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Dental anomalies in subjects with non-syndromic cleft lip and palate

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SUMMARY

Introduction. Cleft lip and palate (CLP) is a common congenital defect where dental anomalies are seen at a higher rate compared to the general population. The study aimed to identify, compare and correlate dental anomalies in non-syndromic cleft lip and palate subjects.

Material and methods. A total of 100 patients with cleft lip and palate were evaluated for the presence of dental anomalies. After informed consent, clinical and radiological examination of the patients was performed. Cone beam computed tomography sections were analysed and dental anomalies present were recorded. The observations were tabulated and the data was subjected to statistical analysis.

Results. Ninety six percent of the cleft population had at least one dental anomaly. Patients with unilateral cleft lip and palate showed the highest prevalence of dental anomalies. Missing teeth (87%) were the most common dental anomaly followed by ectopic eruption, microdontia, enamel hypoplasia and impacted teeth. Other dental anomalies like odontomes and pulp stones were found at a lower prevalence rate.

Conclusion. Patients with CLP are highly susceptible to the occurrence of dental anomalies. Thorough clinical and radiographic examination, careful planning and implementation of specialized services are needed in an effort to provide early diagnosis and comprehensive treatment.

Keywords: cleft lip and palate, cone beam computed tomography, missing teeth, supernumerary teeth, microdontia.

INTRODUCTION

Oral clefts and tooth germ development have a close embryological association in terms of timing and anatomical position, with critical events related to teeth, lip and palate formation occurring almost simultaneously (1, 2). The process of tooth bud formation and calcification is quite sensitive to disturbances that often have lasting impacts on the presentation of the tooth (3). Dental alterations are significantly more frequent in subjects born with oral clefts when compared to the general population (4). In addition, surgical intervention for cleft lip and palate may also cause dental anomalies such as missing teeth which, in turn, may lead to malocclusion, early development of caries, periodontal diseases and aesthetic concerns for the patients. (5) Cone beam computed tomography (CBCT) enables the examiner to view the precise location of teeth and their association with vital structures without superimposition, magnification and distortion, thus making it a favourable imaging modality for patients with CLP (6).

The purpose of this study is to determine the occurrence of dental anomalies in patients with cleft lip and palate by clinical and radiographic examination.

MATERIALS AND METHODS

A total of 100 patients with non-syndromic cleft lip, or cleft lip and palate, aged between 13-50 years, who visited our institute from 2018 to 2020 and required CBCT for further treatment were evaluated for inclusion in the study. The medical and dental records of the patients were retrieved and patients

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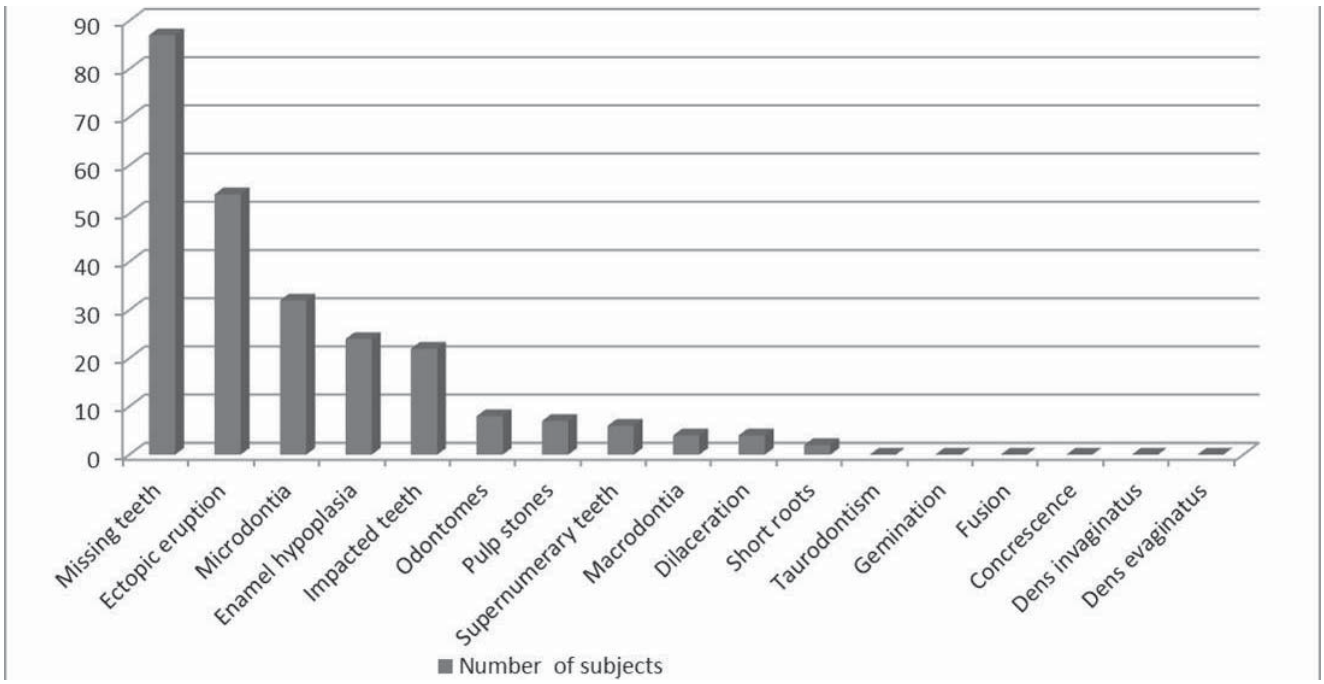

Fig. 1. Graph showing prevalence of dental anomalies among patients with cleft lip and cleft lip and palate. Missing teeth (87%) was the most common dental anomaly followed by ectopic eruption (54%), microdontia (32%), enamel hypoplasia (24%) and impacted teeth (22%). Odontomes (8%), pulp stones (7%), supernumerary teeth (6%), macrodontia (4%), dilaceration (4%) and short roots (2%) were found to be less prevalent.

who fulfilled the inclusion and exclusion criteria were included in the study.

During the period from 2018 to 2020, a total of 795 patients with cleft lip/ cleft lip and palate reported for CBCT. Among these, patients not in the age group of 13-50 years, patients who were diagnosed with syndromes based on clinical examination and medical history, patients with a history of permanent tooth extraction or orthodontic treatment based on their dental records were excluded from the study. All the patients had a history of reparative surgery for cleft lip and cleft lip and palate (CL/P). The study was carried out in accordance with the Declaration of Helsinki and ethical clearance was obtained from the institutional ethics committee. Informed written consent was obtained from all the patients before their participation in the study.

A detailed case history and thorough clinical examination of the study subjects was performed. CBCT scans of the patients with cleft were obtained using Promax 3D Mid (Planmeca, Helsinki, Finland). Standard protocols were followed: Field of view – 12×8 cm, voxel size – 0.1 to 0.2 mm and exposure time – 12 seconds. Romexis digital imaging software version 4.3 was used to evaluate the scans. Clinical examination of the study subjects and evaluation of the CBCT scans was done by a single oral and maxillofacial radiologist. Axial, sagittal, coronal and cross sections were analysed for the presence of the following dental anomalies.

Teeth absent during both clinical and radiological examination were considered as missing teeth. Teeth that were prevented from erupting by some physical barrier in their path of eruption were categorised as impacted. Microdontia was considered when teeth were smaller than the usual limits of variation while teeth larger than the usual limits of variation were considered as macrodontia. Enamel hypoplasia was diagnosed when the dental enamel showed opaque white spots, pits or grooves on the enamel surface. Taurodontism was considered where the body of the tooth was enlarged at the expense of the roots. Teeth present in addition to the regular number of teeth were categorised as supernumer-

Table 1. Distribution of dental anomalies in subjects with cleft lip

Type of cleft		URCL n (%)		ULCL n (%)	
Side		Cleft Side	Non-Cleft Side	Non-Cleft Side	Cleft Side
Missing teeth	A	-	-	-	-
	P	-	-	-	2 (100)
	M	-	-	-	-
Enamel hypoplasia	A	1 (50)	1 (50)	1 (50)	1 (50)
	P	1 (50)	1 (50)	1 (50)	1 (50)
	M	1 (50)	1 (50)	1 (50)	-

URCL – unilateral right cleft lip, ULCL – unilateral left cleft lip, A – Anteriors, P – Premolars, M – Molars. n (%) – Number of cases(Percentage).

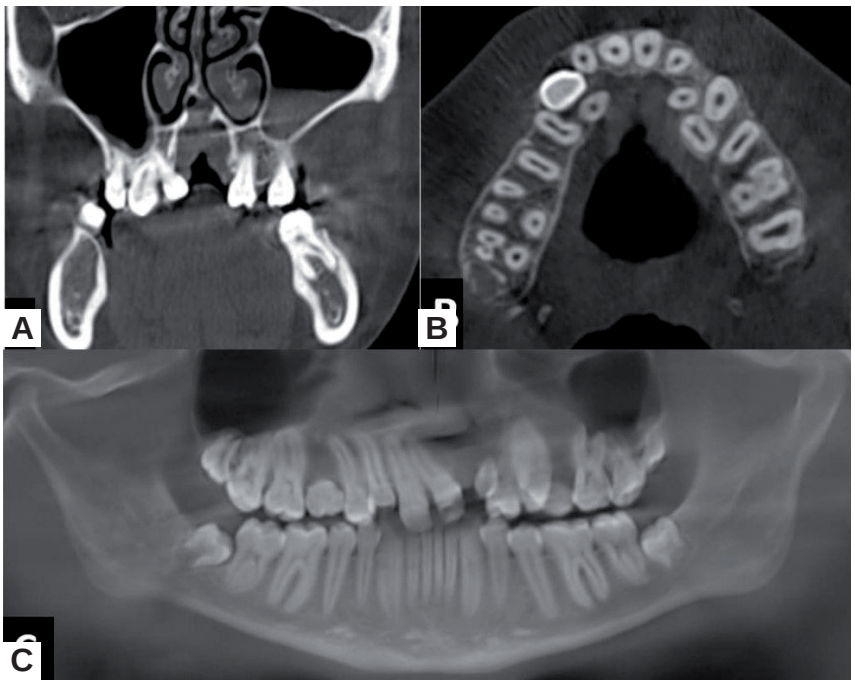

Fig. 2. A – CBCT Coronal section of a patient with cleft lip and palate showing microdontia of the maxillary teeth. B – axial section of a patient showing two supernumerary teeth; one located palatal to upper right canine and the other located palatal to the upper left lateral incisor and canine. C – panoramic reconstruction of another patient showing missing right maxillary premolars and left maxillary second premolar. Maxillary right lateral incisor and canine and maxillary left lateral incisor are reduced in size suggestive of microdontia. Retained maxillary deciduous second molars are also seen bilaterally.

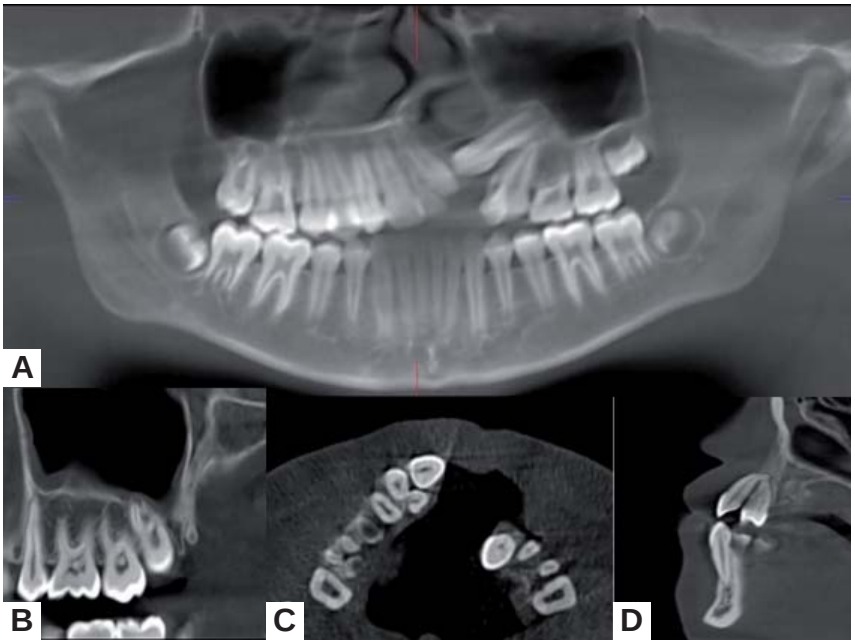

Fig. 3. A – panoramic reconstruction of a patient with cleft lip and palate showing missing maxillary left central incisor and first premolar and retained deciduous maxillary left central incisor and first premolar. Impacted maxillary left canine appears to be obliquely placed with the crown directed inferiorly. B – sagittal section of another patient with well-defined radiopaque masses in the pulp chambers of the maxillary right first and second molar suggestive of pulp stones. C – axial section of a patient showing cleft defect on the left side and missing maxillary left central and lateral incisor. The maxillary right central incisor appears larger in size suggestive of macrodontia. D: Sagittal section of a patient showing a sharp palatal bend of the apical third of the maxillary left canine indicative of dilaceration.

ary and eruption of a tooth in an abnormal position was considered ectopic eruption. CBCT scans were also evaluated for dilacerations, pulp stones, short or blunt roots, odontomes, dens invaginatus, dens evaginatus, germination, fusion and concrescence.

Cleft categorization was based on the side of the cleft (right/left), laterality of the cleft (unilateral/ bilateral) and anatomical involvement (lip/palate). In case of CLP, incisors and canines were considered as teeth inside the area of cleft whereas premolars and molars were considered as teeth outside the region of cleft. The observations were tabulated and analysed.

Statistical Analysis: Data was analysed using SPSS software version 20.0. The association between categorical variables was tested by Fischer's exact probability test.

RESULTS

The study population consisted of 43 males and 57 females with mean age of 19.6±5.3 years. There was no statistically significant difference in the prevalence of dental anomalies between males and females (p=0.226). Patients were categorized into five groups based on the type of cleft present: unilateral right cleft lip (2%), unilateral left cleft lip (2%), unilateral right cleft lip and palate (19%), unilateral left cleft lip and palate (50%) and bilateral cleft lip and palate (27%). The most common cleft type noted in the study population was unilateral cleft lip and palate (UCLP) especially on the left side. Patients with cleft lip (CL) were less affected whereas patients with UCLP showed the highest prevalence of dental anomalies. Table 1 and Table 2 show the prevalence of dental anomalies in different types of cleft lip and palate. At least one dental anomaly was noted in 96% of the study population (Figure 1). Missing teeth (87%) was the most common

dental anomaly followed by ectopic eruption (54%), microdontia (32%), enamel hypoplasia (24%) and impacted teeth (22%). Other dental anomalies like odontomes (8%), pulp stones (7%), supernumerary teeth (6%), macrodontia (4%), dilaceration (4%) and short roots (2%) were found to be less prevalent (Figure 2 and 3). Taurodontism, gemination, fusion, concrescence, dens invaginatus and dens evaginatus showed zero prevalence. In patients with UCLP, teeth on the side of the cleft were more affected. In the area within the cleft the lateral incisors were the most affected teeth while in the area outside the cleft the premolars were most affected.

DISCUSSION

Cleft lip and palate is a congenital defect with a prevalence of 1.76 to 1.81 per 1000 live births (7). Dental anomalies are notably higher in cleft population and prevalence varies across ethnic and racial groups (8-12). The present study observed the prevalence of dental anomalies in non-syndromic cleft lip and palate (NSCLP) population of South India.

In the present study, a total of 100 patients with unilateral cleft lip and unilateral or bilateral cleft lip and palate were evaluated for the presence of dental anomalies. The medical and dental records of the patients were retrieved; clinical examination was done along with evaluation of the CBCT scan of the patient. Dental anomalies seen clinically and radiographically were then recorded, tabulated and analysed statistically.

The average age of patients in the study was 19 years. Letra *et al.* (11) observed a mean age of 17.3 years while Akcam *et al.* (13) and Ajami *et al.* (14) observed a mean age of 14 years in their study population. Thus, the mean age of participants corresponds to the teenage group in whom aesthetics is a major concern. Dental anomalies are also more prevalent in the permanent dentition leading to the need for treatment. (13,14)

In the current study the prevalence of unilateral cleft lip and palate was higher (69%) compared to bilateral cleft lip and palate (27%) which is in accordance with the studies by Gupta *et al.* (15), Ajami *et al.* (14) Paranaiba *et al.* (8) and Sa *et al.* (16). Nagase *et al.* (17) reported that UCLP was common in females, while bilateral cleft lip and palate (BCLP) was common in males. In the present study, we excluded patients who were diagnosed with any

Table 2. Distribution of dental anomalies in subjects with cleft lip and palate

Type of cleft		URCLP		ULCLP		BCLP	
Side		Cleft Side n (%)	Non Cleft Side n (%)	Non Cleft Side n (%)	Cleft Side n (%)	Right n (%)	Left n (%)
Missing teeth	A	15(78.9)	5(26.3)	13(26)	32(64)	19(70.4)	23(85.2)
	P	6(31.6)	4(20)	9(18)	11(12.4)	6(22.2)	5(18.5)
	M	-	-	7(14)	-	-	-
Ectopic eruption	A	2(10.5)	3(15.8)	12(24)	8(16)	7(25.9)	4(14.8)
	P	4(21.1)	7(36.8)	4(8)	13(26)	4(14.8)	4(14.8)
	M	2(10.5)	-	-	-	-	-
Microdontia	A	1(5.3)	4(21.1)	4(8)	10(20)	5(18.5)	4(14.8)
	P	-	2(10.5)	2(4)	6(12)	3(11.1)	2(7.4)
	M	-	-	-	2(4)	2(7.4)	2(7.4)
Enamel hypoplasia	A	9(45)	6(31.6)	9(32.4)	11(20)	2(7.4)	2(7.4)
	P	7(35)	6(31.6)	8(15.3)	10(20)	2(7.4)	2(7.4)
	M	1(5)	-	-	1(2)	2(7.4)	2(7.4)
Impacted teeth	A	2(10.5)	-	2(4)	6(12)	3(11.1)	2(7.4)
	P	-	-	-	-	2(7.4)	1(3.7)
	M	-	-	3(6)	7(14)	1(3.7)	-
Odontomes	A	-	-	-	4(8)	-	2(7.4)
	P	-	-	2(4)	-	-	-
	M	-	-	-	-	-	-
Pulp stones	A	-	-	2(4)	-	-	-
	P	2(10.5)	-	1(2)	2(4)	-	-
	M	2(10.5)	-	-	5(10)	-	-
Supernumerary teeth	A	-	2(10.5)	-	2(4)	2(7.4)	2(7.4)
	P	-	-	-	-	-	-
	M	-	-	-	-	-	-
Macrodontia	A	4(21.1)	-	-	-	-	-
	P	2(10.5)	-	-	-	-	-
	M	2(10.5)	-	-	-	-	-
Dilaceration	A	-	-	2(4)	-	-	-
	P	-	-	-	-	-	-
	M	-	-	-	-	2(7.4)	-
Short roots	A	-	-	-	-	-	-
	P	2(10.5)	-	-	-	-	-
	M	2(10.5)	-	-	-	-	-

URCLP – unilateral right cleft lip and palate, ULCLP – unilateral left cleft lip and palate, BCLP – bilateral cleft lip and palate, A – anteriors, P – premolars, M – molars. n (%) – number of cases (Percentage).

syndromes and limited the study population to patients with non syndromic clefts.

Dental anomalies were more prevalent in the CLP group when compared to cleft lip group. This was also reported by Sa *et al.* (16). Thus, prevalence of dental anomalies varies with the extent and severity of the cleft (10, 13).

In the present study, among the patients with cleft lip and cleft lip and palate, the total number of dental anomalies were greater on the left side than the right side which is in agreement with studies by Gupta *et al.* (15) and Nagase *et al.* (17) The left side of the face in the developing embryo has lesser blood supply compared to the right side thus making the left side more susceptible to dental alterations (14).

In the present study population, 43% patients were males while 57% were females. There was no significant relation between dental anomalies and gender in the present study. Akcam *et al.* (13) and Paranaiba *et al.* (8) showed similar findings.

In the present study, we used a combination of clinical examination and CBCT findings to evaluate the presence of various dental anomalies. The use of CBCT, a method of 3 dimensional imaging, enabled accurate evaluation of dental anomalies. In the present study, the prevalence rate of missing teeth was 87% and was the most common dental anomaly. The lateral incisor was the most common missing tooth. This is similar to the study by Premkumar and Mohan (18) where 88.3% of the patients showed missing teeth and Akcam *et al.* (13) who observed a prevalence of 70.8 to 97.1% missing teeth. The prevalence of missing teeth is 8 times higher in the NSCLP population than the general population (19).

Unilateral cleft patients showed higher prevalence of missing teeth with the maxillary lateral incisor being the most common missing tooth which is similar to previous studies (8,16,20). In the area outside the cleft the most commonly affected teeth were the maxillary second premolars followed by the mandibular second premolars which is in accordance with previous reports (13, 18, 21).

Ectopic eruption was the second most common dental anomaly noted with a prevalence of 54% which is in conformity with the results of Premkumar and Mohan (18) where 41.7% Indian patients with NSCLP presented with ectopically erupted teeth. Al Jamal *et al.* (2) reported a prevalence of 30.8%. However, in the Brazilian population, the prevalence of ectopic eruption ranged from 2.3 to 5.9% (8, 16, 20). Higher prevalence of ectopic eruption was seen in UCLP compared to the other types of cleft. Premolars were more affected followed by anterior teeth and molars. This is in accordance with the study by Al Jamal *et al.* (2) and Sa *et al.* (16). In patients with bilateral cleft lip and palate, anterior teeth were more affected followed by the premolars and molars. This is similar to previous studies (2, 8, 13, 22). Ectopic eruption could be attributed to deficient midfacial growth due to the cleft or secondary to surgical cleft repair causing lack of alveolar continuity or due to some hindrance in the path of eruption (5, 13).

In the present study, the prevalence of microdontia was 32% in the cleft population which is similar to the prevalence reported by Al Jamal *et al.* (2) and Al Kharboush *et al.* (23). Wangsrimongkol *et al.* (24) observed a prevalence of around 41.4% in his study in Thailand. The existing studies have stated that the prevalence of microdontia in the general population ranges from 1.5 to 2% (2). Thus, when compared to the general population, the prevalence of microdontia is approximately 15 times higher in patients with cleft.

In the present study, patients with bilateral cleft lip and palate (BCLP) showed higher prevalence (37%) of microdontia followed by patients with unilateral cleft lip and palate (UCLP) (31%) which is in accordance to the study by Al Jamal *et al.* (2). However, higher prevalence of microdontia in UCLP was reported by few studies (8, 13, 25). Anterior teeth were more commonly affected followed by the premolars and molars. Lateral incisor was the most commonly affected anterior tooth and second premolar affected among premolars which is consistent with studies by Uslu *et al.* (25) and Camporesi *et al.* (26).

Microdontia could be due to insufficient blood supply secondary to surgery or due to the cleft itself. It can also be due to deficiency of mesenchymal tissue (24). Microdontia mainly causes aesthetic concerns which leads to spacing between teeth that needs to be corrected using composite build ups or orthodontic treatment (27).

In the present study, enamel hypoplasia had a prevalence of 24% which is in conformity to the study by Al Jamal *et al.* (2) who reported a prevalence of 30.8%. Premkumar and Mohan (18) found a 5% prevalence of enamel hypoplasia in a South Indian NSCLP population.

Patients with UCLP had a higher prevalence of enamel hypoplasia followed by unilateral cleft lip (UCL) and BCLP. This is similar to the findings of Paranaiba *et al.* (8) but in contrast to the study by Al Jamal *et al.* (2) where patients with BCLP showed a higher prevalence of enamel hypoplasia. Anterior teeth, especially the incisors, showed the highest prevalence of enamel hypoplasia. This is similar to

the studies by Al Jamal *et al.* (2) and Akcam *et al.* (13) and could be attributed to insufficient blood supply following surgical repair of the cleft lip and palate in this area. (5).

Impacted teeth had a prevalence of 22% in this study which is similar to the reports by Premkumar and Mohan (18) who observed a prevalence of 26.7%. In the current study, patients with UCLP had a higher prevalence of impacted teeth which is in divergence to the study by Sa *et al.* (16) in which patients with BCLP had a higher prevalence of impacted teeth (28.6%). Impacted teeth were more commonly seen on the side of the cleft which is similar to studies by Akcam *et al.* (13), Paranaiba *et al.* (8) and Premkumar and Mohan (18). Anterior teeth, mainly the canines, were more affected compared to posterior teeth. Patil *et al.* (28) in his study noted that impacted teeth had a prevalence of 16.8% in the general population which was lower than the cleft population.

In the current study, the prevalence of odontomes was 8%. Studies have rarely evaluated odontomes in the CLP population. Ajami *et al.* (14) found a prevalence of 2.5% in his study with all reported cases in the BCLP group. However, in the present study, the prevalence of odontomes was higher in patients with UCLP compared to other types of cleft. The dental lamina or developing tooth bud could get fragmented during the time of cleft reparative procedures and persist giving rise to odontomes.

In the present study, the prevalence of pulp stones was 7% which was observed only in the UCLP group where posterior teeth were more affected than the anterior teeth which is in conformity to the study by Akcam *et al.* (13) who observed a prevalence of pulp stones ranging from 8.3% to 30% across the cleft types. Turkal *et al.* (29) noted that the prevalence of pulp stones in the general population was 12.7%. Thus, presence or absence of cleft may not affect the occurrence of pulp stones (30).

In the present study, the prevalence of supernumerary teeth was 6% which was in accordance to the findings of Paranaiba *et al.* (8) where 6% of the patients showed supernumerary teeth. Premkumar and Mohan (18) reported that supernumerary teeth had a prevalence of 13.3% in the CLP population of South India. Patients with UCLP showed increased prevalence of supernumerary teeth compared to the other cleft types. Supernumerary teeth were found only in the anterior teeth region most commonly in the lateral incisor region which is in accordance with the study by Tortora *et al.* (22). Lateral incisors are present closest to the cleft defect and are most susceptible to division, modification of epithelial remnant or separation of tooth bud resulting in supernumerary teeth (31, 32). Another hypothesis is that the maxillary lateral incisors have two odontogenic origins, one from the maxillary process and the other from the medial nasal process. Failure of fusion of these processes due to the cleft gives rise to two potential odontogenic regions which can develop into supernumerary teeth (33).

Macrodontia was present in 4% of the cleft population in our study and belonged to the UCLP, which is in conformity with the study by Al-Kharboush *et al.* (23) who reported a prevalence of 3% macrodontia in the CLP population.

In the present study, the prevalence of dilacerations was 4% which is in accordance with the study by Premkumar and Mohan (18) and Wangsrimongkol *et al.* (24). A prevalence of 0.8% dilaceration was seen by Akcam *et al.* (13) while Al Jamal *et al.* (2) reported a prevalence of 19.2%. In the current study BCLP shows higher prevalence of dilaceration compared to other types of cleft which is similar to the results found by Al Jamal *et al.* (2). Dilaceration in the cleft population could be due to idiopathic disturbances during development of the tooth, trauma to the developing tooth during surgical cleft repair, presence of ankylosed or retained deciduous tooth or supernumerary tooth in the path of eruption (13).

In the present study, the prevalence of short roots was 2% with a high prevalence on the cleft side of the patients with UCLP. This is in conformity to the study by Premkumar and Mohan (18) who observed a prevalence of 8% in the South Indian cleft population. Short roots can be attributed to genetic factors, environmental factors or insufficient blood supply due to surgical repair of the cleft lip and palate in this area (34).

Existing studies were mostly retrospective where dental casts, clinical pictures and two dimensional radiographs like panoramic radiograph, intraoral periapical radiographs and bitewing radiographs were used for assessing dental anomalies in the CLP population. The present study was conducted in a prospective manner where both clinical and radiographic examination using CBCT was performed. A three dimensional radiographic examination such as CBCT is useful for the assessment of the alveolar defect, for secondary alveolar bone grafting, for the analysis of morphology of the maxilla and mandible, for assessment of the dentition, and for pre- and post- surgical assessment and can help formulate a better treatment plan in CLP patients. The use of CBCT in patients with CLP is preferred over computed tomography (CT) for its reduced radiation exposure and low cost. The

recent advances in CBCT like spatial resolution, specialized reconstruction algorithms and soft tissue contrast combined with the low radiation exposure has made CBCT the preferred imaging modality in patients with CLP (35).

CONCLUSIONS

The present study was undertaken to evaluate dental anomalies in patients with non-syndromic cleft lip and cleft lip and palate. The majority of the cleft population presented with at least one dental anomaly. Missing teeth was the most common dental anomaly followed by ectopic eruption, microdontia, enamel hypoplasia and impacted teeth. Odontomes, pulp stones, supernumerary teeth, macrodontia, dilaceration and short roots were found to be less prevalent. Patients with unilateral cleft lip and palate showed the highest prevalence of dental anomalies. Maxillary lateral incisors were the most commonly missing teeth within the region of the cleft, while maxillary second premolars were the most commonly missing teeth outside the cleft region. No statistically significant difference was noted in the prevalence of dental anomalies between males and females. Timely identification of dental anomalies in the cleft population is essential for appropriate management.

STATEMENT OF CONFLICT OF INTEREST

The authors state no conflict of interest.

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