Facial Growth of Children with Cleft Lip and/or Palate

SHINJI NAKAMURA, D.D.S., D.D.Sc.
BHIM S. SAVARA, D.M.D., M.S.
DONALD R. THOMAS, B.S.
Portland, Oregon 97201

Introduction

A review of the literature pertaining to the growth of cleft lip and/or palate children and the effects of surgical repair on subsequent growth reveals that controversies exist in this field. Some authors believe that the effects of cleft lip and/or palate are confined to proximal tissues while others believe that such effects can also be observed in more distant structures such as the cranial base. The dimensions of adjacent structures (the mandible and the maxilla) have been reported in a contradictory manner by different workers. Confusing evidence has been presented regarding the effects of surgery, with some authors going so far as to find all differences, if any, of size and growth to be a result of surgery, with unoperated subjects not differing significantly from normal. Considering the need for a reliable account of the effects of clefts of lip and palate on the growth of surrounding or distant structures and the actual effect of surgical intervention on the subsequent growth, it becomes necessary to review the past work to find out why such contradictions exist in cleft lip and palate literature.

The objective of this study is to review the literature and identify the problems associated with cleft lip and palate growth studies and then to test the resolutions to these problems as far as possible with the sample available to us.

Review of Literature

Graber (*1*), studying 45 cleft palate cases, arrived at the conclusion that maxillary growth in cleft palate and cleft lip subjects is deficient in all dimensions while mandibular growth appears normal. Using subsamples of 8 unoperated cases and 14 complete cleft lip and palate cases which had been surgically closed, he also found that the unoperated cases were normal in lateral and vertical dimensions. His sample ranged in age from 7 months to 58 years, with no distinction made as to type of cleft or operation (except in the subsample for comparing operated and unoper-

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ated subjects). No mention was made of sex so presumably the samples contained both males and females. Facial angles and proportional measurements were used to compensate somewhat for the diversity of sizes in the sample.

In a later study, Graber (*2*) reached substantially the same conclusions using a sample of 175 cleft lip and/or palate subjects. This sample was also nonhomogeneous with regard to age, sex, race, type of cleft and surgery, if any.

Jolleys (*3*) confirmed a reduction of maxillary development in operated cleft palate cases in a cross sectional study of 254 subjects, 165 of which were studied in detail. This sample was also nonhomogeneous in all respects except for presence of a cleft.

Harvold (*4*) studied a nonhomogeneous sample of 67 children for asymmetry of the facial skeleton. No mention of surgery was made. He found that the positions of the zygomatic bones were normal, the nasal septum had an abnormal form and the premaxilla was always out of its normal position, and the deformity of the facial skeleton is localized mainly in the alveolar and palatal processes of the lateral parts of the maxillae. From this he concludes that the major part of the deformities seen in these cases cannot be due to reduced growth potentials, although it is not clear how he arrives at this conclusion.

Brader (*5*) studied 23 males and 16 females with varying degrees of cleft palate at ages ranging from 4 to 25 years and found a superior position of the posterior margin of the hard palate in the region of the pterygo-maxillary fissure, a greater quantity of adenoid tissue and a smaller oro-nasopharyngeal area than normal. Whether the sexes were compared separately or together is not made clear nor is the number of cases (if any) with surgery ever stated.

Ortiz-Monasterio, *et al.* (*6*) studied 19 adult cleft palate subjects, 15 of which were unoperated. This sample was also mixed with regard to age, sex and degree of deformity. They found the forward growth of the upper maxillae to be the same or greater than normal, the Frankfort-mandibular angle greater than normal, and the average facial angle comparable to normal. They state that the maxillary protrusion can be explained by the lack of retention by the continuity of the lip, and blame surgery for growth defects in the middle third of the face of operated cases.

Subtelny (*7*) studied the width of the nasopharynx and related structures in 91 unoperated cleft lip and/or palate children less than three years of age. His sample was divided into four groups according to type of cleft (posterior cleft palate only, unilateral cleft lip and palate, bilateral cleft lip and palate, and cleft lip and alveolus), and where possible was divided into three age groups. Comparison was made with 51 normal children, also grouped according to age. It was found that skeletal nasopharynx width of cleft palate children was significantly larger than normal. Maxillary width measurements were all larger than normal, bizygo-

matic width measurement was not significantly different from normal, and all measurements had a high range of variaton.

Coupe and Subtelny (*8*) studied 127 cleft lip and/or palate children less than three years of age. These children were also grouped into three age groups and into four groups according to type of cleft. The article indicates that at least some of the children had surgically repaired cleft lips, but otherwise no indication of surgery, if any, is made. Their results show that clefts involving the palate had a deficiency of hard palate tissue and a strong indication of lateral displacement of the maxillary bones. The bilateral cleft palate children had a greater than normal ZMS width between ages 1 and 3.

Mestre, DeJesus, and Subtelny (*9*) examined facial angles and proportional measurements of 49 adult cleft palate subjects. These were unoperated cleft palate cases, but 21 of them had lip repair. Their results showed that the mandible in these subjects was normal in position relative to the cranium and proportional size. Anterior and posterior maxillary heights, location of ANS and position of the maxillae were also comparable to normal. Anterior-posterior dimension of the maxilla was short in unilateral complete cleft palate subjects but not significantly different from normal. The age range of the sample varied from 15 to 57 years and no division was made according to sex.

Borden (*10*) did a longitudinal study of mandibular growth in cleft palate children. His sample consisted of 27 male infants with varying types of clefts and amount of surgery. Cephalograms were taken at nine intervals from age 15 days to 3 years. He decided that the mandible of cleft palate children tended to be smaller than normal but that the growth rate was the same as normal. No difference was found in size of the anterior cranial base.

Deuschle and Kalter (*11*) also studied mandibular size of cleft palate subjects. Their sample of 23 persons of varying ages, sex, ethnic origin and type of deformity (but all with surgically repaired clefts), were found to have a significantly smaller mandible in anterior-posterior and vertical dimensions.

Moss (*12*) and Ross (*13*) both studied the cranial base of cleft palate subjects and reached contradictory conclusions. Moss found that cleft subjects showed an increased flexure of the cranial base while Ross found no such increased flexure. Comparison of their data though reveals that their cleft groups differ very little while their control groups differ significantly. Moss' sample was grouped according to type of cleft, but no mention was made of surgery, sex, or age (other than a range from 2 to 47 years). Ross used all post-operative subjects, grouped according to type of deformity, age and sex.

Blain (*14*) studied a sample of 443 cleft palate subjects, grouped according to extent of cleft, age, sex and surgical history. Proportional and angular measurements were used. Most subjects with clefts were found to

be deficient in vertical maxillary dimensions, whether or not operated on. Vertical growth did occur, with midline deficiencies tending to be corrected with age. A slight opening of the cranial base angle was seen, and the entire maxillary complex of subjects with clefts appeared to be retroplaced in relation to the anterior cranial base.

Osborne (*15*) studied 25 cleft palate subjects longitudinally during adolescence. The sample was divided according to sex and degree of deformity (all had clefts of the hard and soft palate), but no mention was made of the surgical history of the sample. Essentially, his results were that the face of cleft palate individuals becomes more concave with age.

Aduss (*16*) used a sample of 71 patients with complete unilateral cleft lip and palate for a longitudinal study of facial growth from 4 to 14 years. Primary surgical repair of the lip and palate had been performed on all cases. In general he found that the craniofacial growth of the cleft sample was the same as for the non-cleft populations. The principal differences found were a larger gonial angle and an elevated anterior cranial fossa in the cleft group. While the differences between this and earlier investigations can be attributed mostly to improvements in surgical techniques and skills, the design of this study and the homogeneity of the sample used makes this a more reliable study than many of the earlier studies.

The most obvious point after reviewing the above research is the nonhomogeneous character of most of the samples used. Some of the investigators did attempt to increase sample homogeneity by grouping according to age, sex, and/or type of cleft, but others did not even indicate whether their sample was operated upon or not. Most studies were done using proportional and angular measurements to compensate for differences in size due to age or sex, but shape as well as size may vary with age, sex or degree of deformity. So even using proportional and angular measurements, a nonhomogeneous sample can distort means and variances enough to make comparisons very unreliable.

Material and Methods

The material for this study consisted of serial lateral, frontal and lateral open-mouth cephalograms of cleft lip and/or palate individuals who had been examined at the Special Services Clinic of the University of Oregon Dental School. The sample was composed of 45 boys and 40 girls. All were of predominantly Northwest European ancestry and had been subjected to an initial surgical operation before 3 years of age. Of these children, only two boys and two girls had received orthodontic treatment during the age range studied. One boy with cleft lip was treated with minor tooth movement. The other three orthodontic patients, with clefts involving both the lip and palate, were treated with maxillary expansion devices. (Elimination of these patients makes no significant changes in the results, however.)

The sample was divided into 3 groups according to the extent of deformity:

Group I—clefts of lip (and alveolus),
Group II—isolated clefts of the palate,
Group III—clefts involving both the lip and palate.

Males and females were considered separately. The sample distribution for each group is shown in Figure 1. As control groups, the norms of normal children established by Savara, *et al.* were used (*17, 18, 19, 20*).

Distances between landmarks were calculated by first measuring three coordinates from the lateral and frontal cephalograms, and then, using a computer, correcting the coordinates of each landmark for magnification and distortion, and calculating the three dimensional distances between appropriate landmarks (*21, 22*). A second degree polynominal interpolation was utilized to equally space the data and interpolate for any missing observations.

The cephalometric measurements on the mandible were the following:

ramus height from condylion to gonion,
body length from gonion to pogonion,
maximum length from condylion to pogonion,
bigonial width from right to left gonion, and
bicondylar width from right to left condylion.

The cephalometric measurements on the maxilla were the following:

left to right pterygomaxillary fissure, and
left to right zygomaticomaxillary suture.

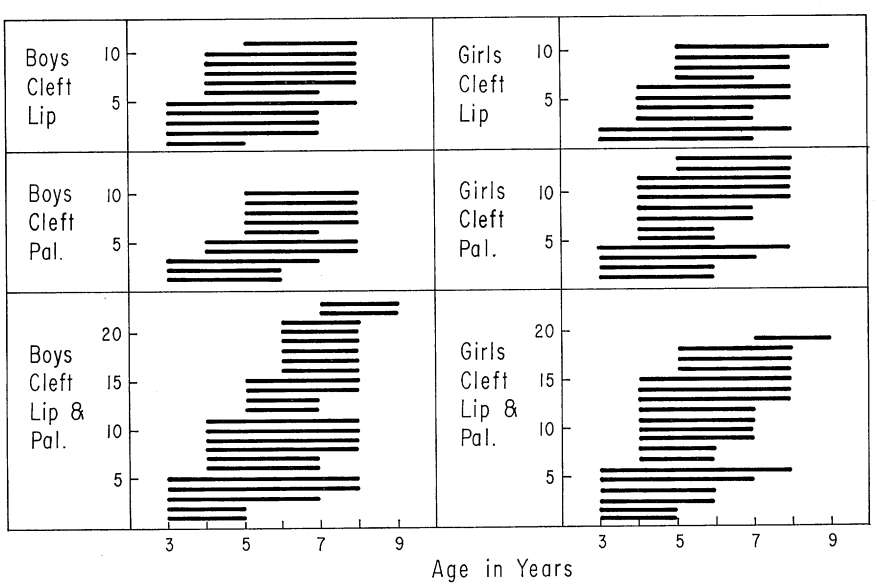

FIGURE 1. Sample sizes and distributions of children with cleft lip and/or palate.

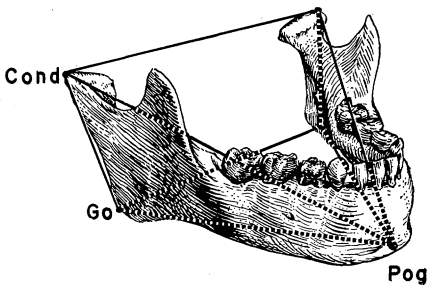

FIGURE 2. Mandible with landmarks and dimensions measured.

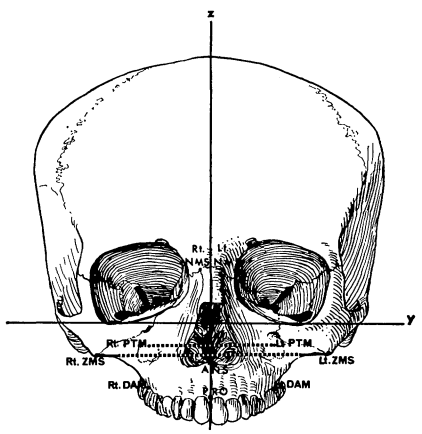

FIGURE 3. Maxilla with landmarks and dimensions measured.

These landmarks and distances are illustrated in Figures 2 and 3. Means and standard deviations for each measurement at each age were calculated for all the groups. "t" tests were done between each cleft lip and/or palate group and the normal group for each measurement at each age.

Findings

Mean sizes and standard deviations for each group at ages 4 through 8 are shown in Tables 1 and 2. Means and standard deviations of annual increments for normal boys and girls and boys and girls with cleft lip and/or palate are shown in Table 3.

Mean sizes of boys combined cleft groups and girls combined cleft groups are plotted with means and standard deviations of normal groups in Figures 4, 5 and 6.

Comparison of the slopes of the means plotted in Figures 4, 5 and 6 show that growth rates of the seven measurements for cleft lip and/or palate groups are similar to growth rates of normal children.

Significant "t" tests of the individual measurements for each group and each age are shown in Table 4. Boys with cleft lip show a significantly larger maxillary width measurement (L-R ZMS). Other measurements of

TABLE 1. Means and standard deviations for five mandibular and two maxillary measurements of normal boys and boys with clefts.

Group	Age (Yrs)	Lt - Rt Con.		Lt - Rt Go.		Con. - Go.		Go. - Pog.		Con. - Pog.		Lt - Rt P.T.M.		Lt - Rt Z.M.S.	
		Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.
Normal Boys	4	8.97	0.52	7.22	0.39	4.30	0.22	6.47	0.26	9.58	0.32	3.96	0.25	7.77	0.39
	5	9.30	0.47	7.43	0.40	4.45	0.27	6.73	0.28	9.92	0.29	4.07	0.24	7.92	0.38
	6	9.53	0.50	7.74	0.47	4.58	0.25	7.03	0.34	10.26	0.33	4.29	0.27	8.10	0.42
	7	9.68	0.47	7.92	0.49	4.76	0.27	7.22	0.34	10.51	0.34	4.47	0.33	8.20	0.44
	8	9.84	0.53	8.12	0.50	4.89	0.29	7.42	0.37	10.78	0.37	4.49	0.30	8.28	0.43
Cleft Lip	5	9.15	0.18	7.56	0.26	4.36	0.21	6.74	0.21	9.87	0.30	4.17	0.16	8.44	0.30
	6	9.44	0.23	7.72	0.24	4.53	0.28	7.06	0.29	10.27	0.32	4.26	0.18	8.62	0.26
	7	9.62	0.27	7.93	0.29	4.72	0.28	7.30	0.28	10.60	0.38	4.39	0.16	8.72	0.23
	8	9.85	0.10	8.12	0.20	4.75	0.22	7.44	0.23	10.75	0.28	4.49	0.19	8.80	0.22
Cleft Palate	5	8.99	0.43	7.58	0.53	4.16	0.31	6.45	0.30	9.58	0.42	3.93	0.29	8.05	0.31
	6	9.17	0.42	7.76	0.54	4.33	0.38	6.67	0.32	9.90	0.54	4.03	0.29	8.18	0.34
	7	9.39	0.40	8.02	0.59	4.53	0.47	7.00	0.33	10.35	0.65	4.14	0.30	8.35	0.36
	8	9.57	0.37	8.19	0.64	4.66	0.50	7.21	0.31	10.64	0.60	4.20	0.35	8.45	0.37
Cleft Lip & Palate	5	9.38	0.30	7.43	0.55	4.32	0.18	6.53	0.29	9.80	0.32	4.13	0.28	8.29	0.31
	6	9.52	0.65	7.65	0.57	4.41	0.35	6.70	0.49	10.01	0.65	4.23	0.44	8.42	0.37
	7	9.65	0.67	7.89	0.57	4.53	0.38	6.89	0.46	10.25	0.61	4.34	0.45	8.59	0.38
	8	9.69	0.76	8.09	0.59	4.53	0.42	7.01	0.48	10.33	0.63	4.42	0.49	8.68	0.50

TABLE 2. Means and standard deviations for five mandibular and two maxillary measurements of normal girls and girls with clefts.

Group	Age (Yrs)	Lt - Rt Con.		Lt - Rt Go.		Con. - Go.		Go. - Pog.		Con. - Pog.		Lt - Rt P.T.M.		Lt - Rt Z.M.S.	
		Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.	Mean	S.D.
Normal Girls	4	8.80	0.44	7.20	0.33	4.09	0.22	6.42	0.23	9.36	0.31	3.86	0.27	7.73	0.34
	5	9.00	0.46	7.43	0.36	4.25	0.22	6.67	0.23	9.67	0.28	4.03	0.24	7.91	0.37
	6	9.28	0.46	7.66	0.36	4.42	0.22	6.90	0.23	9.99	0.28	4.18	0.24	8.04	0.37
	7	9.40	0.48	7.86	0.34	4.53	0.24	7.14	0.26	10.28	0.32	4.26	0.24	8.15	0.36
	8	9.57	0.46	8.04	0.36	4.65	0.26	7.33	0.26	10.50	0.34	4.32	0.26	8.24	0.36
Cleft Lip	5	8.98	0.42	7.37	0.44	4.17	0.26	6.45	0.35	9.45	0.46	4.20	0.26	8.21	0.52
	6	8.95	0.32	7.37	0.48	4.29	0.32	6.55	0.44	9.54	0.56	4.28	0.37	8.26	0.75
	7	9.18	0.38	7.72	0.50	4.37	0.35	6.82	0.35	9.83	0.49	4.39	0.36	8.52	0.71
	8	9.23	0.12	7.87	0.46	4.58	0.38	7.01	0.18	10.27	0.30	4.33	0.37	8.59	0.95
Cleft Palate	5	8.94	0.33	7.37	0.46	4.12	0.25	6.24	0.18	9.31	0.34	3.92	0.26	8.03	0.43
	6	9.12	0.33	7.60	0.52	4.25	0.29	6.44	0.16	9.60	0.29	4.07	0.28	8.15	0.40
	7	9.30	0.32	7.66	0.39	4.33	0.36	6.63	0.16	9.85	0.30	4.14	0.31	8.29	0.38
	8	9.43	0.32	7.96	0.50	4.35	0.30	6.91	0.18	10.14	0.33	4.13	0.38	8.31	0.32
Cleft Lip & Palate	5	9.16	0.44	7.59	0.59	4.41	0.61	6.45	0.33	9.48	0.71	4.09	0.30	8.32	0.40
	6	9.32	0.42	7.64	0.36	4.56	0.33	6.66	0.30	9.97	0.33	4.20	0.26	8.41	0.31
	7	9.50	0.46	7.98	0.31	4.67	0.31	6.93	0.34	10.32	0.36	4.28	0.22	8.51	0.21
	8	9.71	0.50	8.06	0.28	4.69	0.26	6.96	0.34	10.38	0.41	4.37	0.22	8.58	0.26

the boys in this group do not differ significantly from normal boys. The girls in this group show larger maxillary width and smaller mandibular length measurements at the only age tested, but small sample sizes make this an unreliable result.

Children with isolated cleft palate show significantly smaller mandibular length measurements, especially for GO-POG. The boys cleft palate group also show a significantly smaller L-R PTM measurement at some ages, but otherwise all measurements for both sexes do not differ from the normal groups.

Children with cleft involving both the lip and palate show a pattern of

TABLE 3. Means and standard deviations of annual increments for normal boys and girls, and boys and girls with clefts.

Group	Age Interval	LT-RT CON Mean-S.D.	LT-RT GO Mean-S.D.	CON -GO Mean-S.D.	GO- POG Mean-S.D.	CON- POG Mean-S.D.	LT-RT PTM Mean-S.D.	LT-RT ZMS Mean-S.D.
Normal Boys	4-5	0.30 0.14	0.23 0.07	0.19 0.10	0.25 0.09	0.35 0.15	0.16 0.08	0.13 0.07
	5-6	0.21 0.11	0.25 0.09	0.16 0.07	0.28 0.13	0.33 0.15	0.16 0.09	0.14 0.09
	6-7	0.16 0.09	0.18 0.07	0.17 0.08	0.20 0.06	0.26 0.09	0.12 0.07	0.10 0.09
	7-8	0.19 0.12	0.20 0.07	0.15 0.09	0.22 0.01	0.28 0.12	0.09 0.06	0.11 0.06
Boys with Clefts	4-5	0.23 0.11	0.24 0.14	0.16 0.14	0.27 0.14	0.35 0.21	0.16 0.15	0.14 0.07
	5-6	0.17 0.07	0.22 0.08	0.15 0.11	0.22 0.10	0.29 0.13	0.09 0.20	0.04 0.50
	6-7	0.21 0.14	0.21 0.09	0.13 0.12	0.25 0.10	0.33 0.16	0.10 0.09	0.10 0.14
	7-8	0.11 0.09	0.18 0.09	0.11 0.09	0.18 0.08	0.20 0.11	0.03 0.07	0.06 0.10
Normal Girls	4-5	0.24 0.09	0.25 0.09	0.15 0.07	0.26 0.09	0.33 0.10	0.15 0.08	0.16 0.06
	5-6	0.22 0.08	0.25 0.07	0.16 0.07	0.25 0.08	0.32 0.14	0.16 0.08	0.15 0.07
	6-7	0.14 0.07	0.20 0.07	0.11 0.08	0.24 0.10	0.27 0.11	0.10 0.06	0.12 0.05
	7-8	0.19 0.10	0.19 0.07	0.12 0.09	0.21 0.09	0.26 0.09	0.07 0.06	0.11 0.05
Girls with Clefts	4-5	0.27 0.14	0.23 0.10	0.19 0.15	0.26 0.11	0.35 0.13	0.09 0.09	0.11 0.09
	5-6	0.24 0.12	0.20 0.09	0.14 0.12	0.26 0.09	0.32 0.16	0.11 0.10	0.12 0.08
	6-7	0.18 0.12	0.21 0.11	0.18 0.19	0.22 0.12	0.29 0.15	0.13 0.09	0.12 0.11
	7-8	0.16 0.08	0.19 0.08	0.11 0.09	0.20 0.11	0.24 0.19	0.09 0.08	0.05 0.10

a large maxillary width measurement, L-R ZMS, and smaller mandibular length measurements.

Discussion

Our results show that the bizygomaticomaxillary suture width of children with cleft lip is significantly larger than normal, mandibular length measurements of children with isolated cleft palate are significantly smaller than normal, and children with both cleft lip and palate have larger maxillary width measurement L-R ZMS and smaller mandibular length measurements than normal. Several observations should be made concerning these results and the methods used in arriving at them.

From Tables 1 and 2 it can be seen that the standard deviations are generally as large or larger than the differences in means between cleft and normal groups. This simply means that the distributions of the cleft and normal groups overlap to a great extent and that any individual measurement could be a member of either population.

With regard to Table 4, it must be emphasized that a large number of "t" tests were done and the sample sizes for some groups at some ages were small, so not every "t" test can be expected to show the true difference, if any, between the cleft and non-cleft groups. The important thing to observe is the overall pattern for both sexes. This pattern is less likely to be influenced by outlying observations or irregularities in sampling than is any individual "t" test.

Another fact which is apparent from our results is the necessity of using homogeneous groups with respect to sex, age and type of cleft. The differences found were relatively small and any combining of sexes, ages or type of clefts would tend to distort the means and variances, thus possibly obscuring the real differences.

A major point to remember concerning the cleft sample we used is the history of surgery in all cases. Our results, therefore, apply only to the

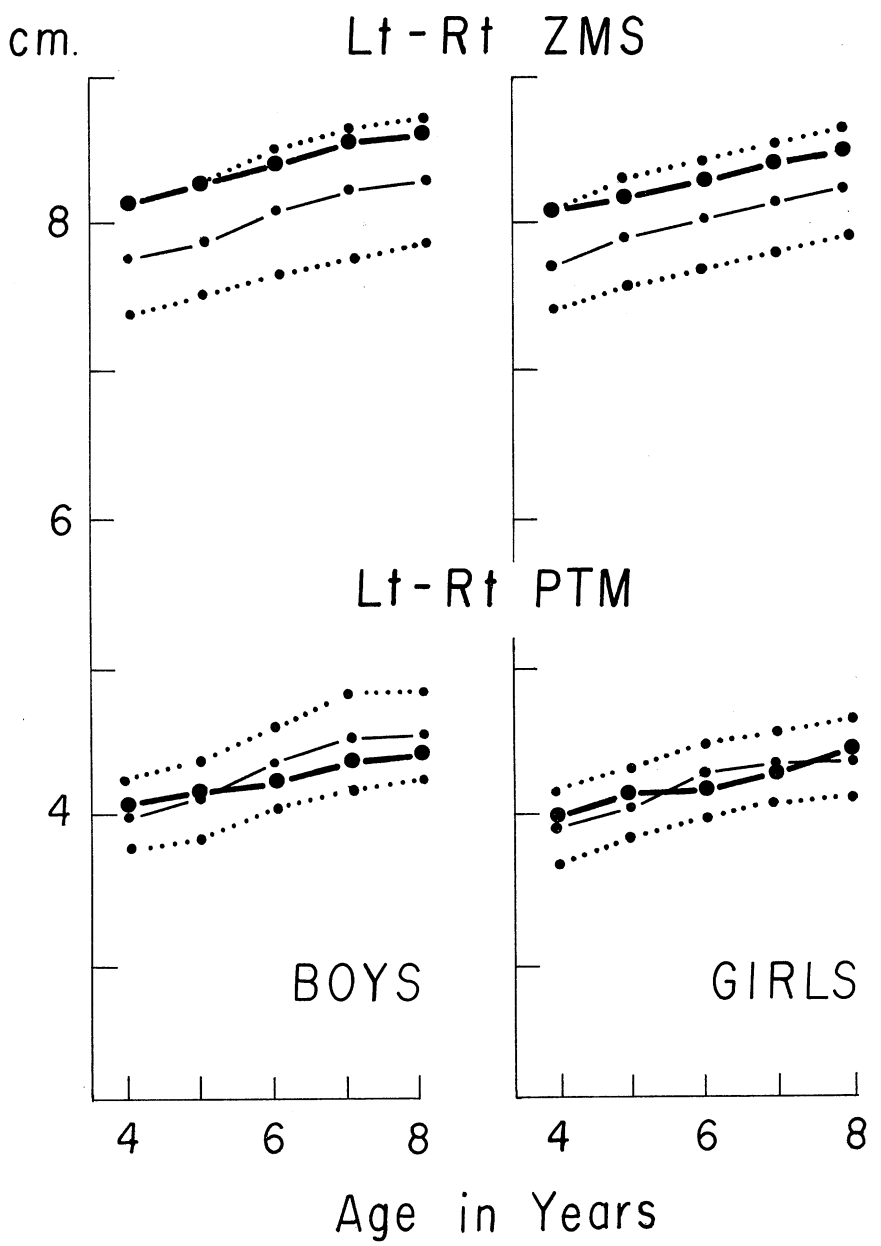

FIGURE 4. Mean sizes of two maxillary width measurements for total cleft groups (heavy lines) with means and ± one standard deviation of normal boys and girls.

population of children with clefts and some form of surgical repair. Two observations can be made though which might be suggestive about the mandible and maxillae of unoperated cleft lip and/or palate children.

First, if the evidence of retarded maxillary growth in postoperative cleft

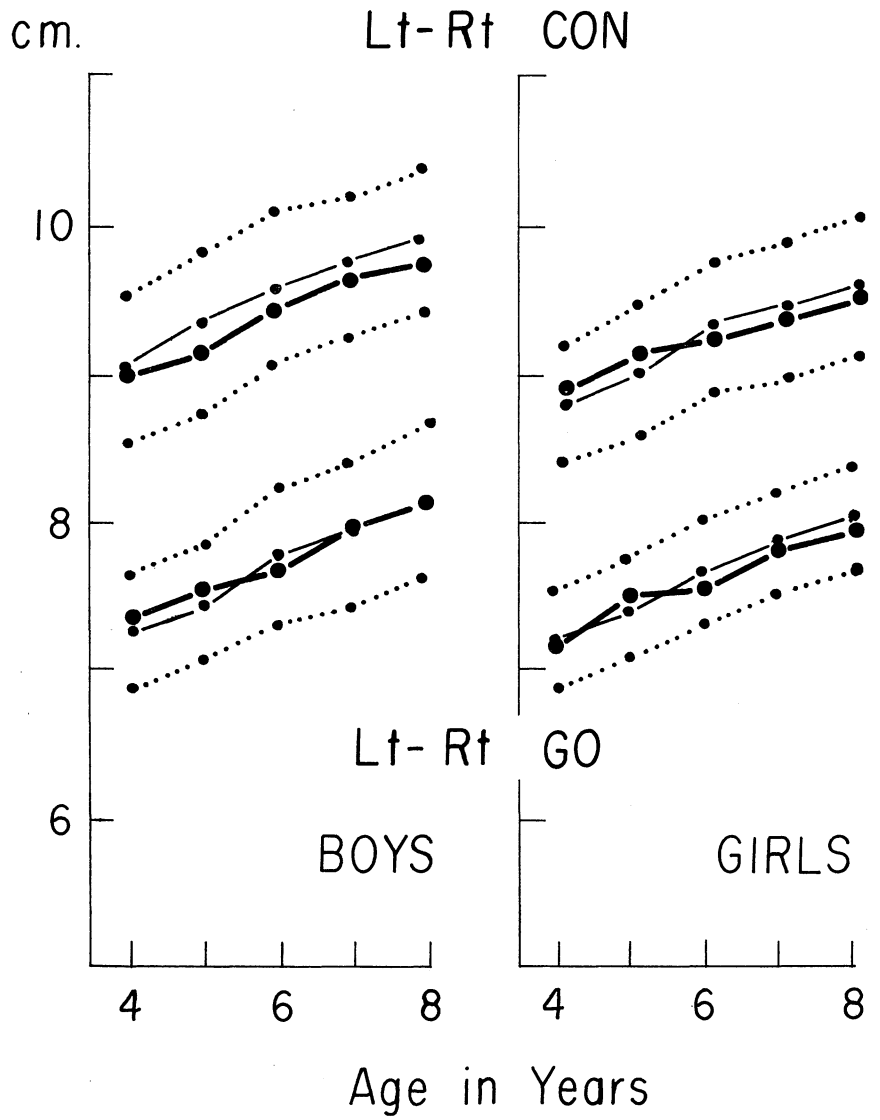

FIGURE 5. Mean sizes of two mandibular width measurements for total cleft groups (heavy lines) with means and ± one standard deviation of normal boys and girls.

lip and/or palate cases is accepted, then how can one explain the normal or larger than normal widths of the maxillae measured between points distant from the site of surgery? The simplest explanation would seem to be that cleft lip is associated with a maxillae normal in some dimensions but slightly wider than normal in other areas. Surgery might inhibit growth in the area immediately adjacent to the site of surgery, especially

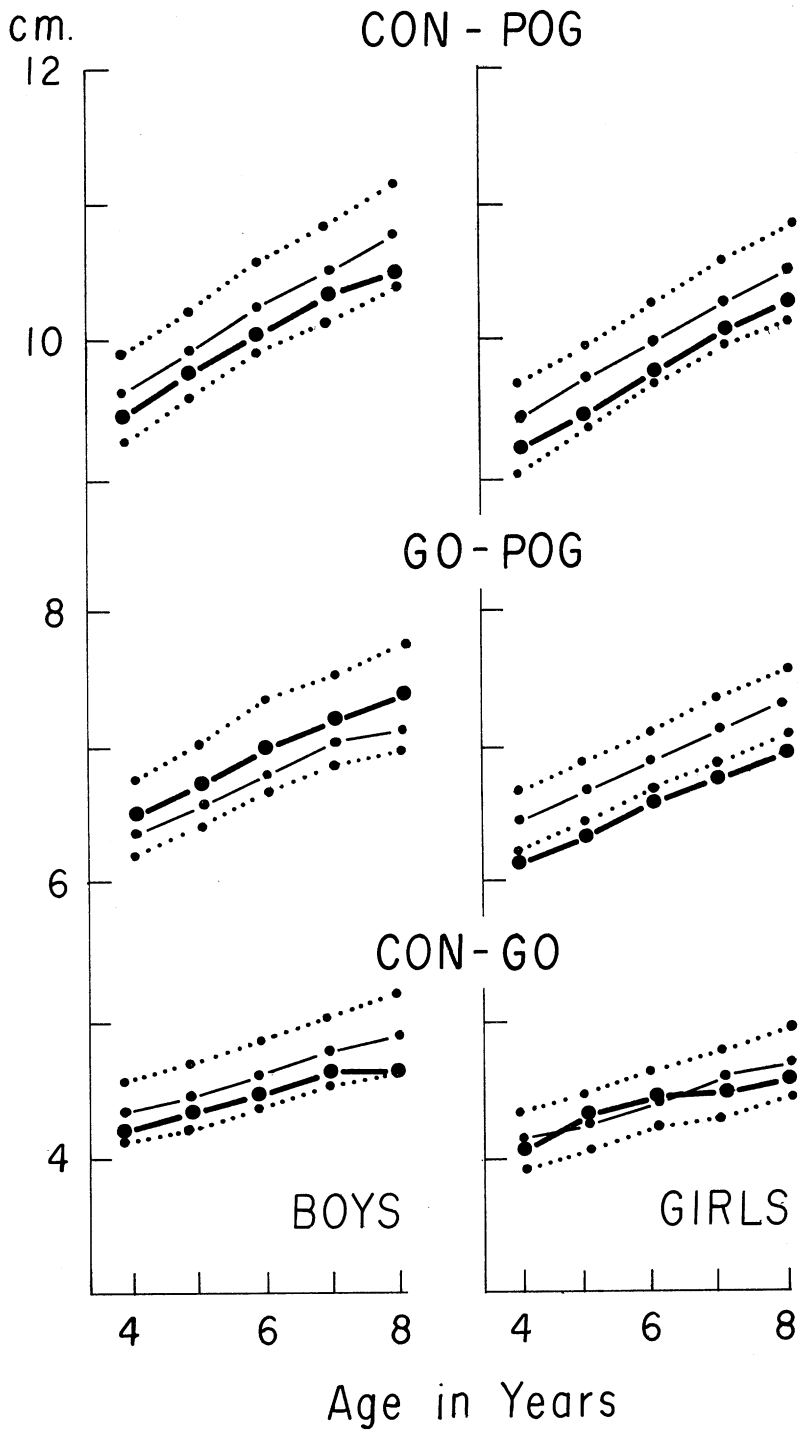

FIGURE 6. Mean sizes of three mandibular length measurements for total cleft groups (heavy lines) with means and ± one standard deviation of normal boys and girls.

TABLE 4. Dimensions of cleft lip and/or palate children which "t" tests show to be significantly different from measurements of normal children.

Group	Age (Years)	BOYS L-R CON	L-R GO	CON -GO	GO -POG	CON -POG	L-R PTM	L-R ZMS	GIRLS L-R CON	L-R GO	CON -GO	GO -POG	CON -POG	L-R PTM	L-R ZMS
Cleft Lip	5							++				-		-	+
	6							++							
	7							++				NO SAMPLE			
	8							++							
Cleft Palate	5	-		--	--	--						--	--		
	6	-		--	--	--	--				-	--	--		
	7			-	-		--				-	--	--		
	8			-			-				-	--	--		
Cleft Lip & Palate	5				-			++				--			++
	6			-	--	-		++				--			++
	7			--	--	-		++			++	-			++
	8			--	--	--		++				--			++

\+ ... Cleft group means significantly larger (.05 level) than normal means
++ ... Cleft group means significantly larger (.01 level) than normal means
\- ... Cleft group means significantly smaller (.05 level) than normal means
-- ... Cleft group means significantly smaller (.01 level) than normal means

anterior-posterior growth, but probably has much less effect on growth at more distant sites.

Secondly, while we found that a short mandible is associated with postoperative cleft palate cases, it is not found in the postoperative cleft lip cases. This might still be explained by the differences in surgical procedures but a question remains as to how the surgery, or a cleft palate for that matter, could be associated with the length of the mandible. The tongue provides a possible explanation. A small mandible could interfere with the descent of the tongue thus preventing full palatal closure, or a clefted palate could fail to provide the necessary purchase for the enlarging tongue to exert a fully effective stimulus for growth on the mandible.

The above conjectures are offered only as explanations which are consistent with our results and most previous work. They may be taken as hypotheses to be tested by future work.

Summary

Mixed longitudinal samples of 45 boys and 40 girls with clefts of the lip and/or palate were used in this study. These children were grouped according to the extent of their deformity, i.e., clefts of the lip (and alveolus), isolated clefts of the palate, and clefts involving both lip and palate. Five mandibular dimensions and two maxillary dimensions were measured using Savara's three-dimensional method and the results compared with the norms for normal children. The comparison showed that children with clefts of the lip have a significantly larger bizygomaticomaxillary suture width measurement than normal children. Children with clefts of the

palate have significantly smaller mandibular length measurements than normal children, while children with clefts of the lip and palate have both smaller mandibles and wider maxillae than normal. Growth rates of the children with clefts appear to be the same as for normal children however.

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