Maxillary Dental Anomalies in Patients with Cleft Lip and Palate: A Cone Beam Computed Tomography Study

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Objective: *To compare the frequency of maxillary dental anomalies in patients affected by unilateral (UCLP) and bilateral (BCLP) cleft lip with palate and to determine whether statistical differences were present or not between cleft and normal sides in UCLP group by using cone beam computed tomography (CBCT). In addition, the frequency of those dental anomalies was compared with previous studies presenting the same population without cleft* ***Study Design:*** *Fifty non-syndromic patients affected by UCLP (28 patients) and BCLP (22 patients) were selected for analysis of dental anomalies by means of CBCT. The frequency of maxillary dental anomalies including tooth agenesis, microdontia of lateral incisor, ectopic eruption and impaction of canine and supernumerary tooth were examined. Pearson chi-square and Fisher's exact tests were performed for statistical comparisons.* ***Results:*** *All patients affected by UCLP and BCLP were found to have at least one maxillary dental anomaly. The most frequently observed dental anomaly was tooth agenesis (92.5% and 86.4%, respectively) in UCLP and BCLP groups. Tooth agenesis and canine impaction were observed more commonly in the cleft side (75.0% and 35.7%, respectively) than in the normal side (57.1% and 14.3%, respectively) in UCLP group ($p>0.05$). All dental anomalies were found to be higher in both cleft groups than in general populations not affected by cleft.* ***Conclusion:*** *Since patients affected by UCLP and BCLP had at least one dental anomaly and higher dental anomaly frequency as compared to patients without cleft, those patients should be examined carefully prior to orthodontic treatment.*

Key Words: *Dental anomaly, cleft lip, cleft palate, cone beam computed tomography.*

INTRODUCTION

Orofacial clefts are among the most common congenital malformations of the craniofacial region.[1] The incidence of cleft lip and palate (CLP) is 1/500–2000 livebirths.[2,3] Despite the fact that clefts have not been completely explained, many genetic and environmental factors have been held responsible for their etiology and pathogenesis.[4, 5]

When compared with the general populations, dental anomalies in number, size, shape, timing of formation and eruption, and enamel dysplasia have been demonstrated to be more commonly observed in patients affected by CLP.[4, 6-10] The association between dental anomalies and CLP might come from their proximate anatomy, timing of dental development and timing of cleft formation.[11] Since dental anomalies observed in patients affected by CLP differ according to the type of cleft, it was suggested to state classification of cleft type in studies performed on patients affected by CLP.[6]

Those patients affected by CLP need multidisciplinary approach and the pediatric dentists and orthodontists represent important members of the interdisciplinary cleft team. A detailed examination of the cleft patients is needed for the presence of dental anomalies, since those anomalies might be complicating factors for orthodontic treatment. The knowledge of dental anomaly presence in patients affected by CLP might provide valuable information for pediatric and orthodontic treatment planning at an early age.[4, 12]

Although there have been many studies on dental anomalies in patients affected by CLP, most of them have used different types of clefts in their study samples; have investigated maxillary and mandibular arches together; and none used cone beam computed tomography (CBCT). The aim of the present study was, therefore, to compare the frequency of maxillary dental anomalies in patients affected by unilateral (UCLP) and bilateral (BCLP) cleft lip with palate and to determine whether statistical differences were present or not between cleft and normal sides in UCLP group by using CBCT. In addition, the frequency of those maxillary dental anomalies was also compared with the previous studies published in the same country in order to compare our results with patients not affected by cleft.

MATERIAL AND METHOD

Fifty patients (33 males and 17 females; age range 12-25 years) affected by UCLP (20 left sides; 8 right sides) and BCLP were consecutively selected from the archieve of the Departments of Orthodontics and Oral and Maxillofacial Radiology of the

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University of Erciyes for the present retrospective study. The sample consisted of 28 patients affected by UCLP (19 males and 9 females; mean age: 13.27±3.68 years) and 22 patients affected by BCLP (14 males and 8 females; mean age: 14.26±2.61 years); who referred for dental and/or orthodontic treatments. All patients included to the study had the same surgical procedure (lip and hard tissue closure) before 3 years of age, no syndromes, no extraction of any permanent tooth, and no history of trauma, previous orthodontic/prosthodontic treatment, or maxillofacial surgery. CBCT, intra-oral photographs, dental casts and dental histories of the patients affected by CLP were used in order to determine maxillary dental anomalies.

CBCT scans of the patients included in this study were part of the diagnostic records collected for impacted teeth cases and/or required orthodontic treatment; the patients were not exposed to any additional radiation. Therefore, approval from the ethics committee was not required for this retrospective archive study. In addition, as a usual protocol, all the patients (or parents) signed an informed consent agreeing to the use of the patients' data for scientific studies. All CBCT images were obtained with the patient in the supine position using the same machine (NewTom 5G; QR, Verona, Italy). Scanning time was 18 seconds, collimation height 13 cm, exposure time 3.6 seconds, and voxel size 0.3 mm^3. Digital Imaging and Communications in Medicine files obtained from the CBCT scans were reconstructed using NNT viewer software (NewTom 5G; QR, Verona, Italy). The following maxillary dental anomalies were investigated:

Tooth agenesis: A tooth, excluding third molar, was registered as congenitally missing when no trace could be found on radiographs and the treatment records confirmed that the tooth had not been extracted.[13]

Microdontia of lateral incisor: an inherited condition that produces smaller tooth.[14]

Ectopic eruption of canine: the eruption of a tooth in an abnormal position.[15]

Impaction of canine: A tooth was accepted as impacted if the tooth was not exposed to the oral cavity and the age of the patient was older than 16 years.[16]

Supernumerary tooth: the existence of an excessive number of teeth in relation to the normal dental formula (20 in the deciduous dentition and 32 in the permanent dentition).[17]

All records were examined by one author in order to reduce error (SKB). Amount of the mesio-distal diameter of the lateral incisor for the presence of microdontia was measured using digital calipers (Mitutoyo, Tokyo, Japan). Any permanent tooth on either side of the alveolar cleft, regardless of morphology, between the canine and central incisor, was considered a maxillary lateral incisor.[18]

Statistical analysis:

The same investigator reassessed the data two weeks after the first examination. The degree of agreement showed no difference between the two examinations. Intra-class correlation coefficient was performed to assess the reliability of the width of maxillary lateral incisor as described by Houston[19] and the coefficient of reliability was 0.98, confirming the reliability of the measurement.

UCLP and BCLP groups were well matched in relation to chronological age (tested by Student's t-test) and gender distribution (tested by Fisher exact test). The data were analyzed with Pearson chi-squared and Fisher exact tests, and the frequency of maxillary dental anomalies in the study sample was compared with control group and published data[13, 14, 17, 20] in the same country.

Statistical analyses were performed using the SPSS software for Windows (version12.0, SPSS, Chicago, Ill). The level of significance for all tests was set at $P < 0.05$.

RESULTS

Table 1 shows chronological age and gender distribution of the patients affected by UCLP and BCLP. The groups were well matched according to chronological age and gender distribution ($p > 0.05$).

Table 1. Mean and standard deviations of chronological ages for each cleft group and gender.

Group	Gender	Age (years)	Number	Gender Comparison
UCLP	Male	13.89±3.71	19	0.197
	Female	11.95±3.44	9	
	Total	13.27±3.68	28	
BCLP	**Male**	13.77±2.74	14	0.252
	Female	15.13±2.28	8	
	Total	14.26±2.61	22	
Group comparison		0.290†	0.754*	

UCLP: Unilateral cleft lip and palate; BCLP: Bilateral cleft lip and palate; *: Results of Pearson chi-square test; †: Results of Student's t-test.

Tables 2 and 3 present distribution and comparison of the maxillary dental anomalies in relation to presence of the cleft in UCLP group and cleft types. Overall, all patients affected by UCLP and BCLP were found to have at least one maxillary dental anomaly (100 % in both groups). The most frequently observed maxillary dental anomaly was tooth agenesis in both UCLP (92.5%) and BCLP (86.4%) groups, followed by canine impaction (42.9%), microdontia of lateral incisor (17.8%), and ectopic eruption of canine (14.3%) in UCLP group and canine impaction (50.0%), ectopic eruption of canine (27.3%), and microdontia of lateral incisor (22.7%) in BCLP group. The most commonly agenesis was found to be maxillary lateral incisor in both UCLP (22/28; 78.6%) and BCLP (15/22; 68.2%) groups (13 unilateral and 9 bilateral; 7 unilateral and 8 bilateral, respectively). Four patients in BCLP group had maxillary second premolar agenesis (2 bilateral and 2 unilateral); while three patients in UCLP group had unilateral maxillary central incisor agenesis and one had bilateral maxillary second premolar agenesis. Two patients affected by UCLP (2/12) and BCLP (2/11) presented bilateral maxillary canine impaction; while remaining patients (10 patients and 9 patients, respectively) had unilateral canine impaction. All cleft patients presenting dental anomaly had unilateral ectopic eruption of maxillary canine (4 patients and 6 patients, respectively), microdontia of maxillary lateral incisor (5 patients in both groups), and supernumerary tooth (2 patients and 1 patient, respectively). Two patients affected by UCLP (7.1%) presented supernumerary tooth in the cleft are, but one (4.5%) in the BCLP group. No statistically significant difference was present for the distribution of dental anomalies in relation to cleft types ($p > 0.05$). On the other hand, tooth agenesis and canine impaction were observed more commonly in the cleft side (75.0% and 35.7%,

Table 2. Comparison of maxillary dental anomalies in cleft groups.

	UCLP (N=28)	BCLP (N=22)	P
Tooth agenesis	26/28; **92.5%**	19/22; **86.4%**	0.768*
Canine impaction	12/28; **42.9%**	11/22; **50.0%**	0.615
Ectopic eruption of canine	4/28; **14.3%**	6/22; **27.3%**	0.432*
Microdontia of lateral	5/28; **17.8%**	5/22; **22.7%**	0.937*
Supernumerary tooth	2/28; **7.1%**	1/22; **4.5%**	0.999*
Dental anomaly	28/28; 100%	22/22; 100%	

UCLP: Unilateral cleft lip and palate; BCLP: Bilateral cleft lip and palate; P: Results of Pearson chi-square test comparing dental anomaly frequency; *: Results of Fisher Exact test

Table 3. Comparison of maxillary dental anomalies between cleft and normal sides in patients with UCLP

	Cleft side	Normal side	P
Tooth agenesis	21/28; **75.0%**	16/28; **57.1%**	0.158
Canine impaction	10/28; **35.7%**	4/28; **14.3%**	0.064
Ectopic eruption of canine	2/28; **7.1%**	2/28; **7.1%**	1.00*
Microdontia of lateral	3/28; **10.7%**	4/28; **14.3%**	1.00*
Supernumerary tooth	1/28; **3.6%**	1/28; **3.6%**	1.00*

P: Results of Pearson chi-square test; *Results of Fisher Exact test

respectively) than in the normal side (57.1% and 14.3%, respectively) in patients affected by UCLP (p>0.05).

Table 4 shows the comparison of the dental anomaly distribution between our study samples and previously published studies presenting the same population without cleft. All dental anomalies were found to be higher in both cleft groups than in normal populations. Those differences were statistically significant at a level of p<0.001 except for the supernumerary tooth presence (p=0.062 for UCLP group and p=0.812 for BCLP group).

DISCUSSION

Males are known to be more commonly affected by CLP.[6,21] Baek *et al* [22] reported the ratio of males to females as 2.75:1 for UCLP. In Korean cleft patients, the ratio of males to females was about 2.52:1.[6] Akcam *et al* [4] reported a ratio of male to female as 1.92:1. In the present study, the ratios of males to females affected by UCLP and BCLP were 2.11:1 and 1.75:1, respectively (p>0.05).

In the present study, patients affected by UCLP and BCLP were assessed for the presence of dental anomalies. Other cleft types including isolated cleft lip, isolated cleft palate, and cleft lip and alveolus were not included to the study. It was underlined that epidemiological studies conducted on patients affected by CLP require classification by cleft type.[6] Since different cleft types could be related to specific patterns of deformities, the cleft type was divided into two groups as UCLP and BCLP.

In this study, patients affected by UCLP and BCLP showed at least one dental anomaly and the most commonly observed maxillary dental anomaly was hypodontia. The rate of overall dental anomaly was found to be 96.7% in the study of Akcam *et al* [4] which was higher in comparison with general populations. In the UCLP group, tooth agenesis and supernumerary tooth were higher than in the BCLP group (p>0.05); however, ectopic eruption and impaction of the maxillary canine and microdontia of the lateral incisor were more common in the BCLP group without statistically significant difference (p>0.05). The most frequently observed maxillary dental anomaly in the present study was tooth agenesis in both UCLP (92.5%) and BCLP (86.4%) groups. In agreement with the previous studies,[4, 11, 12, 24, 25] maxillary lateral incisor is the tooth most frequently missing in the UCLP (22/28; 78.6%) and BCLP (15/22; 68.2%) groups, followed by maxillary second premolar, and maxillary central incisor. The frequency of lateral agenesis in the present study was higher than previous studies[23, 26-28] but close to the finding of Akcam [4] We found that tooth agenesis in the non-cleft side in UCLP group was found to be 57.1% and maxillary lateral agenesis was the most commonly observed type of agenesis (12/28; 42.9%). According to Dewinter *et al* [23] and Brattström and McWilliams,[29] agenesis outside the cleft area was about 28.0%, which is lower that our findings. On the other hand, Akcam et al[4] reported 12.5%-52.8% agenesis in non-cleft side. These differences might be due to the severity of the cleft phenotype, which was previously shown to have correlation with the number of the affected teeth.[23] Previous studies were all performed on panoramic, occlusal films, periapical radiographs, plaster models, and intra-oral photographs. In addition to those conventional diagnosing techniques,

Table 4. Comparisons of the maxillary dental anomalies in cleft patients with previous studies.

	UCLP	BCLP	Previous Studies	P1	P2
Tooth agenesis	26/28; **92.5%**	19/22; **86.4%**	198/3341; **5.9% Celikoglu M 2010**	**<0.001***	**<0.001***
Canine impaction	12/28; **42.9%**	11/22; **50.0%**	109/2215; **4.9% Celikoglu M 2010**	**<0.001***	**<0.001***
Ectopic eruption of canine	4/28; **14.3%**	6/22; **27.3%**	26/3165; **0.8% Kazanci F 2011**	**<0.001***	**<0.001***
Microdontia of lateral	5/28; **17.8%**	5/22; **22.7%**	67/3165; **2.1% Kazanci F 2011**	**<0.001***	**<0.001***
Supernumerary tooth	2/28; **7.1%**	1/22; **4.5%**	33/3491; **0.9% Celikoglu M 2010**	0.062*	0.812*

UCLP: Unilateral cleft lip and palate; BCLP: Bilateral cleft lip and palate; P1: Results of Fisher exact test comparing the prevalence of dental anomalies in patients with UCLP and without CLP; P2: Results of Fisher exact test comparing the prevalence of dental anomalies in patients with BCLP and without CLP.

using CBCT in the present study might be another reason for the potential differences of the dental anomaly frequencies between our results and previous studies. Furthermore, comparison of the dental anomaly frequency in the cleft side and non-cleft side showed no statistically significant difference ($p>0.05$). However, tooth agenesis and canine impaction were higher in cleft side (75.0% and 35.7%, respectively) than in normal side (57.1% and 14.3%, respectively).

Previous studies[1, 4, 8, 9, 11, 27, 28] showed that dental anomalies were more commonly observed in patients affected by cleft. To our knowledge, limited number of studies[12, 25] statistically compared dental anomaly frequency in patients affected by cleft and general populations. Aizenbud *et al* [25] investigated and compared the maxillary and mandibular dental anomaly frequency in 19 patients affected by isolated cleft lip and general populations from different countries. Higher frequencies of dental anomalies in patients with isolated cleft lip were found to be statistically significant. In a controlled study, Camporesi *et al* [12] compared aplasia of lateral and premolar, peg or conoid shape associated with reduced size, supernumerary tooth and enamel hypoplasia frequencies in patients affected by UCLP and BCLP with a control group of 1000 patients without CLP and they found that statistically significant differences were present. In the present study, we found that maxillary tooth agenesis, impaction and ectopic eruption of the maxillary canine and microdontia of maxillary lateral incisor were significantly higher in patients affected by UCLP and BCLP compared to previous studies[13, 14, 17, 20, 30] in the same country.

CONCLUSION

- All patients affected by UCLP and BCLP had at least one maxillary dental anomaly.
- The most frequently observed maxillary dental anomaly was tooth agenesis in both UCLP (92.5%) and BCLP (86.4%) groups, followed by canine impaction (42.9% and 27.3%, respectively) with no significant difference.
- Tooth agenesis and canine impaction were observed more commonly in the cleft side (75.0% and 35.7%, respectively) than in the normal side (57.1% and 14.3%, respectively) in patients affected by UCLP ($p>0.05$).
- All dental anomalies were found to be higher in both cleft groups than in general populations.

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