

Size of The Cranium In Parents and Their Children with Cleft Lip

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Fifty-two children with cleft lip (with or without cleft palate), CL(P), fifty-three non-cleft children, and their respective parents were studied. Area measurements for three craniofacial components of brain case, upper face and lower face were obtained from lateral and frontal roentgenographic cephalograms. The most important finding of this investigation was that the CL(P) patients and their parents had a significantly smaller brain case than did the control subjects. A small brain case may well be one morphological characteristic predisposing toward the cleft anomaly.

The literature is abundant with investigations describing differences in craniofacial relations between cleft and non-cleft populations. These differences have been attributed to the effects of surgery and/or adaptive changes resulting from the mechanical presence of the cleft. The craniofacial complex is however, such a rigid inheritance (Hunter et al., 1970; Saunders et al., 1980; Nakasima et al., 1982) that the difference between the two populations may not be caused by postnatal factors.

Trasler (1965, 1968, 1979) considered that the difference of the susceptibility to cleft lip among several mouse strains was related to the shape of the embryonic primordial face. If facial shape is genetically determined and also related to the predisposition to cleft anomaly, normal parents of children with cleft should have facial dimensions which differ from those of the general populations.

Fraser and Pashayan (1970) suggested that the parents of children with cleft lip (with or without cleft palate) have wider bizygomatic diameters, underdeveloped maxillae and thinner upper lips than the control group. Coccaro et al. (1972) found

that parents of children with CL(P) have less convex faces with a tendency toward mandibular prognathism and a shorter upper facial height than the control group. These findings were supported by the study of Kurisu et al. (1974), as based on measurements from roentgenographic cephalograms. Shibasaki and Ohtsuka (1978) noted the shorter upper facial height and more prognathic mandible as indicated by Coccaro et al. and the thin upper lips as indicated by Fraser and Pashyan in the parents of cleft children. They also found a significantly greater cranial base flexure angle (S-N-Ba) exhibited in the parent of cleft children, although this angle had been reported to be normal in children with clefts (Ross, 1965; Engman et al., 1965; Aduss, 1971a).

Although there are differences in the data heretofore reported, many workers, collectively, have found differences in facial morphology between relatives and non-relatives of cleft probands. Most workers have focused their attention on the face or cranial base. The other portion of the cranium, the brain case has either been tacitly regarded as normal or excluded from consideration. Therefore, it seemed meaningful to examine differences in the size and form of the brain case in CL(P) children, as compared to normal controls.

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Sample

The sample consisted of 52 five-year-old Japanese (25 boys and 27 girls), 12 with cleft lip and 40 with cleft lip and cleft palate, and their normal parents. None of

these children had been diagnosed at birth as having a particular syndrome. The control group consisted of 53 five-year-old Japanese non-cleft children (13 boys and 40 girls) under observation at the Orthodontic Clinic of our University, and their parents.

TABLE 1. Age and Body Stature of Samples

	Proband		Father		Mother	
	Control N = 53	CL(P) N = 52	Control N = 53	CL(P) N = 52	Control N = 53	CL(P) N = 52
	Mean ± S.D.	Mean ± S.D.	Mean ± S.D.	Mean ± S.D.	Mean ± S.D.	Mean ± S.D.
Age (yr)	5.5 ± 0.3	5.3 ± 0.3**	36.0 ± 3.9	35.5 ± 4.7	33.1 ± 3.2	32.5 ± 4.2
Height (cm)	108.9 ± 5.4 (107.3)	106.2 ± 4.1**	166.2 ± 6.8	165.1 ± 6.3	153.1 ± 5.1	154.1 ± 4.5
Weight (kg)	18.1 ± 2.3 (17.4)	16.8 ± 1.7**	62.8 ± 9.4	62.7 ± 9.1	50.2 ± 6.5	51.4 ± 6.1
Breast (cm)	56.7 ± 3.2	55.6 ± 3.2	89.7 ± 5.6	90.4 ± 6.0	83.2 ± 5.4	83.2 ± 6.7

The values within parentheses indicate the corrected ones for age differences of CL(P) children using a regression equation of height or weight on age. Asterisks indicate a significantly different value from the corresponding value in the control group. ** p < 0.01

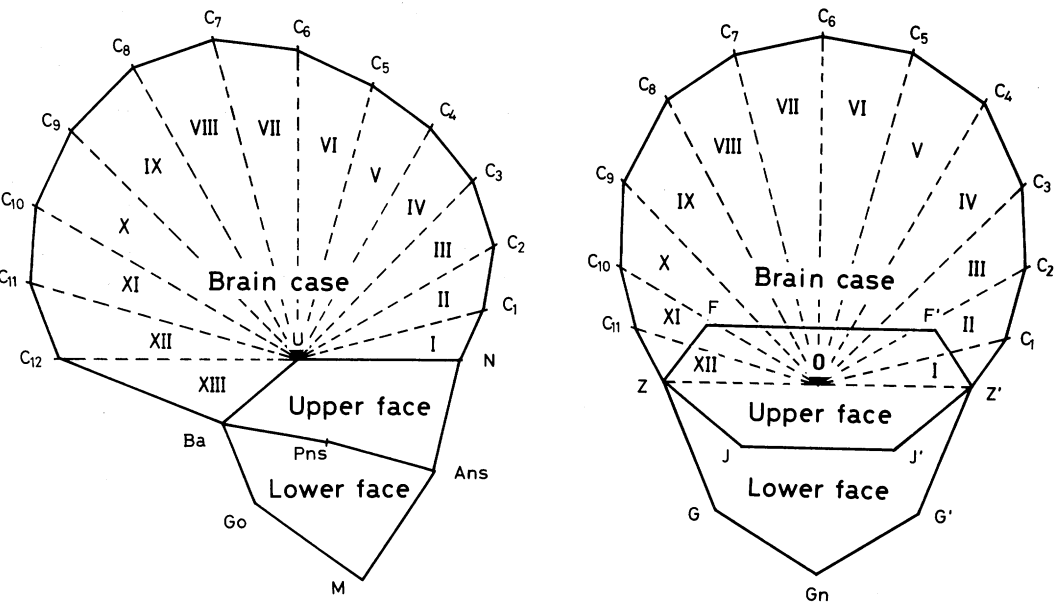


FIGURE 1. Landmarks and areas used in this investigation Lateral view: U, lowest point on outline of hypophyseal fossa; N, nasion; Ans, anterior nasal spine; Pns, Posterior nasal spine; Ba, basion; M, menton; Go, gonion; C₁-C₁₂, the points marked out every 15 degrees on the cranium outline from U-N line; Brain case, Upper face and Lower face were defined as the areas bounded by lines Ba-U-N-C₁-C₂-C₁₂-Ba, U-N-Ans-Pns-Ba-U and Ba-Pns-Ans-M-Go-Ba respectively. Frontal view: Z, Z', most lateral point of zygomatic arch; F, F', mesial border of zygomaticofrontal suture; J, J', intersection of lateral contour of maxillary alveolar process and lower contour of maxillozygomatic process of maxilla; G, G', gonial notch; Gn, lowest point on outline of mandible; O, point of intersection of Z-Z' line and the line connecting the crista galli to anterior nasal spine; C₁-C₁₁, the point marked out every 15 degrees on the cranium outline from O-Z' line; Brain Case, Upper face and Lower face were defined as the areas bounded by lines Z-Z'-C₁-C₂-C₁₁-Z, Z-F-F'-Z'-J'-J-Z and Z-J-J'-Z'-G'-Gn-G-Z, respectively.

Method

The mean values for age, body height and weight of the control children were significantly larger than those of CL(P) children (Table 1). Height and weight, however, were correlative to age, and these values were corrected using a regression equation of height and weight on age of

control children. The differences in body stature between the two groups of children were not significant after correction. Age and body stature in parents were not significantly different between CL(P) and the control group.

Differences in the sex distribution and mean ages (two months) between the two groups of children were ignored because

TABLE 2. Mean and Standard Deviation Values of Measurements of Each Cephalic Component

	<i>Brain case</i>		<i>Upper face</i>		<i>Lower face</i>	
	<i>Control</i> <i>N = 53</i>	<i>CL(P)</i> <i>N = 52</i>	<i>Control</i> <i>N = 53</i>	<i>CL(P)</i> <i>N = 52</i>	<i>Control</i> <i>N = 53</i>	<i>CL(P)</i> <i>N = 52</i>
	<i>Mean ± S.D.</i> <i>cm²</i>	<i>Mean ± S.D.</i> <i>cm²</i>	<i>Mean ± S.D.</i> <i>cm²</i>	<i>Mean ± S.D.</i> <i>cm²</i>	<i>Mean ± S.D.</i> <i>cm²</i>	<i>Mean ± S.D.</i> <i>cm²</i>
Lateral						
Proband	191.0 ± 11.5	187.9 ± 11.2	27.0 ± 1.9	25.5 ± 2.1***	28.6 ± 2.5	28.1 ± 2.3
Father	223.0 ± 14.2	221.0 ± 10.9	43.4 ± 3.0	43.5 ± 3.2	53.5 ± 4.2	52.1 ± 4.8
Mother	206.6 ± 10.9	205.1 ± 11.1	37.8 ± 2.9	37.9 ± 2.2	45.4 ± 4.0	45.4 ± 3.5
Midparent	214.8 ± 8.6	213.1 ± 8.1	40.6 ± 1.9	40.7 ± 2.1	49.5 ± 3.1	48.7 ± 3.2
Frontal						
Proband	184.3 ± 12.7	176.7 ± 11.1**	44.5 ± 4.2	44.3 ± 4.3	36.4 ± 3.6	37.3 ± 2.6
Father	203.2 ± 14.9	197.9 ± 13.2	69.2 ± 5.3	67.2 ± 5.8	70.9 ± 5.4	70.4 ± 4.7
Mother	188.7 ± 12.4	185.9 ± 12.5	61.8 ± 4.3	61.2 ± 4.1	60.6 ± 4.8	60.4 ± 4.4
Midparent	196.0 ± 9.5	191.9 ± 8.4*	65.5 ± 3.5	64.2 ± 3.4	65.7 ± 3.7	65.4 ± 3.5

Asterisks indicate a significantly different value from the corresponding value in the control group.

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

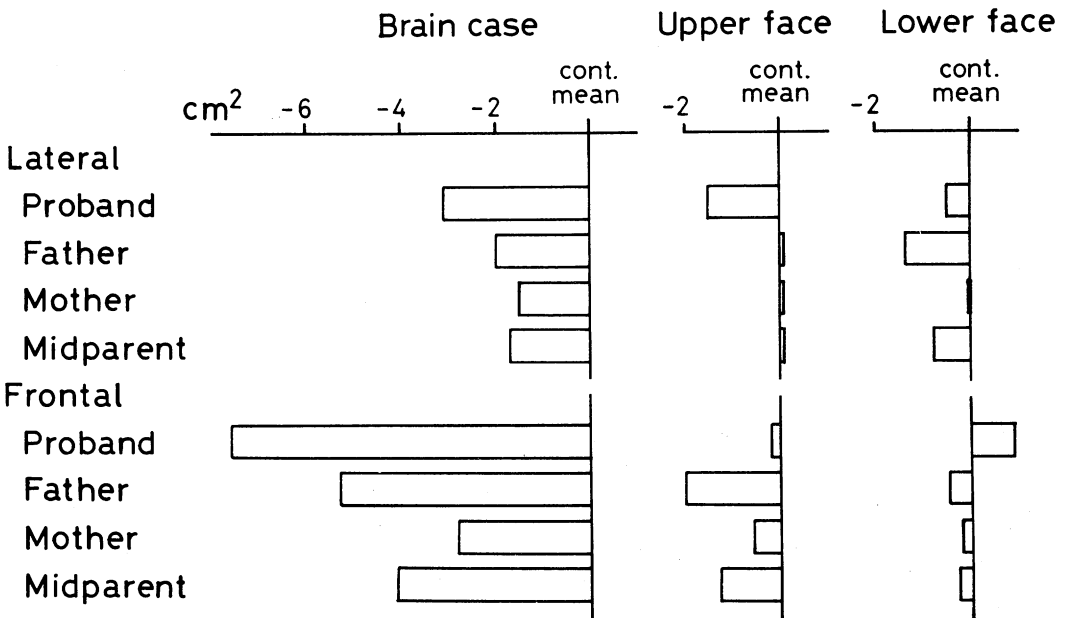


FIGURE 2. Difference in mean values of measurements of cephalic components between the CL(P) and control groups

TABLE 3. Mean and Standard Deviation Values of Brain Case Measurements

	Proband			Father			Mother			Midparent		
	Control N = 53	CL(P) N = 52	Mean $\pm$ S.D. cm ²	Control N = 53	CL(P) N = 52	Mean $\pm$ S.D. cm ²	Control N = 53	CL(P) N = 52	Mean $\pm$ S.D. cm ²	Control N = 53	CL(P) N = 52	
	Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²		Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²		Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²				
Lateral												
I	5.9 $\pm$ 0.4	5.9 $\pm$ 0.7		7.8 $\pm$ 0.7	8.0 $\pm$ 0.8		7.1 $\pm$ 0.6	7.1 $\pm$ 0.6		7.5 $\pm$ 0.5	7.6 $\pm$ 0.5	
II	8.2 $\pm$ 0.8	8.1 $\pm$ 0.9		9.8 $\pm$ 0.8	10.0 $\pm$ 1.0		9.3 $\pm$ 0.9	9.3 $\pm$ 0.9		9.5 $\pm$ 0.7	9.7 $\pm$ 0.6	
III	10.8 $\pm$ 1.1	10.6 $\pm$ 1.2		12.0 $\pm$ 1.1	12.0 $\pm$ 1.2		11.8 $\pm$ 1.0	11.7 $\pm$ 1.1		11.9 $\pm$ 0.8	11.8 $\pm$ 0.8	
IV	12.6 $\pm$ 1.2	12.6 $\pm$ 1.4		13.9 $\pm$ 1.2	14.2 $\pm$ 1.4		13.8 $\pm$ 1.1	13.7 $\pm$ 1.2		13.9 $\pm$ 0.9	14.0 $\pm$ 0.9	
V	14.6 $\pm$ 1.3	14.4 $\pm$ 1.4		16.0 $\pm$ 1.3	16.1 $\pm$ 1.4		15.7 $\pm$ 1.2	15.5 $\pm$ 1.2		15.9 $\pm$ 1.0	15.8 $\pm$ 1.0	
VI	16.8 $\pm$ 1.3	16.7 $\pm$ 1.7		18.3 $\pm$ 1.4	18.3 $\pm$ 1.6		18.1 $\pm$ 1.5	17.7 $\pm$ 1.2		18.2 $\pm$ 1.1	18.0 $\pm$ 1.1	
VII	19.2 $\pm$ 1.5	19.2 $\pm$ 1.8		21.2 $\pm$ 1.5	21.2 $\pm$ 1.8		20.7 $\pm$ 1.8	20.4 $\pm$ 1.4		20.9 $\pm$ 1.2	20.8 $\pm$ 1.3	
VIII	21.2 $\pm$ 1.7	21.1 $\pm$ 1.5		23.5 $\pm$ 1.6	23.2 $\pm$ 1.7		22.4 $\pm$ 1.8	22.2 $\pm$ 1.6		23.0 $\pm$ 1.1	22.7 $\pm$ 1.2	
IX	20.9 $\pm$ 1.7	20.6 $\pm$ 1.3		23.9 $\pm$ 1.9	23.1 $\pm$ 1.8*		22.0 $\pm$ 1.7	21.6 $\pm$ 1.6		22.9 $\pm$ 1.2	22.4 $\pm$ 1.1*	
X	19.0 $\pm$ 1.6	18.5 $\pm$ 1.2		22.3 $\pm$ 2.3	21.6 $\pm$ 2.0		20.2 $\pm$ 1.8	19.9 $\pm$ 1.7		21.2 $\pm$ 1.4	20.8 $\pm$ 1.3	
XI	16.4 $\pm$ 1.6	16.0 $\pm$ 1.3		20.1 $\pm$ 2.2	19.7 $\pm$ 2.0		17.8 $\pm$ 1.8	17.9 $\pm$ 1.6		18.9 $\pm$ 1.4	18.8 $\pm$ 1.3	
XII	13.4 $\pm$ 1.5	12.8 $\pm$ 1.2*		17.0 $\pm$ 2.2	16.6 $\pm$ 2.2		14.4 $\pm$ 1.8	14.5 $\pm$ 1.3		15.7 $\pm$ 1.4	15.6 $\pm$ 1.3	
XIII	12.0 $\pm$ 2.0	11.3 $\pm$ 1.6		17.3 $\pm$ 3.2	17.0 $\pm$ 3.5		13.4 $\pm$ 3.1	13.5 $\pm$ 2.2		15.4 $\pm$ 2.2	15.2 $\pm$ 1.8	
Frontal												
I	5.8 $\pm$ 0.7	5.7 $\pm$ 0.6		8.0 $\pm$ 0.6	7.9 $\pm$ 0.6		7.1 $\pm$ 0.7	7.1 $\pm$ 0.6		7.6 $\pm$ 0.5	7.5 $\pm$ 0.4	
II	8.8 $\pm$ 1.1	8.5 $\pm$ 1.0		10.0 $\pm$ 1.0	9.7 $\pm$ 0.9		9.1 $\pm$ 1.0	9.0 $\pm$ 1.0		9.6 $\pm$ 0.8	9.4 $\pm$ 0.6	
III	13.3 $\pm$ 1.5	12.6 $\pm$ 1.4*		14.1 $\pm$ 1.5	13.5 $\pm$ 1.2*		13.0 $\pm$ 1.5	12.8 $\pm$ 1.3		13.6 $\pm$ 1.1	13.2 $\pm$ 0.9*	
IV	18.4 $\pm$ 1.9	17.4 $\pm$ 1.6**		19.4 $\pm$ 1.7	18.9 $\pm$ 1.6		18.1 $\pm$ 1.6	18.0 $\pm$ 1.6		18.8 $\pm$ 1.2	18.4 $\pm$ 1.0	
V	22.3 $\pm$ 1.8	21.0 $\pm$ 1.5***		23.9 $\pm$ 2.0	23.3 $\pm$ 1.6		22.4 $\pm$ 1.7	22.1 $\pm$ 1.8		23.2 $\pm$ 1.3	22.7 $\pm$ 1.1*	
VI	23.4 $\pm$ 1.8	22.5 $\pm$ 1.7***		26.0 $\pm$ 2.3	25.5 $\pm$ 1.6		24.3 $\pm$ 1.9	23.7 $\pm$ 1.8		25.2 $\pm$ 1.4	24.5 $\pm$ 1.1**	
VII	23.7 $\pm$ 1.7	22.5 $\pm$ 1.6***		25.9 $\pm$ 2.3	25.6 $\pm$ 1.7		24.2 $\pm$ 1.9	23.7 $\pm$ 1.7		25.0 $\pm$ 1.4	24.6 $\pm$ 1.2	
VIII	22.3 $\pm$ 1.6	21.2 $\pm$ 1.5**		24.0 $\pm$ 2.1	23.4 $\pm$ 1.7		22.5 $\pm$ 1.8	22.2 $\pm$ 1.7		23.3 $\pm$ 1.3	22.8 $\pm$ 1.1	
IX	18.3 $\pm$ 1.6	17.7 $\pm$ 1.6		19.7 $\pm$ 1.9	19.1 $\pm$ 1.7		18.4 $\pm$ 1.6	18.1 $\pm$ 1.5		19.1 $\pm$ 1.2	18.6 $\pm$ 1.0*	
X	13.1 $\pm$ 1.5	12.8 $\pm$ 1.3		14.1 $\pm$ 1.4	13.6 $\pm$ 1.4		13.1 $\pm$ 1.2	12.9 $\pm$ 1.3		13.6 $\pm$ 1.0	13.3 $\pm$ 0.9	
XI	8.8 $\pm$ 1.1	8.8 $\pm$ 1.0		10.1 $\pm$ 1.0	9.8 $\pm$ 0.9		9.2 $\pm$ 0.8	9.1 $\pm$ 0.9		9.6 $\pm$ 0.7	9.5 $\pm$ 0.7	
XII	5.8 $\pm$ 0.7	5.9 $\pm$ 0.6		8.0 $\pm$ 0.6	7.9 $\pm$ 0.6		7.1 $\pm$ 0.6	7.1 $\pm$ 0.6		7.6 $\pm$ 0.4	7.5 $\pm$ 0.4	

Asterisks indicate a significantly different value from the corresponding value in the control group.

* p < 0.05, ** p < 0.01, *** p < 0.001

sex is not a significant factor in craniofacial morphology at this age and because no significant correlations were found in preliminary studies between age and each cranial measurement.

Lateral and frontal cephalograms were obtained for each individual using conventional cephalometric roentgenographic techniques. All cephalograms were traced by the same investigator on acetate paper, and nineteen landmarks on the lateral and twenty-two landmarks on the frontal tracings were located. Craniofacial structures were separated into three areas: brain case, upper face and lower face. The brain case, which was of particular interest in the present study, was separated into 13 parts on lateral and 12 parts on frontal cephalograms (Figure 1). Area measurements of individual parts in probands, fathers, mothers and mean values obtained for parents (midparent) in the CL(P) group were compared statistically with the values for the corresponding group of controls.

Results

DIFFERENCE IN SIZE OF EACH CEPHALIC COMPONENT BETWEEN TWO GROUPS. The data and statistical analysis of each cephalic component are summarized and presented in Table 2. Brain case size of proband and midparent in CL(P) group were significantly smaller ($p < 0.01$ and $p < 0.5$ respectively) than those in the control group, as seen on the frontal cephalogram. The upper face size of the probands in the CL(P) group was significantly smaller ($p < 0.01$) than that of the control group on the lateral cephalogram. All other measurements were not significantly different between the subjects of CL(P) and control groups. The differences are presented graphically in Figure 2. The subjects in the CL(P) group showed a general tendency toward possession of a small brain case in both probands and their parents.

DIFFERENCE IN BRAIN CASE SIZE BETWEEN TWO GROUPS. Table 3 shows the

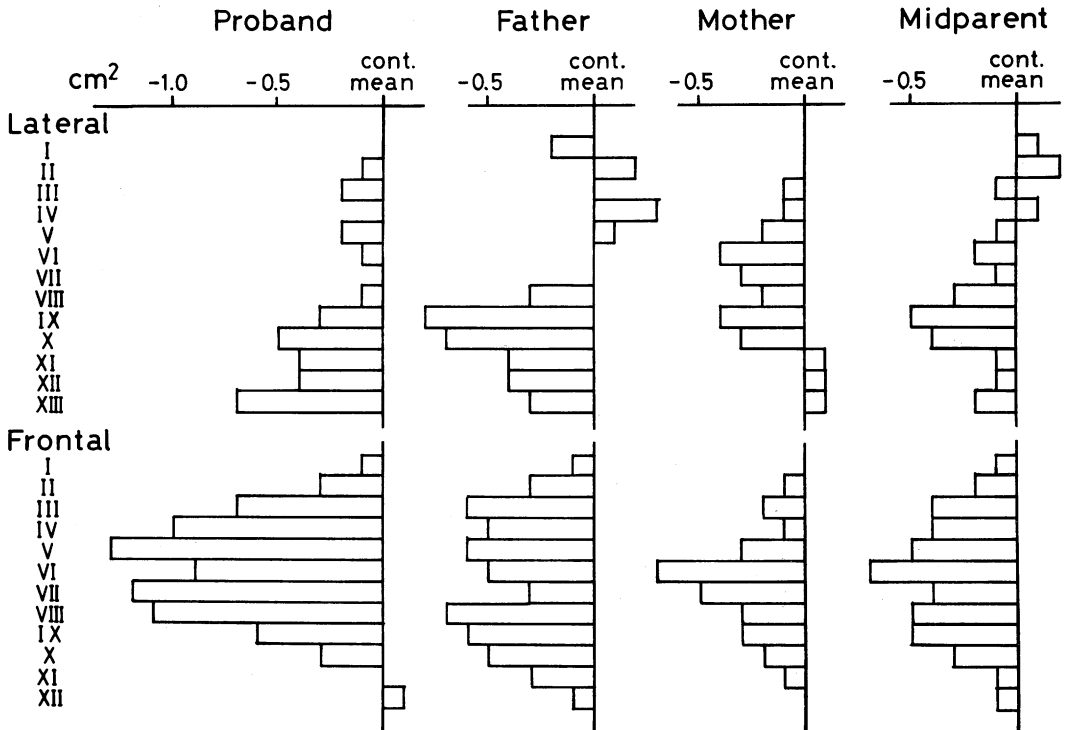


FIGURE 3. Difference between mean values of brain case measurements in the CL(P) and the control groups.

TABLE 4. Correlation Coefficients Between Corresponding Measurements of Brain Case in Proband and Midparent

	<i>Lateral</i>		<i>Frontal</i>	
	<i>Control</i> <i>N</i> = 53	<i>CL(P)</i> <i>N</i> = 52	<i>Control</i> <i>N</i> = 53	<i>CL(P)</i> <i>N</i> = 52
I	0.482***	0.390**	-0.006	0.179
II	0.451***	0.256	0.116	0.135
III	0.461***	0.251	0.128	0.218
IV	0.500***	0.272	0.267	0.143
V	0.500***	0.253	0.246	0.058
VI	0.404***	0.231	0.328*	0.145
VII	0.463***	0.304*	0.260	0.151
VIII	0.354**	0.262	0.107	0.184
IX	0.317*	0.295*	0.270	0.214
X	0.220	0.201	0.343*	0.207
XI	0.163	0.155	0.252	0.051
XII	0.440**	0.141	0.078	0.048
XIII	0.401**	0.371**		
Total	0.426**	0.264	0.384*	0.170

Levels of significance

* $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$

mean and standard deviations of the brain case measurements on both cephalograms, and these differences are illustrated in Figure 3. In the lateral view the differences were not significant. A tendency toward small brain case was noted in CL(P) children on the frontal view and it was highly significant in the parietal region (IV-VIII). Weaker trends were observed for the parents of children with CL(P), as compared to the parents in the control group.

CORRELATION OF BRAIN CASE SIZE BETWEEN PROBAND AND MIDPARENT. Table 4 and Figure 4 illustrate computed correlation coefficients of brain case size between the two sets of probands and their midparents. Significant ($p < 0.05$ and $p < 0.01$) correlation coefficients were found in the total size of brain case in the control group in the lateral and frontal cephalograms. No significant correlation coefficients were

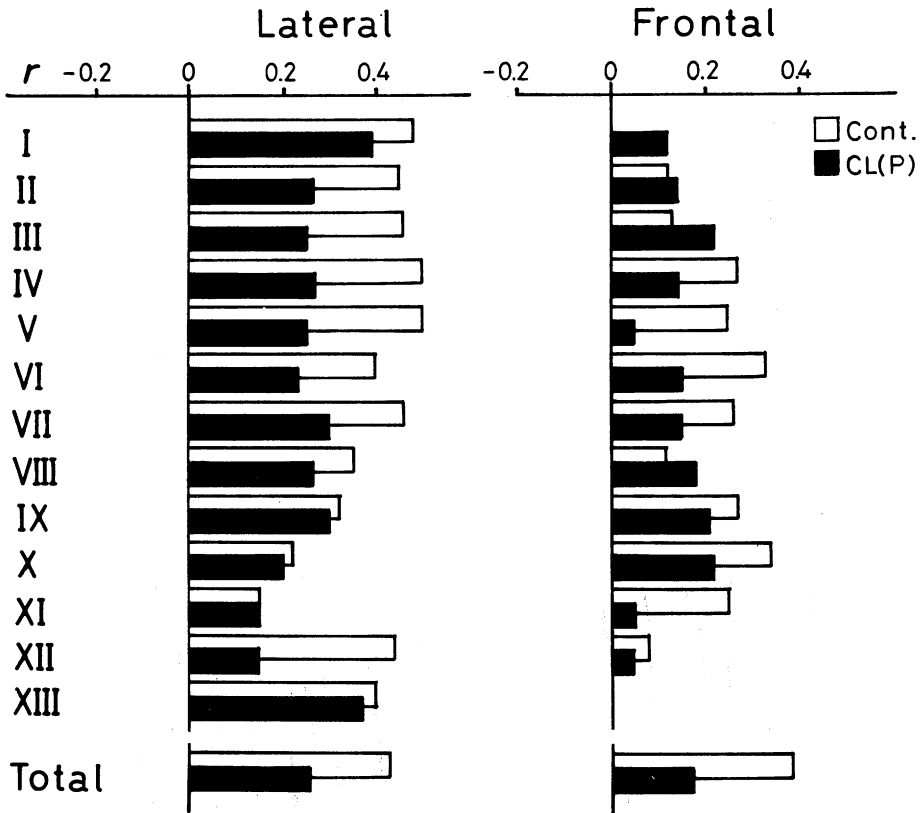


FIGURE 4. Proband-midparent correlation coefficients in brain case size.

TABLE 5. Mean and Standard Deviation Values of the Brain Case Measurements in the University Students

Brain case	Lateral			Frontal		
	Male	Female	Mid-pair	Male	Female	Mid-pair
	N = 100	N = 100	N = 100	N = 100	N = 100	N = 100
	Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²	Mean $\pm$ S.D. cm ²
I	8.0 $\pm$ 0.6	7.1 $\pm$ 0.6	7.5 $\pm$ 0.5	8.0 $\pm$ 0.6	7.1 $\pm$ 0.5	7.5 $\pm$ 0.4
II	10.1 $\pm$ 0.9	9.3 $\pm$ 0.9	9.7 $\pm$ 0.7	10.1 $\pm$ 1.0**	9.4 $\pm$ 0.9*	9.8 $\pm$ 0.6***
III	12.0 $\pm$ 1.2	11.6 $\pm$ 1.1	11.8 $\pm$ 0.9	14.0 $\pm$ 1.4*	13.4 $\pm$ 1.2*	13.7 $\pm$ 0.9***
IV	14.3 $\pm$ 1.4	13.6 $\pm$ 1.2	13.9 $\pm$ 1.0	19.7 $\pm$ 1.6**	18.7 $\pm$ 1.5**	19.2 $\pm$ 1.1***
V	16.3 $\pm$ 1.5	15.6 $\pm$ 1.3	15.9 $\pm$ 1.0	24.2 $\pm$ 1.8**	23.0 $\pm$ 1.8**	23.6 $\pm$ 1.3***
VI	18.5 $\pm$ 1.6	17.6 $\pm$ 1.4	18.1 $\pm$ 1.1	26.2 $\pm$ 1.9**	24.0 $\pm$ 2.0**	25.4 $\pm$ 1.4***
VII	21.6 $\pm$ 1.8	20.3 $\pm$ 1.6	21.0 $\pm$ 1.2	26.6 $\pm$ 1.9***	24.6 $\pm$ 2.1*	25.6 $\pm$ 1.3***
VIII	23.8 $\pm$ 1.8*	22.6 $\pm$ 1.7	23.2 $\pm$ 1.3*	24.6 $\pm$ 1.7***	23.1 $\pm$ 1.8**	23.8 $\pm$ 1.2***
IX	23.7 $\pm$ 1.8	22.0 $\pm$ 1.9	22.8 $\pm$ 1.3*	20.2 $\pm$ 1.5***	18.5 $\pm$ 1.4	19.3 $\pm$ 1.0***
X	21.8 $\pm$ 2.2	20.3 $\pm$ 2.1	21.0 $\pm$ 1.5	14.7 $\pm$ 1.2***	13.1 $\pm$ 1.2	13.9 $\pm$ 0.8***
XI	19.9 $\pm$ 2.3	18.3 $\pm$ 2.1	19.1 $\pm$ 1.5	10.6 $\pm$ 0.9***	9.2 $\pm$ 0.9	9.9 $\pm$ 0.6***
XII	16.8 $\pm$ 2.1	14.9 $\pm$ 1.8	15.8 $\pm$ 1.4	8.2 $\pm$ 0.6***	7.1 $\pm$ 0.6	7.7 $\pm$ 0.4*
XIII	17.6 $\pm$ 3.2	14.2 $\pm$ 2.6	15.9 $\pm$ 2.0*			
Total	224.4 $\pm$ 13.9	207.3 $\pm$ 13.2	215.8 $\pm$ 10.0	207.2 $\pm$ 12.5***	191.7 $\pm$ 12.2**	199.5 $\pm$ 8.5***

Asterisks indicate a significantly different value from the corresponding value in each parent of CL(P) group.
* p < 0.05, ** p < 0.01, *** p < 0.001

noted between the CL(P) group probands and their parents.

Discussion

In the present study, we noted that the brain case in cleft patients and their parents is smaller than in the control subjects especially on the frontal view. This finding seems to be inconsistent with data in earlier studies in which greater facial width such as increased bizygomatic width (Fraser and Pashayan, 1970), interocular width (Fraser and Pashayan, 1970; Dahl, 1970; Aduss et al., 1971b; Nakasima and Ichinose, 1983), nasal width (Nakasima and Ichinose, 1983) and bigonial width (Kurusu et al., 1974; Nakasima and Ichinose, 1983) in the CL(P) pedigree were noted.

An additional study, therefore, using 100 males and 100 females (all students at our University, with an average age of 24.2 and 22.2 years respectively) was carried out to obtain support for the findings in the

present study. Individual brain case measurements for males, females and mean values of random pair of male and female students (mid-pair) were compared with those for fathers, mothers and midparents of cleft children, the same samples used in the present study. The mean and standard deviations of each measurement for students are listed in Table 5 and the differences of mean values for parents of cleft children from those for control students are shown graphically in Figure 5.

The difference in the total area of brain case was not statistically significant in the lateral view. In the frontal view, parents of CL(P) children had a significantly smaller brain case than did students and the difference between two groups was more in evidence in the parietal region. Thus, the results of the additional study were similar to those of the original study.

In our opinion, the findings of a smaller brain case size in the frontal view does not

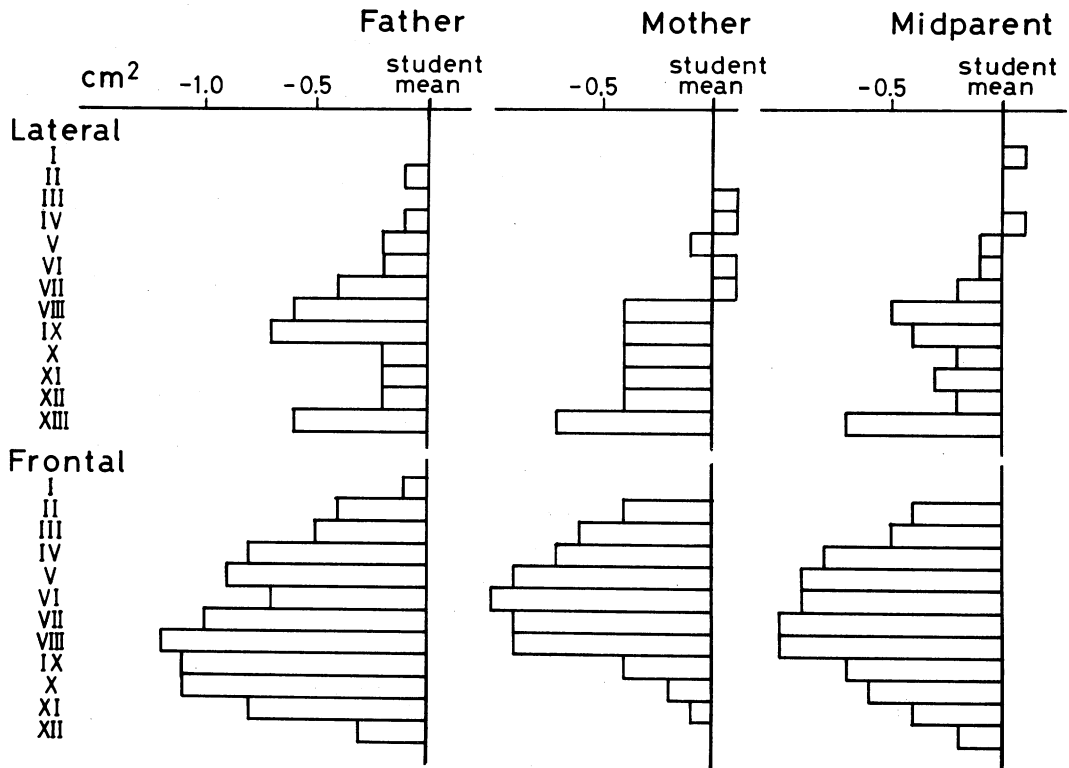


FIGURE 5. Difference between values of brain case measurements of parents of CL(P) children and those for university students.

contradict the findings of greater facial width in earlier investigations. Of particular interest is that the CL(P) pedigree has a smaller brain case, despite an increased width of the face.

Brain case malformations are frequently concomitant with cleft lip and palate, as shown in Table 6. Cohen (1978) listed 154 syndromes with orofacial clefting as a diagnostic aid for clinicians. In about quarter of these syndromes, cleft anomalies are associated with various congenital malformations of the neural skull. The supposedly clinically separate cephalic malformations may not differ in their *kind* but rather in their *degree*. They may also differ in their combinations of individual manifestations. Further analysis of these areas should elucidate how the small brain case size of the

cleft pedigree relates to susceptibility to this facial deformity.

Another interesting finding in our study is that the lower proband-parent correlation coefficients were noted for brain case in CL(P) group. This difference may be related to the surgical intervention on the neural skull growth or adaptive growth change of the skull resulting from the presence of the cleft in CL(P) patients. Thus environmental factors may lead to a decrease in correlation coefficient in the cleft group. Ross (1965) claimed "there are many abnormal environmental influences acting in an individual with a cleft lip or palate which tend to affect the configuration of the face and even the cranial base." We would like to add "and the brain case" to the above sentence.

TABLE 6. Abnormal Shape and Size of Brain Case Found in Syndrome with Cleft Lip and/or Palate

<i>Brain Case Malformation</i>	<i>Syndrome</i>
Microcephaly	Cerebrocostomandibular syndrome (Langer and Herrmann, 1974), Christian syndrome (Christian et al., 1971), Dubowitz syndrome (Gorlin et al., 1976), Fetal alcohol syndrome (Jones et al., 1973), Palant syndrome (Palant et al., 1971), Juberg-Hayward syndrome (Juberg and Hayward, 1969), Say syndrome (Say et al., 1975), Klippel-Feil syndrome (Peters, 1962), Trisomy D1 syndrome (Gorlin et al., 1976), Weaver-Williams syndrome (Weaver and Williams, 1977), Lowry-MacLean syndrome (Lowry and MacLean, 1977), Chromosomal syndrome 4p-(Arias, 1970), 5p-(Sedano et al., 1971), 6q-(Bartoszesky et al., 1978), 10q+(Yunis and Sanches, 1974), 14q-(Muldal et al., 1973), 18q-(Lurie and Lazjuk, 1972)
Microbrachycephaly	deLange syndrome (Berg et al., 1970), Pilotto syndrome (Pilotto et al., 1975), Hermann-Pallister-Opitz syndrome (Herrmann et al., 1969)
Brachycephaly	Beckwith-Wiedemann syndrome (Cohen et al., 1971)
Dolicocephaly or bathrocephaly	Acroosteolysis syndrome (Weleber and Beals, 1976), Marfan syndrome (Gorlin et al., 1976)
Oxycephaly	Apert syndrome (Peterson and Pruzansky, 1974), Lowry syndrome (Lowry, 1972), Chromosomal (14q+) syndrome (Orbeli et al., 1971)
Prominent forehead	Otopalatodigital syndrome (Gorlin et al., 1976), W syndrome (Pallister et al., 1974)
Flat occiput	Apert syndrome (Gorlin et al., 1976), Trisomy 6 syndrome (Gorlin and Pindborg, 1964)
Flattening of saddle angle (S-N-Ba)	Klippel-Feil syndrome (Hellmi and Pruzansky, 1980), OFD (oral-facial-digital) syndrome (Gorlin and Pindborg, 1964)

Conclusion

Lateral and frontal roentgenographic cephalograms were obtained from 52 five-year-old cleft with or without cleft palate (CL(P)) Japanese children and 53 five-year-old non-cleft Japanese children, and both sets of parents. Mean values of area measurements for brain case, upper face and lower face on both cephalograms were compared statistically between the CL(P) group and the control group. Proband-parent correlation coefficients for brain case measurements were calculated within each group.

The results obtained were as follows,

1). Area measurements for brain case of proband and midparent (mean value obtained for parents) were significantly smaller than those in the control group, on the frontal cephalogram. The upper face of the proband in the CL(P) group was also significantly smaller than that of the control group, on the lateral cephalogram.

2). A significantly smaller occipital region on the frontal cephalogram was noted in the CL(P) children. The similar trend was observed for the parents of CL(P) children, as compared to the parents of the controls. The small brain case of CL(P) children is probably an inherited characteristic.

3). The correlation coefficients in the parent-offspring brain case measurements of the control group were statistically significant and higher than those of the cleft group. The environmental factors affecting the neural skull growth of CL(P) patients can probably explain the decreased correlation coefficients in the CL(P) group.

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