Craniofacial Morphology in Twins Discordant for Cleft Lip and/or Palate

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The *craniofacial structure* in 44 pairs of like-sexed *twins discordant* for *cleft* of the *lip* and/or *palate* was investigated using lateral *cephalometric radiographs*. While only a slight rotation of the mandible was evident in the twins having cleft lip, more extensive rotation of the mandible was found in the twins discordant for unilateral cleft lip and palate. Larger, though generally non-significant, changes were recorded for the twins affected with bilateral cleft lip and palate.

In the twins discordant for cleft palate only, statistically significant differences were found for maxillary length, mandibular corpus length, cranial base flexure (Ar-S-Na), horizontal and vertical positioning of the posterior maxilla, mandibular plane angle, and gonial angle. Although the effect of sex on the within twin differences was not statistically significant, the effects of age, zygosity, and cleft type were.

It is suggested that there is an increasing degree of dysmorphism with increasing severity of clefting from cleft lip only to bilateral cleft lip and palate.

Introduction

Twins discordant for cleft lip and/or palate (CL/P) are an uncommon but valuable resource in the study of congenital clefts. Ross and Coupe (1965) reported on the craniofacial characteristics of six pairs of monozygotic twins discordant for CL/P ranging in age from five years to 17 years. They observed that "the entire facial skeleton of the child with a cleft palate or cleft lip and palate is not only retruded, but is rotated relative to the cranial base." Four of the cleft twins exhibited a superior positioning of the gonial area relative to the non-cleft twins.

Of 19 twin pairs discussed by Pruzansky et al. (1970), two monozygotic and 10 dizygotic pairs were discordant for oral or facial clefts. On the basis of their serial data for the twins, they suggested that the degree of within-twin similarity may vary at different ages.

Hunter and Dijkman (1977) reported on the timing of height and weight deficits in 45 sets of twins discordant for CL/P. Harvold (1961), comparing six sets of monozygotic twins discordant for clefting, mentioned the dependence of mandibular morphogenesis on maxillary form.

Although studies by Hunter (1975) and by Shields et al. (1979) also reported on cleft twin samples, they dealt with tooth eruption and incidence of clefting in twins respectively rather than morphology.

The purpose of this study was to examine the effects of clefting on craniofacial structure in 44 pairs of like-sexed twins discordant for CL/P investigated previously by Hunter and Dijkman (1977). Such like-sexed twins, especially monozygotic pairs, provide an unparalleled experimental design for matched controls. Although only one of each pair of monozygotic twins (n=20) is affected, both twins have identical genetic patterns, and it is likely that they both have a predilection for clefting (Shields et al., 1979). The dizygotic twins have, however, varying degrees of genetic similarity, and thus the non-affected twin may or may not have a predilection for clefting. Zygosity as well as sex, age, and cleft type was therefore considered in the evaluation of the data.

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TABLE 1. Distribution of The Sample According to Sex, Zygosity and Type of Cleft

	Males				*Females*				
	cl	*clp*	*cp*	*sub-total*	*cl*	*clp*	*cp*	*sub-total*	*Total*
MZ	6	7 (1)	0	13	3	2 (1)	2	7	20
DZ	2	8 (3)	2	12	4 (1)	2 (1)	6	12	24
Total	8	15	2	25	7	4	8	19	44

* Figures in brackets indicate number of bilateral clefts included in that category.

Sample

Of the 44 sets of like-sexed twins discordant for CL/P, 33 pairs were collected from the Eastern part of the United States, and 11 pairs were obtained from California. The composition of the sample according to sex, zygosity,* and cleft type is shown in Table 1. The sample was categorized into four groups according to the type of cleft for a more detailed examination:

Group 1—cleft of the lip and alveolus (CL/A)

Group 2—unilateral cleft lip and palate (UCLP)

Group 3—bilateral cleft lip and palate (BCLP)

Group 4—cleft palate or cleft of the soft palate (CP)

Although the paucity of males with CP and of females with cleft lip with or without cleft palate CL(P) was not surprising, it did present problems in the analysis of data.

Method

The lateral cephalograms were traced and, for 19 which were registered with teeth apart (8 non-cleft, 11 cleft), the mandibles were rotated into occlusion using dental casts to verify the occlusal relationships. In order to study the shape and size of the maxillary and mandibular structures and the vertical position of the molars, 14 linear and six angular measurements were recorded as shown in Figure 1, using a gauge with dial vernier (reading to 0.05 mm) and a protractor with vernier reading to 0.1 degrees.

* Zygosity of the twins was determined from serologic analysis of 12 factors: ABO, MNS, P, Rh, Kell, Duffy, Jk^a, Lewis, Gm axgfb, Gm In^v, Hp, and Gc. These tests were done by Dr. H. Gershowitz, Department of Human Genetics, The University of Michigan.

All landmarks used are defined in *The Atlas of Craniofacial Growth* (1974) except for PNS′, which is the intersection of palatal plane with a vertical line from S-FMN plane through PTM, and ANS′, which is the intersection of palatal plane with a vertical line from S-FMN plane through A-point. The perpendicular measurements to the maxillary first permanent molars projected to the sagittal plane were made from the distal occlusal contact point. Since FMN point is related to the anterior, superior point of the maxilla, the use of S-FMN plane avoids distortion caused by vertical growth of nasion which may occur in children with clefts (Scott, 1956; Enlow et al., 1969; and Aduss, 1971).

Eighteen lateral cephalograms randomly selected were retraced and measured to estimate the size of the error resulting from landmark selection, tracing, and measuring. The Pearson correlation, the mean error, and the proportion of variation due to measurement error were calculated.

The within-twin differences for the linear and angular measurements were compared visually using composite diagrams, and the data were tested for significance with a two-tailed paired t-test. Similarly, composite diagrams for each cleft group were constructed and the within-twin differences for each group were tested with a two-tailed paired t-test. Differences were considered statistically significant when the P value was .05 or less. Because of the large number of statistical tests, differences that achieved marginal significance should be viewed with caution.

The variances for monozygotic and dizygotic within-twin differences were compared and, because several were found to be significantly greater in the dizygotic twins (Table 4), a weighted multiple linear regression was used to identify the effects of sex, zygosity, age, and cleft types on within twin differences. The CP only group was not included since there is evidence to indicate that CP only is etiologically distinct from CL(P) (Fogh-Angerson, 1942; Saxen, 1975).

Comparisons with the published norms in *The Atlas of Craniofacial Growth* (1974) were made for three measurements for both CP and CL(P) groups. Na-Me, Ar-GoI, and S-Na-Me for cleft and non-cleft twins were averaged separately at two-year intervals from four to 18 years. Appropriate correction factors were used to compare the mean value of Ar-GoI and S-Na-Me for each two-year interval to the respective age norm and standard deviation.

Thus, within-twin differences, in which the value for the non-cleft twin measure was subtracted from the cleft twin measure for each twin pair and accumulated, are reported both for the entire sample and for the various sub-groups (cleft type, sex, age, and zygosity). The extent of the within-twin differences was also compared between sub-groups, and several measures were compared with population norms.

For Figures 2 through 4 and Table 2, the mean values for various groupings of the sample were calculated for cleft twins separately from non-cleft twins. However, for the basic statistical comparisons, the within-twin differences were accumulated as described and tested for significance.

Findings

Error Study. For the 18 replicate measurements, the proportion of the total observed variation that was unrelated ($1-r^2$), and therefore attributable to random error, did not exceed 0.07 (S-Na-palatal plane). Thus, at least 93 percent of the variance associated with any of the recorded dimensions was attributable to true biologic variation. It was concluded that measurement error would have little effect upon the statistical reliability of the results obtained in the main study. The

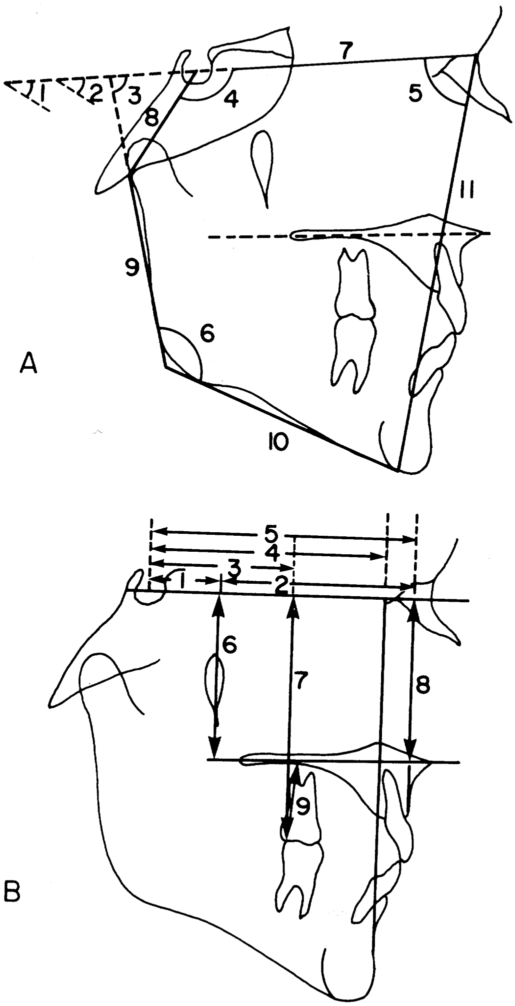

Figure 1. Linear and angular measurements.

A. Angles (degrees)
1. mandibular plane (MP) to sella nasion (S-Na)
2. palatal plane (Pal Pl) to sella nasion (S-Na)
3. articulare gonion (Ar GoI) to sella nasion (S-Na)
4. articulare-sella-nasion (Ar-S-Na)
5. sella-nasion-menton (S-Na-Me)
6. gonial angle (Ar-GoI-Me)

Perimeter (mm)
7. S-Na
8. S-Ar
9. Ar-GoI
10. GoI-Me
11. Na-Me

B. Horizontal (mm)
1. S-PTM (sella-pterygomaxillary)
2. PTM-A
3. S-distal occlusal contact
4. S-B
5. S-A

Vertical (mm)
6. PNS′ to S-FMN plane
7. molar contact to S-FMN plane
8. molar contact to palatal plane.

largest mean errors were 0.56 degrees for SN-palatal plane and 0.5 mm for Na-Me. Measurements associated with S-FMN plane were demonstrated to be reproducible.

OVERALL SAMPLE. (Table 2 and Figure 2). The cleft twins exhibited, on the average, a more posteriorly positioned symphysis with smaller ramus height and corpus length as well as a larger mandibular plane angle than their non-cleft twins. The posterior surface of the maxilla was more posteriorly positioned, and posterior palatal plane was more superiorly positioned in the cleft twins than in the non-cleft twins.

CLEFT GROUPS. (Table 3 and Figure 3). Only statistically significant findings are mentioned here. See Table 3 for complete data.

Cleft lip and alveolus. The cleft group with involvement of the primary palate only (n=15) differed from their non-cleft co-twins on two measurements. They had a 2.3 degree larger angulation of the posterior border of the ramus to SN, and S-Ar was 1.8 mm shorter. Both of these differences are consistent with a clock-wise opening rotation of the mandible in the cleft twins.

Unilateral cleft lip and palate. The UCLP group (n=13) exhibited more extensive within-twin differences than were found in the CL group. The angulation of the palatal plane relative to S-Na was greater in the cleft twins because PNS′ to S-FMN was 3.5 mm shorter. The posterior ramus height also exhibited a within-twin mean deficit of 2.4 mm in the cleft twins. Overall, the cleft twins in this group had smaller faces (total perimeter) characterized by decreased vertical height in both the posterior part of the maxilla and the mandible relative to their co-twins.

Bilateral cleft lip and palate. In the BCLP

TABLE 2. Comparison of Cleft and Non-Cleft Twins

Measurement	*Number*	*Cleft Mean*	*Non-Cleft Mean*	*Mean Difference*
Perimeter				
S-Ar	44	34.61	35.11	−0.50
Ar-Go	44	42.95	44.72	−1.77**
Go Me	44	68.73	70.69	−1.96***
Na Me	44	118.31	116.88	+1.42
S Na	44	72.85	73.48	−0.63
Total Perimeter	44	337.44	340.89	−3.45
Maxilla				
S-Ptm	44	15.72	17.37	−1.65**
S-A	44	64.03	67.08	−3.05**
PNS ⊥ SFMN	43	43.15	45.14	−1.99***
ANS ⊥ SFMN	43	47.29	48.08	−0.79
Ptm-A	44	48.32	49.71	−1.40*
Mandible				
S-B	44	52.30	57.40	−5.10***
Molar				
S-Mo	32	26.12	28.89	−2.77***
Mo ⊥ SFMN	32	65.22	65.86	−0.64
Mo ⊥ Palatal Plane	32	19.60	18.76	+0.85
Angular				
S-Na-Palatal Plane	43	7.83	6.48	+1.35
S-Na-Me	44	72.00	73.97	−1.97**
S-Na-MP	44	36.73	33.21	+3.52***
(S-Na)-(Ar-Go)	44	88.75	87.29	+1.46*
Gonial Angle	44	127.98	125.92	+2.06
Ar-S-Na	44	124.61	123.79	+0.82

* Significant at 0.05 level of significance.
** Significant at 0.01 level of significance.
*** Significant at 0.001 level of significance.

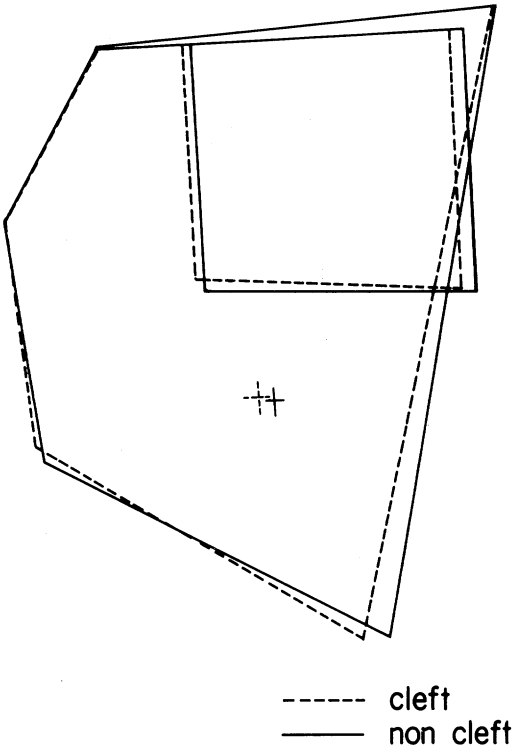

Figure 2. Composite facial diagram of twins discordant for cleft lip and/or palate (n=44).

twins (n=6), the posterior border of the maxilla (PTM) was a mean of 4.0 mm more posterior than in the non-cleft co-twins. Although other within-twin differences were large, they did not achieve statistical significance because of the small sample size.

Cleft palate. For the isolated CP group (n=10), the cranial base angle, Ar-S-Na, was an average of 3.8 degrees larger than in the non-cleft co-twins. Similarly, for this group, the maxilla was smaller, more posteriorly positioned, and rotated higher at the distal portion of the palatal plane than was true for the non-cleft twins. Their mandibles had, on the average, both larger gonial angles and larger mandibular plane angles, and B-point was a mean of 9 mm more posterior than in their non-cleft co-twins. (See Figure 1B.) The measurement representing mandibular corpus length (GoI-Me) was also shorter in the cleft than in the non-cleft twins.

Effect of Sex, Age, Zygosity, and Cleft Type. A multiple regression analysis was carried out to identify the independent effects of sex, age, zygosity, and cleft type.

Sex. In the CL(P) groups, the effect of sex on the within-twin differences was not statistically significant. That is, the differences between the male twin pairs were neither statistically larger nor smaller than those between the female twin pairs for the measures studied.

Age. The effect of age on the within-twin differences in CL(P) groups was statistically significant for three linear measurements (Ar-GoI, Molar to S-FMN, and Molar to palatal plane) and two angular measurements (mandibular plane angle and gonial angle).

The within-twin differences for ramus height, mandibular plane angle, and gonial angle were greater in the older twins. Before the age of ten, the mean distance from S-FMN plane to the occlusal contact of the maxillary molars was greater on the average in the cleft twins, but after the age of ten that dimension was smaller for the cleft twins than for their non-cleft twins. The mean within-twin difference for the distance from palatal plane to occlusal point of the molars was also greater in the cleft twins before the age of ten.

Zygosity. Although the dizygotic twin pairs usually exhibited larger mean within-twin differences as is evident in Table 4, they were not significantly different from the mean monozygotic within-pair differences with one exception. That is, the monozygotic twins exhibited a greater clockwise rotation of the ramus in the cleft twins relative to the non-cleft twins than was found in the dizygotic twin comparisons.

Cleft type. The effect of cleft type on the within-twin differences was statistically significant for five linear measurements (S-Ar, Na-Me, Ar-GoI, S-PTM, PNS′ to S-FMN). S-Ar and Na-Me within-twin differences, which reflect differences in mandibular position, were larger in the BCLP group and indicated a more posterior and inferior positioning of the mandible in these cleft twins than occurred in the other cleft groups. The within-twin deficit for ramus height, Ar-GoI, varied with the cleft type. The greatest within-twin difference for S-PTM increased with the severity of clefting from CL/A to BCLP. Although the within-twin difference for the vertical position of PNS′ was statistically different among the different cleft groups, the greatest intra-twin difference was exhibited in the UCLP group.

Comparison to Published Norms. Generally, comparison with the norms found in *The*

TABLE 3. Comparison of Mean Within Twin Differences for Four Cleft Groups

Measurement *Number of Pairs*	*Cleft Lip and Alveolus* *15*	*Unilateral Cleft Lip and Palate* *13*	*Bilateral Cleft Lip and Palate* *6*	*Cleft Palate or Cleft Soft Palate Only* *10*
Perimeter				
S-Ar	−1.79*	−0.17	+2.63	−0.89
Ar-Go	−0.54	−2.42**	−3.87	−1.53
Go Me	−1.39	−0.93	−3.27	−3.38*
Na Me	+1.35	−1.72	+6.04	+2.86
S Na	+0.78	−1.17	−1.47	−1.55
Total Perimeter	−1.50	−6.41*	+0.07	−4.51
Maxilla				
S-Ptm	−0.14	−1.20	−3.99*	−3.11**
S-A	−0.21	−3.00*	−3.95	−6.83**
PNS ⊥ SFMN	−0.02	−3.49***	−2.43	−2.81**
ANS ⊥ SFMN	−0.71	−2.05	−1.49	+1.34
Ptm-A	−0.07	−1.80	+0.04	−3.72**
Mandible				
S-B	−3.06	−3.14	−7.37	−9.34**
Molar				
S-Mo	−1.51	−2.96	−5.15	−3.55**
Mo ⊥ SFMN	+0.19	−2.02	+1.52	−1.15
Mo ⊥ Palatal Plane	+0.21	+1.33	+2.81	+0.54
Angular				
S-Na-Palatal Plane	−0.75	+2.21*	+1.00	+3.62
S-Na-Me	−2.02	−0.95	−3.47	−2.31
S-Na-MP	+2.48	+1.30	+6.22	+6.35*
(S-Na)-(Ar-Go)	+2.28**	+1.65	−0.02	+0.87
Gonial Angle	+0.20	−0.35	+6.24	+5.48**
Ar-S-Na	−0.45	+0.05	+0.75	+3.79*

* Significant at 0.05 level.
** Significant at 0.01 level.
*** Significant at 0.001 level.

Atlas of Craniofacial Growth (Riolo et al., 1974) at the various ages showed few departures greater than one standard deviation. The exceptions were principally for the 19 pairs of female twins with either CL(P) or CP.

Discussion

Opening rotation of the mandible in the cleft twins relative to the non-cleft twins was present in varying degrees in all the cleft types.

The maxilla was shorter in height posteriorly in the cleft twins relative to their co-twins for all the cleft groups except CL/A. The greatest within-twin deficit for that measure was in the affected twins with UCLP. The different effects of a variety of surgical procedures, often multiple, on maxillary size and position (vertical position of PNS) was not explored because of incomplete medical records. For the same reason, the extent or severity of the cleft is not known precisely. Hence, comparisons with other studies more exact in that respect must be of a general nature.

Cleft lip and alveolus (Figure 3A and Table 3). The twins discordant for CL/A were very similar. The only marked intra-twin difference was an apparent clockwise rotation of the mandible in the cleft twins.

Unilateral cleft lip and palate (Figure 3B and Table 3). In the UCLP group, the greatest difference between the cleft twins and their co-twins was in the total perimeter of the face, which was 6.4 mm smaller on the average in the cleft twins, principally because of the Ar-GoI deficit. The smaller face size may be proportional to general body size as Ross (1965) suggested. The finding of Hunter and Dijkman (1977) that, in this sample, the cleft twins were shorter and lighter than their co-twins after the age of ten, supports that hypothesis.

The smaller mean gonial angle and only

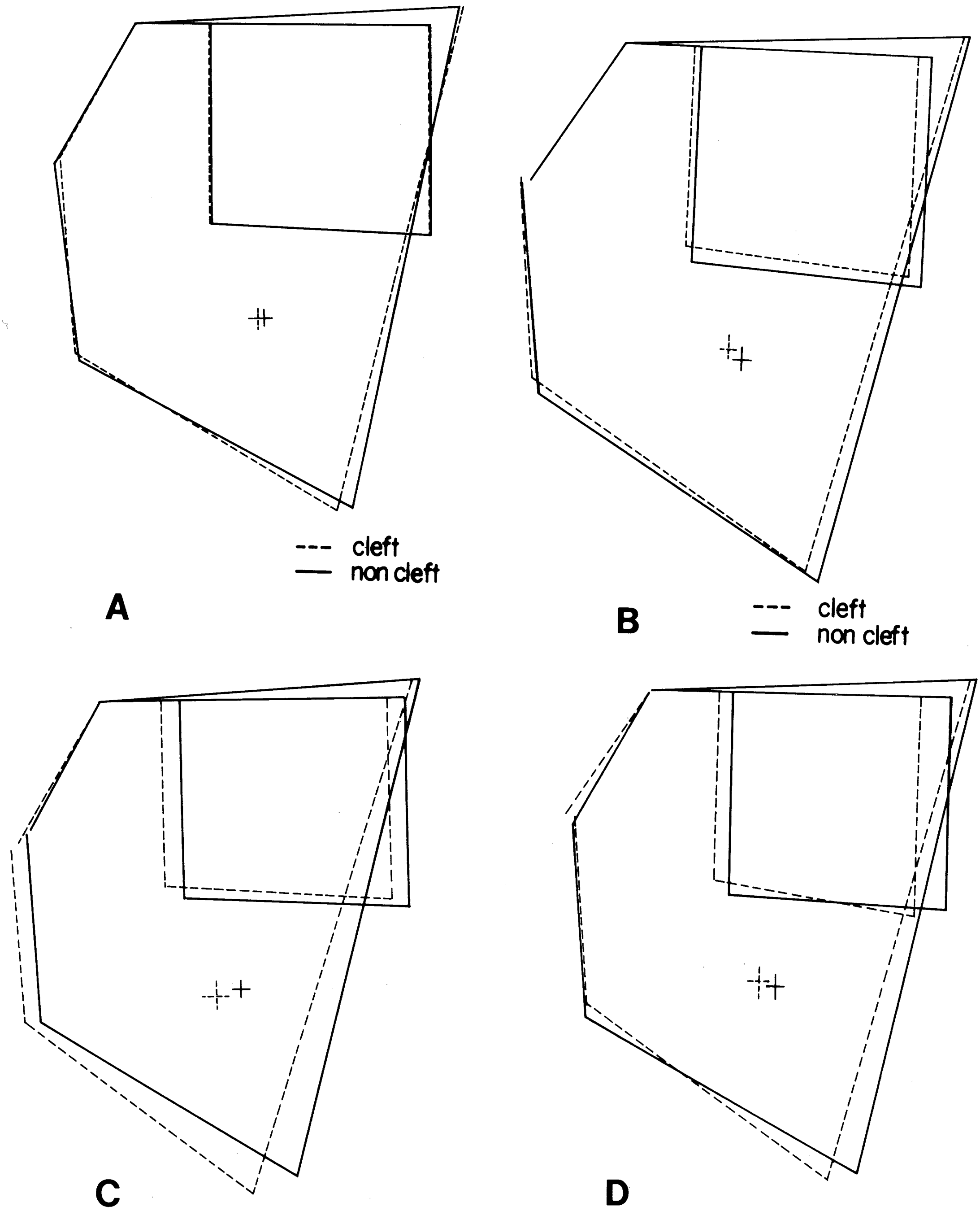

Figure 3. Composite diagrams for four groups of twins discordant for cleft lip and/or palate.
A. Twins discordant for CL/A (n=15)
B. Twins discordant for UCLP (n=13)
C. Twins discordant for BCLP (n=6)
D. Twins discordant for CP (n=10)

TABLE 4. Variance and F-Ratios for All Monozygotic Vs All Dizygotic within Twin Differences

Measurement	*Monozygotic*		*Dizygotic*		*F-Ratio*
	N	*Variance*	*N*	*Variance*	*DZV* / *MZV*
Perimeter					
S-Ar	20	47.97	24	140.26	2.73*
Ar-Go	20	19.69	24	12.39	0.63
Go Me	20	6.41	24	18.62	2.90*
Na Me	20	26.31	24	57.70	2.19*
S Na	20	4.79	24	7.31	1.53
Total Perimeter	20	47.97		140.26	2.92*
Maxilla					
S-Ptm	20	5.05	24	11.86	2.35*
S-A	20	25.35	24	35.51	1.40
PNS ⊥ SFMN	20	8.83	23	9.86	1.12
ANS ⊥ SFMN	20	12.39	23	21.29	1.72
Ptm-A	20	12.82	24	18.53	1.45
Mandible					
S-B	20	44.20	24	73.15	1.65
Molar					
S-Mo	16	15.49	16	24.44	1.58
Mo ⊥ SFMN	16	5.63	16	17.64	3.14**
Mo ⊥ Palatal Plane	16	6.93	16	8.77	1.27
Angular					
S-Na-Palatal Plane	20	18.54	23	25.85	1.39
S-Na-Me	20	12.90	24	16.88	1.31
S-Na-MP	20	34.86	24	55.75	1.60
(S-Na)-(Ar-Go)	20	13.24	24	16.98	1.28
Gonial Angle	20	27.69	24	58.79	2.12*
Ar-S-Na	20	12.51	24	44.25	3.54*

* Where N equals 20 and 24, an F-value of 2.08 or greater is significant at 0.05 level of confidence.
** Where N equals 16 and 16, an F-value of 2.5 or greater is significant at 0.05 level of confidence.

slightly larger mean mandibular plane angle (1.3 degrees) in the cleft twins relative to the non-cleft twins do not support the findings of Aduss (1971) or Hayashi et al. (1976), who reported larger gonial angles and mandibular plane angles in UCLP children as compared with a control group. The similarity in gonial angle and mandibular plane angle may be related to the genetic similarity of the twins.

Bilateral cleft lip and palate (Figure 3C and Table 3). The posterior positioning of the maxilla was the only within-twin difference which achieved statistical significance in the twins discordant for BCLP, partially confirming the findings of Narula and Ross (1970), who, in a study of 47 complete BCLP children from six to 16 years of age, found the lateral maxillary segments to be both posteriorly and superiorly positioned. Although Figure 3C shows many other differences, they were not statistically significant because of the small number in this group (6) and because four pairs were dizygotic with large variances. Possibly, a small difference in the conformation and size of cranial base between the dizygotic twins could have magnified most of the observed differences. Note that compared with their non-cleft twins, mandibular plane angle tended to be large for the cleft twins as did gonial angle.

Cleft palate (Figure 3D and Table 3). The larger cranial base angle in the cleft twins with isolated CP has also been reported by Dahl (1970) but was not supported by Ross (1965). He found only slight angular changes in the cranial base of children with isolated

CP. Dahl (1970), Ross and Coupe (1965), and Shibasaki and Ross (1969) all indicated that the cleft maxilla is smaller than normal, is retruded, and has a backward inclination relative to the cranial base in isolated CP, as was observed here.

Our findings of larger mandibular plane angles and more obtuse gonial angles agree with those reported by Dahl (1970) and Shibasaki and Ross (1969) as characteristic of isolated CP. The relatively large deficits in maxillary length and mandibular corpus length found in the cleft twins with isolated CP were not evident in cleft twins of the CL(P) groups. Ross and Coupe (1964), who made similar observations, suggested that there was an inherent developmental fault in CP only. The larger cranial base angles and the statistically different mandibular morphology evident only in the isolated CP groups further support this hypothesis.

EFFECT OF AGE, ZYGOSITY, AND CLEFT TYPE. *Age.* Larger within-twin deficits in the cleft twins were demonstrated with increasing age. The within-twin difference for Ar-GoI increased with age. After the age of eight, the cleft twins compared with their non-cleft twins exhibited mean deficits in ramus height. Similarly, there was a trend for the within-pair difference in maxillary length (PTM-A) to increase with age (significant at $P < .06$). Similar findings were reported by Hayashi et al. (1976) in a cross-sectional study of 255 UCLP patients compared with 240 control children.

The timing of the within-twin deficit for ramus height in the isolated CP group appeared to be the converse of the pattern evident in the CL(P) group. These findings are in agreement with Shibasaki and Ross (1969), who found that females with isolated CP had less mandibular length than their control sample from nine to 12 years of age and that they continued to have mandibular growth from 12 to 15 years of age, thus reducing the discrepancy between the cleft and the control children.

The increase with age of the within-pair difference for gonial angle and mandibular plane angle may be due to the reduction of these angles with age in the non-affected twins as was suggested by Hayashi et al. (1976).

The dimensions from the distal occlusal contact of the molar to S-FMN plane and palatal plane were greater in the younger cleft twins than in their non-cleft twins. This suggests that the maxillary molars were over-erupted in younger cleft twins. The maxillary molars also appeared to be more posterior on the average in the affected twins in every cleft group. See Figure 3.

The over-eruption of the molars in the cleft twins may be associated with oral respiration and the resultant tongue and mandibular postures. Harvold et al. (1972) concluded that oral respiration can result in over-eruption of the posterior teeth. In addition, Linder-Aronson (1970) has suggested that mouth breathing may result in increased lower face height and thus in increased eruption of the posterior teeth.

The reduction of the within-twin difference for palatal plane to the occlusal point of the maxillary molars in the older twins suggests that continued over-eruption of the maxillary molars was inhibited, perhaps by orthodontic treatment. Another possible explanation is that there could have been a cessation of oral respiration around the age of eight. Warren (1979) has reported that resistance to nasal airflow decreases with age in children with clefts. Thus, the ability to sustain nasal respiration may permit an alteration in neuromuscular function and the cessation of the continued eruption in the non-cleft twins, the within-twin discrepancy decreases.

It is interesting that the larger within-twin deficit for ramus height occurred at the same time that the cleft twins exhibited a smaller mean vertical dimension from the occlusal point of the molars to S-FMN plane relative to their non-cleft twins.

Zygosity. The monozygotic twins exhibited a significantly greater clockwise rotation of the ramus in the cleft twins relative to their non-cleft twins than was found in the dizygotic twin comparisons. The smaller mean within-twin difference for gonial angle in the monozygotic twins as compared to the dizygotic twins suggests that a dissimilarity in inherited mandibular form in the dizygotic twins may account for the zygosity difference. The remodeling changes secondary to mandibular posture that alter gonial angle (Chierici et al., 1973) may have contributed to the lack of statistical significance for the intra-twin difference for gonial angle between the monozygotic and dizygotic twins.

The monozygotic twins in each of the cleft groups were compared visually with superimposed figures (see Figure 4). The differences noted previously for each group were about the same for CL, were amplified in the monozygotic twin comparison for UCLP and BCLP, but were decreased in the monozygotic twins with isolated CP.

Cleft type. The effect of cleft type on the within-twin differences shows a consistent pattern. The mean within-twin deficits for Ar-GoI (a partial measure of ramus height) increased with the increasing degree of clefting from CL/A to UCLP to BCLP. Similarly, S-PTM also demonstrated a progressive increase in the mean within-twin deficits with increasing degree of clefting. The within-twin deficit for anterior cranial base length (S-Na) showed a similar trend of increasing within-twin deficits with increasing degree of clefting although it was not statistically significant. While sella is not the only registration point which could be used for such comparisons, different registrations would alter the location but not the extent of the deficits observed or the sequence of severity seen.

These findings of an increasing degree of dysmorphism with increasing severity of the cleft type for CL(P) could be the result of an increasing genetic component with increasing degree of clefting. That is, the proportion of dizygotic twins in each cleft group increased from CL to UCLP to BCLP reflecting an increasing genetic dissimilarity. On the other hand, the fact that increasing deficits go along with increasing severity of clefting is logical and perhaps does not require further study.

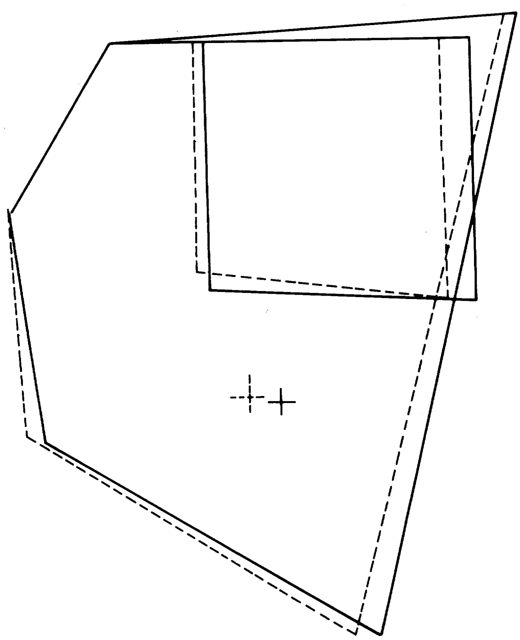

Figure 4. Composite figure for monozygotic twins discordant for UCLP (n=7)

Conclusions

1. Twins discordant for cleft lip or cleft of the lip and alveolus show very little craniofacial difference other than an apparent rotation of the mandible in the cleft twins.
2. Unilateral cleft lip and palate are characterized by decreased face perimeter and smaller posterior maxillary and ramus height.
3. The maxilla in BCLP is more posteriorly positioned than in non-cleft co-twins.
4. Isolated cleft palate is characterized by larger cranial base angles, smaller, more posteriorly positioned maxillae with the palate rotated up distally, smaller mandibular corpus lengths, and larger gonial and mandibular plane angles.
5. There is a trend toward a greater amount of dysmorphism with increasing severity of clefting from isolated cleft lip to complete bilateral cleft lip and palate.
6. In the sample studied, cleft lip and palate were characterized by a reduction in mean ramus height only after the age of eight in both sexes, and the over-eruption of the maxillary molars was not found in the older twins.
7. The change in mandibular structure associated with clefting was at least partly genetically induced.

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