RESEARCH ARTICLE

Evaluation of the cervical vertebral anomalies in patients with cleft lip and palate in Aegean region of Turkey

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Abstract

Objective: To identify the cervical vertebral anomalies in patients with cleft lip and palate, and to compare unilateral and bilateral cleft lip and palate.

Methods: The retrospective cohort study was conducted in 2018 at Department of Orthodontics, Ege University, Izmir, Turkey, and comprised non-deteriorated lateral cephalometric radiographs of non-syndromic patients which showed the entire cervical spine. The radiographs were divided into two groups, with group A having those of patients with cleft lip and palate exposure, and control group B having those with non-exposure. Within group A, unilateral and bilateral cleft lip and palate cases were compared. Data was analysed using SPSS 22.

Results: Of the 220 subjects, 110(50%) were in group A with a mean age of 15±6.3 years, and 110(50%) were in group B with a mean age of 15±2.1 years. Within group A, 56(50.9%) subjects had unilateral and 54(49.1%) had bilateral cleft lip and palate. Cervical vertebral anomalies were found in 71(64.5%) patients and 45(40.9%) controls ($p<0.001$). Among those with bilateral condition, it was found in 41(75.9%) and in unilateral 56(56.6%) ($p<0.05$). Occipitalisation was 21(38.9%) in bilateral and 4(7.1%) in unilateral cases ($p<0.001$). Fusion was higher in bilateral patients 16(63%) compared to 23(41.1%) unilateral ($p<0.05$). Posterior arch deficiencies were found in 30(27.3%) patients in group A and 18(16.4%) controls in group B ($p<0.05$). Fusion was seen in 57(51.8%) group A patients and 33(30%) group B controls ($p<0.001$).

Conclusion: Cervical vertebral anomalies were mostly found in patients with cleft lip and palate. In patients with bilateral condition, more than one anomaly was seen.

Keywords: Cervical vertebral anomalies, Unilateral cleft lip and palate, Bilateral cleft lip and palate.

(JPMA 71: 215; 2021)

DOI: https://doi.org/10.47391/JPMA.213

Introduction

Cleft lip and palate (CLP) are common congenital malformations of the lip and palate caused by genetic and environmental factors. There is an association between CLP and cervical vertebral anomalies (CVA) as aetiological factors can be affected by each other.[1-4] From lateral cephalometric radiographs, cervical vertebra can be recorded and can be used to identify CVA. The radiological images of CVA on standardised lateral cephalometric radiographs have been described in literature according to which, identification of CVAs early is very important for referral to relevant professionals for the correct treatment.[5]

The cervical vertebrae develops from sclerotomes and the sclerotomal paraxial mesodermal sheath appears at about 4 weeks into intrauterine life. By the 8th week of foetal life, ossification begins and is completed at about three years.[3-5] A deficiency of mesenchyme in median palatal process causes cleft lip (CL) anomalies while defective development of the lateral palatal processes causes cleft palate (CP) anomalies. The congenital anomalies of the cervical vertebrae are developed from the same paraxial mesoderm, which may be one of the possible mechanisms for CP development.[6-8] CVAs are divided into posterior arch deficiencies (PADs) and fusions (FUS). PADs are subdivided into spina bifida and dehiscence, while FUS are subdivided into block fusion and occipitalisation.[1,9-11]

The current study was planned to identify CVAs in the Aegean Region of Turkey in CLP patients, and to compare patients with unilateral (UCLP) and bilateral (BCLP) conditions in this regard.

Material and Method

The retrospective cohort study was conducted from the lateral cephalometric radiographs taken between the years of 2015-2018, in 2018 at the Department of Orthodontics, Ege University, Izmir, Turkey. It comprised of non-deteriorated lateral cephalometric radiographs of non-syndromic patients with skeletal Class I anomaly in which the mesiobuccal cusp of the maxillary first molar occludes in the buccal groove of the mandibular first molar which showed the entire

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cervical spine.

After approval from the institutional ethics review committee, the sample size was calculated using G Power software with a significance level of 0.05 and power 80%. Additional archival material was used to increase the power of study to >95%.[12]

Written informed consent for participation and publication of research had been taken from parents of each patient.

Lateral cephalometric radiographs were outlined on an acetate paper with 0.7 graphite pencil and CVAs were noted by a single examiner. All the radiographs were classified as mentioned in literature[1] (Figure).

Various CVAs were defined for the purpose of the study. Spina bifida: Insufficient ossification in the spinous process, breakdown of posterior part of the neural arch; Dehiscence: Spinous process indicating insufficient development of the structures, breakdown of part of a vertebral unit; Fusion: Bony union of one unit with another at the articulation facets, neural arch or transverse processes of vertebrae; Block fusion: Fusion between two cervical vertebrae, and Occipitalization: Assimilation of the atlas with occipital bone either partially or completely.

The radiographs were divided into two groups, with group A having those of patients with CLP exposure, and control group B having those of controls with non-exposure. Within group A, UCLP and BCLP cases were compared.

Data was analysed using SPSS 22. Chi square test was used where applicable. Intra-observer reliability was also evaluated using intra-class correlation coefficients (ICC). The measurements were analysed again after two weeks in randomly selected lateral cephalometric films. P<0.05 was taken as statistically significant.

Results

Of the 220 subjects, 110(50%) were in group A with a mean age of 15±6.3 years, and 110(50%) were in group B with a mean age of 15±2.1 years. Within group A, 56(50.9%) subjects had UCLP and 54(49.1%) had BCLP. CVAs were found in 71(64.5%) patients and 45(40.9%) controls (p<0.001). Among those with BCLP, it was found in 41(75.9%) and in UCLP 56(56.6%) (p<0.05). Occipitalisation was found in 21(38.9%) BCLP and 4(7.1%) UCLP cases (p<0.001) (Table-1). FUS was higher in BCLP patients 16(63%) compared to 23(41.1%) UCLP (p<0.05). PADs were found in 30(27.3%) patients in group A and 18(16.4%) controls in group B (p<0.05). FUS was seen in 57(51.8%) group A patients and in 33(30%) group B controls (p<0.001) (Table-2). In UCLP sub-group, there was one anomaly in 22(39.3%) cases and two anomalies in 8(14.3%) patients. In the BCLP sub-group, there was one anomaly in 23(41.8%) cases, two in

Table-1: Distribution of cervical vertebrae anomalies.

	N	Spina Bifida		P value	Dehiscence		P value	Fusion		P value	Block Fusion		P value	Occipitalization		P value	Total		P value
		N	%		N	%		N	%		N	%		N	%		N	%	
CLASS I	110	18	16.4	0,05	0	0	-	24	21.8	0,069	1	0.9	0,175	10	9.1	0,006	45	40.9	0,000
CLP	110	30	27.3	*	0	0		36	32.7		4	3.6		25	22.7	**	71	64.5	***
UCLP	56	14	25	0,586	0	0	-	18	26.8	0,894	2	3.6	0,970	4	7.1	0,000	30	56.6	0,014
BCLP	54	16	29.6		0	0		18	33.3		2	3.7		21	38.9	***	41	75.9	*

(p<0.05) * (p <0.01) ** (p <0.001) ***

CLP: Cleft lip and palate; UCLP: Unilateral cleft lip and palate; BCLP: Bilateral cleft lip and palate.

Table-2: Distribution of PAD and FUS anomalies.

	N	PAD		P value	FUS		P value
		N	%		N	%	
CLASS I	110	18	16.4	0,050	33	30	0,001
CLP	110	30	27.3	*	57	51.8	***
UCLP	56	14	25	0,586	23	41.1	0,022
BCLP	54	16	29.6		34	63	*

(p<0.05) *(p <0.01) **(p <0.001) ***

PAD: Posterior arch deficiency; FUS: Fusion; CLP: Cleft lip and palate; UCLP: Unilateral cleft lip and palate; BCLP: Bilateral cleft lip and palate.

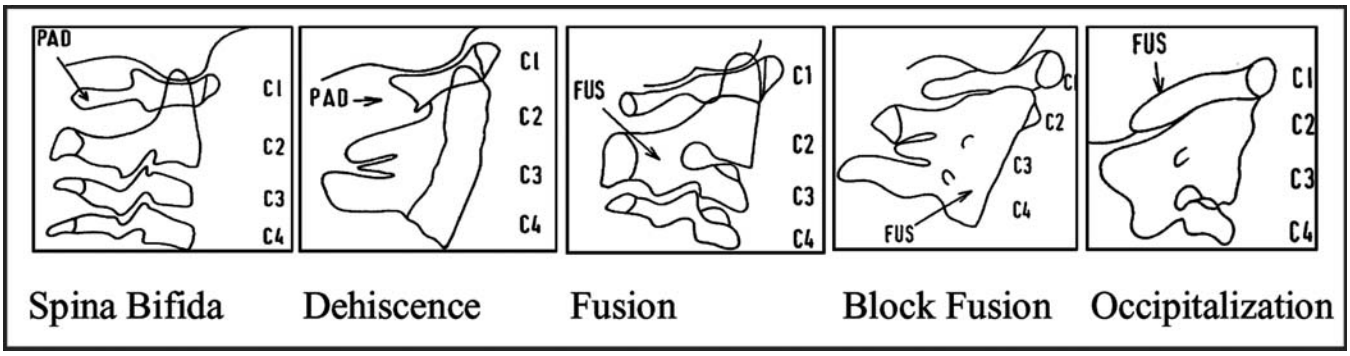

Figure: Cervical vertebral anomalies.[1]

14(25.9%) cases, and 3 in 1(1.9%) patient.

During the two-week analysis of the lateral cephalometric films, 55(25%) were selected and ICC values were highly acceptable with a mean of 0.995±2.4 (range: 0.94-0.99).

Discussion

To the best of our knowledge, the current study is the first in Turkey to have identified and compared CVAs in CLP and non-CLP patients, while also comparing the UCLP and BCLP cases.

The study focussed on patients in the Aegean region of Turkey with minimum 9 years of age. Batwa et al.[10] stated that the onset of the pubertal growth increase was significantly earlier in the non-CLP participants in comparison with UCLP participants. It was suggested that the lower age limit of the patients were 6 years of age because the malformations of the upper cervical vertebrae cannot be confirmed at an earlier age.[3,9] Upper cervical vertebrae malformations cannot be assessed using conventional radiography until complete development has occurred[1]. Some studies[1,9] excluded CLP patients aged <6 years for this reason. Similar was the case with the current study.

Compared to the current study's findings, studies evaluating CVAs have presented different scores for CLP and control groups. Our results are higher than some of those studies.[1,4,9,13]

Compared to some studies in literature,[1,7] the current study evaluated lateral cephalometric radiographs of patients having complete clefts to identify CVA differences between UCLP and BCLP.

The number of male and female subjects in the current study was nearly equal to detect any relationship of gender with CLP. Literature has studies with mixed findings on this association, with some reporting in the affirmative,[14] while others reporting in the negative.[15]

The current study showed that the prevalence of FUS in BCLP was higher than UCLP. The finding is different from some reported earlier,[1,5,16] but is similar to the findings of one study.[17]

More detailed diagnostic methods are needed for evaluating BCLP patients. Further studies are required to establish any association between oral clefts and upper CVA at the genetic level.

Conclusion

CVAs were mostly found in CLP patients. In BCLP patients, more than one anomaly was seen.

Disclaimer: None.

Conflict of Interest: None.

Source of Funding: None.

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