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Examination of the Pattern of Hypodontia in Cleft Lip and Cleft Palate Patients

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Objectives Tooth agenesis or hypodontia is a major problem that may occur in any person in any area of the mouth. However, it is of greater significance in patients with cleft lip and palate (CLP) because of their susceptibility to various problems. This study aimed to investigate the prevalence of tooth agenesis in both jaws among patients with CLP.
Methods In this descriptive analytical study, a total of 150 unilateral CLP and 30 bilateral CLP patients (age range, 4-19 years) were randomly selected, and their panoramic radiographs were examined. The collected data were analyzed using Chi-square test, Mann-Whitney test, Fisher's exact test, and logistic regression analysis.
Results The results of Chi-square test revealed that the prevalence of tooth agenesis was significantly higher in unilateral CLP patients compared to their bilateral CLP counterparts (P=0.039). The maxillary lateral incisors (26%), followed by right maxillary first premolars (20%), accounted for the highest number of hypodontia. The results of Mann-Whitney test did not indicate any significant differences in the number of hypodontia between unilateral and bilateral CLP patients (P=0.158).
Conclusion The frequency of tooth agenesis in the evaluated CLP population of Isfahan (Isfahan Province, Iran) was similar to that of other populations around the world. According to the present findings, unilateral CLP was more frequent than bilateral CLP, and left-sided CLP was more frequent than right-sided CLP and bilateral CLP.
Keywords Hypodontia; Cleft lip; Cleft palate; Prevalence

Introduction

Cleft lip and palate (CLP) is the most frequent congenital craniofacial abnormality, with a mean prevalence of 1:500 to 1:700 in live births. [1] Generally, CLP patients are exposed to multiple problems due to their condition, and alleviation of the functional and aesthetic consequences of CLP is particularly challenging. CLP is accompanied by a wide range of dental anomalies, which have long-term effects on the patient's facial anatomy and self-esteem. Dental anomalies have been shown to occur more frequently in CLP patients compared to the normal population. [2, 3]

Hypodontia, which is described as the congenital absence of one or more primary or permanent teeth, is the most frequently reported dental anomaly in CLP patients, associated with different types of clefts. [4] According to several studies, the incidence of hypodontia is three times higher in patients with CLP compared to the normal population. [5] In the general population, the prevalence of hypodontia (except third molars) varies from 0.027% to 11.3%, with most studies reporting a range of 2% to 7%. [6] A recent review of hypodontia in the normal population reported a prevalence of 0.15-16.2% in permanent teeth, with third molars excluded. [7] On the other hand, studies on CLP patients have reported significantly higher prevalence rates, reaching 29.5% to 77%. [8] The present study aimed to investigate the prevalence of hypodontia in both jaws among patients with CLP.

Methods and Materials

This descriptive analytical study was performed on panoramic radiographs of the patients with non-syndromic unilateral or bilateral CLP, who were referred to Cleft Palate Clinic of Isfahan University of Medical Sciences during 2010-2020. The study sample consisted of panoramic radiographs of 150 patients, including 120 unilateral CLP and 30 bilateral CLP patients (age range, 4- 19 years; average age, 10.3 years) (Table 1).

Table 1 - Frequency and Percentage of hypodontia and cleft type in CLP patients

	Cleft type	Frequency	Percentage
CLP	Right side	55	36.7
	Left side	65	43.3
	Bilateral	30	20.0
	Sum	150	100.0

The following formula was used to determine the sample size:

$$n = \frac{{z_{\alpha/2}}^2 \times (p(1-p))}{\delta^2}$$

The inclusion criteria were as follows: diagnosis of unilateral or bilateral CLP; absence of syndromes or systemic diseases; availability of medical records; and availability of complete patient information, such as age, sex, and history of all treatment procedures and follow-ups. The prevalence of hypodontia was quantified according to the tooth type, mouth quadrant, and maxillary/mandibular area. All panoramic radiographs examined in this study

were investigated by one trained operator. Hypodontia was defined as the lack of any differentially calcified tissue in the area of the corresponding tooth. The third molars were also included in the assessments.

The collected data were analyzed using Chi-square test, Fisher's exact test, Wilcoxon test, Mann-Whitney test and logistic regression analysis in SPSS Version 22, and the significance level was set at 0.05.

Result

The most frequent hypodontia in the CLP population were the maxillary lateral incisors (26%). In unilateral CLP patients, the most frequently reported hypodontia were the maxillary lateral incisors (27.5%) on the cleft side, while in bilateral CLP patients, the maxillary premolars were the most frequent hypodontia (26.7%) (Table 2).

In this study, the results of Fisher's exact test showed no significant difference regarding the prevalence of hypodontia between unilateral and bilateral CLP patients (P>0.05) (Table 3).

Table 2 - Frequency and percentage of hypodontia in maxilla and mandible in CLP patients

Maxilla			Mandible		
Tooth Number	Frequency	Percentage	Tooth Number	Frequency	Percentage
21	4	2.7	34	8	5.3
11	7	4.7	44	11	7.3
22	39	26.0	35	1	0.7
12	39	26.0	45	10	6.7
23	16	10.7	36	1	0.7
13	10	6.7	37	3	2.0
24	29	19.3	47	3	2.0
14	30	20.0	38	20	13.3
25	17	11.3	48	14	9.3
15	23	15.3	46	0	0
26	3	2.0	31	0	0
16	3	2.0	41	0	0
27	7	4.7	32	0	0
17	2	1.3	42	0	0
28	30	20.0	33	0	0
18	20	13.3	43	0	0

Table 3 - Comparison of the frequency and percentage of missing teeth in unilateral and bilateral CLP patients

Tooth Number	Unilateral cleft n=120		Bilateral cleft n=30		P-value
	Frequency	Percentage	Frequency	Percentage	
34	8	6.7	0	0.0	0.358 [b]
44	11	9.2	0	0.0	0.122 [b]
35	1	0.8	0	0.0	1.00 [b]
45	10	8.3	0	0.0	0.213 [b]
36	0	0.0	1	3.3	0.200 [b]
37	3	2.5	0	0.0	1.00 [b]
47	3	2.5	0	0.0	1.00 [b]
38	19	15.8	1	3.3	0.079 [b]
21	3	2.5	1	3.3	1.00 [b]
11	5	4.2	2	6.7	0.627 [b]
22	33	27.5	6	20.0	0.490 [a]
12	33	27.5	6	20.0	0.490 [a]
23	11	9.2	5	16.7	0.317 [b]
13	10	8.3	0	0.0	0.213 [b]
24	22	18.3	7	23.3	0.606 [a]
14	23	19.2	7	23.3	0.615 [a]
25	16	13.3	1	3.3	0.196 [b]
15	15	12.5	8	26.7	0.085 [b]
26	3	2.5	0	0.0	1.00 [b]
16	3	2.5	0	0.0	1.00 [b]
27	7	5.8	0	0.0	0.346 [b]
17	2	1.7	0	0.0	1.00 [b]
28	24	20.0	6	20.0	1.00 [a]
18	17	14.2	3	10.0	0.766 [b]

[a] Chi-square test [b] Fisher exact test

According to the results of Wilcoxon test, in unilateral CLP patients, the number of hypodontia on the cleft side was significantly higher than the non-cleft side (P<0.001), and the number of hypodontia was significantly higher in the

maxillary arch as compared to the mandibular arch (P<0.001) (Table 4).

Generally, in all CLP patients, there was no significant difference in the number of hypodontia on the right and left sides (P=0.166), and the number of hypodontia in the maxilla was significantly higher than the number of hypodontia in the mandible (P<0.001) (Table 4).

In bilateral CLP patients, there was no significant difference in the number of hypodontia between the right and left quadrants (P=0.655). However, the number of hypodontia was significantly higher in the maxilla compared to the mandible (P<0.001) (Table 4).

Table 4- Comparison of the descriptive and analytical statistics of hypodontia in unilateral and bilateral CLP patients

Cleft	Side/Jaw	n	Minimum	Maximum	Mean	Std. deviation	P-value [a]
Unilateral Right side	Right side	65	0.0	6.00	1.42	1.43	<.001
	Left side	65	0.0	5.00	1.02	1.53	
	Maxilla	65	0.0	7.00	2.03	2.01	<.001
	Mandible	65	0.0	4.00	0.40	0.93	
Unilateral Left side	Right side	55	0.0	4.00	0.98	1.05	0.088
	Left side	55	0.0	6.00	1.53	1.50	
	Maxilla	55	0.0	7.00	1.73	1.52	<0.001
	Mandible	55	0.0	4.00	0.78	1.38	
Bilateral	Right side	30	0.0	4.00	0.87	1.11	0.655
	Left side	30	0.0	4.00	0.93	1.11	
	Maxilla	30	0.0	7.00	1.73	1.93	<0.001
	Mandible	30	0.0	1.00	0.07	0.25	
Sum	Right side	150	0.0	6.00	1.15	1.26	0.166
	Left side	150	0.0	6.00	1.19	1.46	
	Maxilla	150	0.0	7.00	1.86	1.82	<0.001
	Mandible	150	0.0	4.00	0.47	1.07	

[a] Wilcoxon test

In unilateral CLP cases, the number of hypodontia was in the range of 0-11 (mean, 2.47±2.44), while in bilateral CLP patients, it was in the range of 0-7 (mean, 1.80±1.99). The results of Mann-Whitney test did not indicate any significant differences in the number of hypodontia between unilateral and bilateral CLP cases (P=0.158) (Table 5).

The results of Chi-square test showed a significant relationship between the presence of hypodontia and the type of CLP (P=0.039); in other words, the frequency of hypodontia was significantly higher in unilateral CLP patients compared to the bilateral CLP cases (Table 6).

Table 5 - Descriptive and analytical statistics of hypodontia in unilateral and bilateral CLP patients

Cleft type	n	Minimum	Maximum	Mean	Std. deviation	P-value [a]
Unilateral	120	0.0	11.00	2.47	2.44	0.158
Bilateral	30	0.0	7.00	1.80	1.9	
Sum	150	0.0	11.00	2.33	2.37	

[a] Mann-Whitney test

Table 6 - Frequency and percentage of hypodontia in unilateral and bilateral CLP patients

hooo T gissii M	Unilateral		Bilateral		muS		P-value [a]
	Frequency	Percentage	Frequency	Percentage	Frequency	Percentage	
oo	26	21.7	12	40.0	38	25.3	0.039
ses	94	78.3	18	60.0	112	74.7	
muS	120	100.0	30	100.0	150	100.0	

[a] Chi-square test

Additionally, the results of Chi-square test did not show asignificant relationship between the presence of hypodontia and the type of unilateral CLP (in the affected side) (P=0.506)(Table 7).

Table 7 - Frequency and percentage of hypodontia in unilateral CLP patients

gissiM hoooT	Unilateral Right side		Unilateral Left side		muS		P-value [a]
	Frequency	Percentage	Frequency	Percentage	Frequency	Percentage	
oo	16	24.6	10	18.2	26	21.7	
ses	49	75.4	45	81.8	94	78.3	0.506
muS	65	100.0	55	100.0	120	100	

[a] Chi-square test

Discussion

Hypodontia may lead to complications, such as aesthetic problems and chewing difficulties. Anterior teeth play a significant role in facial aesthetics, and their absence can have psychological consequences for the individual. Canine teeth also play an essential functional role in dental arches. Besides, the absence of premolar teeth can cause problems, such as space deficiency, physiological drift of adjacent teeth, and ultimately, occlusal disorders. Diagnosis and management of hypodontia should be performed as early as possible to prevent craniofacial developmental disordersand improve the individual's quality of life. [9]

Based on the present results, pertaining to the frequency of CLP types, determination of the most common hypodontia can help dental professionals find a more effective intervention for CLP patients. In this study, 80% of the studied patients had unilateral clefts, while the rest (20%) had bilateral clefts. In other words, the frequency of unilateral CLP was higher than that of bilateral CLP; besides, in unilateral CLP patients, left-sided clefts were more common than the right-sided ones. Although the frequency reported in our study differs from the findings of a study by Ajami et al. [13], it is consistent with the results reported by Neville et al. [12]. It is worth mentioning that in the study by Ajami et al.[13], 45% of patients had bilateral clefts, while 42.5% had unilateral clefts.

According to the current research, the most common hypodontia in CLP patients were teeth 22, 12, 14, and 28, while teeth 35 and 36 showed the lowest frequencies. The most common hypodontia in unilateral CLP patients were the maxillary lateral incisors (27.5%) on the cleft side. On the other hand, in bilateral CLP patients, the maxillary premolars (26.7%) were the most common hypodontia. Although these results are consistent with the findings of some previous studies [5, 10], Camporesi et al. [11] reported that the most common hypodontia in bilateral CLP patients were the lateral incisors; this finding differs from our study, as maxillary premolars were the most frequently reported hypodontia in bilateral CLP patients. Consistent with our results, Ajami et al. [13] showed that the missing lateral incisors on the cleft side were among the most frequent anomalies in CLP patients.

Additionally, in a study conducted in Norway, Shetye et al. [14] concluded that the absence of the left lateral incisor, rotation and absence of the right incisor, and rotation of central incisors were the most common occurrences in CLP patients, which is in line with the current study. The right and left maxillary lateral incisors were the most commonly reported hypodontia; therefore, the results of these two studies are compatible. [15] Moreover, Dahllof et al. [16] found that tooth development was slower in CLP patientscompared to the normal population.

In the present study, hypodontia was found in 78% of CLP patients, which is consistent with the findings of a study by Lisson et al. [17]. In other words, the frequency of hypodontia was significantly higher in unilateral CLP patients as compared to their bilateral CLP counterparts. This finding is inconsistent with the results reported by Ajami et al. [13], which showed that hypodontia was more common in bilateral CLP cases than unilateral CLP cases.

Conclusion

The frequency of hypodontia and cleft types in the studied CLP population of Isfahan was similar to other populations around the world. Based on the present findings, unilateral CLP was more frequent than bilateral CLP, and left-sided CLP was more frequent than right-sided CLP and bilateral CLP. Compared to the general population, CLP patients require special dental care, as well as a more specific treatment plan to meet their aesthetic and functional needs.

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Conflict of Interest

No conflict of interest declared ■

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