Comparison of Craniofacial Morphology in Infants with Incomplete Cleft Lip and Infants with Isolated Cleft Palate

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The craniofacial morphology was compared in 30 infants with incomplete left-sided cleft lip and 30 infants with isolated cleft palate. Cephalometric radiographs were obtained prior to any surgical management at 2–3 months of age in the lateral, anteroposterior and axial projections. Compared to the cleft lip group, the infants with isolated cleft palate showed the following characteristics: short anterior cranial base, short maxilla, reduced posterior maxillary height, reduced dimensions of the mandible—especially mandibular length—, narrow naso- and oropharyngeal airway. The cleft lip group had larger interorbital width and symmetry deviations in the anterior part of the maxilla. It was concluded that already at two months of age and prior to any surgical interference significant differences in craniofacial morphology distinguished the group with clefts of the secondary palate from the group with clefts of the primary palate.

Differences in craniofacial morphology between individuals with clefts of the primary palate only and those with clefts of the secondary palate have previously been demonstrated in children and adults (Ross and Coupe, 1965; Dahl, 1970, Nakamura et al., 1972). Examinations of individuals with unoperated clefts have indicated that some of the craniofacial aberrations are unrelated to the surgical management, and may have an intrinsic relationship to the cleft condition (Dahl, 1970, 1971; Bishara, 1973). The craniofacial morphology in children and adults with operated clefts will accordingly represent the result of an interaction between intrinsic and environmental factors.

The purpose of the present study was to compare the craniofacial morphology prior to surgery in a group of infants with incomplete, left-sided clefts of the primary palate and a group of infants with isolated clefts of the secondary palate.

Sample

During the past five years cephalometric radiographs have been obtained at two to three months of age of nearly all Danish infants with cleft lip and palate. The examination is performed prior to any surgical intervention. This project was made possible by the centralization of the primary surgical management of the clefts to one hospital covering all of the country (Deaconess Hospital, Copenhagen). At present the material comprises 680 infants representing nearly the entire Danish population of infants born with clefts of the lip or palate during this five-year period. From this material were selected at

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TABLE 1. Sample Size, Sex Distribution and Age in Days at Examination

	Total	*Females*	*Males*	*Age*	*Age Range*
CL	30	11	19	71.4	55–103
CP	30	20	10	69.1	49–90

random 30 infants with incomplete left-sided cleft lip (CL) and 30 infants with isolated cleft palate (CP). The CP group included all degrees of isolated cleft palate. The CL group consisted of incomplete clefts. Previous studies have shown that individuals with cleft lip only exhibit minor changes in craniofacial morphology outside the cleft area (Ross and Coupe, 1965; Dahl 1970). However, in unoperated, severe cases the premaxillary region may exhibit distortions. The selection of incomplete CL was made in order to have a group as near normal as possible. All of the children were of Danish ancestry. The age and sex distributions of the sample are shown in Table 1.

Method

A cephalometric unit had been constructed for the project (Kreiborg, Dahl and Prydsø, 1977). The set-up includes a head holder for obtaining cephalometric films in the lateral, anteroposterior, and axial projections. Two X-ray tubes are placed at a fixed distance of 180 cm from the mid-sagittal plane and a plane through the ear-rods. For the lateral projection the enlargement at the mid-sagittal plane was 5.6%. For the anteroposterior and axial projections the enlargement at a plane through the ear-rods of the cephalostat was 8.3%. No correction was made for the enlargements. Exposures were made at 75–85 KV, 280 mA, and 0.06 sec. Primary diaphragm and high speed intensifying screens were used. During the exposure the infants were sedated with N_2O-O supplemented with Halothan for 2–3 minutes.

At the admission to the hospital the weight and body length of the infants were recorded. The birth weight and birth length were obtained from birth certificates. A total of 63 reference points were recorded directly from the radiographs by a D-Mac pencilfollower (Figures 1, 2, 3, 4). Based on the recorded reference points 84 variables were calculated by the program COORD (Solow and Tallgren, 1976). Basic statistical parameters were calculated and the differences between the mean values were assessed by Student's t-test.

Results

BODY MEASUREMENTS. The weight and body length at birth were similar in the two groups (Table 2). At the examination at two months of age the difference in body length was still insignificant, but the CL infants had gained considerably more in weight than had the CP infants.

RADIOGRAPHIC MEASUREMENTS (Table 3).

Cranium. The length and width dimensions of the cranial vault showed insignificant differences between the two groups.

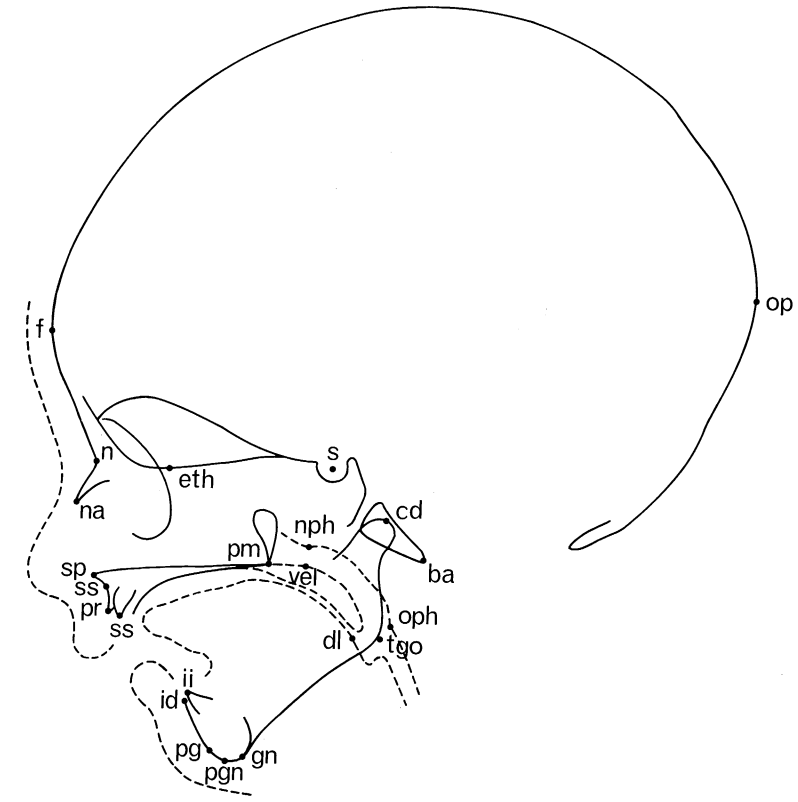
Cranial base. The n-s distance was significantly greater in CL. Other linear and angular measurements in the cranial base showed insignificant differences between the groups.

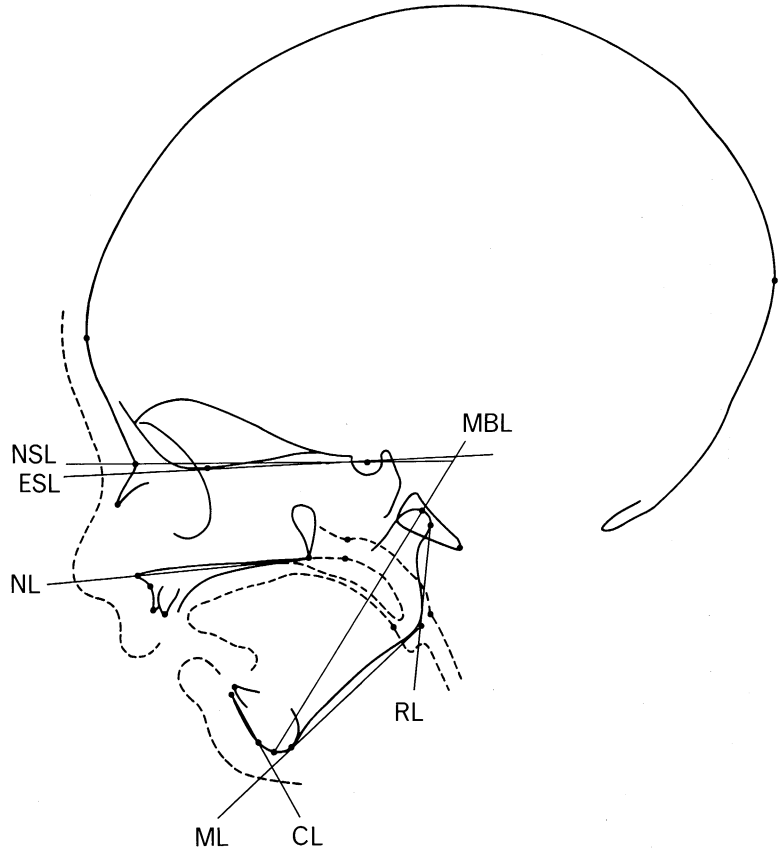
Upper face. The interorbital width was greater in CL both when measured to the medial and to the lateral aspects of the orbits. The total width of the nasal cavity, however, was less in CL because of a smaller distance from the lateral border of the nasal cavity to the nasal septum on the cleft side. The maxilla was longer in the CL group and more prognathic. No significant differences were found in the anterior height of the maxilla, but its posterior height was greater in CL, resulting in a lesser inclination of the NL-plane relative to the cranial base.

Mandible. The total mandibular length and the length of the mandibular body and ramus were significantly greater in the CL group. In this group the measurement of mandibular anterior height was also greater, although no significant differences occurred in mandibular width.

The analysis did not disclose any significant differences in the shape of the mandible in the two groups, but the mandibular retrognathia was considerable in CP, and there was an increased backward inclination of the mandibular line in relation to the cranial base, but not relative to the NL-plane. The difference in mandibular inclination was reduced by a larger degree of mouth opening during the exposure in the CL group (measurement is-ii).

Symmetry aberrations. Significant differences were found between the groups in the symmetry of the anterior part of the maxilla. The anterior nasal spine and the mid-line of the maxillary dental arch were displaced to the





FIGURES 1 and 2. Reference points and reference lines, **ba**: Basion, most posteroinferior point on the clivus; **s**: Sella, center of sella turcica; **n**: Nasion, the intersection between the anterior contour of the frontal bone and the upward extension of the anterior contour of the nasal bone; **na**: Nasal apex, most anterior point on the nasal bone; **sp**: Spinal point, apex of the anterior nasal spine; **ss**: Subspinale, deepest contour on the upper alveolar arch; **pr**: Prosthion, lowest and most prominent point on the upper alveolar margin; **is**: Incision superius, the incisal edge of the most prominent upper central incisor; **ii**: Incision inferius, the incisal edge of the most prominent lower central incisor; **id**: Infradentale, highest and most prominent point on the lower alveolar margin; **pg**: Pogonion, most anterior point on the chin estimated from ML; **pgn**: Prognathion, point on the mandibular symphysis farthest from the cd; **CL**: Chin line, tangent to the chin through id; **cd**: Condylion, most superoposterior point on the condylar head; **pm**: Pterygomaxillare, intersection between the dorsal contour of the maxilla and NL; **vel**: Velum, the point on the nasal surface of velum with the shortest distance to the posterior pharyngeal wall; **nph**: Naso-pharyngeal wall, point on the posterior pharyngeal wall with the shortest distance to velum; **dl**: Dorsum linguae, point on dorsum lingua with the shortest distance to the pharyngeal wall; **oph**: Oro-pharyngeal wall, point on the posterior pharyngeal wall at the shortest distance to dorsum linguae; **f**: Frontale, the most prominent point on the frontal bone estimated from the NSL; **op**: Opisthocranion, most posterior point on the cranial vault defined as the point farthest from the nasion; **NSL**: Nasion-sella line, line joining n and s; **NL**: Nasal line, line through sp and pm; **ML**: Mandibular line, tangent to the lower border of the mandible through gnathion; **LOL**: Latero-orbitale line, line joining the lo point on the right and the left side; **lom**: Midpoint of LOL; **MBL**: Mandibular base line, line connecting pgn and cd; **RL**: Ramus line, tangent to the posterior border of the ramus.

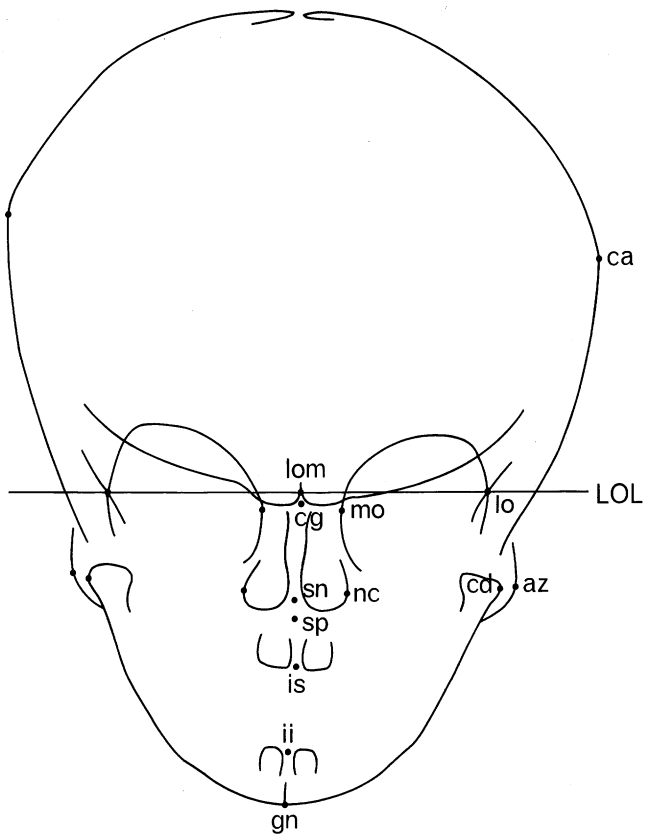


FIGURE 3. Reference points and lines. **ca**: Calvarium, the most lateral point on the cranial vault; **lo**: Latero-orbitale, the intersection between the lateral margin of the orbit and linea innominata; **mo**: Medio-orbitale, the most medial point on the medial orbital margin at the level of the cribriform plate; **cg**: Crista galli, the most inferior point of the crista galli corresponding to the nasal septum; **nc**: Nasal cavity, the most lateral point in the nasal cavity; **sn**: Septum nasi, the most inferior central point on the vertical part of the nasal septum; **sp**: Spinal point, the anterior nasal spine; **is**: Incision superius, mid-point between the upper deciduous central incisors at the level of the incisal edges; **ii**: Incision inferius, mid-point between the lower deciduous central incisors at the level of the incisal edges; **gn**: Gnathion, the most inferior point on the symphysis; **tgo**: Gonion tangent point, point of intersection between the mandibular line and the ramus line.

non-cleft side in CL, and the width of the nasal cavity on the cleft side was reduced. All measurements indicated a symmetric development of the CP maxilla. No symmetry aberrations were revealed in other structures of the cranium, the cranial base, the upper face or the mandible in either of the groups.

Upper airways. The space in the upper airways was significantly restricted in the CP group.

Discussion

The craniofacial morphogenesis in individuals with operated clefts of the lip and palate is determined by many intrinsic and environmental factors. Changes in the postnatal development of the face may occur both as adaptions to intrinsic morphological aberrations related to the cleft and as reactions to environmental changes, principally surgery. By examining infants before surgery at 2–3 months of age the major postnatal growth factors were excluded in the present study.

During the period from birth to the time of examination the CP infants gained less in weight, probably reflecting the feeding problems commonly experienced with cleft palate.

The marked retrognathia of both the maxilla and the mandible in the group with cleft palate represented a major discriminating factor between the two groups. This is in agreement with previous findings in older children and adults (Ross and Coupe, 1965; Shibasaki and Ross, 1969; Dahl, 1970; Bishara, 1973).

The changes of the maxilla in the CP group involved a deficiency of maxillary length and of the posterior maxillary height. Together with the shorter anterior cranial base these findings suggest that an intrinsic deficiency of embryonic chondral or prechondral structures may occur in association with the clefting of the secondary palate. Previous studies of embryos with clefts seem to substantiate this assumption (Stark, 1954; Avery, 1961).

The mandibular retrognathia was primarily caused by a deficiency of the total man-

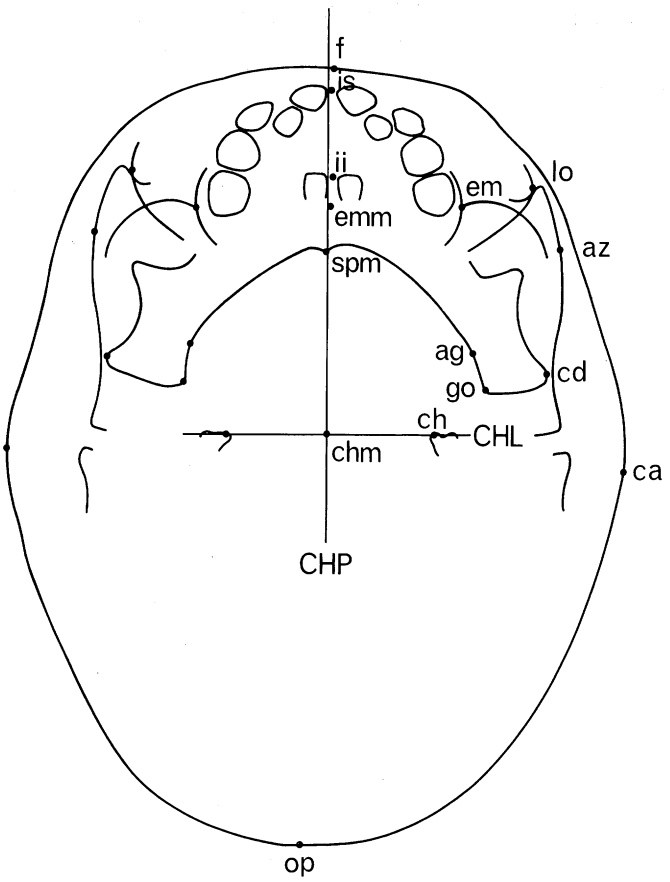


FIGURE 4. Reference points and lines. **ch**: Cochlea, the most anterior and median point of the cochlea; **cd**: Condylion, the most lateral point on the mandibular condyle; **az**: Zygomatic arch, the most lateral point on the zygomatic arch; **em**: Ectomolare, the most lateral point on the maxilla corresponding to the infrazygomatic crest; **go**: Gonion, the most inferior and posterior point on the gonion angle; **ii**: Incision inferius, mid-point between the deciduous central incisors; **is**: Incision superius, the mid-point between the deciduous central incisors; **emm**: Ectomolare mid-point, mid-point on a line connecting the em point on the right and the left side; **CHL**: Cochlea line, line connecting the cochlea point on the right and the left side; **chm**: Midpoint of CHL.

TABLE 2. Body Weight in Kilograms and Body Height in Centimeters of 30 CL and 30 CP Infants.

	CL		*CP*		*Diff.*
	$\bar{x}$	s^2	$\bar{x}$	s^2	*P*
Weight, birth	3.457	0.272	3.413	0.506	n.s.
Weight, 2 months	5.255	0.387	4.759	0.323	0.01
Height, birth	52.30	4.702	51.73	7.170	n.s.
Height, 2 months	59.52	6.560	58.82	7.430	n.s.

dibular length. This dimension can be considered insignificantly influenced by environmental factors during late fetal growth, and it would be reasonable to relate the congenital mandibular retrognathia to a hypoplasia of the mandibular arch cartilage of the embryo. This assumption is supported by experimental studies on cleft palate in animals (Deuschle and Kalter, 1962; Trasler, 1968).

The two groups in the present study exhibited no significant differences in the shape of the cranial base or of mandibular shape. This finding should not be immediately compared to observations in older children and adults, because differences in these structures may arise between the two types of clefts during the future postnatal growth. In the CP individuals an extended head posture can be anticipated when the subjects assume an upright posture because of the facial retrognathia and restricted upper airway (Solow and Tallgren, 1976). This factor might well induce different adaptive growth changes in the two types of clefts resulting in differences in mandibular shape and cranial base angulation at a later stage.

The observation of restricted dimensions of the upper airway in CP shows that some of the factors involved in the respiratory problems seen in infants with the Robin sequence may be potentially present in all infants with isolated cleft palate.

Differences in the craniofacial width dimensions between CL and CP appeared as wider interorbital distances and an asymme-

TABLE 3. Statistical Data for 30 Infants with Incomplete Cleft Lip (CL) and 30 Infants with Isolated Cleft Palate (CP). Variables Indicated with "f" or "a" Represents Measurements on Anteroposterior or Axial Films Respectively.

Variable	*Group*	*Range*		$\bar{x}$	s^2	*Diff.*	*t*
		Min.	*Max.*				
Calvarium							
f-op	CL	137.4	163.8	145.46	42.85		
	CP	132.9	154.5	142.53	24.73	2.93	1.952
ca-ca$_f$	CL	99.3	120.4	109.85	31.53		
	CP	98.4	116.6	108.43	19.03	1.42	1.094
Cranial base							
n-s	CL	43.8	51.2	47.95	3.55		
	CP	40.4	51.3	46.38	4.87	1.57	2.963**
s-ba	CL	23.4	31.2	27.26	3.03		
	CP	23.3	29.4	26.52	2.28	0.74	1.759
ch-ch$_a$	CL	29.8	42.8	36.57	6.83		
	CP	32.0	40.5	36.35	4.53	0.22	0.367
n-s-ba	CL	129.0	148.2	140.36	20.58		
	CP	129.7	151.4	138.41	23.70	1.95	1.605
n-s-cd	CL	103.5	127.5	116.59	37.72		
	CP	102.3	132.5	115.27	49.39	1.32	0.775
Upper face width							
lo-lo$_f$	CL	69.2	83.5	74.19	9.82		
	CP	65.6	76.6	71.29	6.86	2.90	3.889***
mo-mo$_f$	CL	12.1	17.3	14.59	9.32		
	CP	11.9	16.4	13.78	1.54	0.81	2.410*
az-az$_a$	CL	77.3	90.6	82.78	12.87		
	CP	73.4	87.0	81.02	11.28	1.76	1.962
em-em$_a$	CL	43.8	51.9	47.27	4.50		
	CP	41.8	52.6	47.64	7.71	−0.37	0.580
nc-nc$_f$	CL	17.4	24.3	19.92	2.88		
	CP	18.1	27.1	20.87	3.33	−0.95	2.088*
Upper face length							
sp-pm	CL	31.9	42.0	36.25	5.90		
	CP	30.5	38.2	33.45	2.97	2.80	5.149***
ss-pm	CL	31.5	41.1	35.22	6.25		
	CP	28.3	34.6	31.69	2.02	3.53	6.723***
is ⊥ em-em$_a$	CL	18.4	25.2	20.81	3.14		
	CP	12.8	21.4	16.86	3.69	3.95	8.278***
Upper face height							
n-na	CL	8.8	14.3	11.76	2.33		
	CP	9.0	15.3	11.88	1.91	−0.12	0.319
n-sp	CL	21.8	31.3	26.70	3.35		
	CP	21.3	31.5	26.25	4.63	0.45	0.873
n-is	CL	31.3	41.7	36.16	5.16		
	CP	29.6	39.2	35.22	5.31	0.94	1.591
sp-is	CL	9.0	14.1	10.89	1.14		
	CP	8.7	12.8	11.10	0.92	−0.21	0.801
pm-s	CL	22.4	28.4	25.24	2.41		
	CP	21.1	26.4	23.80	1.91	1.44	3.795***
NSL/NL	CL	3.1	13.1	7.87	6.04		
	CP	5.8	18.4	10.68	9.90	−2.81	3.855***
Upper face prognathism							
s-n-na	CL	86.4	119.1	102.34	42.45		
	CP	87.6	120.3	103.30	49.06	−0.96	0.549

TABLE 3—(*continued*)

Variable	*Group*	*Range*		$\bar{x}$	s^2	*Diff.*	*t*
		Min.	*Max.*				
s-n-sp	CL	82.9	100.8	91.89	20.52		
	CP	82.1	99.5	88.68	19.46	3.21	2.781**
s-n-ss	CL	80.4	96.4	88.83	16.84		
	CP	78.7	91.1	84.22	14.31	4.61	4.524***
s-n-is	CL	76.8	88.8	82.23	8.02		
	CP	71.4	82.7	76.52	11.23	5.71	7.128***
pr-n-ss	CL	−5.8	1.6	−2.57	3.23		
	CP	−7.5	−2.4	−4.06	1.99	−1.49	3.572***
n-s-pm	CL	54.4	65.6	59.05	6.05		
	CP	49.4	63.4	57.19	8.65	1.86	2.657*
Mandible length							
cd-pgn	CL	51.1	60.2	55.91	5.22		
	CP	46.2	56.1	52.12	6.78	3.79	5.993***
gn-tgo	CL	30.0	38.3	34.12	5.08		
	CP	27.5	37.4	31.36	6.30	2.76	4.481***
Mandible height							
cd-tgo	CL	19.1	27.8	22.42	3.64		
	CP	17.6	24.8	21.40	4.03	1.02	2.017*
id-gn	CL	15.1	20.2	17.64	1.47		
	CP	14.7	18.6	16.70	1.58	0.94	2.948**
ii-gn	CL	15.6	21.1	18.49	1.88		
	CP	8.5	25.6	17.54	1.34	0.95	2.900**
Mandible width							
cd-cd_a	CL	69.0	83.1	75.67	8.56		
	CP	68.6	83.1	75.62	14.50	0.05	0.057
go-go_a	CL	51.2	62.7	56.17	8.51		
	CP	49.8	64.2	57.19	14.98	−1.02	1.153
Mandible shape							
ML/RL	CL	133.9	151.5	143.95	21.07		
	CP	134.1	151.1	144.60	43.58	−0.65	0.954
MBL/ML	CL	9.0	18.1	13.32	5.17		
	CP	8.2	18.0	13.24	8.21	0.08	0.120
CL/ML	CL	53.8	78.8	66.85	33.34		
	CP	57.9	79.2	66.41	29.28	0.44	0.305
Mandible prognathism and inclination							
s-n-pg	CL	60.6	73.8	67.31	8.17		
	CP	56.5	74.3	64.41	15.20	2.90	3.286**
n-sp-pg	CL	128.3	156.6	141.23	43.68		
	CP	126.8	149.4	141.42	25.81	−0.19	0.125
NSL/ML	CL	39.1	58.5	50.55	22.77		
	CP	35.9	62.9	53.33	34.30	−2.78	2.016*
NSL/MBL	CL	57.3	72.7	63.88	11.85		
	CP	48.3	74.8	66.57	28.03	−2.53	2.194*
NL/ML	CL	30.1	48.7	42.69	18.00		
	CP	27.1	50.6	42.65	30.69	0.04	0.031
NSL/RL	CL	79.3	95.9	86.60	19.74		
	CP	68.3	100.3	88.73	12.63	−2.13	1.786
is-ii	CL	14.6	25.7	21.61	5.42		
	CP	8.5	25.6	19.88	12.90	1.73	2.214*
Symmetry							
lo-lom-sn_f	CL	87.0	94.5	90.12	3.31		
	CP	86.0	93.0	89.93	2.10	0.19	0.447

TABLE 3—*(continued)*

Variable	*Group*	*Range*		$\bar{x}$	s^2	*Diff.*	*t*
		Min.	*Max.*				
lo-lom-sp$_f$	CL	76.0	90.0	85.85	14.82		
	CP	87.5	92.5	89.93	1.30	−4.08	5.589***
lo-lom-is$_f$	CL	83.0	89.5	86.87	3.96		
	CP	87.5	91.5	89.95	0.96	−3.08	7.604***
nc ⊥ cg-sn$_f$	CL	8.0	12.2	10.13	0.83		
(right side)	CP	7.4	14.2	10.29	1.93	−0.16	0.528
nc ⊥ cg-sn$_f$	CL	8.1	12.1	9.76	1.23		
(left side)	CP	8.5	12.9	10.55	1.35	−0.79	2.694**
em-emm-is$_a$	CL	75.0	90.0	83.73	21.34		
	CP	87.0	91.5	89.67	1.24	−5.94	6.843***
ch-chm-is$_a$	CL	83.5	91.5	87.90	4.93		
	CP	87.5	92.0	89.73	0.85	−1.83	4.170***
lo-lom-ii$_f$	CL	86.5	91.5	90.00	0.81		
	CP	86.0	92.5	90.03	1.08	0.03	0.120
lo-lom-gn$_f$	CL	86.5	91.5	90.08	0.92		
	CP	87.0	92.0	90.03	1.05	0.03	0.117
ch-chm-ii$_a$	CL	86.7	93.7	90.37	3.57		
	CP	86.0	93.2	90.58	2.26	−0.21	0.476
Upper airway							
pm-s	CL	22.4	28.4	25.24	2.41		
	CP	21.1	26.4	23.80	1.91	1.44	3.795***
pm-ba	CL	30.4	38.4	34.19	3.77		
	CP	28.0	37.0	32.78	4.13	1.41	2.748**
vel-nph	CL	1.0	6.7	3.89	2.28		
	CP	0.9	5.1	2.37	0.75	1.52	4.783***
dl-oph	CL	3.9	10.0	7.40	2.46		
	CP	0.0	8.7	4.63	5.49	2.77	5.381***

* $p \leq 0.05$, ** $p \leq 0.01$, *** $p \leq 0.001$

try of the nasal cavity in the CL group. No other width dimensions exhibited significant differences. A tendency for hypertelorism in individuals with clefts of the primary palate has been reported previously (Dixon, 1966; Dahl, 1970; Ishiguro et al., 1976) whereas the interorbital distances are considered normal in individuals with isolated cleft palate (Dahl, 1970). Together with the asymmetry of the anterior part of the maxilla the larger interorbital distance may represent an intrinsic factor related to the clefting of the embryonic primary palate. It should be considered that the examined group had only a slight degree of clefting of the primary palate. A group including the entire spectrum of primary palate clefts might well exhibit larger aberrations.

There is some evidence that certain traits of facial morphology might be causally related to the genetic predisposition to facial clefts (Fraser and Pashayan, 1970), and it has been suggested that such phenotypical expressions might serve as indicators of susceptibility for cleft lip and palate (Juriloff and Trasler, 1976; Fraser, 1980). The etiologic basis for the craniofacial changes observed in the present study may well be complex. However, the delineation of a number of morphological aberrations which most probably have an intrinsic relationship to the clefts might be useful in further research on facial shape as an indicator of susceptibility for clefts.

Conclusions

Significant differences in craniofacial morphology were found between infants with clefts of the primary palate only and infants with clefts of the secondary palate only, even at two to three months of age and prior to any treatment of the clefts. Although the effect of growth anomalies in late fetal life cannot be ruled out, the adaptive growth changes can be considered minimal. It was therefore con-

sidered justified to assume that the major part of the observed morphological aberrations reflects abnormalities in early embryonic development. In clefts of the primary palate some width dimensions of the upper face—particularly related to the interorbital distances—were involved together with asymmetry of the anterior part of the maxilla. The characteristic morphologic features in the individuals with clefts of the secondary palate were retrognathia of both the maxilla and the mandible and restricted upper airway.

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