Craniofacial Morphology of Parents with and without Cleft Lip and Palate Children

PETER J. COCCARO, D.D.S., M.S.
RONALD D'AMICO, D.D.S., M.S.
ASHUR CHAVOOR, D.D.S.

The present study concerns the analysis of craniofacial structures in a group of parents, without any history of a cleft lip and palate, who had a cleft lip and palate child.

Genetic contribution of craniofacial structures, which in particular combinations, may favor a predisposition toward cleft lip and palate production served as the basic premise for the study.

Trasler's (*5*) study of early face embryology demonstrated that topography and growth of the facial processes between two strains of mice (inbred A/J) and the (C57BL/6J) are causally related to the A/J predisposition to cleft lip.

Some investigators (*2*) feel that "parents of children with congenital cleft lip should have faces that are on the average of a different shape than those of the general population." Their faith in inherited facial characteristics compelled them to measure superficial dimensions of the face to ascertain differences between two groups of parents—one of which had a cleft lip and palate child.

Other investigators (*4*) have stated "that the morphology of all the bones of the craniofacial complex are under rather rigid control of hereditary forces . . ." and further "that heredity governs morphology but environment in its multitudinous facets has much to say about how these bony elements shall combine to achieve what interests us most—the harmonious (or unharmonious) head and face." In a comprehensive review of cleft lip and palate embryology, pathology, anatomy, and etiology done by Canick (1), he states the following: "The geneticists have shown the influence of genes, whether they be dominant, recessive, incompletely sex linked, of reduced penetrance, and so forth. Their findings deserve the most careful considerations, not because various workers have shown a hereditary disposition up to forty percent of the cases of cleft lip and palate, but rather

Dr. Peter Coccaro is affiliated with Clinical Investigations Branch, National Institute of Dental Research, National Institutes of Health, U. S. Public Health Service, Department of Health, Education, and Welfare, Bethesda, Maryland. Dr. Ronald D'Amico and Dr. Ashur Chavoor are affiliated with Georgetown University, School of Dentistry, Department of Graduate Orthodontics, Washington, D.C.

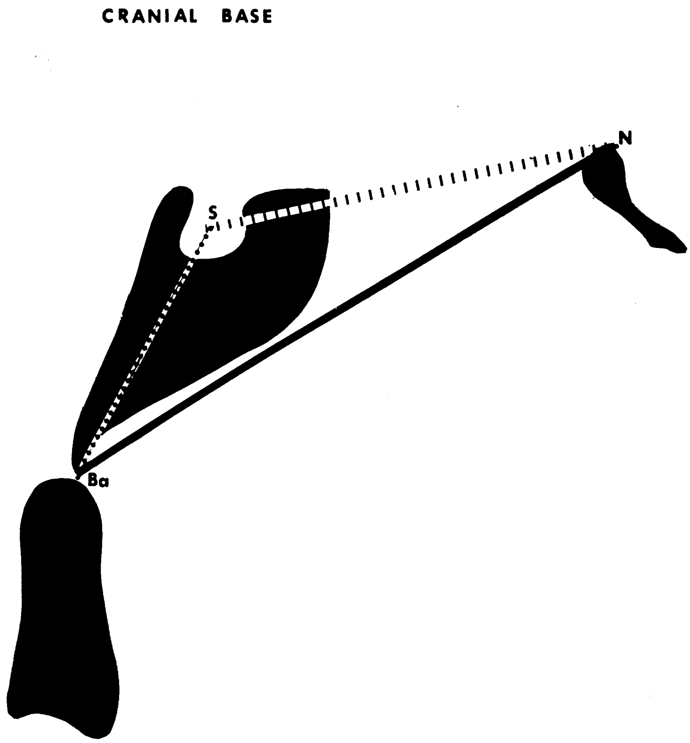


FIGURE 1. CRANIAL BASE.
1. Sella-Nasion (S-N)
2. Sella-Basion (S-Ba)
3. Nasion Basion (N-Ba)
4. Basion-Sella-Nasion (Ba-S-N)

because a strong impression has been gained that there may be a much larger percentage of the anomalous people with hereditary disposition."

Hughes (*3*) points out some interesting observations about hereditary in cranial and facial development. He states that facial asymmetries are almost independent of cranial asymmetries and that the mandible and maxilla are independent of each other with multiple factors being involved in the production of all of the features associated with these bones. It becomes quite apparent at this point that the structures deeper than those more superficially accessible need to be analyzed to give us a better insight into heritable factors in craniofacial morphology.

In searching for the answers to the variables in facial characteristics, it would be desirable to document growth of the individual anatomical structures that go into the formation of the head and face from birth on and determine their effect upon skeletal and soft tissue profiles. Such a course of study becomes even more imperative since individual bones form separate units of growth and in combination truly represent a picture of growth for the head and face. Since this is not possible, we must accept

UPPER FACE

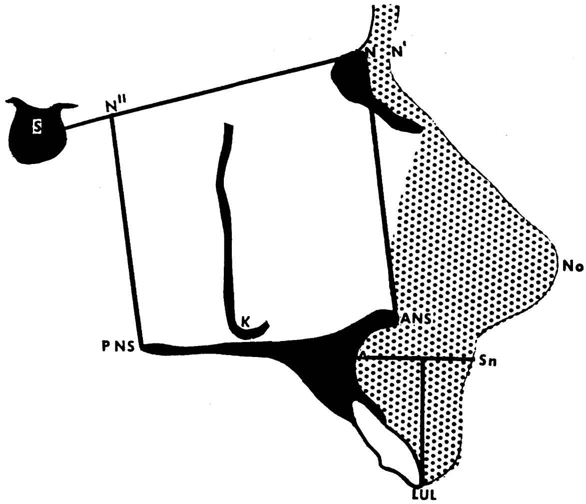


FIGURE 2. UPPER FACE.

1. Nasion-Anterior Nasal Spine (N-ANS)
2. Nasion-Posterior Nasal Spine (N″-PNS)
3. Nasion-Point A (N-A)
4. Anterior Nasal Spine-Posterior Nasal Spine (ANS-PNS)
5. Key Ridge-Anterior Nasal Spine (K-ANS)
6. Key Ridge-Posterior Nasal Spine (K-PNS)
7. Nasion-Soft Tissue Nasion (N-N′)
8. Nasion-Tip of Nose (N-No)
9. Point A-Subnasale (A-Sn)
10. Anterior Nasal Spine-Tip of Nose (ANS-No)
11. Length of Upper Lip (LUL)
12. Sella-Nasion-Anterior Nasal Spine (S-N-ANS) (Angular)

what is possible, as limited as it may be, and that is to study the craniofacial structures in combination with each other as viewed through lateral X-ray headplates.

Thus, the present study was designed to compare the craniofacial structures of parents having a cleft lip-palate child to a group of parents whose children were normal. The principal question to be answered is: Are there any differences in the craniofacial morphology between two groups of parents with the only disparity being the birth of a cleft lip-palate child separating the two groups?

Material and Methods

The sample for this study consisted of two groups of caucasian parents, 20 males and 20 females in each group, one having children with a cleft lip and palate and the other having children without the deformity. Lateral cephalometric radiographs were taken on these parents whose children were included in growth studies at the Clinical Center of the National Institutes of Health. One X-ray was taken on each subject, all of whom were over 30 years of age.

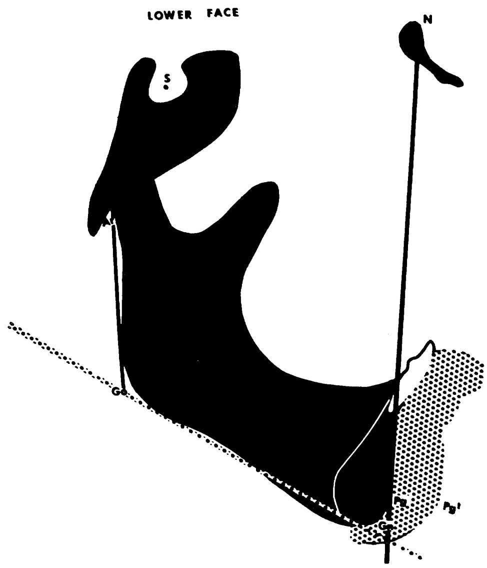


FIGURE 3.
1. Articulare-Gonion (AR-Go)
2. Gonion-Gnathion (Go-Gn)
3. Articulare-Gnathion (Ar-Gn)
4. Pogonion-Soft Tissue Pogonion (Pg-Pg′)

Twenty-five measurements of skeletal and soft tissue structures within the craniofacial areas, taken from the lateral cephalometric X-rays, were made on each subject. The conventional landmarks, reference lines, and angles were utilized in the analysis of the cranial base, upper face, lower face, and facial profile, with the following measurements taken in each of the specific areas studied (Figures 1–4):

Findings

Cranial Base. The cranial base angle (Ba-S-N) measurement was significantly different and proved to be more acute for parents of children with a cleft lip and palate. All other cranial base measurements were not significantly different between the two groups of parents. The cranial base angle (Ba-S-N) was significantly different between all fathers and all mothers. All of the linear measurements were also significantly different between fathers and mothers (Tables I & V) (Figure 5).

Upper Face. The mean values for the measurement nasion-anterior nasal spine (N-ANS) were less for the parents of cleft lip and palate children, indicating a shorter vertical dimension in anterior facial height. No significant difference was noted for posterior facial height (N″-PNS).

The measurement, anterior nasal spine-posterior nasal spine (ANS-PNS) was also found to be less for parents of cleft lip and palate children, indicating a shorter palatal length (antero-posteriorly) for these parents compared to the parents of normal children. To ascertain specific area of palatal plane affected (anterior or posterior) the linear measurement Key-Ridge-Anterior Nasal Spine (K-ANS) was recorded and found to be shorter for parents of cleft lip and palate children. No difference was noted between the two groups in the measurement Key-Ridge-Posterior

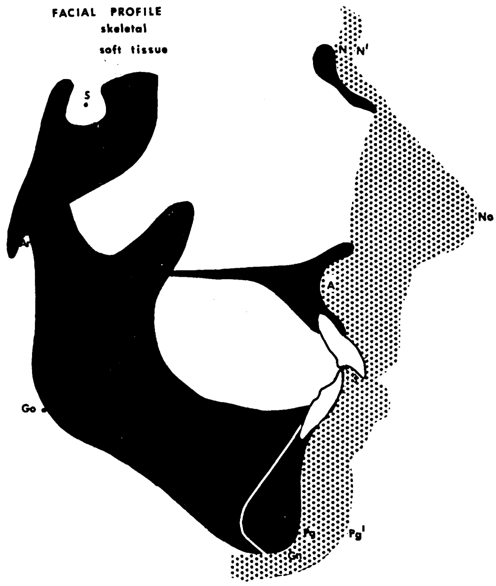


FIGURE 4.
1. Sella-Nasion-Pogonion (S-N-P)
2. Sella-Soft Tissue Nasion-Soft Tissue Pogonion (S-N′-Pg′)
3. Nasion-Point A-Pogonion (N-A-Pg)
4. Soft Tissue Nasion-Subnasale-Soft Tissue Pogonion (N′-Sn-Pg′)
5. Soft Tissue Nasion-Tip of Nose-Soft Tissue Pogonion (N′-NoPg′)
6. Y Axis (A line from sella turcica to gnathion to denote direction of mandibular growth)

TABLE I
ANALYSIS OF VARIANCE

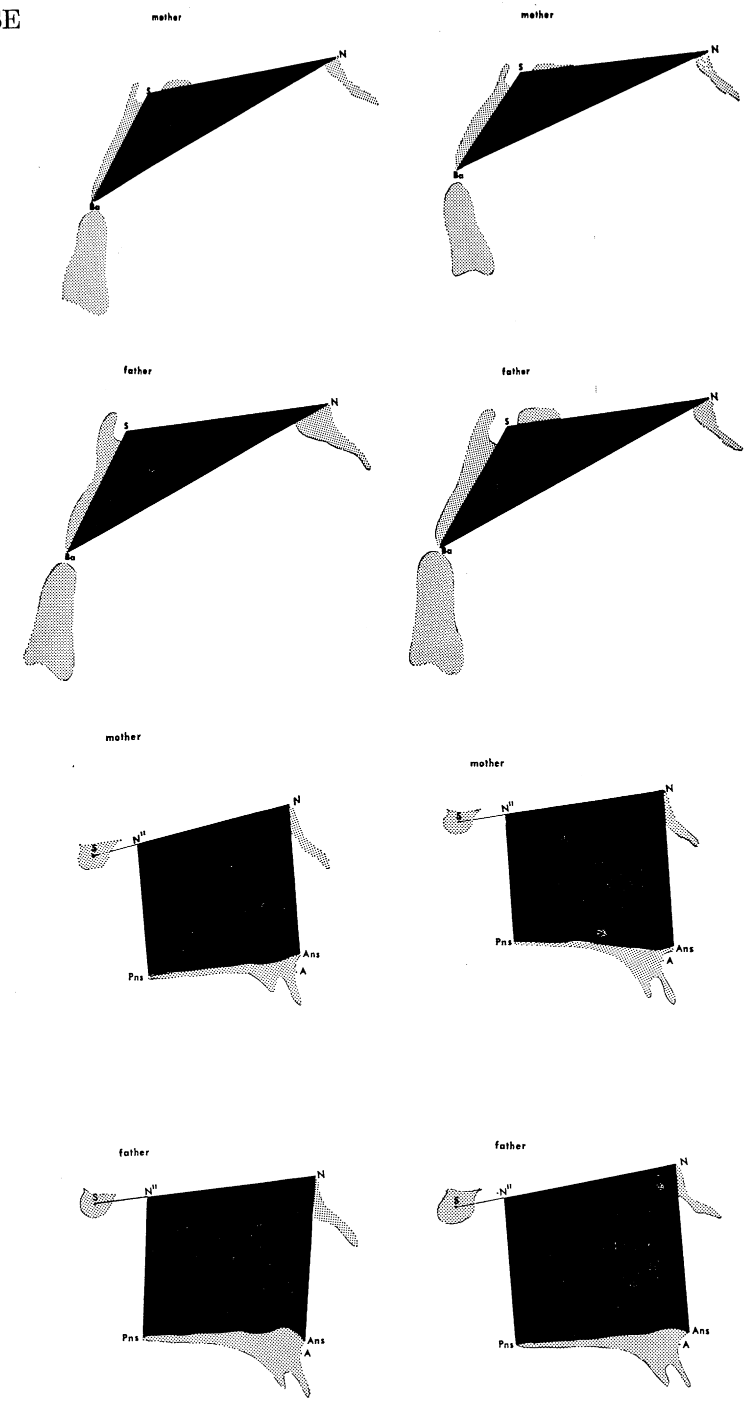
EFFECT OF PALATE				EFFECT OF SEX			
Measurement Angular	Parents (n-c) Mean	Parents (c) Mean	Difference	Measurement Angular	Fathers	Mothers	Difference
Ba-S-N	130.48	126.49	.01 –	Ba-S-N	127.16	129.80	.05
LINEAR				LINEAR			
S-N	72.33	72.85	N.S.	S-N	75.50	69.68	.01 –
S-Ba	46.40	47.11	N.S.	S-Ba	49.63	43.89	.01 –
N-Ba	107.93	108.08	N.S.	N-Ba	112.25	103.75	.01 –

Nasal Spine (K-PNS). Shorter palatal length was more confined to the anterior 2/3rds of the hard palate.

Linear measurements, therefore, of vertical and horizontal dimensions of upper face proved to be significantly different for parents of children with a cleft lip and palate compared to the group of parents with normal children. All linear measurements were significantly different when all fathers were compared to all mothers of both groups.

The linear measurement Nasion-Tip of Nose (N-No) was significantly different between the parents of cleft lip and palate children when compared to the parents of children without the deformity. Minor differences were observed for measurements of thickness of upper lip (A-Sn) and length of upper lip (LUL) but these differences were not statistically significant. However, significant differences were noted for these measurements when all fathers were compared to mothers.

CRANIAL BASE



UPPER FACE

FIGURE 5. Tracings of a composite for parents

.. with cleft lip and palate children	.. with non-cleft lip and palate children

TABLE II
ANALYSIS OF VARIANCE

EFFECT OF PALATE				EFFECT OF SEX			
Measurement Angular	Parents (n-c) Mean	Parents (c) Mean	Difference	Measurement Angular	Fathers Mean	Mothers Mean	Difference
S-N-ANS	86.15	85.83	N.S.	N-ANS	86.46	85.51	N.S.
S-N-A	80.61	80.51	N.S.	S-N-A	80.86	80.26	N.S.
LINEAR				LINEAR			
N-ANS	55.33	53.84	.05	N-ANS	56.90	52.26	.01 –
N"-PNS	46.73	46.50	N.S.	N"-PNS	49.30	43.93	.01 –
N-A	59.68	59.18	N.S.	N-A	61.78	57.08	.01 –
ANS-PNS	56.05	54.20	.02	ANS-PNS	57.10	53.15	.01 –
K-ANS	32.35	30.41	.01 –	K-ANS	32.48	30.29	.01 –
K-PNS	23.68	23.99	N.S.	K-PNS	24.60	22.86	.01 –
N-N'	7.85	7.43	N.S.	N-N'	8.50	6.78	;01 –
A-Sn	17.00	16.26	.10+	A-Sn	18.69	14.58	.01 –
N-No	59.70	57.88	.05	N-No	61.73	55.85	.01 –
ANS-No	30.93	30.88	N.S.	ANS-No	32.75	29.05	.01 –
LUL	23.18	22.18	.10+	LUL	24.15	21.20	.01 –

When the anterior cranial base (Sella-Nasion) (S-N) was related to ANS and to Point A, the angular readings recorded were found to be no different between the two groups of parents. No difference was also noted between all fathers and all mothers (Tables II & V) (Figure 5).

LOWER FACE. Linear measurements for mandibular body length (Go-Gn), only, were significantly different between parents of cleft lip-palate children and parents of non-cleft lip-palate children. However, when mothers were compared to fathers all linear measurements were found to be significantly different (Tables III & V) (Figure 6).

FACIAL PROFILE (Skeletal-soft tissue). Angles for skeletal profile analysis were found to be significantly different between parents of cleft lip and palate children compared to parents of children without such deformity. However, no significant difference was noted between all fathers and all mothers for the same skeletal profile analysis.

Angles for soft tissue profile analysis were significantly different between parents of cleft lip and palate children and parents of children without a cleft lip and palate. No significant difference was observed between fathers and mothers for similar soft tissue profile analysis (Tables IV & V) (Figure 7).

Discussion

The findings in this study demonstrate that parents of cleft lip and palate children differ in their craniofacial morphology from parents of children without a cleft lip and palate. Unusual differences were found in the area of the upper face, particularly in the anterior region where vertical and horizontal dimensions were significantly shorter for parents with cleft lip and palate children. Whether or not this suggests defective development of specific anatomical structures within the upper face or a

TABLE III
ANALYSIS OF VARIANCE

EFFECT OF PALATE				EFFECT OF SEX			
Measurement Linear	Parents (n-c)	Parents (c)	Difference	Measurement Linear	Fathers Mean	Mothers Mean	Difference
Ar-Go	51.00	49.38	.10+	Ar-Go	53.55	46.83	.01 –
Go-Gn	76.25	78.88	.03	Go-Gn	80.88	74.25	.01 –
Ar-Gn	113.48	114.78	N.S.	Ar-Gn	119.50	108.75	.01 –
Pg-Pg′	13.25	13.05	N.S.	Pg-Pg′	14.30	12.00	.01 –

LOWER FACE

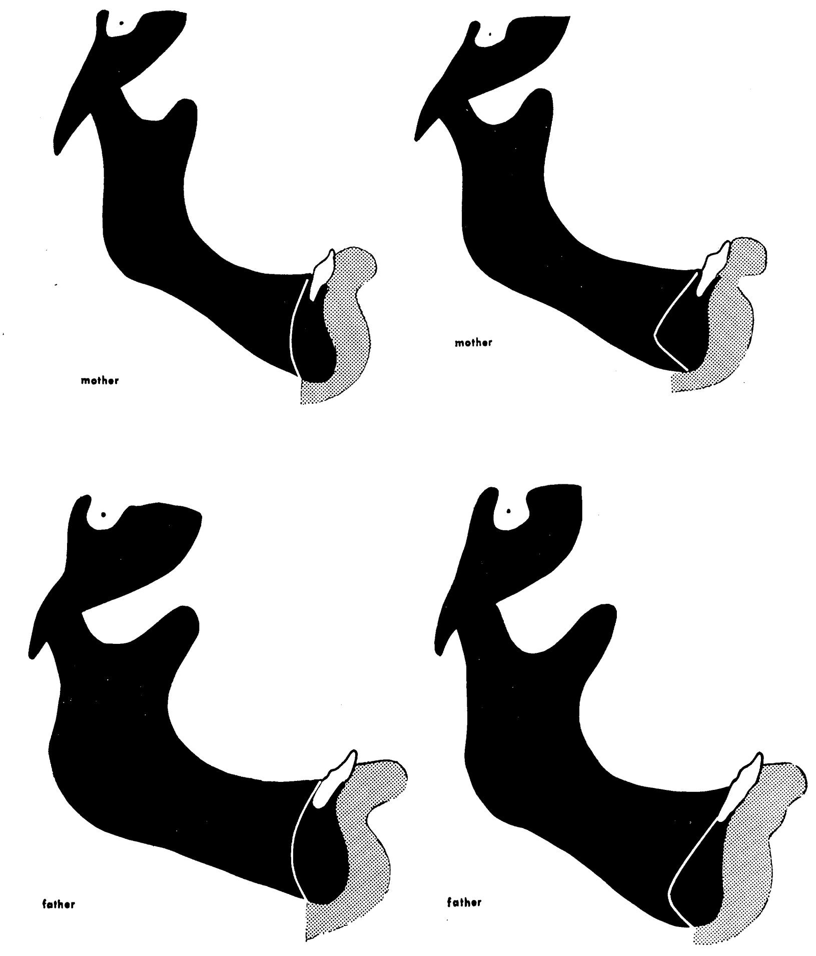


FIGURE 6. Tracings of a composite for parents

..with cleft lip and palate children	..with non-cleft lip and palate children

TABLE IV
ANALYSIS OF VARIANCE

EFFECT OF PALATE				EFFECT OF SEX			
Measurement Angular	Parents (n-c) Mean	Parents (c) Mean	Difference	Measurement Angular	Fathers Mean	Mothers Mean	Difference
S-N-Pg	77.99	80.65	.01 –	S-N-Pg	79.91	78.73	.10+
S-N'-Pg'	81.24	83.24	.025	S-N'-Pg'	82.68	81.80	N.S.
N-A-Pg	5.46	– 0.86	.01 –	N-A-Pg	1.41	3.19	.10+
N'-Sn-Pg'	161.88	167.61	.01 –	N'-Sn-Pg'	164.14	165.35	N.S.
N'-No-Pg'	128.10	131.39	.01 –	N'-No-Pg'	129.63	129.66	N.S.
Y axis	69.66	67.39	.01	Y axis	68.28	68.78	N.S.

generalized smallness of these parts is a matter of conjecture. The other interesting finding is that of mandibular body length, which was found to be longer for parents of children with the facial deformity. Here again, we can hypothesize and consider abnormal development or a generalized largeness of mandible with a genetic factor existing between the two groups studied. A tendency toward mandibular prognathism was noted and was closely identified with the findings of a more concave facial profile in parents of children with cleft lip and palate.

In looking for genetic factors that may be predisposing for the production of a cleft lip and palate child, we might include the significance of the present findings. They could very well be considered as existing unfavorable variations of the upper and lower face which, in particular combinations during embryologic development, may prevent structures from com-

TABLE V
SUMMARY OF MEASUREMENTS

		FATHERS (Non-C/L/P)		FATHERS (C/L/P)		MOTHERS (Non-C/L/P)		MOTHERS (C/L/P)	
		Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.
CRANIAL BASE	S-N	75.0	3.92	76.0	4.69	69.6	3.12	69.7	5.34
	S-Ba	49.6	4.89	49.6	3.36	43.2	3.29	44.5	2.81
	N-Ba	112.5	5.69	111.9	5.66	103.3	5.97	104.2	5.63
	Ba-S-N	128.9	6.37	125.35	7.39	131.9	4.42	127.6	5.45
UPPER FACE	N-ANS	57.4	3.57	56.4	4.04	53.2	3.24	51.2	2.82
	N"-PNS	49.6	3.98	48.9	2.16	43.8	2.53	44.0	2.01
	N-A	61.9	5.51	61.6	4.40	57.4	3.05	56.7	2.96
	ANS-PNS	58.3	3.90	55.8	3.24	53.7	2.95	52.5	3.27
	K-ANS	33.8	2.35	31.1	2.56	30.9	2.71	29.6	3.41
	K-PNS	24.5	2.86	24.7	1.75	22.8	2.21	22.8	2.05
	N-N'	8.7	1.45	8.3	1.56	7.0	1.03	6.5	1.19
	N-No	62.3	4.18	61.1	4.78	57.0	4.38	54.6	3.45
	A-Sn	19.0	2.42	18.3	2.45	15.0	1.72	14.1	2.28
	ANS-No	32.7	2.88	32.7	3.01	29.1	3.13	29.0	4.99
	LUL	24.2	3.13	24.0	3.59	22.0	2.67	20.3	3.11
	S-N-ANS	87.1	3.72	85.8	4.93	85.2	3.93	85.8	4.13
LOWER FACE	Ar-Go	55.1	4.74	51.9	5.97	46.8	4.30	46.8	4.60
	Go-Gn	79.4	3.28	82.3	5.19	73.0	5.81	75.4	4.86
	Ar-Gn	119.0	5.64	119.9	6.28	107.9	5.09	109.6	6.02
	Pg-Pg'	14.3	2.01	14.2	2.15	12.1	1.79	11.8	2.00
FACIAL PROFILE Skeletal-Soft Tissue	S-N-Pg	79.03	4.23	80.8	4.76	76.9	3.75	80.5	3.29
	S-N'-Pg'	81.9	3.61	83.3	4.58	80.5	4.49	83.1	3.64
	N-A-Pg	4.1	5.47	– 1.3	5.88	6.8	4.40	– 0.4	5.36
	N'-Sn-Pg'	161.33	7.73	166.9	5.50	162.4	5.47	168.2	6.20
	N'-No-Pg'	128.00	4.89	131.6	3.84	128.2	3.95	131.1	6.59
	Y-axis	69.0	4.53	67.5	4.38	70.0	4.17	67.2	3.78

FACIAL PROFILE..Skeletal soft tissue

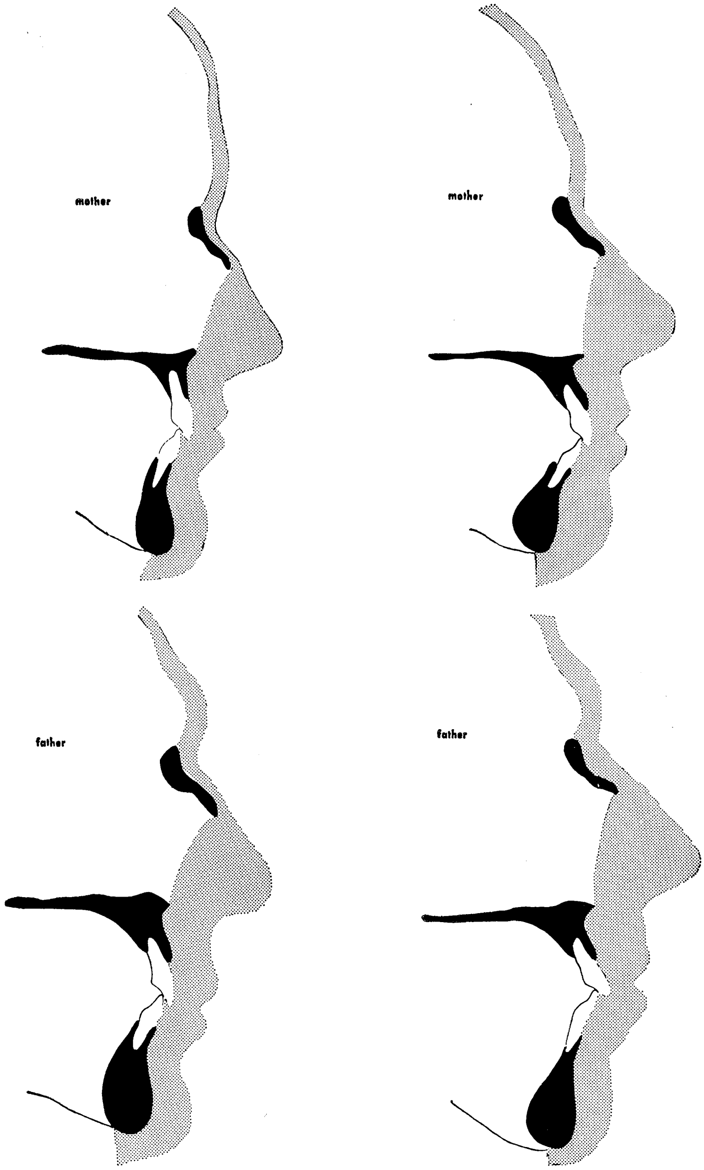


FIGURE 7. Tracings of a composite for parents

..with cleft lip and palate children | ..with non-cleft lip and palate children

ing together normally and also keep them from reaching maximum proportions.

Thus, the quantitative differences of affected parts may be sufficient to create an unfavorable relationship between them and permit them to be displaced and develop abnormally. These parents then could be considered as having, based on the morphological character of the structures, genetic determinants predisposing to the production of a cleft lip and palate anomaly.

Summary

The main purpose of this study was to determine whether any difference existed in craniofacial morphology between parents of children with a cleft lip and palate and parents of children without a cleft lip and palate. Angular and linear measurements of the various regions of cranium and face were obtained from lateral cephalometric roentgenograms of these parents. The means of the measurements were tested for significant statistical differences by a two-way analysis of variance. The total sample was compared for the effect of palate (parents of cleft lip and palate children to parents of children without the deformity) and for effect of sex (all fathers compared to all mothers).

The craniofacial morphology of parents with cleft lip and palate children differed from that observed in parents of children without the facial deformity. The faces of parents with cleft lip and palate children were less convex with a tendency toward mandibular prognathism. Vertical and horizontal measurements of the upper face were shorter and the nose length was also shorter for parents of cleft lip and palate children.

These findings were consistent for both mothers and fathers of cleft lip and palate children when considered collectively. Validity of these differences was supported by the fact that very few differences in relationships existed when all fathers were compared to all mothers.

Further studies are recommended in this area using larger samples to add to the present study and possibly substantiate present findings.

reprints: *Peter J. Coccaro, D.D.S., M.S.*
National Institute of Dental Research
National Institutes of Health
U.S.P.H.S., Department of Health,
Education, and Welfare
Besthesda, Maryland 20014

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