A Longitudinal Study of the Craniofacial Growth Pattern in Children with Clefts as Compared to Normal, Birth to Six Years*

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Foreword

In past years the statement has often been made that surgical intervention in cleft lip/palate cases would have a traumatic effect, i.e., a growthstultifying effect, upon the palato-labial complex, more specifically the palate and the total maxillo-palatine complex (3). It has been our feeling, here at Lancaster, that conservative surgery (properly timed, and offering a minimum of muco-periosteal involvement) should not result in deviant and/or dysplastic maxillo-facial growth.

Hence, we are here testing such an hypothesis, which may be framed somewhat as follows: Operative intervention in cleft palate cases which minimally involves bone-growth potential will guide and facilitate maxillofacial growth in the individual so that post-operative growth, in a *catch-up* manner, will provide for the achievement of an acceptably normal craniofacio-dental growth pattern.

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The Sample

We are reporting on serial lateral roentgenographic cephalometric data from the files of the Institute.* Represented are 59 cleft palate cases and 43 cleft lip and palate cases, analyzed cross-sectionally. In the eight serial visits about one-third of the children had (one) (two) visits missed, irregularly spaced over the B-6:0 period, but most frequently in the later years. Male and female data are pooled. Sample sizes by age and cleft type are as follows:

age (mos., yrs.)	cleft	type
	CP	CL(P)
0-3 mos.	22	30
3-6 mos.	25	30
1 yr.	27	21
2 yrs.	29	23
3 yrs.	27	26
4 yrs.	25	19
5 yrs.	24	17
6 yrs.	18	15

END-POINTS, DIMENSIONS AND ANGLES (Figures A and B). The following craniometric end-points were located on the lateral headfilm:

- 1. **Basion** (Ba:) most anterior point on the anterior contour of the foramen magnum.
- 2. Sella (S): mid-point (by inspection) of the shadow of the sella turcica.
- 3. Nasion (N): the most anterior point on the naso-frontal suture.
- 4. Anterior nasal spine (Ans): tip of the anterior nasal spine.
- 5. **Point A** (A, or subspinale): the deepest point of the concavity of the maxillary shadow below Ans.
- 6. Intradentale superius (Ids): the tip of the alveolar process between *il* | *il* or *Il* | *Il*.
- 7. Intradentale inferius (Idi): the tip of the alveolar process between $\overline{il | il}$ or $\overline{Il | Il}$.
- 8. Gnathion (Gn): the point on the anterior curvature of the chin where the shadow of the mandibular symphysis becomes confluent with the base.
- 9. Menton (Me): the lowest point on the X-ray film shadow of the mandibular symphysis.
- 10. **Gonion** (Go): point on the angle of the mandible determined by the bisection of the angle formed by the intersection of a line tangent to mandibular corporal lower border with a line tangent to the posterior border of the mandibular ascending ramus.
- 11. Articulare(Ar): point of intersection of the outline of the posterior margin of the ascending ramus with the shadow of the external (lower) contour of the cranial base.
- 12. Pterygomaxillary fissure (Ptm): the fissure or space between the anterior margin of the pterygoid process and the posterior margin of the maxillary tuberosity.
- 13. **Posterior nasal spine** (Pns): the tip of the most posterior extension of the bony palate. When in clefting the Pns is absent it can be approximated by extending the axis of Ptm so that it intersects the plane of the palate drawn in via Ans; the point so located is Ptm'.
- 14. **Key ridge** (KR): the lowest point of the zygo-maxillary abutment of the zygomatic arch complex.

* All surgical procedures were by Dr. Harding. Lip was repaired by a triangular flap at two-three months (10 lbs.); palate repair was by vomer flap plus median palatal suture at 14 months (12-16 months range).

Based upon these end-points the following dimensions have been taken:

Ba-S	N-Ans	Go-Gn	
S-N	N-Ids	Ar-Gn	
Ba-N	Idi-Gn	Ar-Go	Ptm'-Ans
			Ptm'-A
		S-Go	Ptm-KR
			KR-Ans

CEPHALOMETRIC DIMENSIONS



FIGURE A. Cephalometric dimensions.



CEPHALOMETRIC ANGLES



FIGURE B. Cephalometric angles.

FIGURE B

Based on these lines the following angles have been calculated:

Ba-S-N	S-N-A
S-N-Ba	Ar-Go-Gn
N-Ba-S	

THE ANTERIOR CRANIAL BASE IN CLEFTING. The anterior cranial base, extending from behind the mid facial complex to above it, is a very complex area. 1) Craniometrically it is bounded by *basion*, behind and below, *sella*, behind and above, and *nasion*, above and anteriorly. 2) Morphologically it involves the bodies of the occipital and sphenoid bones, cribriform plate of the ethmoid, and tabulae interna and externa of the frontal. 3) Functionally it is basically related to brain growth; to naso-respiratory growth (the anterior cranial base also serves as the roof of the nasal area); and to osteomyological structuring. 4) Further, the occipito-sphenoid synchondrosis is an important growth and adjustive locus.

In the discussion that follows references will be made to normal (noncleft) dimensional and angular values. These have been measured on the *Bolton Standards of Roentgenographic Cephalometry*.* We have measured the dimensions and angles pertinent to this study on *Standards* 1:0–6:0, with male and female combined.

In the cranial base the following *dimensions* have been observed:

- 1. Ba-S, which is clival length (bodies of occipital and sphenoid, with their synchondrosis)
- 2. S-N, floor of anterior cranial fossa (body of sphenoid, cribriform plate of ethmoid, frontal)
- 3. Ba-N, functionally a basi-cranial axis or even a craniofacial boundary line (cuts across an occipito-sphenoethmoid complex)

End-points Ba, S, and N establish a triangle, Ba-S-N, the angles of which will reflect the adjustment involved in differential growth rates/amounts in the three sides of the triangle. The following *angles* have been measured:

- 1. Ba-S-N, the sellar angle, reflecting a clival-anterior cranial base relationship
- 2. S-N-Ba, reflecting a cranial base-basic anial axis relationship

3. N-Ba-Sn, reflecting a basicranial axis-clival relationship

In our analysis of the serial data upon which this study is based we superimposed our lateral tracings upon the S-N base line, with S registered.

The cranial base dimensions (in cms.) are in Tables 1–3.*

In the 0:3-6:0 age period the following absolute dimensional increases have occurred (all +):

Dimension	CP	CL(P)
Ba-S	1.35 cm.	1.44 cm.
S-N	1.99 cm.	$2.05 \mathrm{cm}.$
Ba-N	2.72 cm.	2.89 cm.

* These Standards, which are contour modal craniofacial tracings in normae lateralis et facialis, were made available to us through the generous courtesy of B. Holly Broadbent, Sr., D.D.S., and Charles Bingham Bolton, and Case-Western Reserve University of Cleveland, Ohio.

* In all the tables in this study, significance is indicated by the following designations: * = significant at the .05 level, ** = significant at the .01 level, *** = significant at the .01 level, *** = significant at the .00 level, *** = significant at the

TABLE 1. Ba-S

	СР		t-i	t-test		CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	2.67	0.17	50	-0.137	30	2.68	0.24
3–6 Mos.	25	3.06	0.29	53	0.326	30	3.03	0.28
1 Year	27	3.34	0.25	46	0.498	21	3.30	0.28
2 Years	29	3.57	0.20	50	0.332	23	3.56	0.24
3 Years	27	3.71	0.24	51	0.196	26	3.69	0.30
4 Years	25	3.73	0.21	42	-1.489	19	3.84	0.28
5 Years	24	3.93	0.25	39	-1.164	17	4.03	0.29
6 Years	18	4.02	0.29	31	-0.865	15	4.12	0.36

TABLE 2. S-N

	СР		t-test		CL(P)			
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	4.73	0.37	50	-0.049	30	4.74	0.28
3-6 Mos.	25	5.41	0.28	53	-0.591	30	5.45	0.29
1 Year	27	5.97	0.35	46	0.373	21	5.93	0.36
2 Years	29	6.34	0.29	50	0.966	23	6.26	0.28
3 Years	27	6.35	0.27	51	0.187	26	6.33	0.37
4 Years	25	6.47	0.25	42	-0.813	19	6.53	0.26
5 Years	24	6.57	0.26	39	-0.887	17	6.64	0.28
6 Years	18	6.72	0.22	31	-0.743	15	6.79	0.29

TABLE 3. Ba-N

	CP		t-test		CL(P)			
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0-3 Mos.	22	6.87	0.45	50	-0.553	30	6.94	0.43
3–6 Mos.	25	7.79	0.46	53	-0.823	30	7.89	0.46
1 Year	27	8.49	0.47	46	-0.237	21	8.52	0.53
2 Years	29	8.97	0.33	50	0.182	23	8.96	0.37
3 Years	27	9.07	0.33	51	-0.303	26	9.11	0.50
4 Years	25	9.21	0.41	42	-1.781(*)	19	9.44	0.47
5 Years	24	9.43	0.45	39	-1.317	17	9.62	0.45
6 Years	18	9.59	0.39	31	-1.579	15	9.83	0.47



FIGURE 1. Cranial base lengths: normal, CP, CL(P).

For all practical purposes differences are not significant.

At this point it is advantageous to discuss the fact that cranial base complex involves cartilaginous synchondroses, while the facial complex involves intermembranous sutures. They both may be regarded as growth and/or adjustive sites, apart from the resorption-apposition interplay going on upon bone surfaces. Sarnat (10) emphasizes the differing growth roles of synchondroses and sutures. When operated and un-operated sides involving sutural resection were compared there were "revealed no significant growth differences." When septovomeral and mandibular condyle regions were resected there were "profound effects on facial growth." He concluded that "... areas of cartilaginous growth^{*}..., are primary and important growth sites, while areas of sutural growth are secondary or accommodating growth sites." Moss (β) is in substantial agreement: "The sutures are the sites of secondary, compensatory growth of the calvarial bones, following their positive translation as a response to the primary growth impetus of the enclosed neural matrix."

Over-all, as between CP and CL(P), Ba-S (clival length), S-N (anterior cranial base), and Ba-N (basi-cranial axis) have not shown any marked differences in size increase.

The dimensional changes in the cranial base in CP, CL(P) and Normal (*Bolton*) are shown in Figure 1:

^{*} Powell and Brodie (8) accept the spheno-occipital synchondrosis as a major primary growth site in about the first decade and one-half of postnatal cranial base growth.

65

- 1. Ba-S is equal in CL(P) and CP until 3:0; then it is a bit larger in CL(P). Both exceed the Normal.
- 2. S-N is much the same in CP and CL(P); in the 0:3–6:0 period both cleft types exceed Normal values in S-N, a bit more so in CL(P).
- 3. Ba-N shows a similar pattern for CP and CL(P) as did S-N, again, normal values tend to be shorter, though closer to CL(P).

Before attempting a more detailed interpretation of the dimensional data let us look at the angular data, presented (in degrees) in Tables 4–6.

The total absolute changes in these three angles in CP and CL(P) from 0:3-6:0 are as follows:

Angle	CP	CL(P)
Ba-S-N	-9.82°	-9.67°
S-N-Ba	$+4.03^{\circ}$	$+4.19^{\circ}$
N-Ba-S	$+5.78^{\circ}$	$+5.51^{\circ}$

CP		t-lest		CL(P)				
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	134.48	4.79	50	-1.880(*)	30	136.85	4.27
3–6 Mos.	25	132.06	4.43	53	-2.419*	30	135.18	5.02
1 Year	27	129.52	4.72	46	-2.396*	21	132.98	5.25
2 Years	29	127.64	4.37	50	-1.600	23	129.91	5.87
3 Years	27	127.16	5.03	51	-1.311	26	129.05	5.46
4 Years	25	127.30	4.49	42	1.701(*)	19	129.57	4.25
5 Years	24	126.32	4.71	39	0.615	17	127.25	4.77
6 Years	18	124.66	3.63	31	1.740(*)	15	127.16	4.60

TABLE 4. Angle Ba-S-N

TABLE 5. Angle S-N-Ba

	СР			<i>t-</i>	t-test		CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.	
0–3 Mos.	22	16.11	1.79	50	1.718(*)	30	15.29	1.62	
3–6 Mos.	25	16.91	1.88	53	2.387*	30	15.68	1.93	
1 Year	27	17.68	1.99	46	2.071*	21	16.46	2.05	
2 Years	29	18.39	1.85	50	1.140	23	17.73	2.38	
3 Years	27	18.99	2.16	51	1.077	26	18.33	2.28	
4 Years	25	18.76	1.72	42	0.936	19	18.24	1.91	
5 Years	24	19.57	1.90	39	0.225	17	19.43	2.07	
6 Years	18	20.14	1.64	31	0.982	15	19.48	2.20	

	СР			t-l	t-test		CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.	
0-3 Mos.	22	29.42	3.24	50	1.774(*)	30	27.86	3.04	
3–6 Mos.	25	31.03	3.02	53	2.166*	30	29.14	3.37	
1 Year	27	32.80	2.97	46	2.461*	21	30.56	3.32	
2 Years	29	33.96	2.77	50	1.801(*)	23	32.36	3.65	
3 Years	27	33.85	3.14	51	1.389	26	32.62	3.33	
4 Years	25	33.94	2.98	42	2.046*	19	32.18	2.60	
5 Years	24	34.11	2.94	39	0.827	17	33.33	3.03	
6 Years	18	35.20	2.20	31	2.168*	15	33.37	2.66	

TABLE 6. Angle N-Ba-S

Moderately significant differences, especially in the early years, suggest different rates of adjustive basilar growth.

In both cleft types Ba-S-N decreases about the same amount. Figure 2 graphs the age-changes in these angles.

- 1. In all three groups Ba-S-N, the sellar angle, decreases steadily and evenly from 0:3-6:0. In CP it is above the Normal until 3:0, then decreases evenly. In CL(P) it is increasingly below the Normal. As a whole the sellar angle is smallest in CL(P).
- 2. S-N-Ba shows no real difference in the three groups.
- 3. N-Ba-S is larger in CL(P); it is smaller in CP until 4:0, at which age it becomes larger.

Figure 2a is recapitulatory of Figures 1–2. Here the cranial base triangle is shown at 1:0, 3:0, and 6:0 in its entirety:

- 1. In the Normal growth is absolutely pattern-wise. Ba moves evenly down and back, N evenly forward. The Ba-N axis moves evenly down-forward
- 2. In the CP there is a reversal at Ba, which here moves evenly down and forward; in consequence the Ba-S-N angle decreases 3° between 1:0-3:0 and 3° between 3:0-6:0. The Ba-N axis moves evenly downward-forward
- 3. In CL(P) there is also a reversal, but less evenly so. Ba between 1:0-3:0, moves down and forward, and from 3:0-6:0 more downward than forward. The Total Ba-S-N angular change is still 6°, but now 4° from 1:0-3:0, and 2° from 3:0-6:0

DISCUSSION OF THE CRANIAL BASE. The preceding data point to several general conclusions: 1) *dimensionally*, compared to the Normal, clival length (Ba-S) and anterior cranial base length (S-N) are equally longer in the two cleft types, more in CL(P) than in CP; 2) in *angular* relations, compared to the Normal the sellar angle, Ba-S-N, shows greater flexion in clefting, again a bit more in CL(P) than in CP.

We feel that all this points to *basion* and the foramen magnum as the major area of adjustment to CP and/or CL(P) clefting. The reason may well reside in the following deductions:

- 1. CP is probably less strongly entrenched genetically than is CL(P), for CP may have an environmental (teratogenic) etiology.
- 2. Hence, Ba (may) (may not) be involved as a genetic factor in CP.

67



- 3. In CL(P) Ba is more apt to be genetically involved so that the entire foramen magnum (basiocciput)-palatal complex may be quasi-syndromic.
- 4. It follows, then, that angles S-Ba-N and S-N-Ba merely reciprocally relate to the basilar area of growth adjustment, i.e., the Ba-S-N or sellar angle.
- 5. The occipito-sphenoid synchondrosis is probably pivotal here, for it is involved in Ba-S and is the site of the greatest potential growth adjustment.
- 6. In angular age-changes Ba is focal in terms of its relation to S and to N.

The foregoing observations must be regarded as relative, rather than absolute, for methodologically we have fixed S (registration point) and have held the inclination of the S-N line constant (super-imposition). Even with these limiting circumstances the *trend* in dimension and angle separates CP and CL(P) from each other and both from the Normal. Our serial data strongly suggest that in CL(P) the entire anterior cranial base complex, Ba-S-N, is harder "hit," i.e., shows greater divergence from Normal pattern and values.

INTERDEPENDENT GROWTH IN THE CRANIOFACIAL MIDLINE COMPLEX. What has been said up to this point clearly suggests that there is a very close—and possibly reciprocal—relationship between the various components of the craniofacial midline. Specifically, it is very likely that palatal clefting may have repercussions in adjacent bony structures in *both* the cranial base and facial areas, the former involving occipital, sphenoid, and ethmoid bones, the latter the total midfacial complex.

In substance Coccaro and Pruzansky (1) are in agreement with the foregoing generalization when they state that "cleft lip and palate must be considered as an anomaly complex involving organs contiguous to the cleft." Ross (9), however, is not in principle in agreement with our view. He reported a smaller cranial base in cleft children, but felt that it was due to the smaller total size of these children, rather than a "reflection of an abnormality in the cranial base." Shibasaki and Ross (11) seem to suggest a degree of facial component interdependence in older children. In 30 children with isolated CP, age 6:0–15:0 years, they found "evidence of progressive maxillary underdevelopment, but with acceptable facial balance due to positional changes in the mandible."

The problem of *timing*, developmentally speaking, is basic. There is evidence that craniofacial growth dysplasia may be pre-clefting. Moss (5) states that malformations of the skull base in CP "may have a teratogenic period which precedes the onset of chondrification of the skull base," possibly at the beginning of the embryonic second lunar month. Malformation of the cranial base in CP is seen in that the neural skull as a whole is rotated back and down relative to the facial skeleton.

In the facial area Trasler (12) shows a close tie-up between embryonic morphogenesis and CL, which involves facial structural form as a whole. Moreover the relationship is genetically entrenched. Two strains of mice were studied before and during the genesis of CL: 1) A/J, prone to CL with a spontaneous frequency of 12%; 2) C57/BL/6J, which "virtually never has CL." In an A/J destined to have a CL it is early recognized by absence of the fusion normally occurring between the epithelium of the lateral and medial nasal processes, at the posterior end of the entrance to the nasal pit. The medial nasals have an increased prominence generally, and the posterior end of the nasal pit is wider on the side where the CL will occur. The lack of fusion and the deviant orientation of embryonic structures in A/J are the forerunners or predictors of CL.

Clefting is further related to growth-time in cranial vault growth. Moss (7) finds that premature closure of the frontal suture is three times more frequent in clefting than in non-clefting. The possibly wide-spread effect of clefting has been shown experimentally by Gardner and Kronman (2). In Rhesus monkeys the palate was split by an appliance; one week after maxi-

mum expansion of the appliance, the animals were sacrificed. The effect of the palatal splitting was registered in the sutures of the maxillary complex, the vault, and even the spheno-occipital synchondrosis.

There can be no doubt of the genetic, structural, and functional relationship within the total craniofacial complex in CL(P) cases. The cleft palate and/or cleft lip are not isolated defects. They ramify into skull base, skull vault, facial skeleton, on a time-linked basis (genetic) and upon a morphofunctional basis mainly involving sutural systems + remodelling.

Mazaheri *et al.* (4) report that, "Surgical closure of the lip had a significant effect upon reduction of the alveolar and palatal cleft and upon maxillary segmental relationships and positioning."

THE MAXILLARY (MIDFACIAL) COMPLEX IN CLEFTING. We shall first consider anterior facial heights, as follows:

Upper facial height N-Ans N-Ids Lower facial height Idi-Gn

The upper facial heights measure both structural and functional entities: N-Ans includes orbital and naso-respiratory heights; N-Ids includes the foregoing plus maxillary "basal" and alveolar heights.

The lower facial height Idi-Gn is really mandibular symphyseal height, including alveolar height and mandibular body height.

The height data are (in cm.) in Tables 7–9.

The absolute total gain, 0:3-6:0 for each height dimension is as follows (all +):

Dimension	CP	CL(P)
N-Ans	1.65 cm.	1.73 cm.
N-Ids	2.27 cm.	2.37 cm.
Idi-Gn	1.02 cm.	0.93 cm.

TABLE 7. N-Ans

	CP			t-1	test	CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0-3 Mos.	22	2.62	0.36	50	0.663	30	2.56	0.26
3-6 Mos.	25	3.08	0.27	53	0.319	30	3.06	0.23
1 Year	27	3.41	0.28	46	2.276*	21	3.22	0.30
2 Years	29	3.71	0.26	50	1.755(*)	23	3.58	0.27
3 Years	27	3.89	0.25	51	1.730(*)	26	3.77	0.25
4 Years	25	4.04	0.30	42	0.651	19	3.98	0.35
5 Years	24	4.21	0.36	39	0.352	17	4.17	0.30
6 Years	18	4.33	0.30	31	0.447	15	4.29	0.24

		CP		t-i	lest	CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	3.70	0.43	50	0.257	30	3.67	0.30
3–6 Mos.	25	4.27	0.33	53	0.708	30	4.21	0.27
1 Year	27	4.63	0.39	46	0.569	21	4.57	0.37
2 Years	29	5.14	0.33	50	1.011	23	5.05	0.37
3 Years	27	5.43	0.32	51	1.075	26	5.33	0.34
4 Years	25	5.60	0.41	42	-0.755	19	5.69	0.36
5 Years	24	5.82	0.50	39	-0.507	17	5.90	0.32
6 Years	18	5.97	0.32	31	-0.607	15	6.04	0.32

TABLE 8. N-Ids

TABLE 9. Idi-Gn

		CP		t-	test		CL(P)	
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	1.59	0.21	50	-0.601	30	1.63	0.21
3–6 Mos.	25	1.83	0.22	53	0.418	30	1.80	0.26
1 Year	27	2.15	0.24	46	1.217	21	2.06	0.24
2 Years	29	2.40	0.19	50	0.867	23	2.35	0.17
3 Years	27	2.52	0.17	51	2.956**	26	2.36	0.22
4 Years	25	2.61	0.30	42	0.511	19	2.57	0.18
5 Years	24	2.65	0.27	39	0.118	17	2.64	0.21
6 Years	18	2.61	0.37	31	0.431	15	2.56	0.28

Significant differences in upper face height dimensions at 1:0–3:0 may reflect differences in premaxillary positioning.

As far as these data are concerned about all that can be said is that upper anterior facial heights increased a very little bit more in CL(P) than in CP, lower a bit more in CP than in CL(P).

The real story is told in Figures 3 and 4. In Figure 3 it is seen that both upper facial heights are larger in CP and CL(P) than in the Normal, i.e., the face of the cleft child has a greater anterior upper face height than the Normal. Within clefting N-Ans and N-Ids are both about the same in CL(P) and CP. The slightly greater upper face height in CL(P) in N-Ans may well be due to the fact that in CL(P) the anterior maxillary alveolus is involved with a possible forward growth-inhibiting effect against downward growth effect.

Anterior upper face height in general, in both the CP and CL(P) and the Normal, shows an even growth-rate (increase) from 0:3-6:0, with a moderate relative acceleration from 0:3-1:0.





FIGURE 4. Lower face height: normal, CP, CL(P).

Figure 4 shows, as did Figure 3, that anterior facial heights are slightly greater in clefting than in the Normal. In Figure 4 lower face height, Idi-Gn, is slightly greater in both CP and CL(P) than in the Normal. There are really no significant differences in mandibular symphyseal height.

We shall now consider maxillary *lengths* in a p-a or sagittal diameter. The following measurements were taken:

 $Ptm'^{*}-Ans$ is a total maxillary length including the forward projection of the anterior nasal spine

Ptm'—A is a measure of the "basal" (non-alveolar) structure of the maxilla Ptm**—KR is a measure of posterior maxillary length KR-Ans is a measure of anterior maxillary length. (* and **, See Table 10.)

The data for these measurements (in cm.) are shown in Tables 10–13.

The absolute growth-gain in these four length dimensions is as follows (all +):

Dimensions	CP	CL(P)
Ptm'-Ans	1.35 cm.	1.60 cm.
Ptm'-A	1.22 cm.	1.53 cm.
Ptm- KR	0.78 cm.	0.96 cm.
KR-Ans	0.62 cm.	0.64 cm.

TABLE	10.	Ptm'	-Ans
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		CP	CP		-test	CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0-3 Mos.	22	3.31	0.32	50	-2.315^{*}	30	3.55	0.39
3-6 Mos.	25	3.80	0.24	53	-3.125^{**}	30	4.05	0.33
1 Year	27	4.11	0.34	46	-1.432	21	4.26	0.42
2 Years	29	4.36	0.40	50	-2.060*	23	4.57	0.35
3 Years	37	4.41	0.32	51	-4.044^{***}	26	4.75	0.29
4 Years	25	4.56	0.28	42	-3.887***	19	4.86	0.21
5 Years	24	4.62	0.32	39	-3.450**	17	4.99	0.40
6 Years	18	4.66	0.32	31	-3.969***	15	5.15	0.39

 $* \ \rm Ptm'$ is the axial inclination of the pterygo-maxillary fissure projected to the plane of the palate.

** Ptm is from a line tangent to the most anterior border of the pterygo-maxillary fissure.

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СР			t-test		CL(P)			
age	no.	mean	s.d.	degree of freedom	t-value	no.	med	s.d.
0–3 Mos.	22	3.17	0.32	50	-3.046**	30	3.47	0.38
3–6 Mos.	25	3.66	0.22	53	-3.884^{***}	30	3.93	0.27
1 Year	27	3.95	0.32	46	-1.896(*)	21	4.14	0.39
2 Years	29	4.17	0.32	50	-2.829**	23	4.41	0.28
3 Years	27	4.17	0.23	51	-6.256***	26	4.61	0.28
4 Years	25	4.28	0.27	42	-6.050***	19	4.72	0.19
5 Years	24	4.36	0.26	39	-4.824^{***}	17	4.82	0.35
6 Years	18	4.39	0.27	31	-5.378***	15	5.00	0.38

	CP		t t	-test	CL(P)			
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	1.46	0.28	50	0.912	30	1.39	0.27
3–6 Mos.	25	1.72	0.17	53	-0.831	30	1.76	0.17
1 Year	27	1.89	0.24	46	-0.215	21	1.91	0.21
2 Years	29	2.00	0.17	50	-1.438	23	2.08	0.18
3 Years	27	2.08	0.18	51	0.267	26	2.08	0.19
4 Years	25	2.06	0.14	42	-1.756(*)	19	2.15	0.17
5 Years	24	2.12	0.26	39	-1.284	17	2.22	0.27
6 Years	18	2.24	0.23	31	-1.371	15	2.35	0.24

TABLE 12. Ptm-KR

TABLE 13. KR-Ans

		СР		t	-test	CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	1.90	0.23	50	-4.118***	30	2.23	0.33
3–6 Mos.	25	2.11	0.21	53	-3.064**	30	2.34	0.33
1 Year	27	2.27	0.30	46	-1.590	21	2.41	0.31
2 Years	29	2.42	0.33	50	-1.823(*)	23	2.58	0.29
3 Years	27	2.43	0.29	51	-4.629^{***}	26	2.75	0.20
4 Years	25	2.55	0.24	42	-3.685^{***}	19	2.79	0.16
5 Years	24	2.56	0.32	39	-3.087**	17	2.84	0.21
6 Years	18	2.52	0.25	31	-3.817***	15	2.87	0.27

Here is found the greatest (most significant) difference between CP and unilateral CL(P) due almost certainly to greater a-p instability in the latter.

Total maxillary length gains a bit more in CL(P) than in CP. Since the anterior segment, KR-Ans, gains less than the posterior segment, Ptm-KR, in CP and CL(P), it is possible that the total maxillary length difference may be centered upon the displacement anteriorly, where alveolar clefting is a major difference between the two cleft types.

The data of Tables 10–13 are graphed in Figure 5.

- 1. *Ptm'-A* is longer in CP and CL(P) than in the Normal; more so in CL(P); in both CP and CL(P) there is a relatively greater gain to 1:0; CL(P) is longer than CP.
- 2. *Ptm'-Ans* shows CL(P) greater than Normal, CP shorter, probably due to the forward placed premaxilla in CL(P).
- 3. The posterior moiety of maxillary length, *Ptm-KR* contributes slightly less to over-all length than does the anterior moiety, KR-Ans.



FIGURE 5. Maxillary lengths: normal, CP, CL(P).

TABLE 14. Angle S-N-A

	CP		t	-test	CL(P)			
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	82.89	5.31	50	-3.093**	30	87.30	4.92
3–6 Mos.	25	81.64	4.19	53	-2.882^{**}	30	84.70	3.68
1 Year	27	81.15	4.06	46	-1.741(*)	21	83.21	4.10
2 Years	29	80.19	3.90	50	-3.066**	23	83.69	4.33
3 Years	27	79.93	3.31	51	-4.567***	26	84.45	3.88
4 Years	25	79.86	3.59	42	-3.739***	19	83.89	3.49
5 Years	24	78.96	3.36	39	-4.455^{***}	17	83.98	3.83
6 Years	18	78.92	3.37	31	-4.062***	15	83.73	3.41

For the maxillary complex we have calculated but one angle, S-N-A, which relates midface to the anterior cranial base. The data are in Table 14.

The over-all change in this angle is -3.97° for CP and -3.57° for CL(P).

The data for Table 14 are graphed in Figure 6.

The significant differences here are a measure of the tendency to greater retrognathism in unilateral CL(P).

Angle S-N-A is greater in the Normal, 1:0-6:0, than in CL(P), less in CP, i.e., maxilla is further forward in CL(P). It is noteworthy that in the Normal the angle is relatively constant, though it does decrease slightly.

In the cleft face point A is more retrusive than in the Normal, which means that A is a register of some inhibition (or deviation) in the forward



growth of the midface. However, since angle S-N-A is larger in CL(P) than in CP, and since KR-Ans contributes slightly less to maxillary length than does Ptm-KR, it is logical to point to anterior segment (premaxillary) positioning rather than to incremental failure.

THE MANDIBLE IN CLEFTING. Mandibular (symphyseal) corporal height (Idi-Gn) has already been presented in the discussion of anterior facial heights. The following additional measurements have been taken:

- 1. Go-Gn, which is mandibular corporal length
- 2. Ar-Go, which is mandibular ramal height (excluding the condyle)
- 3. Ar-Gn, which is an oblique measurement of ramo-corporal mandibular length.
- 4. S-Go, which is often called "posterior facial height"; it is a very complex measurement, for it involves cranial base (occipito-sphenoid) as well as mandibular ramus. In a sense S-Go is a crude analogue of total anterior face height, N-Gn.

The data for these four measurements are given(in cm.) in Tables 15–18. The total absolute gains in these four measurements are as follows (all +):

Dimension	CP	CL(P)
Go-Gn	2.71 cm.	3.24 cm.
Ar-Go	1.70 cm.	1.79 cm.
Ar-Gn	4.01 cm.	4.06 cm.
S-Go	$2.65 \mathrm{cm}$.	2.70 cm.

Moderately significant differences at 4:0–6:0 in Ar-Go and S-Go point to adjustive growth at Ba (possibly) and in ramal height.

As far as absolute dimensional increase is concerned there's no major difference: corporal length (Go-Gn) increases more than ramal height (Ar-Go).

The data of Tables 15–18 are graphed in Figures 7 and 8.

	CP			t-test		CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	3.40	0.38	50	-0.794	30	3.48	0.35
3–6 Mos.	25	4.03	0.39	53	-0.416	30	4.07	0.35
1 Year	27	4.59	0.38	46	1.021	21	4.49	0.42
2 Years	29	5.03	0.37	50	-0.028	23	5.03	0.32
3 Years	27	5.29	0.32	51	0.439	26	5.25	0.41
4 Years	25	5.63	0.32	42	-0.426	19	5.67	0.32
5 Years	24	5.96	0.40	39	0.623	17	5.88	0.36
6 Years	18	6.11	0.44	31	-0.059	15	6.12	0.45

TABLE 15. Go-Gn

TABLE 16. Ar-Go

		CP		t	-test	CL(P)		
age	n 0.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	1.90	0.26	50	-1.192	30	2.01	0.37
3–6 Mos.	25	2.49	0.38	53	-2.046*	30	2.68	0.29
1 Year	27	3.04	0.36	46	-0.027	21	3.04	0.29
2 Years	29	3.30	0.26	50	-0.506	23	3.34	0.29
3 Years	27	3.51	0.36	51	0.498	26	3.47	0.29
4 Years	25	3.58	0.25	42	-0.634	19	3.63	0.30
5 Years	24	3.57	0.33	39	-1.854(*)	17	3.74	0.23
6 Years	18	3.60	0.25	31	-1.885(*)	15	3.80	0.33

TABLE 17. Ar-Gn

		CP			t-test		CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.	
0–3 Mos.	22	5.01	0.53	50	-0.961	30	5.13	0.44	
3-6 Mos.	25	6.10	0.50	53	-1.411	30	6.28	0.46	
1 Year	27	7.19	0.57	46	1.186	21	7.01	0.49	
2 Years	29	7.82	0.42	50	0.441	23	7.77	0.46	
3 Years	27	8.27	0.51	51	1.347	26	8.09	0.48	
4 Years	25	8.55	0.36	42	-0.589	19	8.63	0.49	
5 Years	24	8.86	0.49	39	-0.326	17	8.91	0.47	
6 Years	18	9.02	0.45	31	-1.030	15	9.19	0.52	

age	СР			t-test		CL(P)		
	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	3.36	0.39	50	-0.967	30	3.46	0.40
3–6 Mos.	25	4.18	0.32	53	-1.189	30	4.29	0.38
1 Year	27	4.80	0.44	46	-0.159	21	4.82	0.39
2 Years	29	5.29	0.36	50	0.216	23	5.26	0.40
3 Years	27	5.54	0.35	51	-0.210	26	5.56	0.41
4 Years	25	5.64	0.26	42	-1.620	19	5.79	0.37
5 Years	24	5.79	0.38	39	-2.357*	17	6.07	0.38
6 Years	18	5.95	0.41	31	-1.382	15	6.16	0.49

TABLE 18. S-Go



FIGURE 7. Corporal length and ramal height: normal, CP, CL(P).

Figure 7 corporal length, Go-Gn, shows no real difference in CP, CL(P), and Normal. Ramal height, Ar-Go is a bit greater in clefting than in Normal but not significantly so. Oblique ramo-corporal, length, Ar-Gn is uniformly slightly greater in clefting, a bit more in CL(P) than in CP.

The foregoing lead to the conclusion that in the mandible both corpus and ramus are slightly larger than Normal. Beyond doubt this still does NOT warrant the tag "micrognathia" to the mandible in the cleft child.

In Figure 8 Ar-Go is repeated to contrast with S-Go. The latter measure-



FIGURE 8. Ramal height and posterior facial height: normal, CP, CL(P).

ment, which we stated above is often called "posterior facial height", is larger after 4:0 in CL(P) than in the Normal, but smaller in CP. There is no marked difference within cleft types. Since Ar-Go in Normal and CP and CL(P) is virtually the same it follows that the slight S-Go difference between Normal and clefting must be in the occipito-sphenoid complex, i.e., growth and repositioning at the O-S synchondrosis; further, it is probable that some rotation in Ba is involved. Thus, once more the basionsellar (clival) growth changes are reflected via adjustment movements in a spheno-occipito-mandibular ramal complex.

In the mandible we took the mandibular angle (ramo-corporal or gonial) as Ar-Go-Gn. The data are given (in cms.) in Table 19.

The absolute changes are as follows:

Angle	CP	CL(P)
Ar-Go-Gn	-4.84°	-2.07°

At 1:0-3:0 significant differences are probably due to adjustive growth movement in the gonial angle and in the TMJ-condylar relationship.

This angle is in large part a measure of differential ramo-corporal growth. The change has been greater in CP than in CL(P).

The data of Table 19 are graphed in Figure 9. The gonial angle is more "open" in clefting than in Normal, and by 6:0 the two cleft types have a similar gonial angle.

Figure 9a summarizes the more or less irregular polygon formed by cranial base length, upper face height, and maxillo-palatal complex (which is the area of immediate focus):

1. In the Normal upper face profile is mildly retrusive at mid-level. However, here the growth pattern is remarkably even: Ptm, Ptm',

	СР			t	test	CL(P)		
age	no.	mean	s.d.	degree of freedom	t-value	no.	mean	s.d.
0–3 Mos.	22	139.93	6.80	50	1.571	30	136.87	7.04
3–6 Mos.	25	137.40	5.56	53	0.594	30	136.45	6.16
1 Year	27	140.41	6.50	46	2.249*	21	136.76	4.07
2 Years	29	138.98	5.10 ·	50	3.038**	23	135.26	3.27
3 Years	27	139.10	4.58	51	3.006**	26	135.29	4.64
4 Years	25	135.62	5.23	42	0.349	19	135.06	5.30
5 Years	24	135.48	4.85	39	0.659	17	134.42	5.39
6 Years	18	135.07	5.62	31	0.140	15	134.80	5.10

TABLE 19. Ar-Go-Gn Angle





KR, and Ans all move uniformly down and forward. Length amounts contributed by Ptm and by KR remain in good balance.

- 2. In the CP upper face profile is mildly retrusive at mid-level. But here growth pattern is quite different: Ptm, Ptm', and Ans move downward (vertical growth); on the other hand KR moves down and forward so that it progressively contributes more to over-all maxillopalatal length.
- 3. In the CL(P) upper face profile is less retrusive generally, Ptm and Ptm' move straight down as in CP but Ptm-Kr and KR-Ans keep in balance, for *both* move evenly downward and forward, almost certainly due to the relative instability of premaxillary involvement.

Figure 9b is a total maxillary complex comparison between non-cleft and cleft cases. In *non-cleft* Ptm and Ptm' move downward-forward, but in *cleft* straight down only. In both *non-cleft* and *cleft* KR moves downward-forward. Ans reflects *type* of cleft: it is totally forward in position at



FIGURE 9a. Midfacial growth changes, 1, 3, 6 years: normal, CP, CL(P).



FIGURE 9b. Summary of growth directions in midface, birth to six years: normal, CP, CL(P).



FIGURE 10. Mandibular growth change, 1, 3, 6 years: normal, CP, CL(P).

1:0 in CL(P) (premaxillary positioning) and moves down and only slightly forward. In CP it is at the same point as the Normal, but the growth movement is straight down. In the Normal it moves evenly downwardforward.

In Figure 10 the AR-Go-Gn mandibular triangle is shown. SN is the standard horizontal reference plane and in each instance Go is on the S-Go vertical to the SN plane. The data are for 1:0, 3:0, 6:0.

- 1. In the Normal Gn is seen to move downward and forward, while in both CL and CL(P) the basic growth movement or change is mostly downward, with very little forward movement.
- 2. The distance of Ar from the S-Go vertical is very revealing. It is about the same in CP and CL(P), but in *both cleft* types it is greater than in the normal. There are probably two processes operative here: a) the increase in the gonial angle in CP and CL(P) is due to the backward "bending" or inclination of the ramus; b) there is some rotation involved, centering in Ba so that the TMJ (temporo-mandibular joint) is also posterior in position. In this manner the two factors, a) and b) are reciprocally related to one another, i.e., if TMJ is slightly posterior in position the gonial angle "opens up" so that

condylar head and TMJ may be in a functionally acceptable relationship.

Summary and Conclusions

- This study is based on tracings of serial lateral roentgenographic cephalometric x-ray films of 59 children with isolated cleft palate and 43 children with unilateral cleft lip and palate, age 0:3 (three months) to 6:0 (six years). Boy and girl headfilms are pooled. The Normal 1:0– 6:0 non-cleft controls were based on the Bolton (Cleveland) Standards of the Bolton Fund, Case-Western Reserve University.
- 2. The Cranial Base: Midline
 - a. Involved here are occipital, sphenoid, and ethmoid bones and their several parts.
 - b. The cranial base grows principally via cartilaginous synchondroses, while the facial skeleton grows principally via intramembranous sutures; in both major areas total remodelling is further achieved via bone resorption-apposition.
 - c. Growth at cartilaginous synchondroses is primary, with sutural centers secondary.
 - d. Cranial base dimensionality.
 - 1) Clival length (Ba-S) is slightly larger in CP than in CL(P): both are above Normal, CP less so.
 - 2) Anterior cranial base (S-N) shows more catch-up growth by 6:0, i.e., a closer approximation to the Normal, in CP than in CL(P).
 - 3) The basic anial axis (Ba-N) has the same growth pattern as does S-N, and again CP is closer to the Normal.
 - 4) The Ba region (anterior foramen magnum) seems to be a focus of adjustive growth, compared to the Normal: in the Normal Ba grows downward-backward, while in CP and CL(P) it grows downward-forward.
 - 5) The angle Ba-S-N (sellar angle) decreases in both cleft types more so than in the Normal after 3:0, and more in CL(P) than in CP.
 - e. It is proposed that CP is less strongly genetically entrenched than is CL(P): the obvious major involvement of Ba, more marked in CL(P), suggests that the foramen magnum-vertebral complex may be related to palatal clefting in a quasi-syndromic manner.
 - f. The serial craniofacial data strongly suggest that the entire cranial base complex, Ba-S-N, is harder "hit" in CL(P) than in CP, i.e., the CL(P) shows a greater divergence from the Normal.
 - g. There is in this study definite evidence that palatal clefting has growth and/or developmental repercussions in the associated cranial base and facial structures, probably a reflection of embryogenic growth-timing, i.e., palatal clefting may be associated with a con-

83

comitant growth dysplasia of basi-facial structures in their formative stages at the time when palatal clefting occurred.

- 3. The Facial Structural Complex: Dimensions
 - a. Upper face height is larger in both CP and CL(P) than in Normal, and slightly larger CL(P) than CP; the latter condition may be due to the fact that in CL(P) the anterior maxillary alveolar area is involved, possibly in terms of a relative forward growth inhibition against downward growth effect.
 - b. Lower face height is somewhat greater in both CP and CL(P) than in the Normal.
 - c. Total maxillary length gains more, 0:3–6:0, in CL(P) than in CP, much of the gain being in the anterior moiety of the maxilla; hence, some of the gain may include the possible forward displacement of the anterior palatal segment (premaxilla) in CL(P).
 - d. Absolutely, maxillary length is slightly less in CP and CL(P) than in the normal.
 - e. The angle S-N-A, which measures the forward growth of the midface relative to anterior cranial base, is greater in the CL(P) than in both CP and Normal.
 - f. In the mandible corporal length (Go-Gn) and ramal height (Ar-Go) are about the same in CP, CL(P) and the Normal. The gonial angle is more obtuse in both cleft types than in the Normal. The TMJ may be slightly retroposition in CP and CL(P), compared to Normal.
 - g. There is no micrognathia in CP and CL(P); to all intents and purposes at 6:0 mandibular size in CP and CL(P) approximates the Normal.
- 4. On the basis of our two serial samples, CP and unilateral CL(P), we have observed that there is a general post-operative *catch-up growth in both cleft types*, more so in CP. It is our conclusion that conservative surgery has facilitated rather than inhibited or deviated growth in both the maxillo-facial skeletal complex and the soft tissues of the labio-facial complex. In the data presented in this study our hypothesis has been substantiated.

References

- COCCARO, P. J., and S. PRUZANSKY, Longitudinal Study of Skeletal and Soft Tissue Profile in Children with Unilateral Cleft Lip and Cleft Palate. *Cleft Palate J.* 2, 1–12, 1965.
- 2. GARDNER, G. E. and J. H. KRONMAN, Cranioskeletal Displacements Caused by Rapid Palatal Expansion in the Monkey. Am. J. Orthodontics 59, 146-155, 1971
- GRABER, T. M., The Congenital Cleft Palate Deformity. J. Am. Dent. Assoc. 48, 375-395, 1954
- MAZAHERI, M., R. L. HARDING, J. A. COOPER, J. A. MEIER, and T. S. JONES, Changes in Arch Form and Dimensions of Cleft Patients. Am. J. Orthodontics 60, 19-32, 1971
- Moss, M. L. Malformations of the Skull Base Associated with Cleft Palate Deformity. Plast. reconstr. Surg. 17, 226-234, 1956

- Moss, M. L. Experimental alteration of sutural area morphology, Anat. Record 127, 569–584, 1957.
- Moss, M. L. Premature Synostosis of the Frontal Suture in the Cleft Palate Skull, Plast. reconstr. Surg. 20, 190-205, 1957
- POWELL, T. V. and A. G. BRODIE, Closure of the Spheno-occipital Synchondrosis, Anat. Record 147, 15-23, 1963
- Ross, R. B., Cranial Base in Children with Lip and Palate Clefts, Cleft Palate J. 2, 157-166, 1965
- SARNAT, B. G. Postnatal Growth of the Upper Face: Some Experimental Considerations Angle Orthodontist 33, 139-161, 1963
- 11. SHIBASAKI, Y., and R. B. Ross, Facial Growth in Children with Isolated Cleft Palate Cleft Palate 6, 290-302, 1969
- TRASLER, D. G., Pathogenesis of Cleft Lip and its Relations to Embryonic Face Shape in A/J and C57BL Mice Teratology 1, 35-50, 1968