**	Id-Gn	28.33	0.33	+1.66†
				N-S-Pgn	71.27	0.47	+2.38†
Upper Face							
N-Rhi	20.53	0.43	+1.34**	S-N-Id	73.07	0.57	−2.89†
N-Sp	41.87	0.47	−1.65**	S-N-Sm	72.17	0.50	−2.57†
Pmp-NSL	34.30	0.42	−3.14‡	S-N-Pg	72.37	0.45	−2.26†
Ss-Pmp	43.83	0.47	−0.39	ML/VL	129.03	0.87	+3.81†
Ptm-VL	11.50	0.34	−1.94‡	ML/RL	133.27	1.03	+2.49
Pmp-VL	11.37	0.44	−2.15‡	CL/ML	71.37	0.89	−3.37†
Pl-VL	13.10	0.45	−1.53**	IIL/ML	66.79	1.17	−5.28†
Pmp-Ba	39.10	0.49	−2.86‡				
S-N-Ss	76.93	0.58	−2.14**	*Soft Profile*			
S-N-Pr	74.73	0.57	−4.34‡	S-N′-Ss′	81.23	0.61	−2.18**
PL/VL	98.57	0.39	+1.68**	S-N′-Sm′	73.17	0.47	−3.20‡
ASL/PL	82.07	1.02	−7.12‡	S-N′-Pg′	74.57	0.49	−2.62†
ISL/PL	68.04	1.68	−5.92†	Prn-Sn	13.03	0.22	−1.04†
				Prn-Sp	18.40	0.32	−1.67‡
Face as a Whole				Sn-Ls	11.93	0.32	−2.26‡
N-Gn	100.57	0.89	+0.24	Sn-Sto	19.13	0.39	−0.98
S-Go	60.87	0.75	−2.09**	Ls-Li	17.17	0.82	+2.69†
Ss-N-Sm	4.77	0.38	+0.44	Ss'_t	13.13	0.32	+1.28†
PL/ML	30.47	0.96	+2.14	Pr_t	15.93	0.35	+3.03‡
Overjet	0.03	0.37	−1.95‡	Id_t	9.52	0.22	+0.78†

* Dif=difference between values for children with clefts and normal (control) children
** Dif=$p<0.05$
† Dif=$p<0.01$
‡ Dif=$p<0.001$

Appendix: The other measured nonsignificant characteristics were: N-Op, Ba-L, S-P, N-S-F, N-S-Br, N-S-L, N-S-Ba, Sp-Is, Sp-Pmp, Ss-Pl, Sp-Pg, Cd-NSL, S-N-Rhi, N-S-Cd, N-S-Ar, RL/NSL, IIL/NSL, CL/NSL, N-Ss-Pg, Ss′-N′-Sm′, N′-Prn-Pg′, N′-Sn-Pg′, N′-Prn, N′-Sn, N′-Sto, N′-Pg′, Sm'_t, Pg'_t and some additional signs used for construction purposes.

TABLE 2 Cephalometric Radiographic Characteristics in the Anteroposterior Projection*

Variable	$\bar{X}$	*SE*	*Dif*	*Variable*	$\bar{X}$	*SE*	*Dif*
Cranium				*Upper Face*			
Eu-Eu	152.37	1.03	−2.67	Zy-Zy	119.97	0.77	−1.03
Lf-Lf	102.45	0.58	−1.65	Mo-Mo	23.60	0.43	+0.71
				Lo-Lo	93.69	0.52	−0.75
Mandible				Ek-Ek	94.90	0.53	−1.32
Go-Go	84.13	0.86	−1.46	Apt-Apt	30.60	0.33	+3.64‡
Ag-Ag	77.53	0.66	−1.21	Index interorb	24.83	0.40	+1.01
Cdl-Cdl	104.57	0.62	−1.24				

* For explanatory notes, see footnotes to Table 1.

without including the vermilion (Sn-Ls v. Sn-Sto). This was suggestive of the greater thickness of the vermilion which was also indicated by an increase in the dimension Ls-Li. Due to the dentoalveolar retroinclination, there was also an increased thickness of the upper lip (Ss'_t, Pr_t), and at the level of the infradental point a similar increase in the thickness of the lower lip (Id_t). This finding corresponded to the very good prominence of the upper lip (Ss′-N′-Sm′). A deficient vertical lip closure during centric occlusion was present in six individuals, with an average gap of 4.8 mm as compared to two individuals from the group of controls.

Only width dimensions were assessed in the anteroposterior projection (Table 2). The single significant deviation observed consisted of a markedly widened nasal cavity (Apt-Apt). The width dimensions of the neurocranium (Eu-Eu), forehead (Lf-Lf), face (Zy-Zy), and mandible (Go-Go, Ag-Ag and base Cdl-Cdl) were unchanged. The interorbital distance (Mo-Mo) showed only a very slight enlargement, which was evident only in relation to other width parameters (interorbital index, $p < 0.1$).

Discussion

Our findings indicate that, with the exception of the normal depth of the maxilla, all other basic deviations of craniofacial configuration in unilateral cleft lip and palate described by us in adults (Šmahel and Brejcha, 1983) were present prior to the primary surgical repair of the palate. The height of the upper face was reduced; the maxilla was displaced backward with a dentoalveolar retroinclination and an increased width of the nasal cavity. The first two deviations were approximately of the same extent as found in adulthood, but dentoalveolar retroinclination was somewhat less and the widening of the nasal cavity was more marked in children. The shortening of the mandibular body and ramus was nearly the same as in the adult, although the compensatory changes of mandibular shape (steeper slope of the body, more obtuse gonial angle, more acute chin angle, increased anterior height of the mandible) were less. However, this comparison showed only a trend of the development of the facial bony framework that was recorded in the earlier described series of adults after surgical repair. It did not anticipate how the development of all of these parameters will proceed in this series of children after their treatment with more recent therapeutic methods.

The reduction of upper face height in children with cleft lip and palate prior to palatoplasty confirmed the early disturbance of vertical development which is considered by some to result from an impairment in the interaction between the maxilla and the growth-regulating nasal septum. Some other authors also believe that this deviation occurs as early as infancy (Bishara et al, 1979; Nordin et al, 1983; Šmahel et al, 1985). It is assumed that the posterior displacement of the maxilla represents an early deviation that is not related to palate surgery. This is suggested also by the fact that we failed to observe this deviation in adult individuals with isolated cleft palate (Šmahel, 1984a), as well as in children prior to primary surgical repair (Šmahel et al, in press). So far it is not possible to establish definitely the cause of this deviation, which was also demonstrated in bilateral cleft lip and palate patients (Šmahel, 1984b).

The deficient growth of the mandible also represents an early disorder which could lead to the development of an isolated cleft of the palate, as was demonstrated in experiments on laboratory animals (Jelínek and Peterka, 1977; Diewert, 1979). The relationship of this disorder to the development of cleft lip and palate has not yet been established. It could represent a teratogenic disturbance of the growth of the mandibular "anlage" at a period when this disturbance simultaneously induces the development of cleft lip and palate (first critical period). In clefts of the lip alone that develop during the same period, an impairment of mandibular growth does not occur (Dahl, 1970; Šmahel 1984c) because of a weaker teratogenic action. This difference could also be the result of a varying duration of the teratogenic impact. In the other two basic skeletal facial deviations seen prior to palatal surgery, the maxillary dentoalveolar retroinclination may have resulted from increased lip tension after cheiloplasty, and the widening of the nasal cavity could be related to the nonunion of the maxillary segments. The widening of maxillary structures prior to palatal repair was demonstrated by Subtelny in 1955.

A substantial maxillary difference was found in these children, compared to adults. In children, the depth of the maxilla was normal prior to palatoplasty. Thus, this shortening, leading to a retrusion of the upper jaw, developed at a later date. Whether it was due exclusively to the sequelae of palatoplasty or whether it represented also a late sequela of a primary disorder in the growth potential of the maxilla could not be definitely established. In order to clarify our findings we have assessed the depth of the upper jaw separately in complete and incomplete clefts, since the observed shortening of the maxilla in adults was much more marked in complete than in incomplete clefts (Šmahel and Brejcha, 1983). In children, we failed to disclose any difference between these two types of clefts (mean values were larger by 0.5 mm in complete clefts). Similarly, we failed to demonstrate a substantial shortening of maxillary depth in patients with isolated cleft palate prior to palatoplasty (Šmahel et al, in press) contrary to our observations in adult patients (Šmahel, 1984a). This evidence indicates that this deviation develops postoperatively. Bishara et al (1979) observed during longitudinal studies of complete clefts that, at the age of 5 years, the maxilla shows even a greater depth than the norm. During subsequent years, the difference gradually decreased because of the more exuberant anterior growth of the maxilla in controls. Throughout the entire observation period, the angle SNA (S-N-Ss) was more acute, which, in the presence of an increased depth of the maxilla, could be accounted for by the displacement of the maxilla backward. Examinations of adult patients who were not treated by surgery also showed an adequate anterior growth of the maxilla (Ortiz-Monasterio et al, 1959, 1966; Pitanguy and Franco, 1967; Bishara et al, 1976). Dahl (1970), however, demonstrated a shortening of the depth of the maxilla in adults without palatoplasty.

The other described skeletal deviations resulted from the basic deviations previously described (retrusion of both jaws, limited anterior growth rotation of the face, shortening of the depth of the nasopharyngeal bony framework, and anomalies of occlusion) or were of no practical importance. We have observed an elongation of the nasal bones similar to that found in adults. Although there was evidence of its correlation with maxillary retrusion (Šmahel and Brejcha, 1983), this does not serve as a full explanation of its cause. In two facial characteristics, the relation between the preoperative and final findings remained unclear. In adults, we failed to observe a reduction in the posterior height of the upper face or a retroinclination of the palatal plane. However, information reported in the literature shows that these characteristics can undergo changes with age. A trend toward these variations was seen (Hayashi et al, 1976). Further, we did not observe the significant increase in the interorbital distance that has been demonstrated repeatedly in the literature (in children, by Graber, 1964; Aduss et al, 1971; Figalová et al, 1974). We have stated that such an increase was apparent in relation to facial width parameters, especially in the orbital region (interorbital index, $p < 0.1$). Contrary to that which was seen in adults, our children had neither a shallower slope of the forehead nor posterior rotation of the cranial vault. The cranial base showed a shortening of its postsellar part. The other characteristics of the cranium yielded similar findings in both children and adults.

Of the variations seen in the soft tissues, the height of the upper lip showed a similar reduction both in adults and in children prior to palatoplasty. This shortening resulted primarily from a tissue deficiency. The observations differed between adults and children in the thickness of the upper lip. It was always narrower in adults, thus making the retrocheilia even more conspicuous. The higher mean values recorded prior to palatoplasty resulted from the retroinclination of the maxillary dentoalveolar component. At that age the lip appeared visually to be hypertrophic, with good prominence. This increased thickness resulted from the stretching of the lip into the vestibular sulcus, which was increased because of retroinclination of the anterior maxillary segment.

In the literature available, we failed to find a study dealing in more detail with an analysis of the craniofacial deviations in cleft lip and palate patients prior to palatoplasty. Therefore, we did not have data at our disposal that would allow a comparison with our findings. The main features of our results were, however, in agreement with present knowledge.

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