Comparative Study Between Children with and without Cleft Lip and Cleft Palate, Part 1: Cephalometric Analysis

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Objective: **This study was conducted to compare craniofacial relationships, position, and curvature of the cervical spine between children with cleft lip and cleft palate who had been operated on and children without clefts.**

Method: **This study was performed in 28 children with mixed dentition. They were divided into two groups. The study group included 14 children with unilateral operated cleft lip and cleft palate, ranging in age from 6 to 12 years, who clinically presented with a short upper lip, abnormal lip seal, and inhibition of sagittal development of the midface that was radiographically assessed. The control group included 14 children without clefts, ranging in age from 8 to 11 years. All of them had normal lip seal, nasal breathing, and a clinically normal body posture.**

Design: **A lateral craniocervical radiograph in a self-balanced natural head position in an erect posture, and without using a head holder, was taken for each child of both groups, with the mandible in maximum intercuspation and lips in habitual posture. The true vertical was marked on all the films. Specific angular and linear dimensions were used to assess the craniocervical relationships, as were the position of the cervical spine, its curvature, or both.**

Results and Conclusions: **The study group presented a significant increase in the extension of the head on the neck, forward position of the cervical spine, and a decrease in the curvature of the cervical spine in comparison with the children without clefts. These results are more relevant considering that the study group also presented higher significant values of lower facial height than children without clefts.**

KEY WORDS: *anterior facial height, cephalometry, cleft lip, cleft palate, craniocervical relationships, spinal curvature, spinal position*

Several studies have shown the presence of morphological cervical spine anomalies in patients with cleft lip and cleft palate (Sandham, 1986; Hoenig and Schoener, 1991, 1992). Moreover, cephalometric studies have shown that there are differences concerning facial relationships in populations with and without clefts. These differences have been attributed to the management of the lip, palate, or both, functional changes resulting from the mechanical presence of the cleft, genetic pattern, or a combination of these factors (Bishara et al., 1976).

It is well known that children who have been born with cleft lip and palate need the current techniques for the surgical repair aiming for restoration of shape and muscle function as a fundamental condition for preventing or minimizing secondary skeletal changes of the midface (Kernaham and Bauer, 1983; Joos, 1987).

Bardach and Eisbach (1977) stated that primary lip repair always results in a certain degree of labial tension that is transferred as pressure to the underlying maxilla, which may significantly interfere with normal maxillary growth. Bardach (1990), in a retrospective review of his previous clinical and experimental research, restated his original hypothesis that cleft lip repair, and not palate repair, should be considered to be the major cause of the maxillofacial deformities observed in the population with clefts.

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It has been shown that a characteristic feature in the soft tissue profile of operated children with unilateral cleft lip and palate is a short upper lip (Smahel and Mullerova, 1986). The causal relationship between lip pressure following lip repair and facial growth disturbances has been confirmed in experimental studies on rabbits and beagles (Bardach et al., 1982; Bardach and Mooney, 1984) and on humans (Eisbach et al., 1978; Bardach, 1990).

We know that patients with cleft lip who have undergone surgical correction during childhood often show considerable inhibition in the anteroposterior development of the midface after completion of growth. Semb (1991) suggested that retrusion of the maxilla is a common finding in most patients with cleft palate or with cleft lip and palate that have been surgically repaired during childhood.

Given that several studies have pointed out a close interrelationship among the different components of the cranio-cervico-mandibular system (Rocabado, 1984; Kraus, 1988), it could be relevant to know whether children with cleft lip and cleft palate who had been operated on who presented abnormal lip seal, short upper lip, and maxillary retrusion present a significant difference in craniocervical relationships and in the position, curvature, or both of the cervical spine in comparison with a sample of children without clefts as a control group.

Many studies in the population without clefts have shown that a large craniocervical angle is related to a vertical facial development (Solow and Tallgren, 1976; Marcotte, 1981; Solow et al., 1984; Hellsing et al., 1987). Afterward, Solow and Siersbaek-Nielsen (1982), while studying predictive relationships in children without clefts, found an association between craniocervical angulation and development of the lower face. They showed that children with a large craniocervical angle and an upright position of the upper cervical column displayed a more vertical subsequent facial growth pattern than children with a small craniocervical angulation and a backward inclination of the upper cervical column.

Based on the above considerations, the purpose of the present work was to compare some specific craniofacial and craniocervical measurements, as well as measurements of the position of the spine, its curvature, or both, between children with cleft lip and cleft palate who had been operated on and children without clefts.

Materials and Methods

This study included 28 children with mixed dentition. They were divided into two groups: a study group and a control group. The study group included 14 children, eight girls and six boys aged 6 to 12 years with an average age of 9.5 (Table 1). All had undergone lip surgery during the first year of life. For the purpose of this study, all children with unilateral operated cleft lip and cleft palate selected presented a clinical short upper lip; abnormal lip seal (Carvajal et al., 1994; Carvajal et al., 1995); and inhibition of anteroposterior development of the midface, which was assessed radiographically (see below). The control group included 14 children without cleft lip and palate or dentomaxillary anomalies, four girls and 10 boys aged 8 to 11 years, with an average age of 10.14 (Table 1). All of them had normal lip seal, nasal breathing (checked through an aerophonoscopic test), normal craniofacial development (see below), and a clinical normal body posture.

TABLE 1 Characteristics of Children Studied

Study Group			*Control Group*		
Child	*Sex*	*Age (y)*	*Child*	*Sex*	*Age (y)*
1	M	9	1	M	11
2	M	12	2	M	10
3	F	9	3	M	11
4	F	12	4	F	11
5	F	6	5	M	11
6	M	9	6	M	10
7	M	8	7	F	11
8	M	12	8	M	10
9	M	13	9	M	11
10	F	9	10	F	9
11	F	11	11	F	8
12	F	7	12	M	10
13	F	10	13	M	10
14	F	6	14	M	9

A lateral craniocervical radiograph in a self-balanced, natural head position in erect posture, and without a head holder, was taken for each child of both groups, with the mandible in maximum intercuspation and the lips in habitual posture. A free-hanging plumb line, located behind each child's head, was mounted in front of the cassette to indicate a true vertical on all of the films (Solow and Tallgren, 1971; Siersbaek-Nielsen and Solow, 1982; Cooke, 1990; Huggare and Cooke, 1994; Moya et al., 1994; Miralles et al., 1997). In this study the radiographic equipment used was the Paloceph model (Siemens Corporation, Bensheim, Germany). The focus median plane distance was 155 cm, standardized 65 Kv, 20 mA for 0.8 seconds of exposition, and the radiographic film used was Kodak TMG-1 (24 $\times$ 30 cm). A sheet of transparent acetate was placed over the radiographs and the anatomical structures were outlined. Cephalometric points used in this study are shown in Figure 1. Angular and linear dimensions were carried out using a protractor and millimeter ruler.

Figure 2 shows angular and linear dimensions used in this study, according to previous works (Carvajal et al., 1994; Moya et al., 1994; Miralles et al., 1997). In order to minimize methodological errors, two outlines and measurements were made on each roentgenogram in the study group as well as in the control group by two examiners. The mean value of both measurements was used.

Comparison of angular and linear dimensions between the study group and the control group was performed by means of a *t* test for independent samples.

In the study group, a simple linear correlation analysis was made between facial convexity (selection criteria for the sample chosen) and the angular and lineal measurements that showed significant differences when comparing the study group versus the control group.

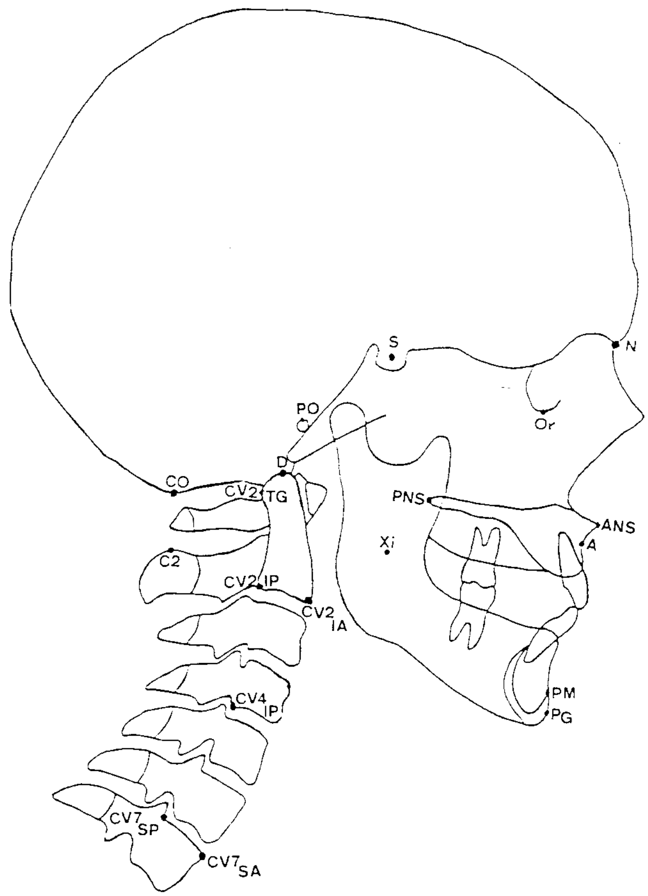

FIGURE 1 **Diagram of points used in the present study: N = nasion, the most anterior point on the frontonasal suture. S = sella, the center of the sella turcica. ANS = spina nasalis anterior, the apex of the anterior nasal spine. PNS = spina nasalis posterior, the apex of the posterior nasal spine. OR = orbitale, the lowest point on the external border of the orbital cavity. PO = porion, a point located at the most superior point of the external acoustic meatus. A point = the deepest point selected of the curve of the maxilla between the anterior nasal spine and the dental alveolus. PM = protuberance menti or suprapogonion, a point selected at which the curvature of the anterior border of the symphysis changes from concave to convex. PG = pogonion, the most anterior point on the mandibular symphysis. XI point = a point located at the geographic center of the ramus. C0 = the most inferior and posterior point of the occipital bone. C2 = the most superior point of the spine of the second cervical vertebrae. D = dens, upper point of the odontoid apophysis. CV2TG = tangent of the second cervical vertebra, the point most superior and posterior of the odontoid apophysis. CV2IP = the most inferior and posterior point of the second cervical vertebra. CV2IA = the most inferior and anterior point of the second cervical vertebra. CV4IP = the most inferior and posterior point of the fourth cervical vertebra. CV7SP = the most superior and posterior point of the seventh cervical vertebra. CV7SA = the most superior and anterior point of the seventh cervical vertebra.**

TABLE 2 Group Mean Values and Standard Deviation of the Craniofacial Angular and Linear Measurements in the Samples Studied

	Study Group		*Control Group*	
	Mean	*SD*	*Mean*	*SD*
Facial convexity	−3.10	3.02	4.88	1.98
Facial depth	87.62	5.46	88.64	4.52
Lower facial height	53.14	3.26	46.93	3.88

** $p < .01$ (t-test for independent samples).

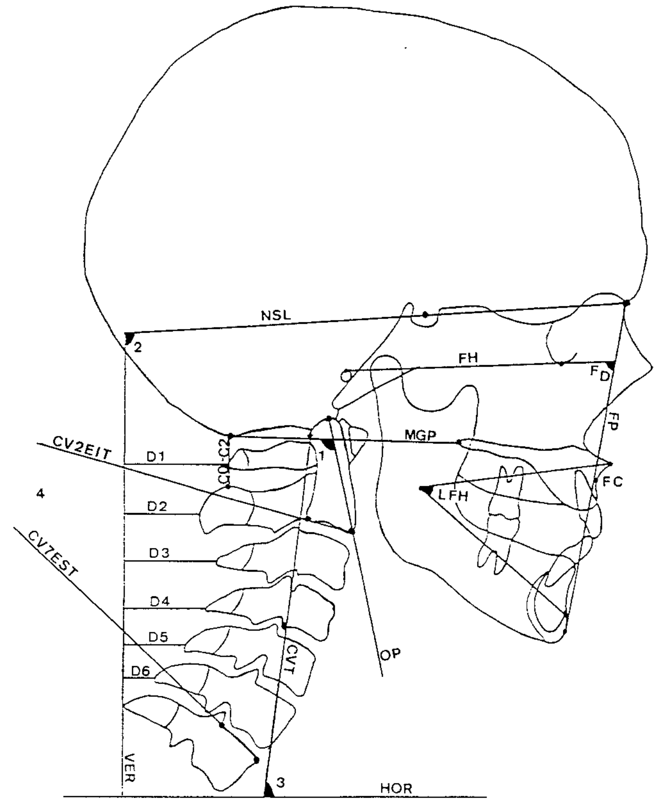

FIGURE 2 **Angular and linear dimensions used in the present study: NSL = nasion-sella line. FH = Frankfort plane, line through PO and OR. FP = facial plane, line through nasion and pogonion. MGP = MacGregor plane, line through PNS and C0. OP = odontoid plane, line through D (dens) and CV2IA. VER = true vertical plane. HOR = true horizontal plane. CVT = cervical vertebra tangent, line through CV2TG and CV4IP. CV2EIT = tangent line of the inferior edge of the second cervical vertebra. CV7EST = tangent line of the superior edge of the seventh cervical vertebra. Angle 1 = MGP/OP, angle between MacGregor plane and odontoid plane. Angle 2 = NSL/VER, angle between nasion-sella line and the true vertical plane. Angle 3 = CVT/HOR, angle between cervical vertebra tangent and the true horizontal plane. Angle 4 = CV2EIT/CV7EST, angle between tangent line of the inferior edge of the second cervical vertebra and tangent line of the superior edge of the seventh cervical vertebra. FD = facial depth, angle between Frankfort plane and facial plane. LFH = lower facial height, angle between ANS/XI/PM. FC = facial convexity, distance between A point and the facial plane. C0–C2 = distance between C0 and C2. D1 to D6 are the distances between the true vertical (tangent to the most posterior point of the spinal apophysis of the seventh cervical vertebra) to the most posterior point of each of the first to sixth spinal apophysis of the cervical vertebra.**

Statistical procedures related with the simple linear correlation analysis and simple regression analysis were performed using statistical software Stata (Stata Corporation, College Station, TX).

Results

Table 1 shows study and control group children's age and sex. Comparison according to the age of the children showed no significant differences between both groups ($t = -0.974$; $p > .339$).

TABLE 3 Group Mean Values and Standard Deviation of the Craniocervical Angular Measurements in the Samples Studied

	Study Group		*Control Group*		
Angle	*Mean*	*SD*	*Mean*	*SD*	*p Value*
1, MGP/OP	90.60	10.26	106.21	8.63	.000**
2, NSL/VER	97.71	7.59	100.59	8.33	.348
3, CVT/HOR	77.89	8.75	90.03	6.85	.000**
4, CV2EIT/CV7EST	11.02	13.03	21.18	10.97	.035*

* $p < .05$ (*t*-test for independent samples).
** $p < .01$ (*t*-test for independent samples).

Table 2 presents the group mean values and standard deviation of the craniofacial angular and linear measurements in the study group and control group. It is possible to observe a significant difference in facial convexity and lower facial height between both groups, whereas no significant difference was observed in facial depth.

Table 3 shows the group mean values and standard deviation of the craniocervical angular measurements in the study group and control group. Significant differences in the angles MGP/OP; CVT/HOR; CV2EIT/CV7EST (angles 1, 3, and 4, respectively) were observed between both groups. The angle NSL/VER (angle 2) did not differ significantly between groups.

Table 4 presents the group mean values and standard deviation of the craniocervical linear measurements in the study group and control group. A significant decrease in the distance C0–C2 was observed in the study group in comparison with the control group. Distances D1 to D6 were taken from the projection of the true vertical to the most posterior point of the spinal apophysis of the seventh cervical vertebra. Each distance was measured from the true vertical projection to each spinal apophysis for each one of the first six cervical vertebrae. A significant increase in the distances D1, D2, and D3 in the study group was observed, whereas distances D4, D5, and D6 did not present significant changes in either group.

In the study group a correlation analysis was performed between facial convexity (selection criteria for the sample chosen) and the significant variables observed when comparing the study group versus the control group (see Tables 3 and 4). In Table 5 it is possible to observe that we found significant negative correlation only between facial convexity and angle 1 as well as between facial convexity and C0–C2 distance and a significant positive correlation between facial convexity and distance D3 among all the variables shown in Tables 3 and 4.

Table 6 shows a higher correlation among the predictor variables in the study group.

TABLE 4 Group Mean Values and Standard Deviation of the Craniocervical Linear Measurements in the Samples Studied

	Study Group		*Control Group*		
Distance	*Mean*	*SD*	*Mean*	*SD*	*p Value*
C0–C2	13.73	6.00	21.55	5.03	.000**
D1	32.41	12.30	19.43	9.09	.004**
D2	23.98	9.78	17.57	6.14	.048*
D3	26.39	7.87	20.73	5.33	.035*
D4	23.86	5.76	21.23	4.52	.189
D5	19.66	3.85	17.96	3.40	.226
D6	11.68	3.40	11.52	3.08	.897

* $p < .05$ (*t*-test for independent samples).
** $p < .01$ (*t*-test for independent samples).

TABLE 5 Simple Linear Correlation Between Facial Convexity and Angular or Linear Dimensions

		Angle 1	*C0–C2*	*D3*
Facial convexity	*r*	−0.65**	−0.69**	0.59*

* $p < .05$ (*t*-test for independent samples).
** $p < .01$ (*t*-test for independent samples).

Discussion

In the analysis of the results in the present study, it is important to emphasize the technique used to take the lateral craniocervical radiographs. It is well known that self-balanced natural head position with erect posture, not using a head holder, and the incorporation of a true vertical line in the film, ensures not only a natural head position but also a natural position of the cervical column (Siersbaek-Nielsen and Solow, 1982), which is relevant to consider in any study involving the relationships between the different components of the craniocervical-mandibular system.

With respect to the values of facial convexity observed (maxillary retrusion) in the study group in comparison with the control group, it is important to note that there were not significant differences in the facial depth values between both groups, which means that the maxillary retrusion observed in the children with cleft lip and cleft palate is not due to a forward mandibular position.

From a physiological point of view, results of the present study support the idea of several studies that have pointed out that there is a close interrelationship between the different components of the cranio-cervico-mandibular system (Rocabado, 1984; Kraus, 1988). In fact, children with cleft lip and cleft palate who underwent operations (with abnormal lip seal, short upper lip, and maxillary retrusion) presented a significant increase in the extension of the head on the neck (decreased angle 1 and C0–C2 distance), forward position of the cervical spine (decreased angle 3, and increased distances D1, D2, and D3), and decreased curvature (angle 4) of the cervical spine in comparison with a sample of children without clefts.

From a clinical point of view, it is important to be aware that craniocervical changes observed in the study group could be the cause for cervical signs or symptoms that these children could be presenting during clinical examination.

TABLE 6 Correlations Between Predictor Variables

	Angle 1	*C0–C2*
C0–C2	0.90**	
D3	−0.84**	−0.84**

** $p < .01$.

The significant changes in craniocervical relationships as well as in position, curvature, or both of the cervical spine in the study group are also relevant if we consider that the children of that group presented a significant increase of the lower facial height in comparison with the control group. This result agrees with many studies in a population without clefts that have shown that a large craniocervical angle is related to vertical facial development (Solow and Tallgren, 1976; Marcotte, 1981; Solow et al., 1984; Hellsing et al., 1987). Moreover, this also agrees with a study of predictive relationships in children without clefts of Solow and Siersbaek-Nielsen (1992) who found an association between craniocervical angulation and development of the lower face. They showed that children with a large craniocervical angle and an upright position of the upper cervical column displayed a more vertical subsequent facial growth pattern than children with a small craniocervical angulation and a backward inclination of the upper cervical column.

It is interesting to note that in the present study, the magnitude of significant correlation found between facial convexity (selection criteria of the children chosen) and angle 1, distance C0–C2, and distance D3 presented a moderate magnitude (0.59 to 0.69). The decrease of angle 1 reveals a head extension over the cervical column, which is in a close relationship with the significant decrease observed in the C0–C2 distance. Both measurements are also deeply related to the forward position of the cervical spine (demonstrated by the increase of distance D3) as a compensation mechanism upon head extension on the upper cervical spine.

From a research point of view, the finding of significant correlation between facial convexity and angle 1 and the position, curvature, or both of the cervical spine raised some interesting questions: (1) Is it possible that the maxillary retrusion may be the causal factor in determining a decrease of the angle 1 in children without cleft lip and cleft palate? (2) Is it possible that the maxillary retrusion may be a causal factor in determining the position and curvature of the cervical spine observed in the children with cleft lip and cleft palate? (3) Is it possible that the increase of the lower facial height observed in the children with cleft lip and cleft palate may be caused by the decrease of the angle 1 or by the position and curvature of their cervical spine? (4) Is one or more than one of these factors the reason that could be relevant in the long-term craniofacial development of these children?

Results of the present study did not allow us to answer some of these questions. Nevertheless, a current study is being conducted to elucidate some answers to these questions.

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