Cephalometric Studies of the Mandible in Individuals with Clefts: Part I. A Review

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Various authors (*30, 31, 33*) have essentially agreed that the underdevelopment of the maxilla as seen in individuals with cleft lip and/or palate may be to some extent due to surgical procedures, especially when surgery is performed at a very early age. On the other hand, there has been much disagreement about whether there is an accompanying underdevelopment of the mandible and a malpositioning of the mandible in relation to other facial and cranial structures. The purpose of this report, the first of two, is to critically review the literature of North America regarding the size and position of the mandible in individuals with clefts of the lip and/or palate. In a later report, research will be reported describing the size and position of the mandible in individuals with clefts of the lip and/or palate.

Review of the Literature

Presented in Table 1 are data from several studies relating to mandibular position. Gilley (*9*) was one of the first to evaluate objectively the facial deformity in cleft tip and palate individuals. He conducted a study of the lateral cephalometric roentgenograms of 20 subjects, 14 males and 6 females, ranging in age from four years to 30 years. Of these, there were 10 subjects with unilateral clefts of the lip and palate and seven with bilateral clefts of the lip and palate. Gilley reported a definite increase in the mandibular-Frankfort plane angle in the cleft individuals which he thought might be due to a short ramus. The other three angles that he used to evaluate the mandible (N-S-Gn, S-N-Gn, and N-S-Go), when compared with normal values, showed differences which were not significant

Graber (*10*) studied 45 male and female subjects with deformities varying from simple alveolar cleft to complete bilateral clefts of the

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TABLE 1. A summary of findings from previous studies of mandible position. Items are angle used to assess position, investigator, mean value of the angle, and type of subject.

Angle	*Investigator*	*Mean angle*	*Subjects and age*
Sella-nasion-gnathion	Riedel (*32*)	76.93	normal, children
		79.29	normal, adults
	Mayne (*22*)	79.99	normal, adults
	Gilley (*9*)	76.6	cleft, 4 to 30 years
	Graber (*10*)	80.0	cleft, 7 months to 58 years
	Graber (*11*)	78.84	cleft, 2 weeks to 73 years
	Graber (*13*)	74.75	cleft, 2 weeks to 77 years
	Ponterio (*29*)	75.82	cleft, 4 to 18 years
	Snodgrasse (*34*)	73.4	cleft, 2 to 18 years
	Tong (*38*)	55.36–77.94 (Range of means for his various cleft subgroups)	
Nasion-sella-gnathion	Riedel (*32*)	67.69	normal, children
		67.90	normal, adults
	Mayne (*22*)	67.05	normal, adults
	Gilley (*9*)	67.9	cleft
	Graber (*10*)	68.9	cleft
	Graber (*11*)	68.19	cleft
	Graber (*13*)	71.01	cleft
	Ponterio (*29*)	68.12	cleft
	Snodgrasse (*34*)	71.7	cleft
Frankfort-mandible plane	Downs (*6*)	21.9	normal, 12 to 17 years
	Mayne (*22*)	22.51	normal, adults
	Gilley (*9*)	28.5	cleft
	Graber (*10*)	24.3	cleft
	Swanson (*36*)	33.0	cleft
	Snodgrasse (*34*)	29.7	cleft, boys
		37.5	cleft, girls
	Tong (*38*)	24.67–39.40 (Range of means for his various cleft subgroups)	
	Ortiz-Monasterio (*27*)	28.4	cleft, 15 to 43 years

lip and palate. The subjects ranged in age from seven months to 58 years. He analyzed lateral cephalometric roentgenograms and compared his findings with those of various studies which had been conducted with children and adults having normal or 'ideal' occlusions. He reported that 'on the whole the mandible appeared normal'. Later (*11*), he enlarged his sample to include 60 subjects, 39 male and 21 female (ranging in age from two weeks to 73 years) using the same methods employed in the first study. His conclusions in regard to the mandible were essentially the same as in his first study, that there was 'no significant mandibular underdevelopment'. In a third report (*13*), Graber increased his sample size considerably to 175 subjects (98 male and 77 female) ranging in age from two weeks to 77 years. He found that the means of the angles NS-GoGn, S-N-Gn, and N-S-Gn in the cleft groups were significantly different from the means of the corresponding angles in the groups of normal individuals who had clinically normal occlusion. He concluded from these findings that there was 'a definite mandibular underdevelopment in individuals with clefts of the lip and palate'. While these findings may indicate a retrusion of the mandible in the cleft group, it is not felt that Graber has sufficient evidence to state there is 'an underdevelopment'.

Ponterio (*29*) conducted a cephalometric study of 41 cleft individuals, 26 males and 15 females, ranging in age from four years, one month to 18 years, eight months. He made tracings of each radiograph and from the tracings made 12 linear measurements and nine angular measurements. He compared the cleft groups with control groups from other studies. These so-called 'control' groups, however, consisted mainly of children having excellent occlusion and a much narrower age range than that of the cleft group. For example, Riedel (*32*) studied children whose age ranged from seven to 11 years. Petraitis (*28*) studied children from 11½ to 13½ years of age. Ponterio's major finding relative to the mandible was that the total mandibular length as measured from the condyle to pogonion was much smaller in the cleft group than in the control group. However, on the last page of his discussion, he recognized the importance of the age difference, and stated that 'due to the wide age range of the cleft palate sample, it was felt no valid comparisons could be made with linear measurements against controls.'

Snodgrasse (*34*) described the mandibular growth in cleft lip and cleft palate patients as compared to subjects that are described as having 'clinically excellent occlusions.' His subjects were six boys and five girls, ranging in age from two years three months to 18 years, with defects varying from cleft lip only to unilateral and bilateral clefts of the lip and palate. He concluded that there was evidence of a tendency toward mandibular underdevelopment and that the female mandible inclined to be the more underdeveloped.

Swanson (*35*) statistically evaluated the growth of the face in 100 cleft lip and/or cleft palate subjects that had undergone surgical re-

pairs of the palate. The lateral cephalometric roentgenograms of the 39 female and 61 male subjects, ranging in age from 3½ to 17 years, were evaluated by employing Downs' analysis. His most significant findings were related to the mandible. The chin point was retruded with a mean facial angle 7° less than Downs' 88° normal, the Frankfort-mandibular plane angle was 10° greater than Downs' mean, and the Y-axis was found to have a mean of 6° greater than the standard. Swanson ends the report of his study by pointing out that there are certain fallacies introduced by statistically comparing this group of patients with ideal normals, since it seems logical to assume that most factors producing malocclusion in the general population are also influencing the development of children with cleft palates. Relative to this point, Swanson, in two later studies (*19, 36*), found that even though the 'composite' skeletal profile of the cleft lip and/or palate group was distinctly different from children with 'ideal-normal' occlusions, the cleft group differed only in minor detail from 25 children which he chose at random as being representative of the 'average-normal' population. No age range or sex breakdown is given for this 'control' group of 25 children.

Borden (*2*) investigated mandibular growth in cleft lip and cleft palate infants and attempted to determine if this growth differed from that in normal infants. His cleft sample consisted of 27 male infants ranging in age from 15 days to three years. The control group consisted of the records of 21 male infants ranging in age from five months to three years (material previously reported by Brodie in 1941). Tracings were made of films taken at 15 days, and at three, six, nine, 12, 18, 24, 30, and 36 months. Various measurements of the mandible were made on the tracings and the significance of the differences between the cleft palate and normal samples was tested. Borden presented the following conclusions:

> The rates of growth and type of growth pattern exhibited by the mandibles of the cleft palate and normal infants of this study were almost identical. This substantiates the contention that the growth of the mandibles of cleft palate individuals appears normal. In this study, however, the actual difference in size, although small, could not be ignored. All linear measurements, except incisor to gnathion, exhibited a difference which proved to be statistically significant. This indicates that some factor or factors have affected mandibular growth in the cleft palate sample.
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> The results of this study indicate that it would be desirable to differentiate between pre- and post-natal influences of cleft palate pathology upon mandibular growth. Such a differentiation would have been possible in this study had cephalometric material been available to serve as a control. Records of cleft palate newborns were available to the author, but the only material of normal newborns available was of nonsedated infants and in many cases the mandibles were blurred beyond acceptable accuracy.

Since none of the infants in the control group had been sedated at any age that was studied for comparison, some question might be raised as

to the reliability of the linear measurements taken on the control group mandibles used in his statistical analysis.

Kim (*17*) conducted a cephalometric study on 42 white children with surgically repaired clefts of the lip and/or palate. The age range of the cleft group was three to 11 years. The subjects were divided into three age groups, in order to minimize dimensional differences of facial structures resulting from growth. The greatest number of subjects studied in any one of the three age groups was 17 subjects, 11 boys and six girls, in the three to five year age group. A normal sample, established for comparative purposes, included 42 white children who were fairly well matched to the cleft sample according to age, sex, and angular configuration and dimension of the cranial base. He states that since a major purpose of his study was to analyze the facial proportions of cleft children by comparing them with noncleft or normal children, sex difference was not considered. He found that, regardless of the age group studied, there was no statistically significant difference between the normal and cleft samples either in mandibular body length or in mandibular ramus height. He concluded that the mandible is not abnormal in size in either cleft lip and/or palate cases.

There have been two recent reports (*25, 27*) of comparative cephalometric studies carried out on nonoperated cleft palate adults and normal adults. Ortiz-Monasterio and associates (*27*) studied 19 Mexican subjects, 12 male and seven female, ranging in age from 15 to 43 years. Three angles were measured and the findings were compared with the 'ideal-normal' sample of Downs. They found that the Frankfort-mandibular plane angle was 6° steeper, on the average, than in the controls, but the average facial angle was comparable to the normal angle, indicating an adequate craniofacial proportion. Mestre (*25*) studied lateral cephalometric roentgenograms of male and female adults living on the island of Puerto Rico. The groups studied were 29 subjects with unilateral clefts of the palate, 28 subjects with posterior clefts of the palate, and 30 subjects without cleft palate. Proportional linear and angular measurements were made and averages were obtained for purposes of comparison. The means obtained from the linear measurements showed no significant difference between the cleft palate and control groups.

Tong (*38*) compared 79 subjects (41 females and 38 males, ranging in age from four to 18 years) who had clefts that had been operated upon by the Brophy-Shearer method with standards that had been compiled in previous studies on individuals with normal occlusions. He found that the cleft groups aged four years, five months to nine years, six months showed a tendency to be statistically similar to the normal group in the measurements relating to the mandible. However, the subjects aged nine years, seven months, to 18 years showed a statistically significant deficiency in the total effective horizontal length of the mandible. He determined this by expressing the distance from articulare to pogonion as

a percentage of the distance from basion to nasion and comparing this percentage with the mean percentage found by Coben (*4*) in his study on normals that were 15 to 17 years of age. The largest sample size of any of the samples compared with the normals over this age range was eight subjects, and in one instance a sample including only three subjects was statistically compared to the normal individuals. Tong mentions that some of the difficulties of interpreting the statistical data in this study were probably due to the small size of the samples. He suggested that further study be made with a larger size sample.

Levin (*18*) studied 53 lateral cephalometric roentgenograms of cleft lip and/or cleft palate children who were judged to have an anteroposterior deficiency in the middle third of the face. The criterion for a deficiency was the presence of an SNB angle as large as or larger than the SNA angle. The values obtained for the cleft group, aged seven to nine years, were compared with values for noncleft children of comparable ages, derived by Coben. The following significant differences were found in regard to the mandible: the body of the mandible (gonion-pogonion) was shorter, the lower face depth (basion-pogonion) was deficient, the mandibular ramus (articulare-gonion) was shorter, and the posterior face height (sella-gonion) was shorter in all cleft groups studied. The validity of applying these findings in an attempt to describe the cleft population as a whole is questionable in view of the fact that the main criterion used for selection of the cleft sample was that the person had an anteroposterior deficiency in the middle third of the face.

Deuschle and Kalter (*5*) have provided the most recent report on observations of the mandible in association with defects of the lip and palate. Their subjects were 23 persons with surgically repaired clefts of the lip and palate that were chosen at random from their files without considering age, sex, ethnic origin, or degree of malformation. The mean age of the cleft patients was 9.1 years. The control group consisted of 25 noncleft individuals selected to conform to criteria for well-developed, nonprognathic faces and fairly good occlusion. The mean age of the control group was 11.5 years, which the authors state is not significantly different from the age of the cleft group. Maxillofacial triangles, as described by Margolis (*21*), were traced on the lateral cephalometric roentgenograms of the subjects. A comparison of the means of the three angular measurements taken on the two groups of subjects indicated to the authors that in the cleft sample there was a shortening in the anteroposterior dimension of the mandible and a developmental failure in the vertical growth of the mandible.

Critical Analysis of Literature

In general, the following conclusions may be made regarding the literature reviewed here:

a) In 10 of the 15 studies reviewed, it was found that the cleft lip

and/or palate subjects were compared with control groups which had been selected on the basis of their having 'ideal-normal' occlusion. In seven of these 10 studies, there was reported to be 'mandibular underdevelopment' in the cleft group. However, since most of the studies included comparison of the angular measurements of cleft subjects with those obtained on normal subjects, it cannot be stated that there is a true 'mandibular underdevelopment' because the only accurate method to assess the size of the mandible would be to compare linear measurements made on the mandible. As Swanson (*35*) pointed out, an important point to consider in attempting to determine whether or not there is any fallacy in using a group of individuals with 'ideal-normal' occlusions for comparison with a cleft sample is the criteria used for selection of the 'ideal-normal' sample. Many orthodontists include in their concept of an 'ideal-normal' not only the position of the teeth but also the profile of the individual. Since a 'straight' or a 'slightly concave' profile is considered by many to be more ideal than a 'convex' profile, it would be expected that an 'ideal-normal' sample would exclude those individuals with a small or retrusive mandible and include those individuals with a prominent mandible. Therefore an 'ideal-normal' sample would most likely include individuals with a larger mean mandibular size than that of a random sample selected from the population without regard to occlusion or facial profile.

For the above reasons it is suggested that any control sample used for comparison with a cleft lip and palate sample should be chosen without regard as to the type of occlusion or profile the individuals possess.

b) The fact that both male and female subjects were grouped together for analysis was not considered in 13 of the 15 studies reviewed. Higley (*15*), in his compilation of data of the means of 17 linear and 20 angular measurements determined from oriented profile roentgenograms of 25 to 30 boys and an equal number of girls from the fourth through the eighth year, showed that there is a sex difference between the means of the male groups and the female groups. Therefore, the sex of both the cleft group and the control group should either be the same or include the same proportions of males and females. This would be especially true if it was expected that there might be only a small real difference between the means of the measures that are to be compared.

c) Twelve of the studies reviewed had an age range of cleft subjects exceeding 13 years. The extreme was in the study by Graber (*13*) in which there was an age difference of 77 years between the youngest and oldest subject. It must be pointed out that changes which are incident to growth result in increments in the dimension of the mandible. Recognizing this, it is evident that any comparative skeletal study of the mandible while it is undergoing growth changes requires that the groups being compared have relatively small age ranges.

d) The majority of the studies reviewed included individuals with

both cleft lip and palate and cleft palate only in their group of cleft subjects. There are several reasons why it might be desirable to separate those subjects having clefts of both the lip and palate from those subjects having clefts of the palate only for comparative purposes. Fogh-Andersen (*7*) and Fraser and Baxter (*8*) have reported that the two types of defects, cleft lip and palate and cleft palate only, are etiologically different. Several authors (*7, 39*) express the view that if one part of the body is malformed, there is a tendency for other parts also to be malformed. The degree and extent of the malformation result basically from the differences in the modifying cause, the intensity of the modifying cause, and also the time of its action. So if the modifying cause is different in the two types of defects, then a difference might be expected in the types of associated malformations. Another reason why it might be desirable to study the subjects with the two types of defects separately is that the physical management of the two types of defects varies somewhat. It is the type of physical management that the subject has received that is often credited as being responsible for the obtained result, good or bad.

Summary

Presented in this paper is a review and a critical analysis of the literature of North America regarding the size and position of the mandible in individuals with clefts of the lip and/or palate. Additional research is indicated which is designed to determine a) the degree of reliability of certain measures which may be used to assess the size and position of the mandible, and b) whether the mandibular depth of the cleft lip and palate or cleft palate only subjects differ significantly from a sample of normal subjects.

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