Comparison of Midfacial Development of Children with Clefts with Their Siblings

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Craniofacial morphology in individuals with cleft lip and/or cleft palate has been studied cross-sectionally and compared to the morphology of normal groups by Graber (*4*), Moss (*7*), Mestre, DeJesus, and Subtelny (*6*), Harvold (*5*), and others. Illuminating as they are, such studies do not provide satisfactory answers to such important questions as the following. a) Assuming the cleft had been absent at birth, what would have been the normal pattern of facial development for the individual? b) If the pattern of development deviates from an expected normal, should the difference be attributed to the congenital insult or to the effect of all types of treatment instituted after birth?

The problem is essentially one of establishing controls. Sassouni (*8*) has proposed that the ideal procedure to investigate this and similar problems in humans would be the study of identical twins, one of whom is treated and the other of whom could be utilized as control. Siblings would form the next best control group, differing from identical twins only in their dissimilar genotypes. Of a similar genetic pool and of the same environmental background, such sibling groups would be useful in the evaluation of the cleft palate child during his growth and development.

This is a pilot study utilizing the method of using siblings as controls. No attempt will be made to compare the results obtained with cephalometric studies of previous investigations at this time.

Materials and Method

Lateral roentgenographic cephalometric plates of a group of cleft palate children and their normal siblings were obtained and traced. Twenty-seven normal siblings of the children with clefts who were

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of comparable ages were selected as controls. The 27 cleft palate subjects consisted of 12 children with Veau Type III clefts (unilateral complete cleft of lip and palate) and 15 children with Veau Type IV clefts (bilateral complete cleft of lip and palate). All had been treated surgically. They were then divided into two groups according to their age (see Table 1). Data regarding the subjects under study were obtained at the Lancaster Cleft Palate Clinic, Lancaster, Pennsylvania.

Method of Deriving Composites

The composite technique used in the present study is based upon the work of Broadbent (*2*) and Sassouni (*9*), and has been used in the establishment of growth and development standards at various age levels. Generally, the technique consists of superimposing pairs of lateral tracings at the closest resembling areas and deriving an 'average' outline of the midline of all structures by bisecting the two originals. A composite is then formed which is again paired with another original to obtain another composite, et cetera, until the last single tracing which is obtained represents the geometric average of the original group. In this study, separate composites were obtained from the Type III and Type IV cleft subjects, as well as for the normal matching siblings, for the various age groups. The composite tracings representing each type were superimposed with their matching normal siblings composite in such a manner as to obtain the maximum similarity at the cranial base. This method permits the evaluation of differences and resemblances within each group.

Findings

The following comparisons were made: all normals at two age levels; related normals to Type III clefts at two age levels; and related normals to Type IV clefts at two age levels.

Normal to Normal

Age Seven. The composites of the two normal groups (siblings of Type III clefts and siblings of Type IV clefts) in the younger age

TABLE 1. Subjects used for study, according to cleft type and age.

cleft type	*younger age group*		*older age group*		*total*
	range	*M*	*range*	*M*	
(Veau)	6 to 9	7:6	10 to 14	11:0	
Type III (unilateral)	4		8		12
Type IV (bilateral)	8		7		15

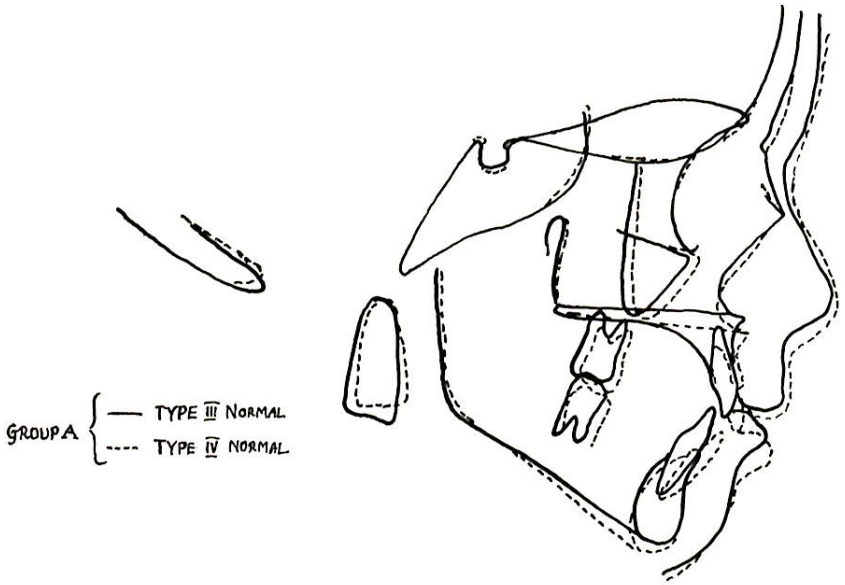

FIGURE 1. The composites of the two seven year old normal groups (siblings of Type III clefts and siblings of Type IV clefts) were superimposed. The two composites were judged to approximate each other relatively closely, indicating that the technique is sufficiently reliable to be useful.

group were superimposed at the points of their greatest concordance (Figure 1). The two composites matched each other closely, within mm divergence of any contour, indicating the reliability of the composite tracing technique.

Age Eleven. The superimposition of the two eleven-year-old normal groups shows a close similarity, just as that shown for the two younger groups (Figure 2). The greatest difference (4 mm) is seen at nasion.

Age Seven versus Age Eleven. This comparison permits the evaluation of average growth changes in size and direction between the two normal age groups (Figure 3). The findings here are in general agreement with findings by Broadbent (*1*), Brodie (*3*), and Sassouni (*9*). The palatal planes generally are parallel. ANS grows downward and forward while PNS grows straight downward. There is little difference in orbital growth after the age of seven years, six months. When the DeCoster line is used for superimposition (planum-cribriform, internal frontal contour), the behavior of nasion is erratic: upward and forward to downward and forward.

Type III Cleft versus the Normal

Age Seven. There is no appreciable difference between these two composites at cranial base size or angulation, orbito-malar contour,

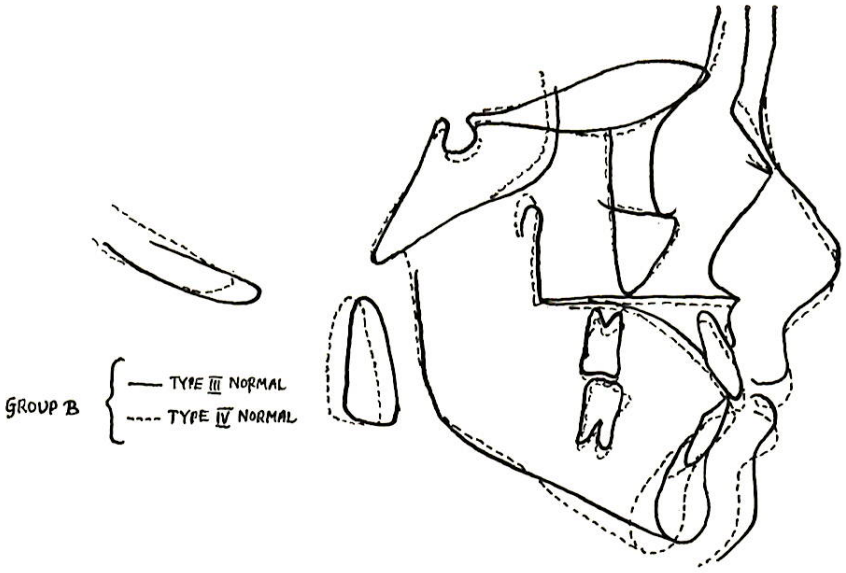

FIGURE 2. The composites of the two groups at age eleven shows they are similarly close as the two seven-year-old groups.

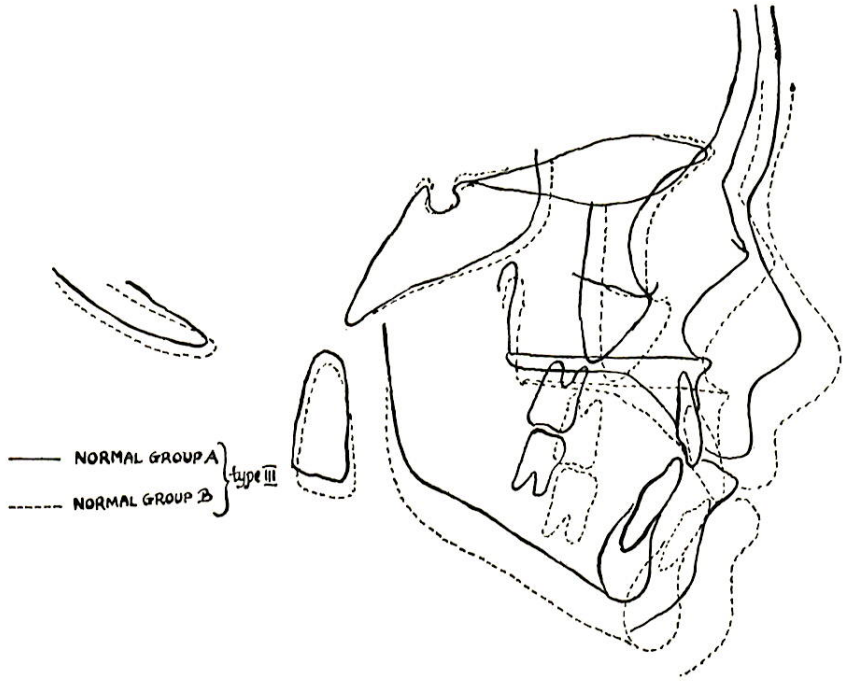

FIGURE 3. To evaluate the average growth changes for normals, the two sibling groups (age seven and eleven) were superimposed. The palatal planes, on the average, are parallel. ANS grows downward and forward, while PNS grows straight downward.

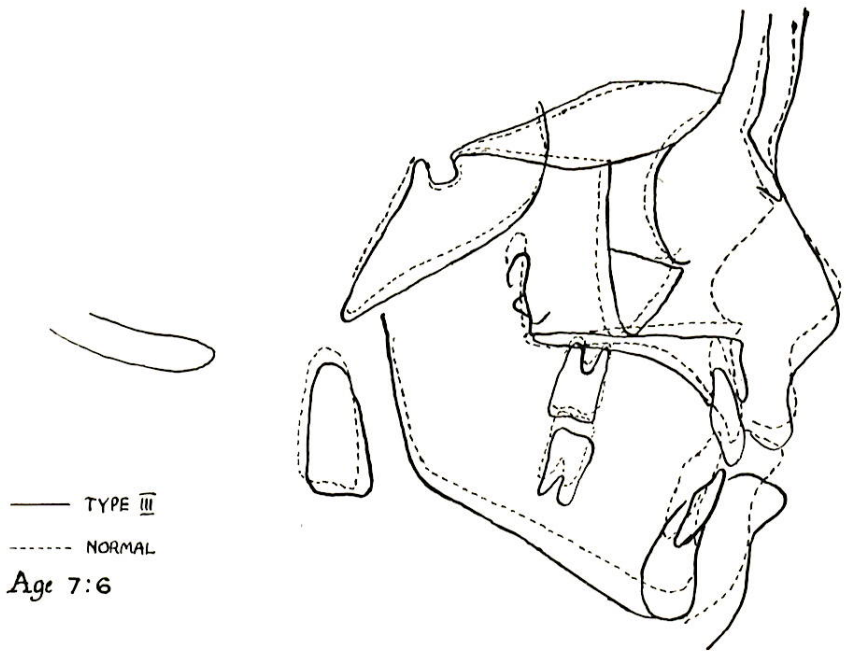

FIGURE 4. Composite Type III cleft compared to normal at 7 years, 6 months. There is no appreciable difference in cranial base or palatal size or position.

palatal size or position (Figure 4), although there is a suggestion that the palate in the cleft group is more tipped downward at ANS.

Age Eleven. There is no appreciable difference in size or in proportions between the two composites (Figure 5). The palate of the cleft group has the same general size and anteroposterior position as that of the normal group. However, there is an indication that it might be under-developed vertically (situated at a higher level).

Type IV Cleft versus the Normal

Age Seven. There is no difference in the average size of position of skeletal structures in the composites (Figure 6). The palates are parallel and of a similar size. However, the palate of the cleft group is about 3 mm higher, a position which may be indicative of a slight vertical underdevelopment.

Age Eleven. This is the group in which the most marked differences exist (Figure 7). The cranial bases of the two groups in size, shape, and angle match very closely. PNS is in the same anteroposterior level. ANS is retrusive by an average of 6 mm. This means that the overall length of the palate is short by 6 mm, on the average. The angulation of the palatal planes is different; the cleft group has the palate tipped up at PNS. This should be interpreted as a vertical posterior deficiency.

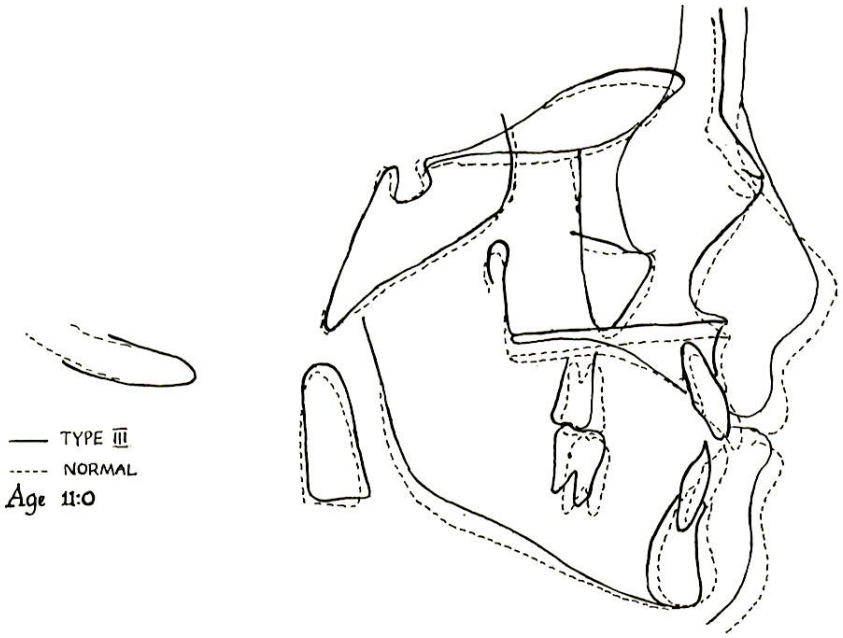

FIGURE. 5. Composite Type III cleft compared to normal at eleven years. The palate has the same size and anteroposterior position as that of the normal group. However, there is an indication that it might be under-developed vertically.

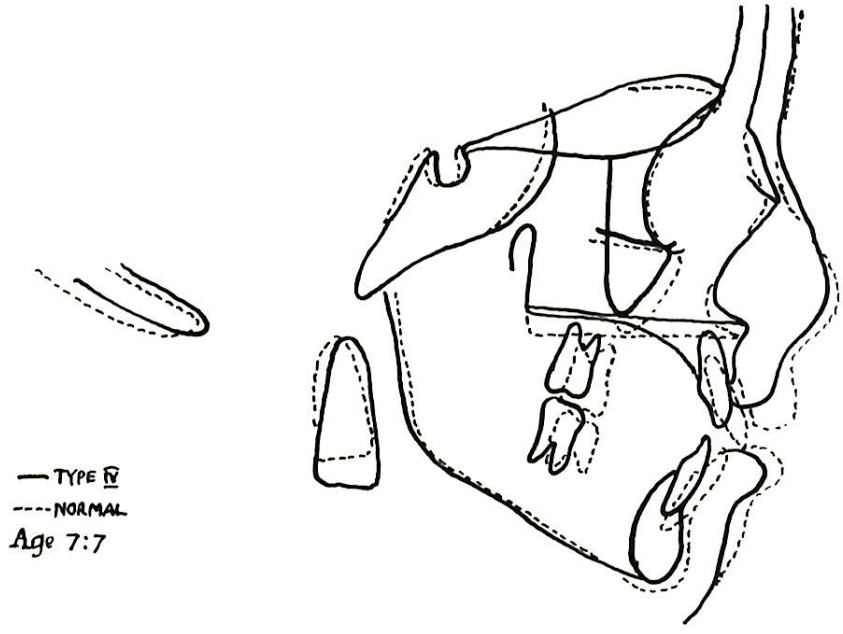

FIGURE 6. Type IV cleft and normal composites compared at seven years. There is no appreciable difference in the average in size or position of the skeletal structures. There is an indication of slight vertical under-development of palate.

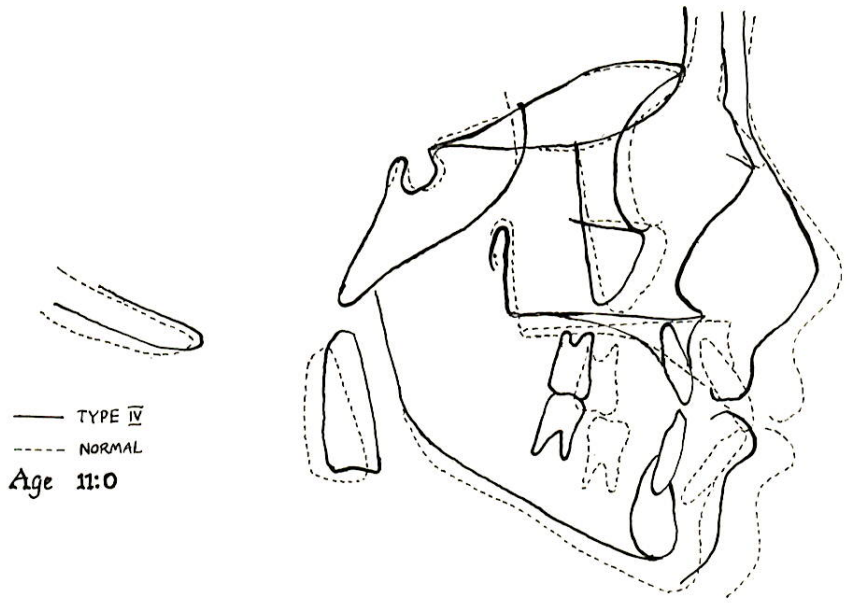

FIGURE 7. Type IV cleft compared to normal at eleven years. The PNS is at the same anteroposterior level, while ANS is retrusive by an average 6 mm. The palatal plane is tipped up at the PNS.

The malar bone follows the anteroposterior deficiency of the palate, indicating that this lack of development may not be confined to the premaxilla.

Facial Growth

Type III Cleft. As suggested by the above findings, there does not seem to be major skeletal retardation in growth in Type III between seven years, six months and 11 years, with the possible exception of a slight vertical slowing process.

Type IV Cleft. There is a major growth retardation (even a stagnation) in the Type IV group between seven years, seven months to eleven years. While nasion is growing during this period (6 mm), no change in size and anteroposterior position could be detected during this three and one-half year period. For all practical purposes, the palate stopped growing.

Discussion

The present study of the midfacial development of the cleft palate children as compared to that of normal siblings has only been exploratory in nature and cross-sectional in design. Probably the major contribution of this study is the introduction of a control group which

has a similar genetic and envrionmental background as the cleft palate group.

The method of composite has been used before and it has some advantages from the computed arithmetic standards in that it provides for comparison of not only selected landmarks, but also of the contours of all structures. The method is not influenced by and limited to certain measurements. The number of subjects studied employed here is not large enough to provide reliable standards for any cleft population, but it is sufficiently large to demonstrate the usefulness of the method. The comments concerning the growth should be taken only as indications that longitudinal studies are needed, not only on cleft population but on the normal siblings as well and over the total period of growth.

Summary

In this investigation, 54 lateral roentgenographic cephalometric plates of 27 cleft palate children and 27 of their normal siblings were used. A composite technique was used to obtain tracings for the normal and cleft palate groups at two age levels. The growth pattern of the midfacial region was also subjectively evaluated. The results show that there is no obvious skeletal retardation in growth in individuals with Type III clefts; however, in individuals with Type IV clefts, there is a major growth retardation. There was no change in size and anteroposterior position for a period of three and one-half years. The present study illustrates the need for longitudinal studies of normal and cleft palate siblings.

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