Craniofacial pattern of parents of children having cleft lip and/or cleft palate anomaly

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The cleft lip and/or cleft palate anomaly is one of the most frequently encountered congenital malformations. Race affects the incidence of this disorder with the Mongoloid race having a higher incidence than Caucasians and Caucasians a higher incidence than Negroids.[1,2]

A cleft of the lip and/or palate is multifactorial in origin. Some clefts are caused by single mutant genes, some are due to chromosomal aberrations, and some are caused by specific environmental agents; the great majority are caused by the interaction of genetic and environmental factors, each with a relatively small effect. There is evidence to suggest that isolated clefts of the secondary palate are both genetically and developmentally different from clefts of the primary palate or complete clefts.[3] Ward et al.[4] found a substantial genetic component in at least one of the parents in many cases of sporadic cleft lip/palate. Trasler[5,6,7] believed the difference in susceptibility to cleft lip among several mouse strains was related to the shape of the embryonic primordial face.

Based on these findings, Fraser and Pashayan[8] inferred that if facial shape is genetically determined and also related to a predisposition to the cleft anomaly, then the parents of children with cleft lip/palate should have facial dimensions different from those of the general population. Parents of cleft lip/palate children do have greater bizygomatic width, an underdeveloped maxilla and a thinner upper lip than the general population.[8]

Other dissimilarities in facial form between par-

Abstract

The craniofacial patterns of 38 sets of parents who had children with cleft lip and/or cleft palate anomalies (experimental group) were compared with the 24 sets of parents of healthy (noncleft) children (control group). Using a computerized program, 248 cephalograms (124 lateral and 124 frontal) were digitized and analyzed. The parents in the experimental group exhibited a distinct craniofacial morphology, including a significant decrease in upper anterior facial height (N-Ans) and total anterior face height (V-Gn). Anterior nasal spine (Ans) and maxillary alveolar process (A) were positioned more anteriorly and superiorly in the experimental group, which contributed to a significant increase in the length of the palate (Ans-Pns) and an anterosuperior rotation of the palatal plane. The cranial base angle in the experimental group was significantly obtuse and the articular angle was smaller than that of the controls. The counterclockwise rotation of the mandible was mitigated by a significant increase in the gonial angle. Parents in the experimental group also tended to have faces which were smaller in both transverse and vertical dimensions.

Key Words

Cleft • Parents • Craniofacial morphology • Cephalogram • Face height • Cranial base angle

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Table I
Mean and age range of parents (years)

Parent	Parents age at child's birth				Age at visit examination			
	Control		Experimental		Control		Experimental	
	Range	Mean	Range	Mean	Range	Mean	Range	Mean
1 Mother	19-37	22.61	18-39	23.46	21-40	27.04	21-45	28.97
2 Father	21-40	30.92	20-42	27.81	21-44	35.13	25-45	33.3
Mid parent	19-40	26.76	18-42	25.64	21-44	31.08	21-45	31.04

ents of cleft and non-cleft children have also been reported.[9,10,11] Coccaro et al.[9] observed a less convex face with a tendency towards mandibular prognathism and a short upper facial height in the parents of cleft children, findings that were later supported by Kurisu et al.[10] and Nakasimo and Ichinose.[11] People of different racial backgrouds would express these observations by exhibiting different rates of cleft lip and palate anomaly.

To further corroborate these findings, the present study was undertaken among parents of children having cleft lip/palate anomaly and parents of children without clefts. The objective was also to correlate the findings with those of other investigators and to throw some light on the predisposition of the anomaly as related to the morphological structures. Any relationship between craniofacial morphology and susceptibility to the cleft lip/palate anomaly might broaden our concept regarding the etiology of this malformation.

Table II
Distribution of Probands according to cleft types

S.N.	Type of cleft	M	F	Total
1	Complete bilateral cleft lip and palate	10	1	11
2	Complete unilateral cleft lip and palate	10	10	20
3	Unilateral cleft lip only	-	1	1
4	Unilateral cleft lip and alveolus	3	2	5
5	Bilateral cleft lip and primary palate	1	-	1
	Total	24	14	38

Materials and methods

The subjects for the study were 38 sets of parents (38 mothers and 38 fathers) of children with cleft lip/palate anomaly. These parents constituted the experimental group. The control group consisted of 24 sets of parents of children without clefts. All the subjects of the study were drawn from the same geographical location in India. The sample selection was limited to one region so as to avoid any discrepancies in population groups which might otherwise influence the craniofacial form.

The age group of the parents of children with the cleft anomaly ranged from 21 to 45 years at the time of the investigation. The mean age of the mothers at the time the cephalograms were taken was 28.97 years; the mean age at the time of birth of the affected children was 23.46 years. The age of the fathers ranged from 25 to 45 years with a mean of 33.3 years at the time of the investigation. The mean age of the fathers when their children with cleft were born was 27.81 years. The mid parent mean age was 31.04 years at the visit examination and 25.64 years at birth of their probands (Table I).

The 38 sets of parents constituting the experimental group had no evidence of any types of cleft while their progenies exhibited different types of clefts (Table 2). Parents with children having isolated cleft palate were not considered for this study.

Criteria of selection for the control group were:

a) Parents whose children had

i) no anomaly of a skeletal, genetic, endocrinal or any other nature.

ii) no gross skeletal defect, although malocclusion was acceptable.

iii) a full complement of teeth from second molar to second molar in both jaws.

b) Individuals who had no disease of a skeletal, genetic or endocrinal nature.

The age of these parents ranged from 21 to 45 years to match the experimental group (Table I). The mean age of the mothers at the time of the study was 27.04 years and at the time of the birth of their children was 22.61 years. The mean age of

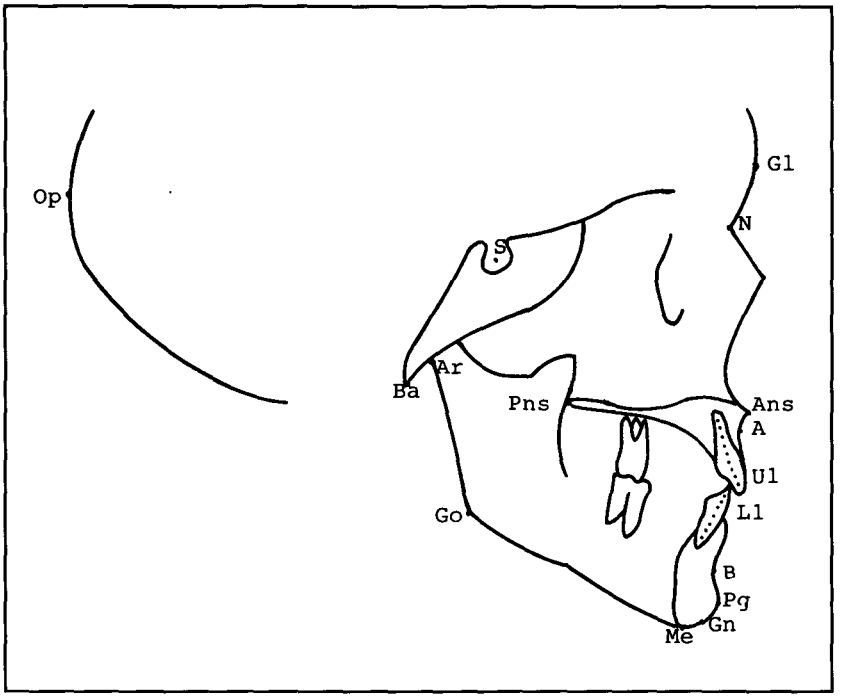


Figure 1

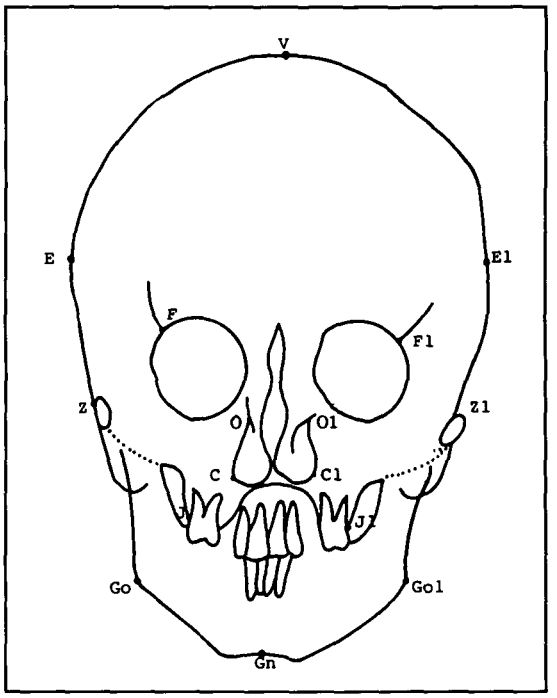


Figure 2

Figure 1
Diagrammatic representation of the lateral cephalogram showing the landmarks used.

Figure 2
Diagrammatic representation of the frontal cephalogram showing the landmarks used.

the fathers at the time of the study was 35.13 years and at birth of their children was 30.92 years (Table I).

Lateral and frontal cephalograms were taken using a standardized method. A total of 248 (124 lateral and 124 frontal) cephalograms of 38 sets of parents of children with clefts (Experimental group) and 24 sets of parents of healthy children (Control group) were taken. The cephalograms were digitized by one investigator and measurements were recorded using a Houston Hipad Digitizer interfaced with a computer. Ten cephalograms were digitized again on a separate occasion by the same investigator. The intra investigator error for the linear and angular measurements was calculated using Dahlberg's[12] formula. The mean error was 0.12 mm for linear measurements and 0.41° for angular measurements. The error was insignificant for purposes of statistical analysis.

Landmarks for the lateral cephalograms are shown in Figure 1; Figure 2 shows the landmarks for the PA cephalograms. Some of the landmarks used on the P.A. cephalograms are not commonly used. These include:

E Euryon: The most lateral point on the side of head in the region of parietal bone

F: Point on the mesial border of zygomaticofrontal suture

O Orbitale: The most median point of optic foramen

C: The most lateral point on the lateral wall of the nasal cavity

J: Intersection of the lateral contour of the maxillary alveolar process and the lower contour of the maxillo-zygomatic process of maxilla.

A mid parent value for each of 30 parameters was arrived at for the 38 sets of parents in the experimental group and for the 24 sets of parents in the control group. The experimental and control groups were compared for differences in craniofacial morphology.

Ethnic variability of craniofacial form in the sample was eliminated by restricting the study to subjects drawn from one region only. The 30 variables from the lateral and frontal cephalograms of the experimental and control groups were compared (Tables III, IV). These were subjected to statistical evaluation using Student's t Test (unpaired). The statistically significant differences between the two groups were evaluated by calculating the p value.

Results

The mean and standard deviation of the linear and angular cephalometric variables of parents of cleft lip/palate children (experimental group) and of parents of healthy children (control group) are presented in Tables III and IV. The most significant differences in craniofacial morphology between the two groups were mainly expressed in the variables of the cranial base, upper face and facial profile. There was a significantly ($p<.01$) larger cranial base angle (Ba-S-N) in parents in the experimental group and the articular angle (S-Ar-Go) was significantly smaller ($p<.001$). The lengths

Table III
Mean and S.D. of cephalometric variables of control and experimental groups

Variable	Control N=24		Exp. Group N=38	
Cranial Base	Mean	SD	Mean	SD
1. Ba-S-N	126.9	± 2.9	129.7	± 4.7**
2. S-N	77.5	± 2.9	76.9	± 1.9
3. N-Ba	114.8	± 4.0	114.9	± 4.7
4. S-Ba	50.0	± 3.0	49.6	± 3.5
5. S-Ar-Go	145.7	± 4.9	138.9	± 6.1***
Upper Face				
6. N-Ans	58.7	± 2.8	56.3	± 2.3***
7. S-Pns	51.9	± 2.2	50.1	± 2.2**
8. N-A	63.0	± 3.3	61.8	± 2.7
9. Ans-Pns	56.0	± 2.9	60.4	± 3.5***
10. Gl-Op	202.4	± 13.5	206.8	± 5.6
11. S-N-A	80.4	± 3.0	81.1	± 3.1
12. S-N-Ans	82.9	± 2.9	86.3	± 3.9***
Lower Face				
13. Ar-Go	53.4	± 3.1	54.7	± 4.7
14. Go-Gn	82.2	± 4.6	81.8	± 4.5
15. Ar-Gn	117.3	± 4.9	118.0	± 6.0
16. Ans-Me	71.7	± 4.8	72.3	± 4.3
17. SNB	77.7	± 2.7	78.7	± 3.4
Facial Profile				
18. S-N-Pg	78.5	± 2.8	79.7	± 3.1
19. N-A-Pg	172.3	± 4.0	175.1	± 3.6
20. N-S-Gn	68.3	± 2.5	66.8	± 3.0
21. Ar-Go-Me	119.1	± 4.4	121.9	± 4.7**
22 U1/L1	122.9	± 6.6	125.5	± 12.8

* = p < 0.05, ** = p < 0.01, *** = p < 0.001
Linear measurements are recorded in mm.

Table IV
Mean and S.D. of frontal view measurements in control and experimental groups

Variable	Control N = 24		Exp. Group N = 38	
	Mean	SD	Mean	SD
Upper Face				
MHW (E-E1)	163.7	± 11.5	150.7	± 5.7***
IOW (0-01)	25.1	± 2.4	25.2	± 2.4
FW (F-F1)	99.8	± 6.2	94.5	± 3.6***
NW (C-C1)	34.8	± 3.2	37.4	± 3.9**
ZW (Z-Z1)	144.8	± 11.4	138.5	± 5.4**
Lower Face				
AW (J-J1)	73.3	± 5.5	70.2	± 3.4**
GW (G-G1)	109.8	± 8.4	104.6	± 5.3**
Total Facial Height				
TFH (V-Gn)	247.9	± 20.1	232.1	± 11.3***

* = P<0.05, ** = P<0.01, *** = P<0.001

of anterior cranial base (S-N) and posterior cranial base (S-Ba) were almost the same in both groups. In the upper face most of the parameters showed significant differences. There was a significantly (p<.001) shorter anterior facial height (N-Ans) as well as posterior facial height (S-Pns) (p<.01) in parents of children with the cleft anomaly. These parents also had a significantly(p<.001) larger palate (Ans-Pns) and larger S-N-Ans angle. The depth of the forehead (Gl-Op) was not different in the two groups.

The measurements of the lower face and dental pattern did not show any significant differences between the two groups. The length of the mandible (Ar-Go, Go-Gn, Ar-Gn) and angle S-N-B did not show any significant differences between the two groups. The gonial angle was significantly larger (p<.01). Though the mean value of inter incisal angle was higher in the parents in the experimental group, the difference was not statistically significant.

All measurements taken on the frontal cephalograms were linear, seven in the horizontal plane and one in the vertical plane. The maximum head width (E-E1), bizygomaticofrontal suture width (F-F1) and bizygomatic width (Z-Z1) were significantly shorter in the experimental group whereas the nasal width (C-C1) was significantly larger. The orbital width (O-O1) did not show a significant difference between the two groups. Both the variables of the lower face, the alveolar width (J-J1) and bigonial width (G-G1) were significantly (p<.01) smaller in parents of children with clefts. The total facial height (V-Gn) was also significantly smaller (p<.001) in these parents.

Discussion

The craniofacial morphology of parents of children having cleft lip and/or palate anomaly is distinctly different from that of parents of children without clefts.[8] Most of the differences are located in the upper face, cranial base, face width and face height (Figures 3-10). The cranial base

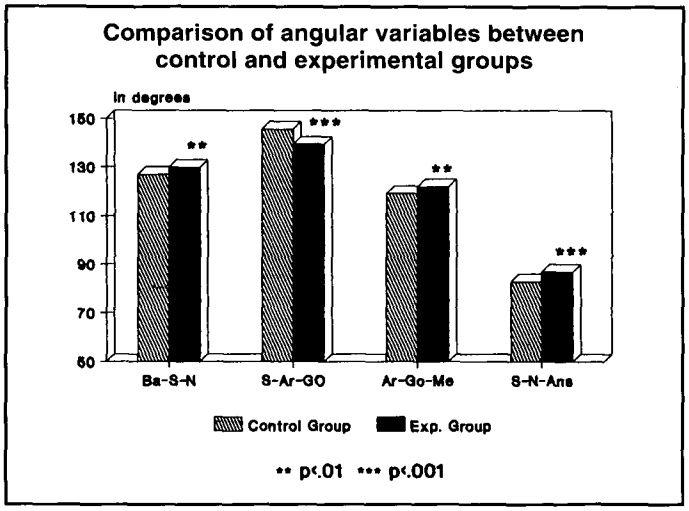
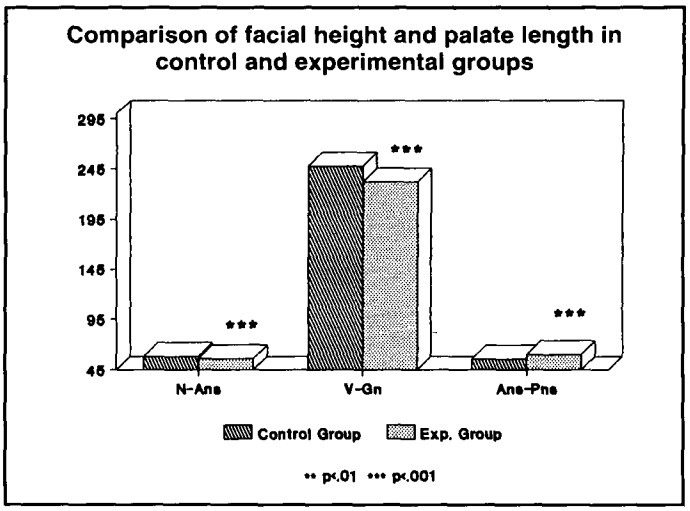


Figure 3



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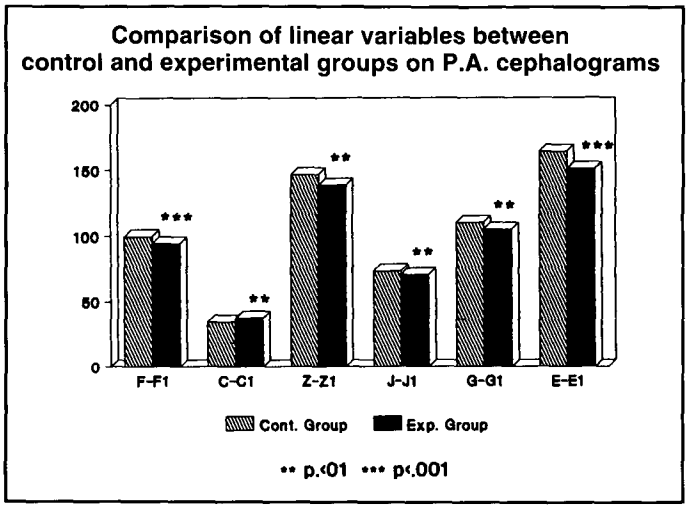


Figure 5

Figure 3
Histogram showing differences in angular variables between Experimental and Control groups on the lateral cephalogram.

Figure 4
Histogram showing differences in linear variables between Experimental and Control groups on the lateral cephalogram.

Figure 5
Histogram showing differences in linear variables between experimental and control groups on the frontal cephalogram.

angle of the parents in the experimental group is more obtuse ($p<.01$). These findings are in contrast to the findings of Coccaro et al[9] who found a decrease in this angle. Keeping in view the hereditary component of the craniofacial complex, similar or near similar measurements between parents and their cleft children are expected. Dahl,[13] and Cronin and Hunter[14] found an obtuse angle of the cranial base in children with clefts. Moss[15] made similar observations in children with clefts and also conducted experiments on rats to alter the rotation of the cranial bones.[16] He concluded that anterior placement of the foramen magnum and the lowering of the mid sagittal point contributed to the increased flexure of the cranial base which was expressed as a backward and downward rotation of the neural skull relative to the facial skeleton.

The upper anterior facial height in the experimental group was significantly shorter ($p<.001$) than in the control group. The posterior upper facial height also decreased significantly ($p<.01$) although not to the same degree of severity. These measurements were similar to the findings of Coccaro et al.,[9] Kurisu et al.,[10] and Nakasimo and Ichinose.[11] The decrease in vertical facial height in the sample points to a significant deficiency of the anterior as well as posterior height of the maxilla. The height of the maxilla as measured at N-A was also smaller in the experimental group than in the control group. Similarly, Coccaro et al.[9] observed a decrease in facial height (N-A) which was not significant. Thus the parents of children with clefts have a smaller maxillary height and a superiorly placed anterior nasal spine along with the deficiency of the maxillary alveolar bone. A significant ($p<.001$) increase in angulation of S-N-Ans and an increase in angle S-N-A reflect a protrusive anterior nasal spine as well as the maxillary alveolar bone. The vertically short but anteriorly positioned maxilla contributed to the anterior upward rotation of the palatal plane. The

Figure 6
Diagrammatic representation of the cranial base and upper face in the two groups with superimposition on S-N line and registration at S.

Figure 7
Diagrammatic representation of the cranial base and upper face in the two groups with superimposition on S-Ba and registration at S.

Figure 8
Diagrammatic representation of the lower face in the two groups with superimposition on S-Ar and registration at Ar.

Figure 9
Diagrammatic representation of the facial profile in the two groups with superimposition on S-N and registration at S.

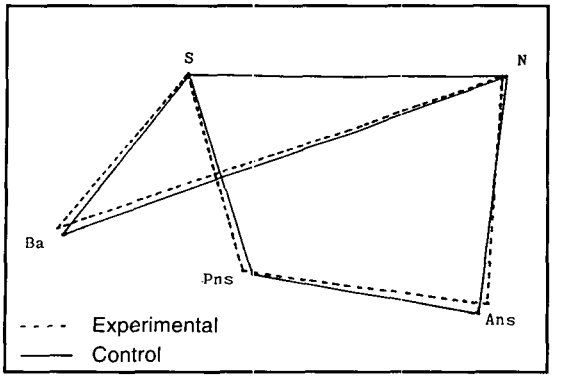
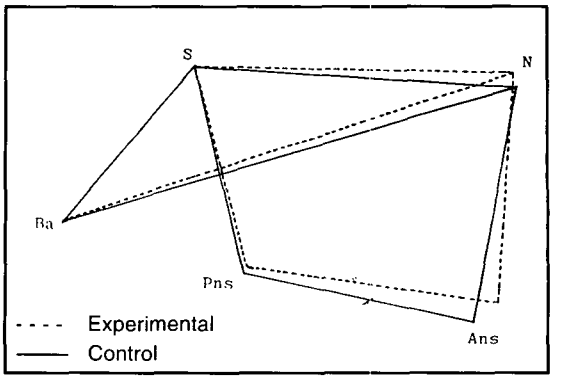


Figure 6



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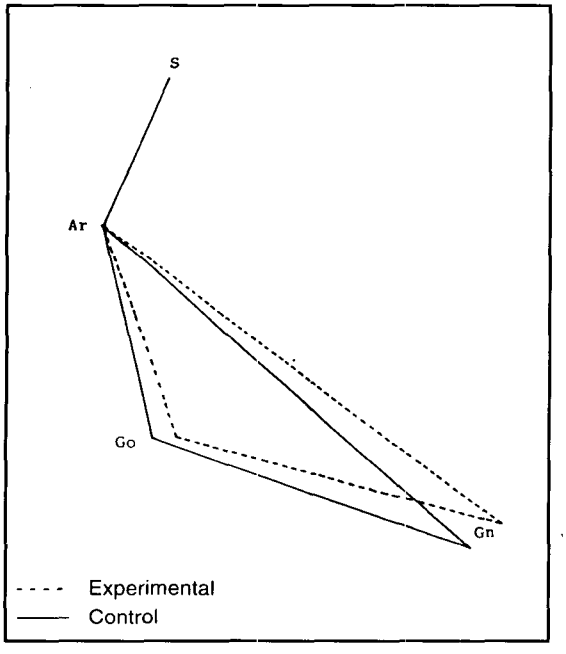

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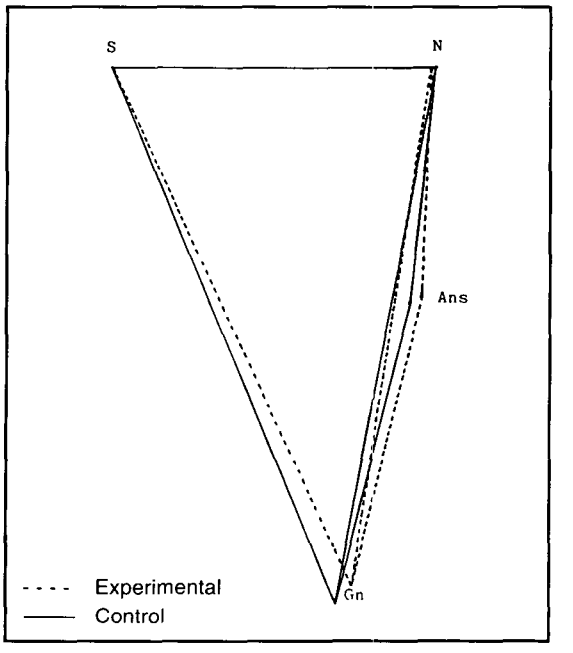

Figure 9

anterior upward swing of the palatal plane could also be due to a significantly increased cranial base angle. A similar relationship between the two has been proposed by Moss[15] following experiments on rats. The increase in the angles S-N-Ans and S-N-A was associated with a significant ($p<.001$) increase in the palatal length as measured at Ans-Pns (Figure 6). Ward et al.[4] also noted a longer palate in parents of children with clefts. This was in contrast to the findings of Coccaro et al.[9] Nakasimo and Ichinose,[11] measuring from point Ptm to Ans, also found a decrease in palatal length. The increased palatal length in this sample is accounted for by the anteriorly placed nasal spine. An increase in the palatal length noted in children with cleft lip (palate) anomaly by Friede and Pruzansky,[17] Krogman et al.,[18] Bishara et al.,[19] and Dahl[20] has been attributed to the protrusiveness of the premaxilla. Friede and Pruzansky[17] held the view that the protrusion of the premaxilla was linked to an overgrowth of the premaxillary vomerine complex rather than that of a deficiency in palatal shelves or of their relative spatial retrusion. It is thus postulated that parents of children with cleft lip/palate anomaly have a tendency towards an increased length of the palate and a more protrusive palate, which is manifest in their cleft offspring.

A significant shortening of the posterior height of the face could also be associated with a significant ($p<.001$) decrease in the articular angle causing a counterclockwise rotation of the mandible. A decrease in the Y axis was due to a forward and upward positioning of the mandible. An analogous finding was seen by Coccaro et al.[9] and Nakasimo and Ichinose.[11] The gonial angle increased significantly ($p<.01$) in the experimental sample. Nakasimo and Ichinose[11] did not observe a significant difference in this angle in their investigation. An increased gonial angle is manifest by posterior rotation of the mandible with the condylar growth directed posteriorly. This was miti-

gated by the small articular angle and Y axis. Thus the degree of mandibular prognathism was not fully expressed in the experimental group. The total anterior facial height was reduced in parents of children having cleft lip/palate anomaly as compared to parents of normal children. This tallied with the findings of Coccaro et al.[9] and Kurisu et al.[10] In the upper part of the face, parents from the experimental group had smaller outer dimensions. Their maximum head and facial widths were significantly smaller ($p<.001$) than those of the parents in the control group, as was the bizygomatic width. This was in contrast to the findings of Fraser and Pashayan[8] and also Nakasimo and Ichinose[11] who found larger values for the bizygomatic width in their samples. The inter-orbital width in the experimental group parents increased although not to a significant level. This finding coincided with the findings of Fraser and Pashayan,[8] Kurisu et al.[10] and Nakasimo and Ichinose.[11] Nasal width increased significantly ($p<.001$) in the experimental group, however, bigonial width and inter-alveolar width in the mandible decreased significantly ($p.<01$). These findings are in agreement with the observations made by Kurisu et al.[10] Nakasimo and Ichinose[11] differed by reporting a greater value of the intergonial width in their sample. Thus, the parents of cleft children showed a general tendency towards a smaller face in horizontal as well as vertical dimensions. The nasal widths were significantly larger.

The variant form of craniofacial morphology of parents of children with cleft lip/palate anomaly was manifest in a combination of alterations in angular and linear measurements on the frontal and lateral cephalogram. The distinct morphological features of these parents were cephalometerically visualized as a deficiency in total anterior facial height, especially upper anterior facial height. The anterior nasal spine was placed in an anterior and superior position resulting in a greater length of the palate, thus leading to an counterclockwise rotation of the palatal plane. A similar rotation of the mandible, which was already slightly prognathic, was also produced. These parents had small dimensions of skull and face i.e. their maximum head width, bizygomaticofrontal suture width, bizygomatic width, bigonial width and alveolar widths were smaller. They had a greater width of the nasal cavity and a longer palate.

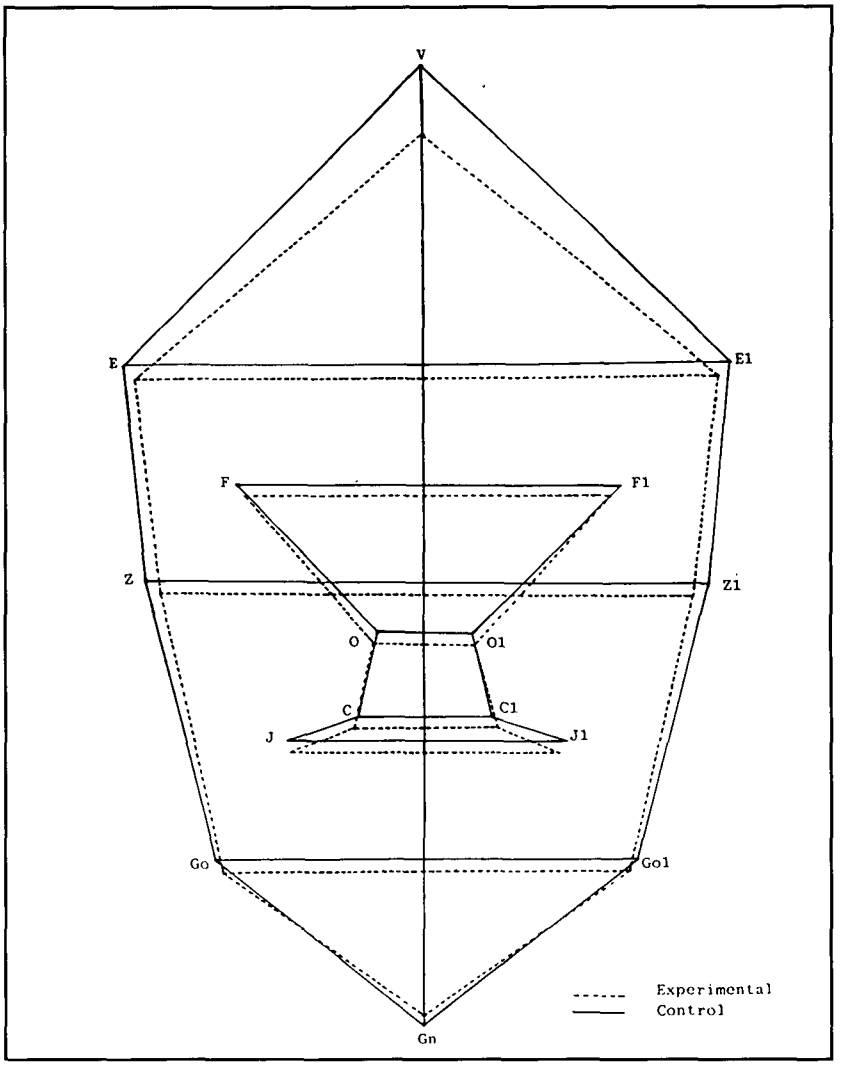

Figure 10

Conclusions

The parents of children having cleft lip/palate anomaly differed from parents of noncleft children in the following ways:

1. They had smaller facial dimensions as a whole both in transverse and vertical dimensions.
2. The cranial base angle showed an increased obtuseness with a tendency towards a shorter length of the cranial base.
3. There was an increased length of the palate with an anteriorly and superiorly placed anterior nasal spine.
4. The upper anterior as well as upper posterior facial heights were significantly shorter.
5. The biparietal, bizygomatic, bigonial and bizygomaticofrontal suture widths were significantly smaller.
6. They displayed larger nasal widths.

Figure 10
Diagrammatic representation of the frontal measurements in the two groups with superimposition on V-Gn.

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