A Roentgenographic Cephalometric Study of Identical Twin Females with Cleft Lips and Palates

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Cleft palate is a congenital malformation which occurs in about one in every 800 live births in the United States (*8*). It poses complex problems not only in cosmetics and speech, but also in the psychosocial status of the patient.

A set of 21-year-old identical twin females is enrolled at the Institute of Logopedics, Wichita, Kansas. Both have operated bilateral complete cleft lips and palates. A roentgenographic cephalometric study was done to determine whether or not similarities existed in development of the maxillae. Instances of cleft lip and palate in both of identical twins are very rare. Metrakos, Metrakos, and Baxter (*7*) reported that among 29 sets of identical twins with cleft lips and palates (27 reported in the literature and two from their own study) concordance was seen only in nine sets.

Patients

Neither the parents nor an older brother has any congenital malformation. No congenital malformation or mental retardation has been reported among uncles, aunts, or cousins.

The mother was 35 years old when the twins were born. There was no previous history of stillbirth or miscarriage. The mother suffered from acute food poisoning about two weeks before delivery. This was the only prenatal incident except that at the third or fourth month the father contracted severe pneumonia which caused the mother great concern.

The patients were born on December 13, 1941. Gestation was nine months. Birth weights were five pounds for twin J and six pounds for twin E. There is nothing remarkable in their general anamneses. Surgical histories are shown in Table 1. Neither has any congenital malformation other than the cleft lip and palate. Figures 1 and 2 show profile views.

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TABLE 1. Surgical histories of twins E and J.

Surgery	*E*		*J*	
	Age	*Operator*	*Age*	*Operator*
Cleft Lip	2 mos	Dr. P.	2 mos	Dr. P.
	10 mos	Dr. P.	5 years	Dr. B.
	2 years	Dr. P.		
	19 years	Dr. B.		
Cleft Palate	5 years	Dr. B.	5 years	Dr. B.
Mandibular Prognathism			19 years	Dr. B.

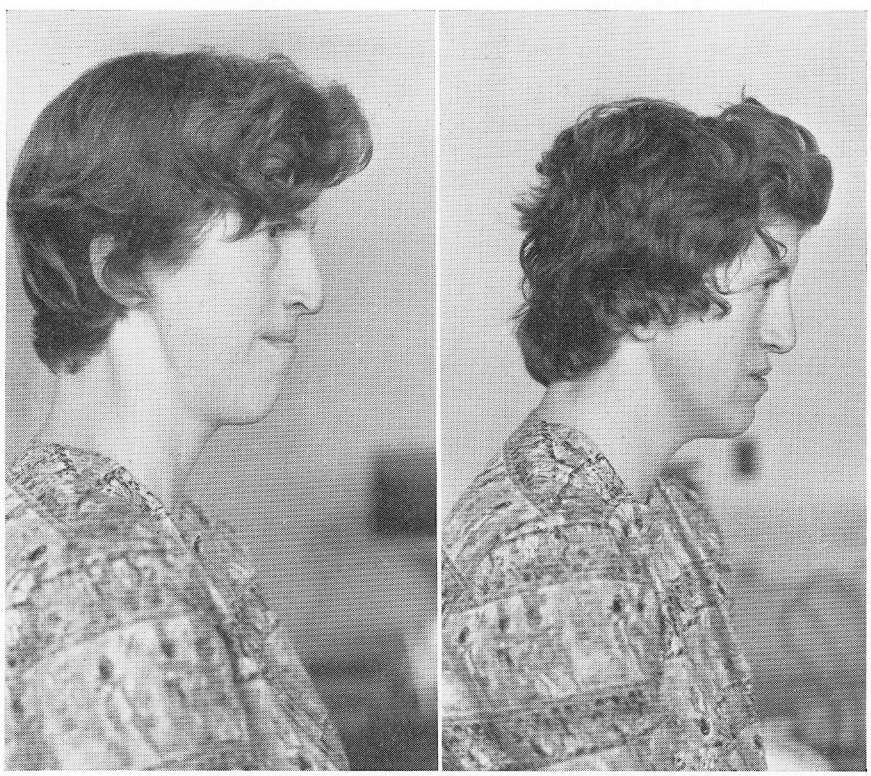

FIGURE 1. Profile view of twin J.
FIGURE 2. Profile view of twin E.

The x-ray films show that twin J has an impacted tooth in the medial portion of the maxilla while E has an impacted maxillary third molar. Verification of monozygoticity has been made from the color of skin, iris, and hair; shape of the ear; presence of mid-digital hair; and patterns of fingerprints (*5*).

Method and Results

On each twin a lateral cephalometric roentgenogram was taken in a manner suggested by Graber (*4*). The tracings are shown in Figures 3 and 4. Analyses of the tracings are shown in Table 2.

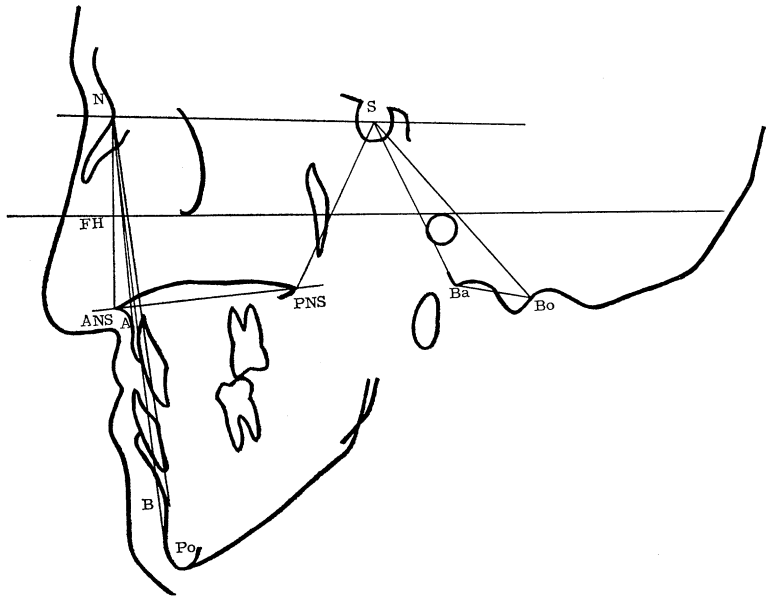

FIGURE 3. Tracings of lateral cephalometric roentgenogram taken of twin J.

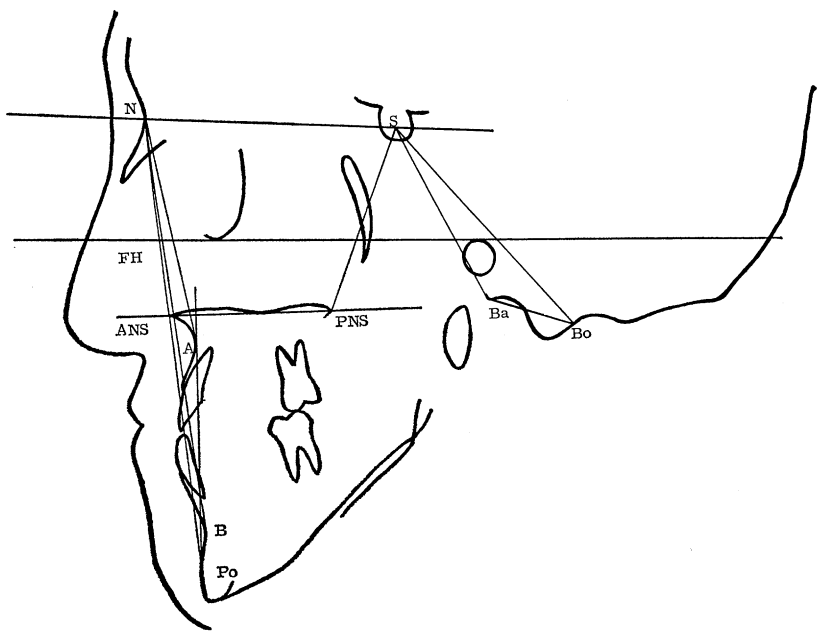

FIGURE 4. Tracings of lateral cephalometric roentgenograms taken of twin E.

TABLE 2. Measurements from roentgenograph tracings. Landmarks are: A, subspinale; ANS, anterior nasal spine; B, supramentale; Ba, basion; Bo, Bolton point; FH, Frankfort horizontal plane; N, nasion; PNS, posterior nasal spine; Po, pogonion; and S, sella turcica.

Measurement	*E*	*J*	*Normal (1, 3)*		*SD*
			Mean	*Range*	
Cranial Base					
N-S-Ba angle	120.2°	119.2°	134.7°	122.5°–150.5°	
S-Ba-Bo angle	134.0°	125.1°	137.1°	120.5°–154.0°	
Maxilla: anterior portion					
S-N-A angle	74.9°	82.1°	82.0°		3.9
A-N-Po angle	6.9°	1.3°	1.6°		4.8
A-N-B angle	−4.7°	2.0°	2.0°		
Maxilla: posterior portion					
N-S-PNS angle	73.0°	66.0°			
Ba-S-PNS angle	47.3°	53.1°			
Palatal Plane angle with FH	2.4°	7.9°			
S to PNS length on FH	8.2 mm	13.7 mm			
ANS to PNS length	53.1 mm	55.9 mm			

Discussion

Stein, Kelley, and Wood (*10*), superimposing tracings from lateral cephalometric roentgenograms, reported that striking likenesses were shown in three pairs of identical twins who were females and who had malocclusions. Kraus, Wise, and Frei (*6*) in a study of six sets of triplets speculate that the morphology of all the bones of the craniofacial complex are under the rather rigid control of hereditary forces. Comparisons between the set of identical twins being reported are complicated by the fact that each had a different number of operations for cleft lip and one had a mandibular resection to correct prognathism. However, some comparative observations can be made. These are as follows:

a) Development of their cranial bases may be said to be essentially similar and within the normal range since the N-S-Ba angles in each twin are nearly the same and are only a little less than the minimum normal value (*1*) and the S-Ba-Bo angles are within the range of normal standards (*1*).

b) Twin J has values within the normal range (*3*) for angles S-N-A, A-N-Po, and A-N-B. Data for her twin E is far below the normal. It may be recalled that twin E had her second lip operation at age 10 months, the third operation at age two years, and the fourth at age 19 years. Twin J had the second lip operation at the comparatively advanced age of five years. The same surgeon performed initial lip repair at age two months. This restriction of the anterior growth of the maxilla due to more lip operations at earlier ages corroborates the study by Graber (*3*).

c) In twin E the values of the palatal plan angle with FH plane, the

Ba-S-PNS angle, and the distance between S and PNS are smaller than those of her sister. The value of the N-S-PNS angle is larger. This means that PNS is situated more posterosuperiorly in E than in J.

d) The ANS-PNS length is nearly the same; for twin J it is 55.9 mm and for twin E 53.1 mm.

It may be assumed, then, that each twin had the same maxillary developmental potential, but that in twin E, because of her lip operations, the maxilla could not develop anteriorly and so developed posteriorly.

Many factors have been considered to be potential causes of cleft palate (*8*): adhesion of the amnion sac to the embryo face; insertion of the fingers into the embryonic mouth; infectious disease or toxemia in the mother in the first three months of pregnancy; maternal avitaminosis or other nutritional deficiencies; hormonal disorders caused by stress; and heredity. Fogh-Anderson (*2*) suggests that heredity may be an etiological factor since the concordance of cleft lip and palate among identical twins (33%) is higher than that of fraternal twins (5%).

The authors cannot state that heredity is the cause in this case simply because both twins have cleft lips and palates. Physical factors or medications could not be considered as etiologies. But some stress such as the mother's worry about her husband's illness might possibly alter her endocrine balance (*11*) sufficiently to cause hypoproliferation of fiber tissues in both twins and result in malformation (*9*).

Summary

The authors studied a very rare case of 21-year-old identical twin girls, both of whom have bilateral complete cleft lips and palates. Surgical repairs of the cleft palates were done at age five years by the same surgeon. The first repairs of the cleft lips were done by different surgeons when the twins were two months old. Lateral roentgenographic cephalometry showed: a) Anatomical craniobase measurements are nearly the same in both cases and are within the normal range. b) The twin who had a greater number of early lip operations demonstrated poorer development in the anterior part of the maxilla, but showed apparent compensatory growth in the posterior position. Her PNS was located more posteriorly and superiorly. Antero-posterior dimensions of the maxillae were about the same for both girls. Heredity or a hormonal disturbance in the mother due to severe stress in the first trimester of pregnancy might be the cause of the malformation in this instance.

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References

1. Brader, A. C., A cephalometric x-ray appraisal of morphological variations in cranial base and associated pharyngeal structures; implications in cleft palate therapy. *Angle Orthod.*, *27*, 179–195, 1957.

2. FOGH-ANDERSON, P., *Inheritance of Harelip and Cleft Palate.* Copenhagen: Nyt Nordisk Forlag, 1942.
3. GRABER, T. M., The congenital cleft palate deformity. *J. Amer. dent. Assoc., 48,* 375–395, 1954.
4. GRABER, T. M., Implemention of the roentgenographic cephalometric technique. *Amer. J. Orthod., 44,* 906–932, 1958.
5. INOUE, E., Futago. In N. Izeki (Ed.), *Iden-Igaku.* Tokyo: Kanehara, 1960.
6. KRAUS, B. S., WISE, W. J., and FREI, R. H., Heredity and the craniofacial complex. *Amer. J. Orthod., 45,* 172–217, 1959.
7. METRAKOS, J. D., METRAKOS, K., and BAXTER, H., Clefts of the lip and palate in twins: including a discordant pair whose monozygosity was confirmed by skin transplants. *Plastic reconstr. Surg., 22,* 109–122, 1958.
8. OLIN, W. H., *Cleft Lip and Palate Rehabilitation.* Springfield: Charles C Thomas, 1960.
9. STARK, R. B., The pathogenesis of harelip and cleft palate. *Plastic reconstr. Surg., 13,* 20–39, 1954.
10. STEIN, K. F., KELLEY, T. J., and WOOD, E., Influence of heredity in the etiology of malocclusion. *Amer. J. Orthod., 42,* 125–141, 1956.
11. STREAM, L. P., and PEER, L. A., Stress as an etiologic factor in the development of cleft palate. *Plastic reconstr. Surg., 18,* 1–8, 1956.