Morphology of Facial Bones in Skulls with Unoperated Unilateral Cleft Palate

J. DAVID ATHERTON, F.D.S., D.D.O
Liverpool, England

This paper describes the appearance of skulls affected by a complete unilateral cleft of the primary and secondary palate. The skulls range in age from birth to old age. They were of persons who had received no surgical correction of the deformity. Although they are of different races and ages, the skulls have many features in common, attributable to the cleft. They probably represent the way a unilateral cleft palate baby would grow if it was left untreated.

The unilateral cleft is particularly interesting as it represents a natural experiment in which the maxilla on the cleft side has been separated by the cleft from several structures with which it would normally be in continuity, and which may play an important part in its development. The bones on the noncleft side are used in this paper as a control for those bones on the cleft side.

Review of the Literature

Studies of the appearance, growth, and development of untreated clefts fall into two general groups.

The first group of studies consider the untreated living patient. Innes (*4*) presented a group of patients of the Dusan tribe in Borneo. Eleven of the patients were unilateral cleft lip and palate (in six of these, the alveolus only was involved). Innes described in these patients a medial palatal collapse of the lateral alveolar segment associated with diminished downward (vertical) growth of the alveolus resulting in cross bite, lateral open bite, and anterior open bite. Other workers have made cephalometric studies of the face of similar patients. Monasterio and associates (*7*) studied 14 untreated complete unilateral cases in Mexico. Law and associates (*5*) studied patients in Puerto Rico but included the seven untreated patients with the treated. Other studies refer to patients in which the lip has been closed surgically (De Jesus, *2*, Mestre and associates, *6*). These cephalometric studies, although including numbers of treated cases and different types of cleft, indicate that there is no difference in the dimensions of facial development between unilateral cleft patients and appropriate controls, apart from the region of the cleft.

The second group of studies deals with the morphology of untreated

Dr. Atherton, formerly Research Associate at the University of Pittsburgh Cleft Palate Research Center, is Lecturer in Orthodontics, University of Liverpool. Part of the investigation for this paper was conducted at Pittsburgh, supported by PHS Research Grant DE 01697, National Institute of Dental Research.

skulls. Veau (*12*) describes a number of skulls. Psaumbe (*9*) further describes these same skulls. Van Limborgh (*13*) studies cleft skulls of different ages and types. His conclusions were that in individuals with clefts the growth processes may always proceed more slowly than in normal individuals but that the growth potentials are essentially the same, except in the region of the cleft. Regarding the complete unilateral adult skulls, he observes a slight rotation in the region of the premaxilla. On the cleft side, his interpretation is that the maxilla is developed in its lower parts to the same extent as the other side but is displaced in a posterior direction. The pterygoid process on the affected side is larger than on the unaffected side and its direction is more posterior.

Material

The material studied consists of 17 unilateral cleft palate skulls. These skulls were examined in various museums and, as far as is known, the individuals had not received treatment. They were of different and sometimes unknown racial origin. The portion of the skull preserved varied from the facial part alone, to the whole skull. The lower jaw was present on only two skulls. The skulls are described in Table 1. Unless otherwise stated, the cleft was a complete unilateral of the primary and secondary palate.

Description of the Skulls with Complete Clefts

At birth the unilateral cleft palate skull (Figure 1) shows certain deviations from the normal which are characteristic of this congenital ab-

TABLE 1. Description of the 15 skulls used in this study.

specimen	*museum*	*age (approx.)*	*description of cleft*
V11	Vrolik	birth	complete
V12	Vrolik	birth	complete
42	University College Hos. London	9 months	complete
V17	Vrolik	4 years	primary and partial secondary
293252	Smithsonian	9 years	complete
V22	Vrolik	12 years	primary
266052	Smithsonian	adult	partial primary secondary
43	Royal College Surg. London	adult	complete
V14	Vrolik	adult	Simonart's bar
V16	Vrolik	adult	complete
V18	Vrolik	adult	primary
V13	Vrolik	adult	complete
316482	Smithsonian	adult	partial primary secondary
V15	Vrolik	old age	complete
41	Royal College Surg. London	old age	complete

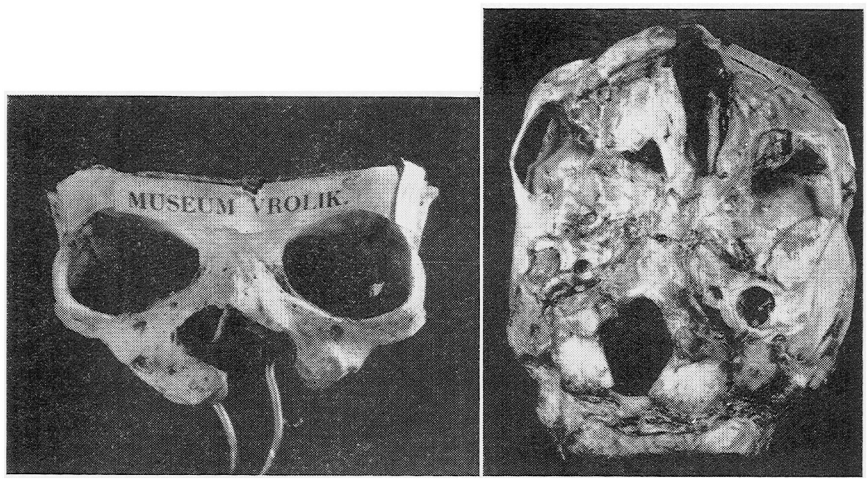

FIGURE 1. Specimen VII. The skull of a newborn infant with a complete unilateral cleft.

normality. From the frontal aspect, there is a distortion of the nasal aperture. The anterior nasal spine is displaced away from the center line in the direction of the noncleft side. The nasal bones are markedly distorted. On the cleft side, the nasal bone is flattened and, on the noncleft side, this bone is curved convexly outward. The effect on the nasal bones continues into the frontal process of the maxilla on each side. When it is present, the cartilaginous nasal septum can be seen extending downwards and laterally from the internasal suture to the anterior nasal spine.

The palatal view of the palate shows the two parts of the palate widely separated. They are equally displaced from the midline. The maxilla on the cleft side lies slightly retroplaced in relation to the opposite side. The premaxilla on the cleft side is very much reduced in size, is rotated forwards, and is largely composed of the alveolar bone around the developing teeth. The occlusal plane of the incisors slopes progressively upwards from the noncleft to the cleft side, so that the incisor adjacent to the cleft lacks a considerable degree of vertical development. The anterior open bite that is created by this failure of vertical development (*4*) is reduced in size by the adaption to the cleft area by the alveolus and, to a lesser extent, the body of the mandible (Figure 2). The failure of vertical development appears to be due to a shortage of basal bone rather than alveolar bone since the teeth appear to induce alveolar bone formation. The palatal shelves are reduced in size, the one on the cleft side being the more severely affected. The difference in size between the shelves is much more marked in the specimen shown in Figure 2 than in others of this age. The vomer deviates laterally from its articulation with the nasal septum so that it is almost horizontally disposed.

At nine months (Figure 3), the appearance is very similar to that at birth. By nine years (Figure 4), certain changes have taken place. The nasal bones show less deviation in form (this improvement is exaggerated by the fracture of the lower border of these bones). The frontal processes of the maxillae are still deviated. From the palatal aspect, the general features described from the newborn are present. The palate is abnormally wide. The palate shown in Figure 4, at nine years, was wider than

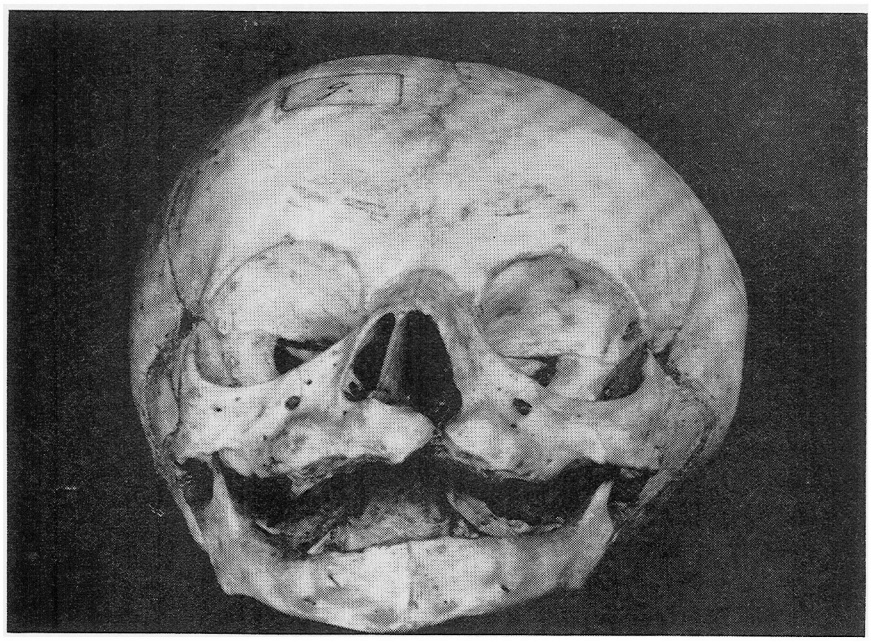

FIGURE 2. Specimen V12. A skull at birth showing the adaption of the mandible to the cleft.

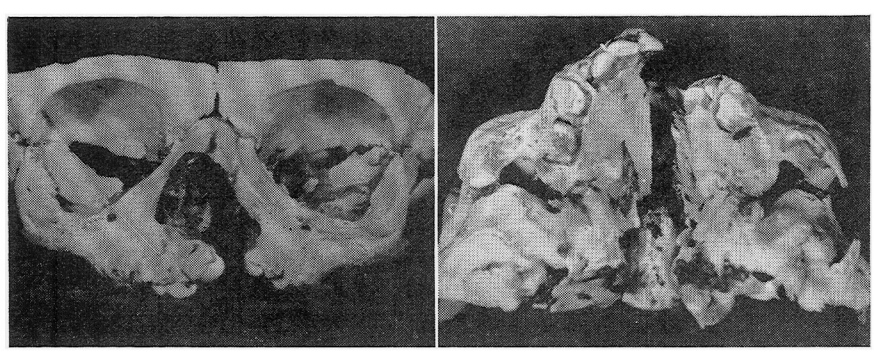

FIGURE 3. Specimen 42. A nine month old infant skull with a complete unilateral cleft.

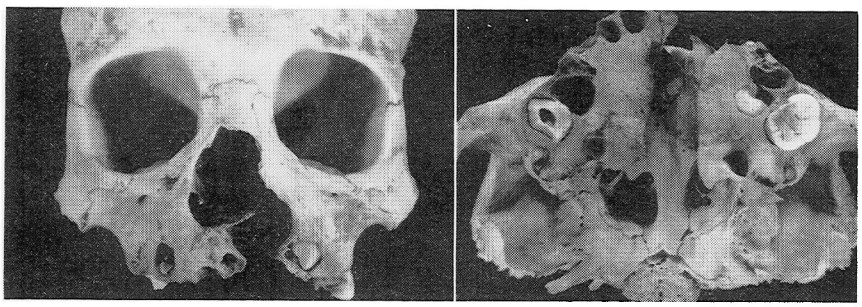

FIGURE 4. Specimen 293252. A nine year old (American Indian) skull with a complete unilateral cleft.

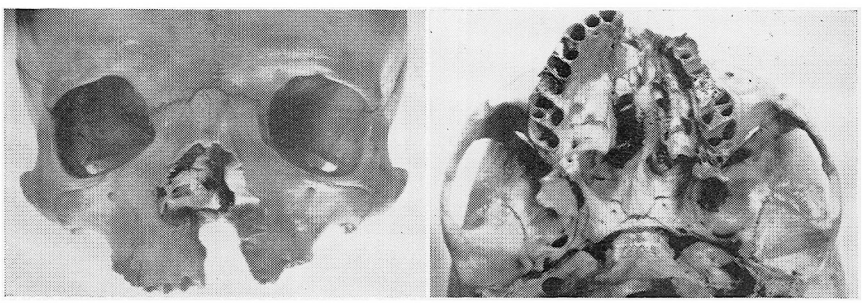

FIGURE 5. Specimen 43. An adult (South Pacific Islander) skull with a complete unilateral cleft.

that shown by the skulls of several adult members of the same American Indian tribe. The vomer is markedly displaced. Its articulation with the bony and cartilaginous nasal septum lies well over on the cleft side of the midline and, from there, the vomer slopes sharply laterally to the palatal shelves. The palatal shelves themselves are underdeveloped compared with noncleft specimens. The palatal shelf on the cleft side in this specimen (Figure 4) and in most of the others show a degree of development which is only slightly less than that of the opposite side. Vertically, the palatal shelf on the cleft side is lower than on the noncleft side. This feature is common to skulls in which the vomer retains continuity with the noncleft side. The vomer appears to restrict the vertical descent of the palatal shelf on that side (Figure 7). The maxillae in specimen 293252 are symmetrically placed. There is little to suggest that the maxilla and palatine bone are retroplaced on the cleft side except for the region of the posterior palatal spine.

The effect of the palate being abnormally wide is shown in the pterygoid region. In the normal skull, the medial pterygoid plates are almost parallel. In the untreated clefts, the medial pterygoid plates slope laterally to a greater or lesser extent (Figure 7).

Figure 5 shows the appearance of an adult skull which is typical for

the age. The nasal aperture looks much more normal than in the younger specimens. Deviations in the nasal bones are slight or even absent. The frontal processes of the maxillae still show some distortion. This particular specimen shows a metoptic suture and abnormally widely separated orbits; no other skulls showed these features. This view shows well the perpendicular plate of the ethmoid. This bone deviates toward the cleft side and meets the vomer at an angle. The junction between the two bones is much nearer the cleft side than the noncleft side. This is why patients suffer a greater degree of nasal obstruction and inflammation on the cleft side.

From the palatal aspect, features seen in Figure 5 are similar to those of the preceding specimens. The maxilla and palatine bones of the cleft side lie slightly more retroplaced than the noncleft side. This position is representative of the adult specimens with complete clefts. The position of the maxilla and palatine bones on each side of the cleft varies greatly from almost complete symmetry to a marked displacement. In specimen V13, for example, bones are perfectly symmetrical while in specimen V16, there is a marked retroplacement of the maxilla on the cleft side.

Specimen V15 (Figure 6) is an edentulous skull, probably an aged adult. The general features are similar to the other adult skulls. The vomer is well shown on this specimen. It is much less horizontal than in the younger age group and is typical of the adult skulls.

A Comparison of the Cleft and Noncleft Sides

It was anticipated when this study was initiated that there would be a marked difference between the cleft and noncleft sides (excluding the premaxillary area), that the noncleft side would be relatively normal, and that the noncleft side would serve as a control for the cleft side. However, the development of both sides appeared to be affected by the cleft, and the differences between them appeared small. Measurements were

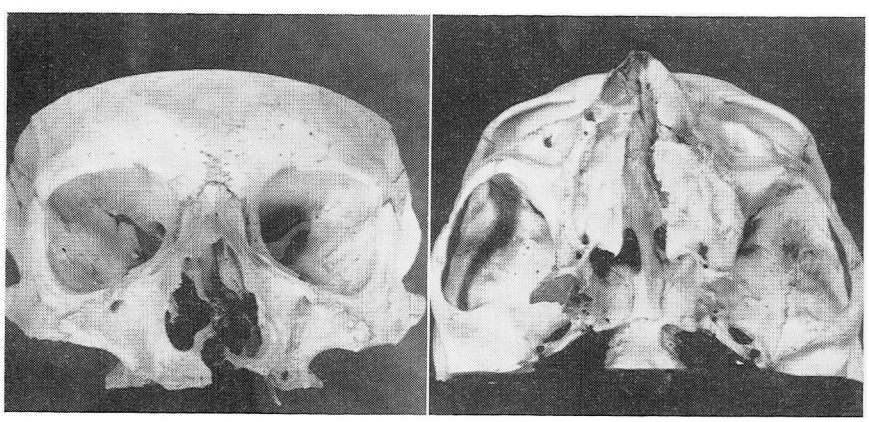

FIGURE 6. Specimen V15. Edentulous aged skull with a complete cleft.

TABLE 2. Measurements from the cleft and the noncleft sides of seven skulls. Some measurements were not available (na).

specimen	*height of maxilla (cm)*		*height of pterygoid process (cm)*		*length of maxilla (cm)*		*palatal index*
	cleft	*noncleft*	*cleft*	*noncleft*	*cleft*	*noncleft*	
42	1.55	1.38	na	na	2.38	2.4	na
43	na	na	na	na	na	na	124
293252	2.92	3.0	1.8	1.8	3.3	3.7	na
V14	4.11	4.02	3.72	3.51	4.23	4.32	109
V16	4.96	4.98	2.86	2.76	4.43	4.53	96
V13	4.63	4.61	2.87	2.96	3.4	3.9	96
V15	2.50	2.66	3.21	3.07	na	na	na

taken to compare the development of the maxilla and palatine bones on either side of the cleft. Unfortunately, and for various reasons (such as damage or protection of the specimen in a sealed box), it was not possible to take all measurements on all the specimens. The vertical development of the body of the maxilla was measured from the lower orbital margin to the alveolus in molar regions. No difference was observed between the two sides in that dimension (Table 2). The height of the pterygoid processes was measured from the base of the scaphoid fossa to the base of the hamular process and there was no difference in that dimension, either. The maxilla and palatine bones therefore achieve a similar degree of vertical development on each side of the cleft. While the body of the maxilla and palatine bones developed similarly on each side of the cleft, there appeared to be some lack in development in the immediate area of the canine on the cleft side.

The length of the maxilla on the two sides was measured from the canine crypt to the tuberosity (Table 2). There is a slight reduction in the length of the maxilla on the cleft side. The difference is small (a mean reduction of 5.6%) and probably not of clinical significance.

Difficulty was experienced in choosing suitable measurements to establish whether these skulls had a wider palate than nonclefts. The skulls were of extremely diverse origin ranging from Polynesian (specimens 43 and 41) to American Indian (specimens 266052 and 253256). Several were of unknown origin. Many skulls were undoubtedly wider than normal (for example, Figures 1, 2, 3, and 4). In these skulls, the width of the palate considerably exceeds the length. The palatal index was measured by dividing the width between the second molar sockets by the palatal length (*3*). Only four skulls had the necessary bony landmarks. They were used for study (Table 2) but the results should be viewed as being inconclusive because of the small number of observations. Two of the skulls are much wider than and the other two are within the means

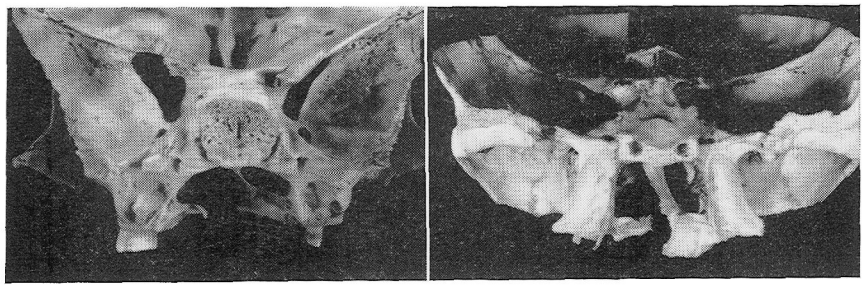

FIGURE 7. View of the pterygoid plates, vomer and palatal shelves, left, a cleft skull of nine years (specimen 293252), and right, an aged cleft skull (Specimen V15).

given in Goose's study of different British populations. The mean palatal index in the Goose study ranged from 91 to 97.

The segments were equally placed from the midline in the molar area. The canine region on the cleft side is somewhat nearer the midline than the opposite side. This appearance is enhanced by the position of the lateral incisor which remains palatal to the canine (see superior view, Figure 9). The crypt of this tooth is originally formed in this position (Figure 7); on the noncleft side, it moves forward to its place on the alveolar ridge. On the cleft side, it remains in the original crypt position.

Incomplete Unilateral Clefts

Several of the skulls had clefts which did not completely penetrate the primary or secondary palates. The 'typical' appearance so far described is, in these clefts, modified. When the primary palate only is affected, the maxillae develop normally and symmetrically. Figure 8 shows an interesting example of a complete cleft of the primary palate associated with an incomplete cleft of the secondary. Although fusion of the palate is only partially completed, symmetrical development of the maxillae and palatine bones has taken place. The area of the primary palate and nasal aperture in this type of cleft appears similar to skulls where there is a complete cleft of the primary and secondary palates.

Specimen 266052 (Figure 9) has a complete cleft of the secondary palate on the left side and a partial cleft of the alveolus on that side. The outline of the palate is good. The posterior palatal spine is probably retroplaced on the cleft side when compared with the noncleft side (unfortunately this area is fractured); otherwise, the symmetry is good. It is interesting to note that the palatal shelf on the cleft side is fused to the suture between the vomer and ethmoid bones. The frontal aspect shows a marked step in the alveolus of the cleft side.

From the appearance of the premaxilla on the cleft side of specimen V14 (Figure 10) it was assumed that there must have been a Simonart's

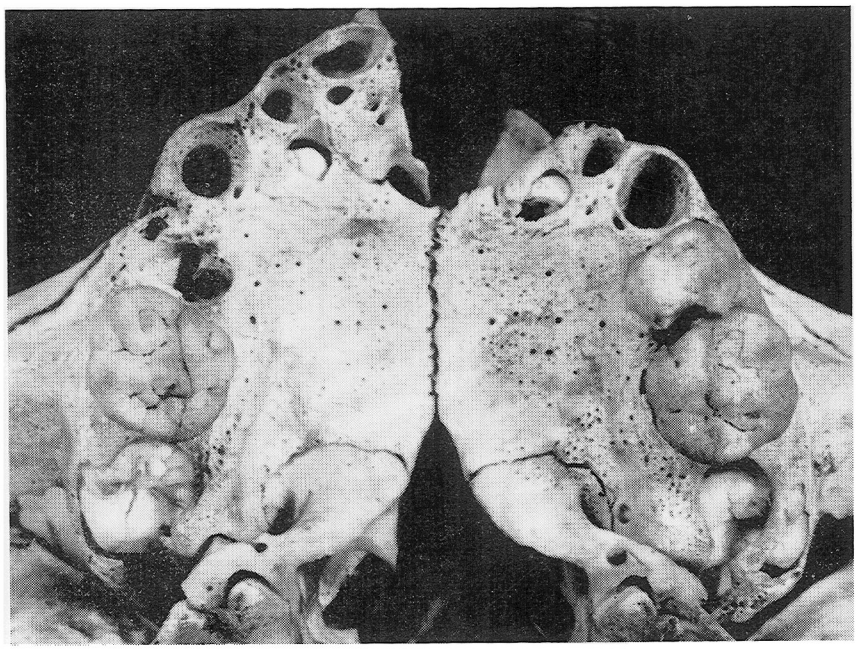

FIGURE 8. Specimen V17. A four year old with a complete cleft of the primary and an incomplete cleft of the secondary palate.

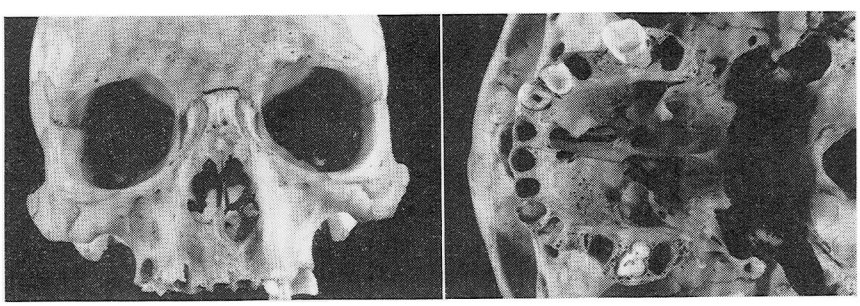

FIGURE 9. Specimen 266052. An adult skull presenting a complete cleft of the secondary palate and an incomplete cleft of the primary.

bar, which is very different from specimens with a complete cleft. There is very much more bone present along the floor of the pyriform fossa than in the complete cleft specimens where the bone is reduced to a thin lamina extending from the anterior nasal spine to the vomer. This skull shows very much more asymmetry in the position of the maxillae than do the other adult skulls. The maxilla is markedly displaced distally. The appearance of rotation inwards at the canine region is accentuated by the position of teeth palatal to the canine (first bicuspid and lateral incisor).

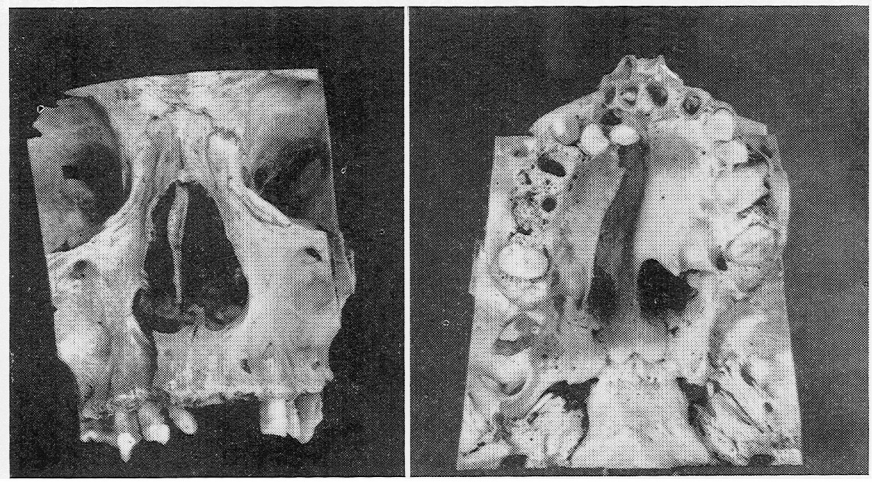

FIGURE 10. Specimen V14. An adult skull with a complete cleft of the bony parts of the primary and secondary palates. Probably in life there was a Simonart's bar present.

Discussion

The findings of this paper are in general agreement with previously published work. The development of the facial bones (apart from the immediate area of the cleft) is good. The bones show an improvement in appearance with age, certain deviations being most apparent in the younger specimens.

The nasal bones and frontal processes of the maxilla are markedly deviated in form at birth. These deviations, although present in the adult, are in many cases so reduced as to be almost undetectable. The vomer, which is almost horizontal in the unilateral cleft at birth, becomes much more upright in the adult. The change in the shape of the nasal and vomer bones seems to be associated with the nature of the original deformity and to the changes in the proportion of the face with age. The deviations seen at birth are established early in fetal life, as reported elsewhere (*1*). The deviations at birth of the nasal bones, the anterior part of the cartilaginous nasal septum, and the vomer are attributable to several factors, the most important of which are the increased width of the palate so that the nasal septum and vomer pass downwards and laterally. The reduction in width of the palatal shelf and possibly the premaxilla increases this tendency. The nasal cavity at birth is short and wide, less than one-third of the total lying at a level below the orbits. During postnatal growth, the nasal cavity increases little in width but very much in depth so that, in the adult, fully one-half of its depth lies below the level of the orbits. The anterior part of the nasal septum and vomer which passes at an angle across the short, wide, nasal cavity at

birth becomes much more upright in the adult as it traverses the much greater depth of the adult nasal cavity. The nasal bones, too, adapt to these changes, thus becoming more mature in form. The effect may be summarized in the following way. With the great increase in depth and with the little increase in width of the nasal cavity during postnatal life, the parts most severely affected by the cleft are carried away from the upper part of the face, giving rise to an improvement in shape of the nasal bones, the vomer, and the anterior part of the cartilaginous nasal septum.

Little difference in size or form was found between the bodies of the maxillary and palatine bones on either side of the cleft. The bones on the cleft side are somewhat reduced in size as compared with the noncleft side in three sites: a) the palatal shelf, b) the length of the maxilla, and c) the region of the canine. In other respects, these bones appear similar in shape and size.

The palatal shelf is consistently smaller on the cleft side. The reduction in size was not progressive with age. The palatal shelf on the cleft side of the adult shows a remarkable degree of development and remains only slightly smaller than the shelf of the opposite side. The shelf on the cleft side shows no reduction in vertical development; indeed, it lies at a lower level than that of the noncleft shelf. The attachment of the vomer exerts a slight restriction to the vertical development of the shelf on the noncleft side.

When the distance between the canine crypt and tuberosity was measured on each side, it was found that the distance on the cleft side was slightly smaller than on the noncleft side. That reduction in size was found during fetal life also (*1*). The reduction appears to be very small, the body of the maxilla on the cleft side averaging 94.5% of that of the noncleft side. It is doubtful, however, whether this is of clinical significance. There was a consistent lack of development in the region of the canine tooth on the cleft side. In each skull, the canine did not quite erupt to the same occlusal level as the rest of the teeth in the arch. It is possible that the lack of development in this region is responsible for the slight shortness of the maxilla on the cleft side.

It is apparent that, although the maxilla and palatine bones on the cleft side are separated from tissues with which they would normally be in continuity, these bones grow extremely well. Scott (*10*) has suggested that the nasal septum is the organ which, in early postnatal life, produces growth of the bones in a downward and forward direction by applying tension at the facial sutures. In the cleft face, the maxilla on the cleft side is separated from direct continuity with the nasal septum. Growth nevertheless proceeds to substantially the same degree on this side as compared to the other side which retains its continuity. However, the nasal septum may still play a part in the growth of the maxilla and of the palatine bones by applying tension through the alar of the nose. The slight retroplacement of the maxillary and palatine bones on the cleft side tends

to support the role of the nasal septum in the production of facial growth. This retroplacement, which is very characteristic of the unilateral cleft does not become more severe with age.

While it is possible to compare and contrast the bones on one side of the cleft with those of the other, it is much more difficult to assess the development of the cleft skull as a whole and compare it with the noncleft skull. It is well documented that the width of the palate and nasopharynx is increased in cleft palate infants (*8, 11*) and in the unilateral cleft fetus (*1*). Many of these skulls appeared to be wider than normal also, with a palatal index in excess of 100, but such was the diversity of origin and age of the skulls that a statistical comparison with the noncleft was not thought to be practicable.

The nasal cavity is less deep and is wider in the cleft fetus (*1*). The adult skulls show a nasal cavity which is well developed in depth. It seems possible that during fetal life the tongue influences the position of the palatal shelves and that following birth and the increase in the height of the lower part of the face, the tongue falls away from the roof of the mouth and allows the normal descent and horizontal positioning of the palatal shelves.

The typical appearance of the facial bones in the complete unilateral cleft is modified when the cleft is incomplete. Union of the bones across either the secondary or primary palate leads to an improvement in the symmetry of the maxilla and palatine bones (Figures 8 and 9), but a union of only soft tissue (Figure 10) may lead to a high degree of asymmetry.

Summary

The appearance of fifteen unilateral cleft palate skulls of approximate age from birth to aged adult is described. The general development of the facial bones of these skulls, except for the region of the cleft, was good. The deviations in the nasal and vomer bones which are marked at birth become less extensive with age. This improvement is attributed to the much greater depth of the nasal cavity in the adult skull, rather than to any improvement in the growth potential of the tissues.

A comparison of the size of the maxilla and palatine bones on each side of the cleft showed the bones on the cleft side to be smaller in the following dimensions; a) the width of the palatal shelf, b) the length of the maxilla, and c) the vertical development of the canine. The difference in size was small and did not become progressively greater with age. In other respects, the form and size of the bones on each side of the cleft was similar. It was apparent that, whatever the growth mechanisms are which determine the size of the facial bones, the bones on the cleft side achieve a remarkable degree of development.

When viewed from the palatal aspect the palate presents features which are consistent to all the skulls with complete clefts. The maxilla and pala-

tine bones on the cleft side lie slightly distal (retroplaced) to those on the noncleft side, particularly in the region of the posterior palatal spine. This characteristic appearance does not change with age. Individual skulls appeared to have a very wide palate. It was not possible, however, to prove statistically that the cleft skulls had a wider palate than the noncleft. The maxilla and palatine bones were equidistant from the midline on each side of the cleft, except in the canine region where the cleft side was somewhat nearer to the midline. The characteristic appearance of the completely cleft skull is modified when either the secondary palate or primary palate is partially or completely fused.

reprints: *Dr. J. D. Atherton*
Dental School
Liverpool University
Liverpool 3, England

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