

Abstract

Background: Cleft lip and palate (CLP) is the most common orofacial congenital malformation in live births. CLP can occur individually or in combination with other congenital deformities. Affected patients experience a number of dental, aesthetic, speech, hearing, and psychological complications and have a higher incidence of severe dental conditions. The purpose of this study is to characterise the different types of dental anomalies that are frequently associated with CLP patients based on a literature survey.

Methods: By literature survey, this study characterises the different types of dental anomalies that are frequently associated with cleft lip and palate patients.

Results: Common dental anomalies associated with CLP are supernumerary tooth, congenitally missing tooth, delayed tooth development, morphological anomalies in both deciduous and permanent dentition, delayed eruption of permanent maxillary incisors, microdontia, and abnormal tooth number.

Conclusion: The incidence of certain dental anomalies is strongly correlated with Cleft lip and palate, a finding that is consistent with previous studies.

Keywords: dental anomalies, cleft lip palate, hypodontia, supernumerary tooth

Introduction

The overall incidence of cleft lip and palate (CLP) is approximately 1 in 700 live births (1), making CLP the most common orofacial congenital malformation (2). Affected patients suffer a multitude of problems, and alleviating the functional and aesthetic consequences of CLP is particularly challenging. CLP is accompanied by a wide variety of dental anomalies, which also have a long-term impact on the patient's facial anatomy and self-esteem (3). Dental anomalies are considered a contributing factor in cleft formation (4).

The incidence of dental anomalies is markedly increased in children with CLP compared to the general population (5). Generally, the specific anomaly varies according to the CLP category (6). Studies have shown that both permanent and deciduous teeth may be affected, and that dental anomaly occurs more frequently on the cleft side (7). The maxillary lateral incisors are the most susceptible to dental anomalies within the cleft region (3).

The purpose of this study is to describe the different types of dental anomalies frequently associated with CLP.

Methods

Considering the importance of dental anomalies in cleft lip and palate cases, a literature search was conducted using several electronic databases including Medline-PUBMED, Science Direct, and Google Scholar search engines. The MeSH search terms were cleft lip and palate, and dental anomaly. Title search terms were cleft lip palate and one of the following: anomalies, malformations, supernumerary, missing, congenital, delayed eruption, ectopic, hypodontia, impaction, transposition, and microdontia. There was no language preference. Both original research articles and literature reviews were included. In addition, the Cleft Palate Craniofacial Journal was manually searched, and the reference lists of relevant publications were comprehensively searched to identify any previously overlooked articles.

Results

The most common dental anomalies found in CLP patients are: multiple missing teeth/hypodontia/agenesis (usually the maxillary lateral incisors); ectopic teeth; impaction;

supernumerary teeth; microdontia; maxillary canines and premolars transposition; delayed development; crown and root malformation; and multiple decayed teeth.

The results of the literature survey of dental anomaly in CLP patients are summarised in Table 1.

Discussion

According to cleft severity, the three most common dental anomalies were missing maxillary lateral incisors, supernumerary teeth, and missing lower incisors. The maxillary lateral incisor was the most frequently affected tooth in the cleft area (6). In a study evaluating dental anomalies in Brazilian cleft patients, male patients had a higher incidence of CLP, agenesis, and supernumerary teeth than did female patients. In cases of complete CLP, the left maxillary lateral incisor was the most commonly absent tooth. Supernumerary teeth were typically located distal to the cleft (8).

A parallel study was conducted comparing the dental anomaly prevalence in the primary and permanent dentition of patients with unilateral cleft lip and palate (UCLP) and bilateral cleft lip and palate (BCLP). A total 96 cases (67 UCLP cases and 29 BCLP cases) were examined, and a high prevalence of dental anomaly was observed in both primary and permanent teeth. A total 93% of UCLP cases and 96% of BCLP cases presented with at least one dental anomaly, and the patient groups showed significant differences in the prevalence of single missing tooth (UCLP = 39%, BCLP = 14%), multiple missing teeth (UCLP = 22%, BCLP = 54%), and anterior malocclusion (UCLP = 15%, BCLP = 41%) (9).

Menezes and Vieira (10), reviewed the radiographic and medical records of 146 subjects with CLP. In 47 (32.19%) subjects, at least one dental anomaly (such as agenesis, microdontia, impaction, and structural anomalies) was identified outside of the cleft region, and subjects with complete CLP showed more dental anomalies than those with incomplete CLP. A higher rate of mandibular premolar anomalies was observed in cleft palate patients than in those with CLP. The most commonly affected teeth were the maxillary lateral incisors and maxillary premolars. The agenesis incidence was also elevated in the maxillary second premolar of subjects with BCLP or UCLP, and 12.5% of subjects showed dental anomalies in the maxillary lateral incisor of the non-cleft side.

In a study of Jordanian subjects, the prevalence of dental anomaly was higher in

CLP patients than in normal subjects. Missing teeth were observed in 66.7% of patients, with maxillary lateral incisor as the most frequently affected tooth. Supernumerary teeth were observed in 16.7% of patients; other findings included microdontia (37%), taurodontism (70.5%), transposition or ectopic teeth (30.8%), dilacerations (19.2%), and hypoplasia (30.8%). The incidence of microdontia, dilaceration, and hypoplasia was significantly higher in bilateral CLP patients than in unilateral CLP patients, and none of the anomalies showed any significant sexual dimorphism (11).

In another study, the data analysis showed that the dental anomalies in cleft patients were located in various neural crest regions. There was a vast variation in the incidence of agenesis, supernumerary teeth, and malformation between the different cleft types. Supernumerary lateral incisors were considerably more frequent in patients with cleft lip than in other cleft types. Malformed roots were significantly more frequent in those with cleft palate. In patients with both cleft lip and palate, the number and type of dental anomalies differed significantly from those in other cleft types; for example, CLP patients had significantly more agenesis (12).

In a study performed in Athens, Greece on children and adolescents, the oral health profile and dental anomaly incidence were compared between CLP patients and control subjects. In the CLP patients, 9.8% had at least one supernumerary tooth, while none of the control group showed supernumerary teeth (13). A clinical study of 76 patients with right unilateral transforamen cleft (15.8%), left unilateral transforamen cleft (40.8%), bilateral transforamen cleft (32.9%) and post-foramen cleft (10.5%) conducted by the Brazilian Dental Association in the Paraíba state in north-eastern Brazil found agenesis in 31.6% of patients, conical teeth in 28.9%, supernumerary teeth in 13.2%, ectopic teeth in 5.3%, rotated teeth in 13.2%, impacted teeth in 14.5%, and a twin tooth in 1.3% (one patient) (14).

Consistent with previous reports, a study conducted in Sweden, Holland, and Norway analysing 240 panoramic radiographs of patients with complete BCLP found agenesis affecting at least one tooth in 59.8% of patients, and observed a higher frequency in lateral incisors and second premolars (15). Shetty et al. (16) analysed the incidence of incisal anomalies in 113 untreated UCLP patients. They reported a higher percentage of absent left lateral incisors (48.7%), rotated right lateral incisors (22.1%), missing right lateral incisors (21.2%), and rotated central incisors

Table 1: Incidence of dental anomalies in patients with cleft lip and/or palate based on a literature survey

Author name and year	Cleft type *	No of subjects	Dental anomalies found
Menezes and Vieira (10), 2008	Complete CLP	146	Agenesis
	Incomplete CLP		Microdontia
	CP		Impacted tooth
	UCLP		Structural anomalies
	BCLP		
da Silva et al. (17), 2008	Complete BCL	150	Hypodontia
	Incomplete BCL		a. Complete BCL (31.6%)
			b. Incomplete BCL (26.8%)
			Supernumerary teeth
	a. Complete BCL (28.2%) b. Incomplete BCL (29.2%)		
Parapanisiou et al. (13), 2009	CLP	41	Supernumerary tooth (9.8% in CLP)
	HLP	41	
Al Jamal et al. (11), 2010	UCLP	78	Agenesis (66.7%)
	BCLP		Supernumerary teeth (16.7%)
			Microdontia (37%)
			Taurodontism (70.5%)
			Transposition and/or ectopic teeth (30.8%)
			Dilacerations (19.2%)
Hypoplasia (30.8%)			
Menezes et al. (8), 2010	CLA	200	Agenesis (66.5% overall; MLI affected in 78.5% of lesions)
	CLP		
	CP		
Bartzela et al. (15), 2010	BCLP	240	Agenesis (59.8%)
Tereza et al. (18), 2010	Complete BCLP	205	Hypodontia (70.2%)
			Supernumerary teeth (11.7%)
Al-Kharboush (19), 2010	CLP	200	Hypodontia (46.5%)
			Microdontia (31.6%)
			Ectopic eruption (10.4%)
			Supernumerary teeth (9%)
			Macrodontia (2.4%)
Wu et al. (6), 2011	UCLP	83	Missing MLI
	UCL	20	a. BCLP (65.8%)
	UCLA	31	b. UCLP (56.7%)
	BCLP	38	c. UCLA (35.5%)
	CP	20	d. UCL (20%) e. CP (10%)

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(Table 1 continued)

			Missing lower incisors
			a. UCLP (19.2%)
			b. CP (10%)
			c. BCLP (7.6%)
			Peg laterals
			a. UCLA (61.3%)
			b. BCLP (58%)
			c. UCLP (48.2%)
			d. UCL (45%)
			e. CP (10%)
			Transposition
			a. BCLP (10.6%)
			b. UCLP (3.6%)
Qureshi et al. (9), 2012	UCLP	67	Single missing tooth
	BCLP	29	a. UCLP (39%)
			b. BCLP (14%)
			Multiple missing tooth
			a. UCLP (22%)
			b. BCLP (54%)
			Anterior malocclusion
			a. UCLP (15%)
			b. BCLP (41%)
Costa et al. (14), 2012	RUTC	12	Agenesis (31.6%)
	LUTC	32	Conical teeth (28.9%)
	BTC	25	Supernumerary teeth (13.2%)
	PC	08	Ectopic teeth (5.3%)
			Rotated teeth (13.2%)
			Impacted teeth (14.5%)
			Twin tooth (1.3%)
Shetty et al. (16), 2013	UCLP	113	Missing MLI (48.7%)
			Rotated maxillary right lateral incisors (22.1%)
			Rotated maxillary right central incisors (18.6%)
			Missing right lateral incisors (21.2%)
Riis et al. (12), 2014	CL	30	Agenesis (significantly higher in CLP)
	CP	30	Supernumerary teeth (significantly higher in CL)
	CLP	30	Malformation (significantly higher in CP)

Abbreviation: CP = cleft palate; CLP = cleft lip and palate; UCLP = unilateral cleft lip and palate; BCLP = bilateral cleft lip and palate; UCL = unilateral cleft palate; UCLA = unilateral cleft lip and alveolus; HLP = control group; CL = cleft lip; RUTC = right unilateral transforamen cleft; LUTC = left unilateral transforamen cleft; BTC = bilateral transforamen cleft; PC = post-foramen cleft; BCL = bilateral cleft lip; MLI = maxillary lateral incisor; No-number.

(18.6%).

da Silva et al. (17) measured the number of dental anomalies in the permanent dentition of patients with bilateral cleft lip (BCL). The prevalence of hypodontia was higher in patients with complete BCL, and the most frequently affected tooth was the maxillary lateral incisor (26.6%), followed by the mandibular second premolar (8%) and maxillary second premolar (4.6%). They also reported that the prevalence of supernumerary teeth was 28.2% in male subjects with complete BCL and 29.2% for incomplete BCL. In female subjects, they reported incidences of 17.5% and 46.6% for complete BCL and incomplete BCL, respectively.

A high prevalence of hypodontia and supernumerary teeth was observed in a study of 205 CLP patients. Hypodontia was diagnosed in 144 patients (70.2%), and the maxillary lateral incisor showed the highest prevalence. The lesions were primarily located distal to the cleft (25%) when both lateral incisors were intact (43%). Supernumerary teeth were observed in 11.7% of patients (18).

In another study, Al-Kharboush (19) reported hypodontia as the most commonly observed dental anomaly, affecting 134 of 288 cleft individuals (46.5%), followed by microdontia (91 patients, 31.6%), ectopic eruption (30 patients, 10.4%), supernumerary teeth (26 patients, 9%), and macrodontia (7 patients, 2.4%). In a study assessing the craniofacial morphology of Japanese patients with UCLP, pushback palatoplasty was the frequently selected repair for maxillary and dentoalveolar malformation (20).

A systematic review of the literature is needed to establish the globalized norms and trends in dental anomalies associated with CLP. Understanding these norms will better our understanding of CLP cases complicated by dental anomaly and improve therapeutic planning. Therapy should emphasize prompt management and close monitoring of CLP patients with dental anomaly. Many complications can be easily treated or prevented if detected early, saving the patient from invasive procedures. An inter-disciplinary approach is required in some cases and can completely redirect the treatment plan in cases of impaction, ectopic eruption, or hypodontia. Timely examination with a keen eye for potential complications can be highly beneficial when incorporating other specialties to better the outcome.

Summary of literature survey

The results of the literature survey of dental anomaly incidence in CLP patients are summarised in Table 1.

Conclusion

The incidence of certain dental anomalies is strongly correlated with CLP, a finding that is consistent with previous studies. Further evidence is needed to accurately establish the nature of this relationship according to each particular dental anomaly. Our literature survey summarises the absolute prevalence of dental anomalies in CLP patients and is an essential step in determining the association of each dental anomaly with cleft palate and lip. Based on our survey, the most common dental anomalies in CLP patients are as follows: multiple missing teeth/hypodontia/agenesis (usually the maxillary lateral incisors); ectopic teeth; impaction; supernumerary teeth; microdontia; maxillary canines and premolars transposition; delayed development; crown and root malformation; and multiple decayed teeth. Specific methodology and inclusion and exclusion criteria are needed to assess the global prevalence of specific anomalies. Statistical analysis of resulting data is required to establish global norms.

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Conflict of Interest

None.

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Authors' Contributions

Conception and design, drafting of the article, critical revision of the article for important intellectual content, final approval of the article, collection and assembly of data: MKA, SH

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References

1. Mossey P, Castillia E. *Global Registry and Database on Craniofacial Anomalies*. Geneva (CH): World Health Organization; 2003.
2. Al Omari F, Al-Omari IK. Cleft lip and palate in Jordan: birth prevalence rate. *Cleft Palate Craniofac J*. 2004; **41(6)**:609–612. doi: <http://dx.doi.org/10.1597/03-034.1>.
3. Cassolato SF, Ross B, Daskalogiannakis J, Noble J, Tompson B, Paedo D. Treatment of dental anomalies in children with complete unilateral cleft lip and palate at SickKids hospital, Toronto. *Cleft Palate Craniofac J*. 2009; **46(2)**:166–172. doi: <http://dx.doi.org/10.1597/07-239.1>.
4. Stahl F, Grabowski R, Wigger K. Epidemiology of Hoffmeister's "genetically determined predisposition to disturbed development of the dentition" in patients with cleft lip and palate. *Cleft Palate Craniofac J*. 2006; **43(4)**:457–465. doi: <http://dx.doi.org/10.1597/04-156.1>.
5. Shapira Y, Lubit E, Kuftinec MM. Congenitally missing second premolars in cleft lip and cleft palate children. *Am J Orthod Dentofacial Orthop*. 1999; **115(4)**:396–400. doi: 10.1016/S0889-5406(99)70258-1.
6. Wu TT, Chen PKT, Lo LJ, Cheng MC, Ko EW. The characteristics and distribution of dental anomalies in patients with cleft. *Chang Gung Med J*. 2011; **34(3)**:306–314.
7. Camporesi M, Baccetti T, Marinelli A, Defraia E, Franchi L. Maxillary dental anomalies in children with cleft lip and palate: a controlled study. *Int J Paediatr Dent*. 2010; **20(6)**:442–450. doi: 10.1111/j.1365-263X.2010.01063.x.
8. Menezes LM, Rizzato SMD, Azeredo F, Vargas DA. Characteristics and distribution of dental anomalies in a Brazilian cleft population. *Rev Odonto Cienc*. 2010; **25(2)**:137. doi: <http://dx.doi.org/10.1590/S1980-65232010000200006>.
9. Qureshi WA, Beiraghi S, Leon-Salazar V. Dental anomalies associated with unilateral and bilateral cleft lip and palate. *J Dent Child (Chic)*. 2012; **79(2)**:69–73.
10. Menezes R and Vieira AR. Dental anomalies as part of the cleft spectrum. *Cleft Palate Craniofac J*. 2008; **45(4)**:414–419. doi: 10.1597/07-064.1.
11. Al Jamal GA, Hazza'a AM, Rawashdeh MA. Prevalence of dental anomalies in a population of cleft lip and palate patients. *Cleft Palate Craniofac J*. 2010; **47(4)**:413–420. doi: 10.1597/08-275.1.
12. Riis LC, Kjær I, Mølsted K. Dental anomalies in different cleft groups related to neural crest developmental fields contributes to the understanding of cleft aetiology. *J Plast Surg Hand Surg*. 2014; **48(2)**:126–131. doi: 10.3109/2000656X.2013.831767.
13. Parapanisiou V, Gizani S, Makou M, Papagiannoulis L. Oral health status and behaviour of Greek patients with cleft lip and palate. *Eur Arch of Paediatr Dent*. 2009; **10(2)**:85–89.
14. Costa CH, Diniz LV, Lacerda RH, Forte FD, Sampaio FC. Prevalence of dental anomalies in patients with cleft lip and palate, Paraiba, Brazil: clinic and radiographic study. *Acta Odontol Latinoam*. 2012; **25(2)**:181–185.
15. Bartzela TN, Carels CE, Bronkhorst EM, Rønning E, Rizell S, Kuijpers-Jagtman AM. Tooth agenesis patterns in bilateral cleft lip and palate. *Eur J Oral Sci*. 2010; **118(1)**:47–52. doi: 10.1111/j.1600-0722.2009.00698.x.
16. Shetty A, Rai K, Hegde AM. Incisal abnormalities in children with Unilateral cleft lip and palate. *Sch J App Med. Sci*. 2013; **1(4)**:233–235.
17. daSilva A, Costa B, deCarvalho, Carrara C. Dental anomalies of number in the permanent dentition of patients with bilateral cleft lip: Radiographic study. *Cleft Palate Craniofac J*. 2008; **45(5)**:473–476. doi: 10.1597/06-099.1.
18. Tereza GPG, Carrara CFC, Costa B. Tooth abnormalities of number and position in the permanent dentition of patients with complete bilateral cleft lip and palate. *Cleft Palate Craniofac J*. 2010; **47(3)**:247–252. doi: <http://dx.doi.org/10.1597/08-268.1>.
19. Al-Kharboush GH. Dental anomalies associated with cleft lip and palate in a Saudi sample [master's thesis]. [Saudi Arabia (SC)]: College of Dentistry, King Saud University, 2010.
20. Alam MK, Iida J, Sato Y, Kajii TS. Postnatal treatment factors affecting craniofacial morphology of unilateral cleft lip and palate (UCLP) patients in a Japanese population. *Br J Oral Maxillofac Surg*. 2012; **51(8)**:205–210. doi: 10.1016/j.bjoms.2012.10.001.