

Original article**Prevalence and Characteristics of Developmental Dental Anomalies in Iranian Orofacial Cleft Patients**Shabnam Ajami¹, Hamidreza Pakshir¹, Hedyeh Samady²¹ Orthodontics Research Center, Dept. of Orthodontics, School of Dentistry, Shiraz University of Medical Sciences, Shiraz, Iran.² Undergraduate Student, Student Research Committee, International Branch, Shiraz University of Medical Sciences, Shiraz, Iran.**KEY WORDS**Dentition;
Abnormalities;
Cleft lip;
Cleft palate;
Prevalence;**ABSTRACT****Statement of the Problem:** Individuals with oral clefts exhibit considerably more dental anomalies than individuals without clefts. These problems could initially be among the symptoms of their disease and/or they may be the side effect of their treatments. Pushback palatoplasty could cause some interference during the development of teeth and result in tooth defects.**Purpose:** The study was performed to assess the prevalence and characteristics of developmental dental anomalies in orofacial cleft patients who attended Shiraz Orthodontics Research Center-Cleft Lip and Palate Clinic. We managed to compare dental anomaly traits based on gender and cleft side.**Materials and Method:** Eighty out of 121 cleft patients were included in this cross-sectional study. All the patients used pushback palatoplasty in their palate closure surgeries. Intraoral photographs, panoramic and intraoral radiographs, cone-beam computed tomography (CBCT) and dental and medical histories were examined and recorded by two observers. Data were analyzed using SPSS PC version 20.0. The differences in the side of cleft and dental anomalies were compared using the Mann-Whitney test.**Results:** The mean age of patients was 14.27 years (SD=5.06). The most frequent cleft type was unilateral cleft lip and palate (50%) followed by bilateral cleft lip and palate (43.75%), cleft palate (2.5%) and cleft lip (1.25%). Male predominance (70%) was observed. 92.5 percent had at least one developmental dental anomaly. The most prevalent anomalies were hypodontia (71.25%) followed by microdontia (30%), root dilacerations (21.25%) and supernumerary teeth (15%).**Conclusion:** The most prevalent cleft types were unilateral and bilateral cleft lip and palate with male and left side predominance. Hypodontia, microdontia, dilacerations and supernumerary teeth were the most prevalent developmental dental anomalies among Iranian southwestern cleft patients. The surgical technique used to repair their cleft palate may have played a role in developmental dental defects.**Corresponding Author:** Samady H., Naderkazemi Dental Clinic, International Branch of Shiraz University of Medical Sciences, Shiraz, Iran. Email: hedyeh.samady@gmail.com
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Introduction

Orofacial clefts are the most frequent congenital abnormalities of head and neck. [1] Orofacial clefts occur as a result of a failure in fusion of nasal and maxillary processes (cleft lip) or palatal shelves (cleft palate) in the

6th week of pregnancy. [2] The incidence was reported 1.03 per thousand live births in Iran. [3] The sufferers undergo many surgeries, vocal and dental treatments in their childhood. Dental problems are one of their major issues ranging from malocclusions to dentoalveolar

growth issues as well as dental anomalies and malformations.

These problems could initially be among the symptoms of their disease and/or they may be the side effect of their treatments. Operation could lead to retrognathic maxilla and tooth bud damage especially in the anterior region. Genetic and environmental factors cause higher incidence of developmental dental defects in these patients compared to general population. [4] Similarly, Eslami *et al.* [5] has reported higher prevalence of dental anomalies in cleft patients than normal population. Akcam *et al.* [4] reported 96.7% of cleft patients to have at least one dental anomaly.

The most common technique of palatoplasty in cleft lip and palate patients was Veau-Wardill-Kilner pushback palatoplasty till a few years back. In this technique, V-Y procedure retroposes the whole mucoperiosteal flap and soft palate and lengthens the palate. However, it leaves an extensive raw area along the alveolar margin to heal with secondary intention. It results in alveolar arch deformity and dental mal-alignment. [6] This method could cause some interference during the development of teeth and result in tooth defects. [7]

In a study on the Taiwanese cleft patients it was shown that the frequency of dental anomalies is associated with the cleft severity. [2] Some studies suggest that the anomalies' side is associated with the cleft side. [5] Some studies have reported hypodontia as the most frequent dental developmental anomaly in cleft patients especially in the maxillary lateral incisor area. [8-9] In cleft patients, maxillary lateral incisor area was the most common area for the supernumerary teeth, too. [10]

Investigation of these defects and their association with the cleft palate repair method is valuable in dental treatment planning. Since dental anomalies may be complicating factors in dental as well as orthodontic treatment, a detailed examination to determine the existence of anomalies is required before the initiation of orthodontic correction. This is especially true with regard to orthodontic treatment that involves extractions, which relies on healthy remaining teeth and roots to accommodate force application. [4] There is no study on dental defects of the orofacial cleft patients in south of Iran despite its high incidence. In view of the above considerations and to provide a guide for researchers and clinicians, the aim of this study was to evaluate the

prevalence of different dental anomalies and defects in different cleft types in orofacial cleft patients admitted to The Cleft Lip and Palate Clinic of Orthodontic Research Center- Shiraz University of Medical Sciences. Thus, this is not an epidemiological study.

Materials and Method

This exploratory cross-sectional analysis was carried out in accordance with the ethical standards of the 1964 *Helsinki* declaration. All patients' records were kept confidential (ethical code#Ir.sums.rec.1394.s716). We did not prescribe any radiography or CT-scan for this study and excluded patients without such records. We collected cone beam CT, panoramic, occlusal and periapical radiographs, photography and dental history prior to dental treatments of 122 non-syndromic cleft patients (79 male and 43 female). Subjects were from southwestern provinces of Iran. All the patients were operated with the Veau-Wardill-Kilner pushback palatoplasty method in two main centers in Shiraz-Iran under supervision of expert oral and maxillofacial surgeons. No extraction or trauma was reported in their dental history. All the records were examined by two observers and checked with the radiologist reports if there were any disagreements. Patients with systemic, metabolic, developmental or mental problems were considered syndromic and three patients were excluded from the study to eliminate the possible effect of the syndrome on dentition. These criteria were assessed by the physician and were recorded in their medical history. Patients were classified into 6 groups based on the following criteria [2]:

1. Unilateral cleft lip (UCL): Lip was affected in one side and the alveolar process and palate were not affected.
2. Unilateral cleft lip and alveolus (UCLA): Lip and alveolar process were involved in one side and the palate was intact.
3. Bilateral cleft lip and alveolus (BCLA): Lip and alveolar process were involved in both sides and the palate was intact.
4. Complete unilateral cleft lip and palate (UCLP): Lip, alveolar process and palate were affected in one side.
5. Complete bilateral cleft lip and palate (BCLP): Lip, alveolar process and palate were affected in both sides.
6. Cleft palate (CP): Intact lip and alveolar process with

the palate as the only affected part. [2]

We checked all the patients' permanent teeth except third molars from their records. This examination was in a standard order from upper right quadrant to the lower right quadrant by using a negatoscope in a dark room. Recorded anomalies included hypodontia, supernumerary teeth, dilacerations, microdontia, macrodontia, crown malformation, pulp stone, taurodontism, follicular cyst, tooth transposition, and odontoma. Definitions of these anomalies were considered as follows:

1. Hypodontia: when at least one developmentally missing tooth was observed.
2. Supernumerary teeth: any additional tooth more than normal series in any region of the patient's jaw. [11]
3. Dilaceration: a sudden change in the axial inclination of root or between the crown and the root of a tooth. [12]
4. Microdontia: one or more disproportionately smaller teeth.
5. Macrodontia: one or more disproportionately larger teeth. [11]
6. Crown malformation: any abnormality in the crown structure other than normal variations.
7. Pulp stone: a calcified mass in the pulp of a tooth.
8. Taurodontism: when the body of the tooth is enlarged and the roots are decreased in size. [13]
9. Follicular cyst: an increase in the size of the dental follicle of an unerupted tooth.
10. Transposition: a positional interchange of two permanent teeth within the same quadrant of the dental arch. [14]
11. Odontoma: a benign tumor composed of normal dental tissue grown in an irregular way.

Statistical Analysis

Two investigators analyzed the data blindly at the same time. Kappa score was used for inter-observer agreement. In few cases of disagreement, the radiologist's report was considered as the third party. Data were analyzed using the statistical package SPSS PC version 20.0. The differences in side of cleft and dental anomalies were compared using the Mann-Whitney Test. We considered $p < 0.05$ as statistically significant.

Results

The descriptive analysis was performed on 80 patients

who had full records. There was almost complete inter-observer agreement (Kappa value of .95). The mean age was 14.27 years (range: 8-32 years) (SD= 5.06). The most frequent defect type among these patients was UCLP (50%) followed by BCLP (43.75%), CP (2.5%) and CL (1.25%). Veau-Wardill-Kilner pushback palatoplasty was performed on all the patients at the mean age of 1.4 years. 92.5% of our samples had at least one dental developmental anomaly in their dentition. Distributions of the number of dental anomalies are presented in Table 1.

Table 1: Distribution of the number of dental developmental anomalies

Affected Teeth	0	1	2	3	4	5	6	7	Total
n	6	18	23	12	9	5	4	3	80
%	7.5	22.5	28.8	15	11.3	6.3	5	3.8	100

Hypodontia in the premolar region was observed in 30% of our cases with a total number of 42 missing second premolars, of which 27(64.3%) were maxillary and 15(35.7%) were mandibular. Hypodontia especially in the anterior region was the most frequent developmental anomaly. The most affected tooth was the upper left lateral incisor (UL2) with 32.5% of all the tooth missing. (Tables 2 and 3)

Table 2: Maxillary hypodontia

	Maxillary						Total
	UR5	UL5	UR2	UL2	UR1	UL1	
Female	4	5	11	13	1	1	35 (30.7%)
Male	9	9	14	24	4	3	63 (55.3%)
Total	13 (11.4%)	14 (12.3%)	25 (21.9%)	37 (32.5%)	5 (4.4%)	4 (3.5%)	98 (86%)

Table 3: Mandibular hypodontia

	Mandibular				Total
	LR5	LL5	LR2	LL2	
Female	3	3	0	1	7(6.1%)
Male	5	4	0	0	9(7.9%)
Total	8(7%)	7(6.1%)	0(0%)	1(0.8%)	16(14%)

We noticed that the majority of missing maxillary lateral incisor cases were on the left side (Fisher's one-sided $p = 3.2E-11$, FDR= 1.8E-09). Accordingly, second premolar missing cases were mostly observed on the right side (Fisher's one-sided $p = 4.7E-4$, FDR= 0.02). (Figure 1)

Supernumerary teeth were mostly seen in the m-

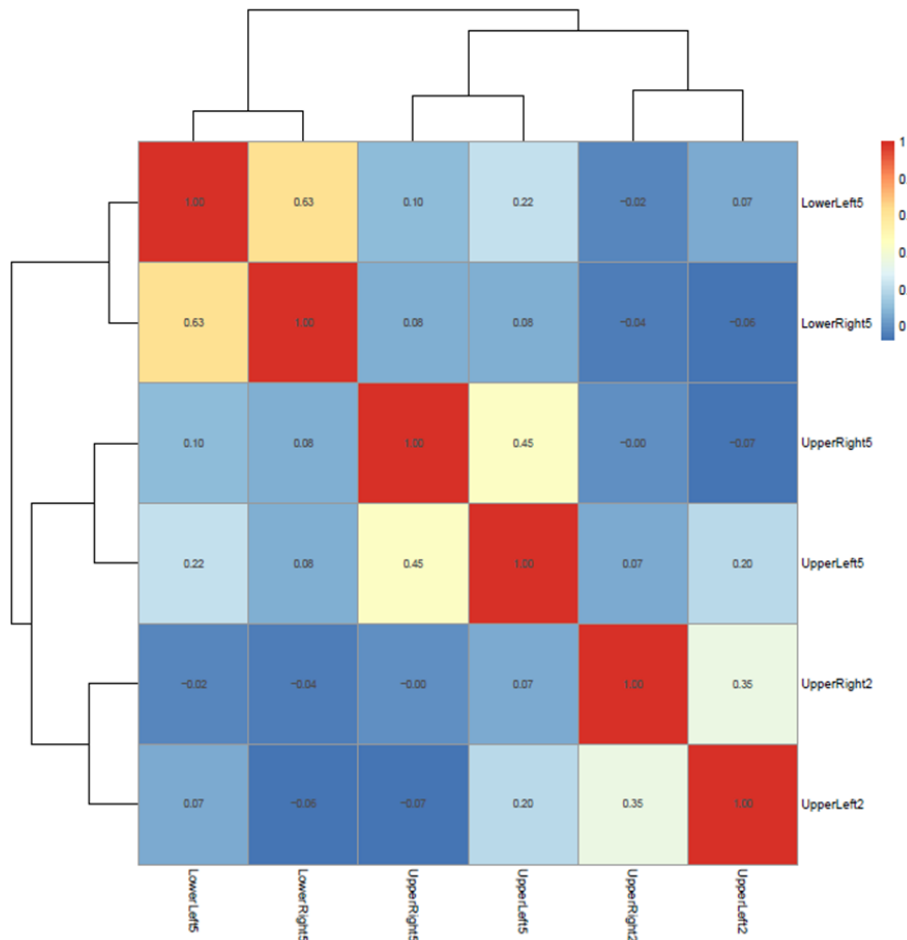


Figure 1: Correlations of the hypodontia occurrences

axillary lateral incisor area and in the male cases.

Supernumerary teeth were seen equally in both sides of the maxilla. Among microdontia cases, peg laterals were the most detected anomaly; it was nearly equal in both sides of the maxilla. (Table 4)

Table 4: Microdontia

	UR2	UL2	UR5	Total
Female	2(25%)	6(75%)	0(0%)	8(21.6%)
Male	16(55.2%)	11(37.9%)	2(6.9%)	29(78.4%)
Total	18(48.6%)	17(45.9%)	2(5.4%)	37(100%)

Table 5 shows the distribution of dental anomalies in unilateral and bilateral cleft lip and palate groups which were the most frequent cleft types among our samples. Developmental dental anomalies had higher rates in BCLP group compared with the UCLP group. Mann-Whitney test showed no significant difference between the sides of the anomalies and the cleft side ($p=0.291$).

Discussion

The prevalence and characteristics of developmental

dental anomalies in southwestern Iranian cleft patients undergone pushback palatoplasty was described in the present study. The patients in this cross-sectional study were mostly males and this is in accordance with the results of Rajabian and Aghaei’s study [15] who observed a male predominance in Iranian southwestern cleft patients. Similarly in Mashhad, a North-Eastern city of Iran, the male/female ratio was 2.3. [16] In this study, we focused on defects in permanent dentition; therefore we excluded patients younger than 8 years of age. Cleft lip and palate (CLP) were the most abundant cleft types. More than half of the defects were unilateral, 45% bilateral and 2.5% were isolated cleft palate. The unilateral defects in our samples were mostly on the left side as it was shown in Jews and Arab cleft patients of Israel. [1] This could be explained by the greater blood supply to the right side of the embryo’s face compared to the left side. [1]

As it was stated by previous studies, developmental anomalies in CLP patients occur more frequently than in non-cleft patients. [17] In the current

Table 5: Distribution of dental anomalies in the unilateral right /left cleft lip and palate (URCLP) (N=14), (ULCLP) (N=26) and bilateral cleft lip and palate (N=36)

Anomaly	Region	Right (side)		Left (side)		Right (side)	Left (side)
		URCLP n (%)	ULCLP n (%)	URCLP n (%)	ULCLP n (%)	BCLP n(%)	
Hypodontia	Incisor premolar	3 (2.6%)	7 (6.1%)	5 (4.4%)	14 (12.3%)	19 (16.7%)	22 (19.3%)
		4 (3.5%)	4 (3.5%)	4 (3.5%)	4 (3.5%)	12 (10.5%)	13 (11.4%)
Supernumerary	U2	3 (20%)	0 (0%)	2 (13.3%)	1 (6.7%)	3 (20%)	3 (20%)
Dilaceration	U1	1 (4.3%)	0 (0%)	1 (4.3%)	2 (8.7%)	5 (21.7%)	3 (13%)
	U2	1 (4.3%)	0 (0%)	1 (4.3%)	0 (0%)	2 (8.7%)	2 (8.7%)
	U3					1 (4.3%)	3 (13%)
Microdontia	U2	4 (10.8%)	2 (5.4%)	1 (2.7%)	8 (21.6%)	12 (32.4%)	8 (21.6%)
	U5	0 (0%)	2 (5.4%)	0 (0%)	0 (0%)		
Crown malformation	U1	1 (12.5%)	3 (37.5%)	0 (0%)	2 (25%)	1 (12.5%)	1 (12.5%)
Taurodontism	Molar	1 (25%)	0 (0%)	1 (25%)	0 (0%)	1 (25%)	1(25%)
Pulp stone	U1	0 (0%)	1 (33.3%)	0 (0%)	2 (66.7%)	0 (0%)	0 (0%)
Follicular cyst	U3	1 (50%)	0 (0%)	1 (50%)	0 (0%)	0 (0%)	0 (0%)
Macrodontia	ant	0 (0%)	2 (100%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Transposition	U3,4	0 (0%)	0 (0%)	0 (0%)	0 (0%)	2 (66.7%)	1 (33.3%)
Odontoma	anterior	0 (0%)	0 (0%)	0 (0%)	0 (0%)	2 (100%)	0 (0%)
Mesiodens	anterior	0 (0%)		0 (0%)		2 (100%)	

n= number of teeth

study, hypodontia was the most common anomaly especially in the incisor region and on the cleft side. Bartzela *et al.* [9] showed there were more missing teeth in the cleft quadrant than non-cleft quadrants. Similarly in this study, RUCLP patients were less likely to have missing teeth compared to LUCLP patients. [9] There were 114 missing teeth in 71.25% of the cases. This percentage was 7.66% for non-cleft orthodontic patients in Shiraz. [18] Missing of maxillary lateral incisor mostly occurs in left side, while the missing of second premolars mostly occurs in the right side. Both maxillary and mandibular second premolars tend to be missed bilaterally.

Veau-Wardill-Kilner pushback palatoplasty could have a role in the high incidence of dental developmental disruptions since it interferes with the blood supply of the developing premolars at a critical age that the permanent tooth buds are forming. In a study performed by Carpentier *et al.* [7] only the patients who had undergone pushback palatoplasty showed defects on maxillary premolars. Moreover, Lekkas *et al.* [19] investigated the absence of maxillary teeth in adult cleft lip and palate patients who did not undergo any palate repair surgeries. There was no missing of permanent teeth in the maxillary post canine area. As a result, surgery for the cleft palate repair in early childhood is the most important etiological factor for the hypodontia outside the cleft area. [19]

Today, even in the most isolated parts of Iran, cleft lip and palate patients undergo surgery within the first two years of their life, and there is no possible way for comparison with an un-treated group. However, further studies are recommended to confirm these results.

In the present study, supernumerary teeth were mostly maxillary laterals and almost all of them were observed in the male patients. Fifteen percent of the patients had at least one supernumerary tooth while the prevalence of this defect in Iranian general population was 0.74%. [20] Moreover, 3.75% of the patients had mesiodens while the prevalence of mesiodens in general population has been reported between 0.13-1.4 percent in literature. [21-24]

Microdontia was the second most common anomaly after hypodontia affecting 30% of the cases, mostly presented in the form of peg laterals. Microdontia does not seem to be among prevalent developmental anomalies in the non-cleft population. [25-26]

Root dilaceration in posterior teeth is more prevalent than anterior teeth in general population. [12] However in cleft patients, not only it is mostly in anterior teeth but also more prevalent (21.25%) than non-cleft patients. Tooth bud disturbances in the cleft area and surgery can justify this noncompliance.

Moreover, crown malformations were reported in both cleft side and non-cleft side particularly in the anterior region and on the cleft side. [4, 10] In the cur-

rent study, 7.5% of the patients had crown malformations in their maxillary central incisors.

Both taurodontism and pulp stones affected 2.5% of our patients. The rates of taurodontism and pulp stones have not been more than those in non-cleft patients. [13, 27-29] In Syrian general population, pulp stone prevalence was found to increase with age; older individuals had more pulp stones than younger individuals. [30] Racial factors may possibly affect these types of dental anomalies.

Tooth transposition has a low prevalence in the general population and it primarily affects maxillary canines and premolars. [31] Similarly, all three transposed teeth in our cases were maxillary canines and premolars.

Although follicular cysts and odontomas have been observed in current study, there does not seem to be any correlation between cysts and tumors and CLP. Also, no correlation has been mentioned in the literature and this requires an expanded investigation in cleft patients.

We did not detect a significant correlation between cleft side and tooth anomaly side. A study conducted in France showed that all dental anomalies were found in proportionately higher frequencies as the severity of the cleft increased, and they found left side predominance for hypodontia ($p < .01$) irrespective of cleft sidedness. [32]

This study is somewhat limited due to the missing data which affected the results and reduced the sample size. Extending the information sources to the dental histories and charts, intraoral photos, panoramic and intraoral radiographs and interpreted CBCTs helped obtain more accurate data and reduced errors. However with this small group, we could not generalize the results. Collaboration between centers in any region would result in a pool data and obtain more accurate results. Since it was the only accepted and tutored method of palatoplasty for the last 15 years in this center, we did not have patients with any other repair methods than pushback palatoplasty to evaluate the impact of different types of surgical repair on the type of dental anomalies. Due to the shifts of this center and many others in the methods of palatal repair in last few years, this can be proposed for the future studies.

Conclusion

In this study, we found that clefts were more frequent in left side, and most prevalent in men. The majority of the cleft types were CLP, both unilateral and bilateral. In patients with CLP, dental development is affected more frequently than in non-cleft population. The surgical technique used to repair their cleft palate may have played a role in developmental dental defects. Hypodontia and microdontia were the most associated dental developmental anomalies with cleft lip and palate. There was no significant correlation between the side of detected dental anomaly and the cleft side.

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Conflict of Interest

The authors of this manuscript certify no financial or other competing interest regarding this article.

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