

Prevalence of Maxillary Dental Anomalies and Related Factors in Children with Cleft Lip and Palate in Sari

Parastoo Namdar¹, Abbas Mesgarani², *Atena Shiva³

¹ Assistant Professor, Department of Orthodontics, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran Dental Research Center, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran.

² Associate Professor, Department of Endodontics, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran Dental Research Center, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran.

³ Associate Professor, Department of Pathology, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran Dental Research Center, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran.

Abstract

Background: Individuals with cleft lip and palate (CLP) experience a number of dental anomalies, which affect the patient's facial anatomy and self- esteem. Dental complications are considered as a contributing factor in cleft formation. The purpose of this study was to evaluate the prevalence of various kinds of maxillary dental anomalies in a group of children with CLP in the city of Sari, Iran.

Methods: This cross-sectional study was performed to evaluate the panoramic radiographs of 84 patients (7-19 years old) with CLP, who attended private and public dental and/or orthodontic clinics in Sari during 2020- 2021. The participants were selected by census method. The frequency of maxillary dental anomalies including hypodontia, microdontia and macrodontia, ectopic eruption, rotation and supernumerary tooth were examined by orthodoncy and radiology experts. Standard Pearson chi-squared tests were used for all dental anomalies. Data were analyzed using SPSS software through the Mann-Whitney and chi-squared tests.

Results: At least one maxillary dental anomaly was reported in 83.3% of patients with CLP. The unilateral CLP (61.9%) was the most common type of cleft. Tooth agenesis was observed in 63.09% of subjects with more frequencies on the left side (35.7%) followed by rotation (35.71%), microdontia (34.5%), supernumerary teeth (11.9%), and macrodontia (5.9%). No difference was observed in the prevalence of these anomalies between genders.

Conclusion: According to our results the most prevalent cleft type was unilateral cleft lip and palate (UCLP) with left side predominance. Hypodontia, rotation, and microdontia were the most prevalent developmental dental anomalies among children with CLP in Sari.

Key Words: Cleft lip, Cleft palate, Cleft lip and palate, Dental anomalies.

<u>*Please cite this article as</u>: Namdar P, Mesgarani A, Shiva A. Prevalence of Maxillary Dental Anomalies and Related Factors in Children with Cleft Lip and Palate in Sari. Int J Pediatr 2021; 9 (10): 14600-14607. DOI: **10.22038/ijp.2020.53798.4363**

Received date: Apr.13,2021; Accepted date: Apr.14,2021

^{*} Corresponding Author:

Atena Shiva, Atena Shiva, Associate Professor, Department of Pathology, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran Dental Research Center, Faculty of Dentistry, Mazandaran University of Medical Sciences, Sari, Iran. Email: atenashiva@yahoo.com

1- INTRODUCTION

Orofacial cleft lip/palate (CLP) is the most congenital malformation of craniofacial region present during birth. This defect occurs as an incomplete fusion of maxillary prominence and nasal processes (cleft lip) or palatal shelves (cleft palate) [1] It was reported that the overall incidence of CLP is 1.42 in 1000 live births [2] and 1.03 per 1000 live births in Iran [3]

The etiology of this malformation is multifactorial and scientists believe that genetic and environmental factors (e.g., ethnic, racial, geographic, socioeconomic, maternal illness, drugs and malnutrition factors) are involved in the occurrence of this anomaly [4]. The most common types of dental anomalies have been observed to be multiple missing teeth, hypodontia, supernumerary teeth, impaction, ectopic teeth, maxillary canines and premolars transposition, delayed tooth development, microdontia, crown and root malformation, and multiple decayed teeth [4]

These anomalies have ruinous effects on the dentition resulted in esthetic problems, improper phonation and impairment of mastication [5]. Additionally, CLP causes long- term disability in children as well as emotional tremendous and financial problems for affected individuals and families. Therefore, the knowledge of dental anomaly presence in CLP children could provide valuable information for pediatric and orthodontic treatment planning at an early age.

Considering the little information on dental defects of the orofacial cleft patients in north of Iran, this study was designed to evaluate the prevalence of different dental anomalies and defects among various cleft types in orofacial cleft patients at private and public dental and/or orthodontic clinics in Sari, Iran during 2020-2021.

2- MATERIALS AND METHOD

2-1. Study design and population

This retrospective cross-sectional study was performed to evaluate the panoramic radiographs in patients with CLP (age 7-19 years-old) in Sari, Iran during 2020-2021.

2-2. Inclusion and exclusion criteria

The inclusion criteria included having CLP and being in the age range of 7-19 years. Having syndromic CLP, incomplete set of dental records, extraction of the permanent tooth, history of trauma, dental implant and denture and poor quality of panoramic radiography were the exclusion criteria.

2-3. Method

The samples were selected by census method from among individuals who aged under 19 and suffered from different types of cleft palate in Sari, Iran. Initially, the records of 135 cases with CLP were found in the archive of the private and public orthodontic departments in Sari. From among this pool, 84 patients who suffered from CLP (non-syndromic CLP) and aged between 7-19 years-old were selected for the purpose of the study. The eligibility criteria were assessed by an orthodontist and recorded in their medical and dental history. Radiographies were performed by orthodontist and maxillofacial an radiologist.

Patients were classified into 3 groups based on the following criteria (14).

Diagnoses of macrodontia, microdontia and ectopic eruption were established according to criteria reported by Mallya and Lam (21).

1. and 2. Complete unilateral cleft lip and palate (UCLP) in right or left side: Lip, alveolar process and palate were affected in one side.

3. Complete bilateral cleft lip and palate (BCLP): Lip, alveolar process and palate were affected in both sides

2-4. Measuring tool: Laboratory measurements

The dental panoramic radiographs were investigated in a darkened room utilizing an illuminated x-ray viewing box and the following maxillary dental anomalies were recorded:

a. Hypodontia or tooth agenesis: It is an inherited condition characterized by the developmentally missing of 1 or more teeth [7].

b. <u>Tooth rotation</u>: Mesiolingual or distolingual intra alveolar movement of the tooth around its longitudinal [8].

c. Microdontia: When one or more teeth appear disproportionally smaller than the usual limits of variation [9].

d. Macrodontia: When one or more teeth appear disproportionally larger than the usual limits of variation [10].

e. Supernumerary teeth: Additional teeth, or hyperdontia, is defined as an excessive number of teeth as compared to normal series in any region of the patient's jaw [11].

f. Ectopic eruption: It is a trouble in which the tooth does not erupt in its normal position [12].

2-5. Ethical consideration

The study protocol was approved by the Ethics Committee of AJUMS, Sari, Iran (IR.MAZUMS.REC.1399.7521). All data were coded in order to maintain the confidentiality of information.

2-6. Data Analyses

Data were analyzed using the statistical package SPSS PC (version 11.5; SPSS Inc., Chicago, IL, USA). The differences between categorical variables were compared using the Mann-Whitney Tests. Standard Pearson chi-squared tests were used for all dental anomalies. We considered 0.05 statistically p< as significant.

3- RESULTS

This retrospective analysis was performed on 84 patients (51(60.7%) girl, 33 (39.2%) boy), who had full records. The mean age was 14.51 years (range: 7-19 years) (SD= 6.06). 52 (61.9%) subjects had UCLP (36 (42.8%) left, 16 (19%) right), 20 (23.8%) subjects had BCLP and 6 (7.1%) subjects had only cleft lip or palate separately. Patient's demographic information was presented in **Table 1**.

In general, 83.3% of our samples had at least one dental developmental anomaly in their dentition. The percentage and distributions of the number of dental anomalies are presented in **Figure 1**.

The most frequent defect type among these patients was maxillary hypodontia which occurred in 53 (63.09%) subjects; and left side (30 (35.7%)) was more affected than the right side (9 (10.7%).

Rotation was the second most commonly observed dental anomaly, occurring in 30 (35.71%) subjects. This anomaly was more prevalent in subjects with UCLP (21(25%)) than in those with BCLP (9 (10.7%)).

Microdontia was the third most commonly observed dental anomaly, occurring in 29 (34.5%) subjects, followed by ectopic eruption which was observed in 14(16.6%) subjects.

Supernumerary teeth were observed in 10 (11.9%) subjects. This anomaly was found more frequently in subjects with UCLP (7(8.3%)) than in those with BCLP (3(3.5%)). It was observed in 4 (4.7%) individuals with right UCLP and 3 (3.5%) individuals with left UCLP. Macrodontia was observed in 5 (5.9%) subjects. A total of 140 dental anomalies were observed in 70 subjects with CLP. **Table 2** presents the prevalence of dental anomalies and their distribution in clefts groups

characteristics	Unilateral left(n=36)	Unilateral right(n=16)	Bilateral (n=20)	cleft palate only (n=6)	cleft lip only (n=6)	P Value
Age(year)	13.8 ± 3.5^{1}	14.1 ± 3.4^{1}	13.7 ± 3.7^{1}	13 ± 3.3^{1}	13.8 ± 3.9^{1}	0.54^{2}
Gender(male frequency)	$12(33.3\%)^3$	7(43.7%) ³	$10(50\%)^3$	$2(33.3\%)^3$	$2(33.3\%)^3$	0.7^{4}

Table-1: Patients' Demographic information at baseline

1=Mean±SD

2= One way Anova tast with post hoc sheffe

3= Frequency

4=Chi square test



Fig. 1: Percentage of dental development anomalies

Table-2: The prevalence of dental anomalies and their distribution in cleft groups

Anomaly	Unilateral left(n=36)	Unilateral right(n=16)	Bilateral (n=20)	cleft palate only (n=6)	cleft lip only (n=6)	P value
Hypodontia	30 (83.3%)*	9 (56.25%)*	14 (70%)*	0	0	0.0001**
Supernumerary	3 (8.3%)*	4 (25%)*	3 (15%)*	0	0	0.6**
Microdontia	10 (27.7%)*	5 (31.25%)*	13 (65%)*	1 (16.6%)*	0	0.9**
Ectopic eruption	8 (22.2%)*	4 (25%)*	2 (10)*	0	0	0.053**
Macrodontia	3 (8.3%)*	2 (12.5%)*	0	0	0	0.16**
Rotation	10 (27.7%)*	11 (68.7%)*	9 (45%)*	0	0	0.27**

* Frequency

** Chi square test

UCLP: Unilateral cleft lip and palate BCLP: Bilateral cleft lip and palate

Pearson chi-squared tests showed no significant difference between the sides of the anomalies and the cleft side, except for hypodontia and the cleft side (p=0.0001);

post hoc test showed that there is a significant difference in hypodontia between unilateral left and both cleft palate only and cleft lip only (p=0.0009). Also

there is a significant difference in hypodontia between unilateral left and right (p=0.037). The relationship between anomalies and gender in children referred

to orthodontic and radiographic clinics is shown in the **Table 3**. No significant differences were observed between gender and any dental anomaly (**Table 4**).

Table-3: The association between anomalies and gender in children and adolescents referred to orthodontic and radiographic clinics

Anomaly	Boy	Girl	P value
Hypodontia	22 (26.1%)*	31 (36.9%)*	0.64**
Supernumerary	4 (4.76)*	6 (7.1%)*	1**
Microdontia	11 (13%)*	18 (21.4%)*	1**
Ectopic eruption	6 (7.1%)*	8 (9.5%)*	0.77**
Macrodontia	3 (3.5%)*	2 (2.3%)*	0.37**
Rotation	11 (13%)*	19 (22.6%)*	0.81**

* Frequency

** K square test

Table-4: The association between anomalies and gender in children and adolescents referred to orthodontic clinics

Anomaly	Sub- group	Hypodontia (n=53)	Supernumerary (n=11)	Microdontia (n=29)	Ectopic eruption (n=29)	Macrodontia (n=5)	Rotation (n=30)	P- value
Gender	Boy	22(42.3%)*	4(36.3%)*	12(41.3%)*	$6(20.6\%)^{*}$	3(60%)*	11(36.6%)*	0.58**
	Girl	31(58.4%)*	7(63.6%)*	$17(58.6\%)^{*}$	8(27.5%)*	2(40%)*	19(63.3%)*	

* Frequency

** K square test

4- DISCUSSION

This study was conducted to evaluate the prevalence of various kinds of maxillary dental anomalies in a group of children with CLP in the city of Sari, Iran. The prevalence of UCLP (61.9 %) was more than BCLP (23.8%) followed by isolated cleft lip and isolated cleft palate (7.1%). The unilateral defects were mostly on the left side in our subjects as it was also shown by Ajami et al, in southwestern Iranian cleft patients [13] and in Jew and Arab cleft patients [14]. Although the reason is not understood well, but it might be explained due to greater blood supply to the right side of the embryo's face compared to the left side [14]. It was accepted that the prevalence of oral clefts is related to gender. CLP is known to occur more frequently and severely in

male individuals, whereas isolated cleft palate is more common in females [15-17]. However, in the present study, no significant differences in dental anomalies were observed between males and females. In the same line, Al-Kharboush et al. did not report any gender difference in dental anomalies in a group of Saudi CLP patients [18]. Similarly, no statistically significant differences between genders were determined for any of the criteria studied [19]. It was believed that the of mechanisms controlling dental development are independent of sexual and somatic maturity, but may be influenced by the same factors that cause clefting [20]

As it was indicated by several studies, the dental anomalies in CLP patients occur

more frequently than in non-cleft patients [18, 19, 21].

As reported in the results, 83.3% of our samples had at least one dental developmental anomaly in their dentition and a total of 140 dental anomalies were observed in 70 subjects with CLP. Akcam et al. [22] reported dental anomalies in of cleft patients. A higher 96.7% prevalence of dental anomalies in cleft patients in comparison to the normal population was reported by Eslami et al. [23].

In the present study, the most commonly observed maxillary dental anomaly was hypodontia (63.09%), which mostly occurred in ULCP in left side. Furthermore, a significant difference was observed in hypodontia between UCLP in left and both cleft palate only and cleft lip only (p= 0.0009). Likewise, Bartzela et al. showed children with UCLP on the right side were less likely to have missing teeth compared to UCLP patients on the left side [24]. In another study, researchers indicated that by increasing the severity of cleft, all dental anomalies were found in proportionately higher frequencies and they found a left side predominance for hypodontia (p< .01) irrespective of cleft sidedness [25]. Hypodontia is a common trait in CLP patients. It was reported as the most common anomaly among Iranian southwestern cleft patients [13]

In addition, the frequency of genesis in the permanent maxillary lateral incisor varies according to the population studied [26-28]. The tooth missing in the permanent maxillary lateral incisor was the most frequent anomaly in the cleft area in UCLP patients in Italian population [29]. Fattahi and Zarvani reported the tooth missing in the upper lateral incisor in the cleft side as the most dental anomaly in Shiraz city [28].

Tooth agenesis may be explained by severe impairment of the embryonic

structures as early as the dental development phase (39). It can be caused by a deficiency in blood supply or by a surgical treatment in the cleft area [30, 31].

The tooth prevalence of rotation in subjects with UCLP (21(25%)) was more than that in BCLP subjects (9 (10.7%)). The rotation of the central incisors is caused by a lack of space at the end of the alveolar segment which implies that the premaxilla has insufficient space to accommodate the central incisors [32]

It was found that tooth agenesis is related to an overall reduction in tooth size. Consequently, hypodontia and microdontia tend to occur in the same subjects [33]. Microdontia anomaly affects 34.5% of the cases (29 subjects out of 84), mostly presented in the form of peg laterals. The prevalence of macrodontia was observed in 5 (5.9%) of UCLP subjects.

Supernumerary teeth were observed in 10 (11.9%) subjects and the frequency of supernumerary teeth was higher in subjects with UCLP (7(8.3\%)) than in those with BCLP (3(3.5\%), which is in accordance to Ajami et al.'s report [13]

Overall, according to our results, the maxillarv prevalence of rotation, microdontia, macrodontia, ectopic eruption and supernumerary teeth was more common in UCLP subjects without statistically significant differences. This study was somewhat limited due to the small sample size. Only patients with maxillary dental anomalies with CLP were included in this study and we could not generalize the results. A larger multicenter investigation is needed to obtain more accurate results.

5- CONCLUSION

In conclusion, we found that UCLP was more frequent than BCLP; and it was more prevalent in the left side among the present sample of patients from Sari. Hypodontia, was the most observed dental developmental anomaly in CLP cases. There was no significant correlation between gender and the prevalence of dental anomaly.

Patients with CLP need extensive dental care due to the presence of various dental anomalies, which makes the health care interventions more complicated. These patients should be examined carefully prior to orthodontic treatment.

6- CONFLICT OF INTEREST

The authors have no conflict of interest to declare.

7- REFERENCES

1. Keerthana, S.V., D. Saravanan, and D. Kannan, DENTAL ANOMALIES IN CLEFT LIP AND PALATE. European Journal of Molecular & Clinical Medicine, 2020. 7(4): p. 1749-1752.

2. Vyas, T., et al., Cleft of lip and palate: A review. Journal of Family Medicine and Primary Care, 2020. 9(6): p. 2621.

3. Rajabian, M.H. and M. Sherkat, An epidemiologic study of oral clefts in Iran: analysis of 1669 cases. The Cleft palate-craniofacial journal, 2000. 37(2): p. 191-196.

4. Mednick, L., et al., Causal attributions of cleft lip and palate across cultures. The Cleft Palate-Craniofacial Journal, 2013. 50(6): p. 655-661.

5. Hardin-Jones, M.A. and D.L. Jones, Speech production of preschoolers with cleft palate. The Cleft palate-craniofacial journal, 2005. 42(1): p. 7-13.

6. Stewart, C., J. Michael, and F. White, Principles of oral radiology in 2004. Translated by Talaei Pour A. Tehran, Shayan Chart.

7. Al-Ani, A.H., et al., Hypodontia: an update on its etiology, classification, and clinical management. BioMed Research International, 2017. 2017.

8. Baccetti, T., Tooth rotation associated with aplasia of nonadjacent teeth. The Angle Orthodontist, 1998. 68(5): p. 471-474.

9. Laverty, D. and M. Thomas, The restorative management of microdontia. British Dental Journal, 2016. 221(4): p. 160-166.

10. Pace, A., P.J. Sandler, and A. Murray, Macrodont management. Dental Update, 2013. 40(1): p. 18-26.

11. Rao, P. and M. Chidzonga, Supernumerary teeth: literature review. Cent Afr J Med, 2001. 47(1): p. 22-6.

12. Yaseen, S.M., S. Naik, and K. Uloopi, Ectopic eruption-A review and case report. Contemporary Clinical Dentistry, 2011. 2(1): p. 3.

13. Ajami, S., H. Pakshir, and H. Samady, Prevalence and characteristics of developmental dental anomalies in Iranian orofacial cleft patients. Journal of Dentistry, 2017. 18(3): p. 193.

14. Shapira, Y., et al., Prevalence of nonsyndromic orofacial clefts among Jews and Arabs, by type, site, gender and geography: a multi-center study in Israel. The Israel Medical Association Journal: IMAJ, 2014. 16(12): p. 759-763.

15. Al-Balkhi, K.M., The distribution and classification of clefts in patients attending a cleft lip and palate clinic in Riyadh, Saudi Arabia. Saudi medical journal, 2008. 29(5): p. 739-742.

16. Baek, S.-H. and N.-Y. Kim, Congenital missing permanent teeth in Korean unilateral cleft lip and alveolus and unilateral cleft lip and palate patients. The Angle Orthodontist, 2007. 77(1): p. 88-93.

17. Menezes, L.M.d., et al., Characteristics and distribution of dental anomalies in a Brazilian cleft population. Revista Odonto Ciência, 2010. 25(2): p. 137-141.

18. Al-Kharboush, G.H., K.M. Al-Balkhi, and K. Al-Moammar, The prevalence of specific dental anomalies in a group of Saudi cleft lip and palate patients. The Saudi dental journal, 2015. 27(2): p. 75-80.

19. Celikoglu, M., et al., Maxillary dental anomalies in patients with cleft lip and palate: a cone beam computed tomography study. Journal of Clinical Pediatric Dentistry, 2015. 39(2): p. 183-186. 20. Demirjian, A., H. Goldstein, and J.M. Tanner, A new system of dental age assessment. Human biology, 1973: p. 211-227.

21. Lai, M.C., N.M. King, and H.M. Wong, Abnormalities of maxillary anterior teeth in Chinese children with cleft lip and palate. The Cleft Palate-Craniofacial Journal, 2009. 46(1): p. 58-64.

22. Akcam, M.O., et al., Dental anomalies in individuals with cleft lip and/or palate. The European Journal of Orthodontics, 2010. 32(2): p. 207-213.

23. Eslami, N., et al., Prevalence of dental anomalies in patients with cleft lip and palate. Journal of Craniofacial Surgery, 2013. 24(5): p. 1695-1698.

24. Bartzela, T.N., et al., Tooth agenesis patterns in unilateral cleft lip and palate in humans. Archives of oral biology, 2013. 58(6): p. 596-602.

25. Matern, O., et al., Left-sided predominance of hypodontia irrespective of cleft sidedness in a French population. The Cleft Palate-Craniofacial Journal, 2012. 49(3): p. 1-5.

26. Aasheim, B. and B. Ögaard, Hypodontia in 9-year-old Norwegians related to need of orthodontic treatment. European Journal of Oral Sciences, 1993. 101(5): p. 257-260.

27. Horowitz, J.M., Aplasia and malocclusion: a survey and appraisal. American journal of orthodontics, 1966. 52(6): p. 440-453.

28. Fattahi, H. and N. Zarvani, A Study of Dental Anomalies in Patients with Cleft Lip and/or Palate in Few Dental Clinics of Shiraz in 1383. Journal of Dentistry, 2004. 5(1, 2): p. 79-87.

29. Camporesi, M., et al., Maxillary dental anomalies in children with cleft lip and palate: a controlled study. International Journal of Paediatric Dentistry, 2010. 20(6): p. 442-450.

30. Lekkas, C., et al., The adult unoperated cleft patient: absence of maxillary teeth outside the cleft area. The Cleft palate-craniofacial journal, 2000. 37(1): p. 17-20.

31. Jiroutova, O. and Z. Müllerová, The occurrence of hypodontia in patients with cleft lip and/or palate. Acta chirurgiae plasticae, 1994. 36(2): p. 53-56.

32. Šmahel, Z., M. Tomanová, and Ž. Müllerová, Position of upper permanent central incisors prior to eruption in unilateral cleft lip and palate. The Cleft palate-craniofacial journal, 1996. 33(3): p. 219-224.

33. Baccetti, T., A clinical and statistical study of etiologic aspects related to associated tooth anomalies in number, size, and position. Minerva stomatologica, 1998. 47(12): p. 655-663.