Dental Anomalies Associated With Unilateral and Bilateral Cleft Lip and Palate

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ABSTRACT

Purpose: The purpose of this study was to compare the prevalence of dental anomalies in the primary and permanent dentition of patients with unilateral (UCLP) and bilateral (BCLP) cleft lip with or without palate.

Methods: One hundred two complete clinical records were randomly selected for review from a university-based cleft palate clinic. Only nonsyndromic UCLP and BCLP cases were further selected for analysis of dental anomalies. The prevalence of 9 dental categories, including anomalies in number, crown structure, position, and maxillary-mandibular relationship, was assessed and compared between UCLP and BCLP cases using Fisher's exact test.

Results: Of the 102 charts evaluated, there were 67 cases of UCLP and 29 cases of BCLP for a total of 96 cases. There was a high prevalence of dental anomalies in primary and permanent teeth; 93% of UCLP cases and 96% of BCLP cases presented with at least 1 dental anomaly. Significant differences ($P<.05$) were only found in the prevalence of anodontia of a single tooth (UCLP=39%, BCLP=14%), multiple anodontia (UCLP=22%, BCLP=54%), and anterior malocclusion (UCLP=15%, BCLP=41%).

Conclusions: There is a high prevalence of dental anomalies associated with orofacial clefts regardless of whether they are unilateral or bilateral cleft lip with or without palate. (J Dent Child 2012;79(2):69-73)

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Restoring the functional and esthetic occlusal impairment resulting from orofacial clefts is one of the most challenging clinical conditions that pediatric dentists face due to the wide range of dental anomalies accompanying the cleft and its long-term impact on the patients' facial appearance and self esteem.[1]

Cleft lip, with or without cleft palate (**CLP**), is the most common congenital defect, with a prevalence varying from 1 in 500 to 1 in 2,500 live births, depending on the geographic origin and ethnic background.[2-7] Worldwide, this prevalence means that approximately every 2 minutes an infant with a cleft is born. In the United States, approximately everyday, 20 children are born with the same condition.[3,8]

The increase in frequency of dental anomalies in CLP patients seems to be the result of a close genetic, embryologic, and anatomical relationship in the development of the lip, primary and secondary palate, and tooth buds.[4,6,9-16] Facial development starts during the fourth week of the embryonic development, when neural crest cells migrate from the bilateral neural folds as mesenchymal tissue and combine with the mesoderm to form the facial primordia and give rise to several other cells and tissues, including neural, skeletal, and dental tissues.[2,6] A complex sequence of events, genetically and/or environmentally mediated during the fourth to eighth weeks of the embryonic period, can be disrupted, resulting in different types of clefts, which can affect normal tooth development.[6] Tooth abnormalities,

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including number, position, morphology, structure, and eruption pattern, affect the primary and permanent dentition more frequently in CLP children. These dental abnormalities have been reported to be more prevalent in these patients than in their non-affected siblings as well as in the general population.[11,15,17-22]

Surgical procedures for lip and palate repair have also been reported as possible unintended causes of tooth anomalies, malocclusion, and deficient maxillary growth.[15,16,23-26] The proximity of maxillary incisors to the cleft area may predispose these teeth to a higher frequency of anomalies, particularly in permanent lateral incisors, as a consequence of primary or secondary tissue damage.[18,21,27-34] Dental anomalies are not exclusive to the cleft area, however, and can be present in noncleft segments of the dental arch.[11,35,36]

According to previous studies on CLP, the severity of tooth anomalies in a patient is directly proportional to the severity of the cleft.[11,12,17,21,35] Hence, a clinician may expect to find more teeth with severe dental anomalies in bilateral CLP because, in these cases, the alveolar process is usually more severely affected than unilateral CLP. Few studies, however, have actually compared the prevalence of dental anomalies of unilateral (**UCLP**) and bilateral (**BCLP**) cases.[20-22,37] Vichi and Franchi[20] (1995) did report that the frequency of dental anomalies may differ in these 2 CLP types. According to their results, the prevalence of supernumerary lateral incisors in the primary maxillary dentition of UCLP and BCLP cases was different (39% and 11%, respectively), whereas in the permanent dentition it was similar.

Since the presence of dental anomalies affecting both primary and permanent teeth has a great effect on the functional and esthetic long-term outcomes for orofacial cleft patients, we considered it important to evaluate the frequency of dental anomalies of position, number, and structure in UCLP and BCLP patients using a population seen longitudinally at a university-based cleft palate team.

METHODS

A total of 102 records, including clinical notes, cephalometric and panoramic X-rays, and cone beam computed tomography (**CBCT**), were randomly selected for review and analysis from the University of Minnesota Cleft Lip and Palate Clinic database. Patients in this clinic are typically evaluated yearly, from birth to adulthood, by a team that includes a pediatric dentist, orthodontist, oral surgeon, and prosthodontist, among others. Charts of patients not established or with incomplete clinical records were not included for the initial review. Therefore, all 102 clinical records reviewed contained information regarding both the primary and permanent dentition. To be included in the study, patients had to have a confirmed diagnosis of UCLP or BCLP involving the primary palate only (incomplete) or the primary and secondary palate (complete), without any previously diagnosed syndrome. The patients also had to be at least 2 years old when data were collected. The sample was divided into 2 groups, according to CLP type: (1) UCLP; and (2) BCLP. Gender was not considered for sample grouping or data analysis. Exception approval from the Institutional Review Board of the University of Minnesota, Minneapolis, Minn, was obtained before starting data collection.

The presence or absence of 9 separate categories of dental anomalies: 1)single anodontia, 2) multiple anodontia, 3) supernumerary tooth 4) anterior malocclusion, 5) posterior malocclusion, 6) enamel hypoplasia, 7) other enamel defects, 8) discolored tooth 9) rotated/ectopic tooth (Table 1) was primarily obtained from the clinical dental charts. The charts included: a complete odontogram obtained at each patient's visit; the interdisciplinary team evaluation report; and prior records of orthodontic, surgical, and restorative dental evaluations and treatments, if any. All available radiographs, including periapical, occlusal, panoramic, and cephalograms, as well as the CBCT, were analyzed by the same operator to verify the information found in the clinical charts. The cases of tooth agenesis were divided into 2 main groups: (1) single, when only 1 tooth was missing; and (2) multiple, when more than 1 primary or permanent tooth was missing. Cases of crossbite malocclusion were divided into anterior and posterior and analyzed separately. Cases with overlap or ambiguity in interpretation were forced into 1 category or the other. A Fisher's exact test at a level of significance of 0.05 was used to compare the prevalence of dental anomalies between groups.

RESULTS

Of 102 cases evaluated, 53% (54) were male patients and 47% (48) were female patients with ages ranging from 26 months to 54 years old. Left-sided UCLP (52 patients; 51%) was most prevalent, followed by BCLP (29 patients; 28%) and right-sided UCLP (15

Table 1. Comparison of the Dental Anomalies in Unilateral and Bilateral Cleft Patients

Anomaly	Bilateral (N=29) N (%)	Unilateral (N=67) N (%)	*P*-value*
Single anodontia	4 (14)	26 (39)	<.02
Multiple anodontia	15 (54)	15 (22)	.004
Supernumerary tooth	10 (34)	25 (37)	>.82
Anterior malocclusion	12 (41)	10 (15)	.008
Posterior malocclusion	3 (10)	2 (3)	.16
Enamel hypoplasia	4 (14)	7 (10)	.73
Other enamel defects	5 (17)	12 (18)	1.00
Discolored tooth	0 (0)	2 (3)	1.00
Rotated/ectopic tooth	3 (10)	9 (13)	1.00

** Fisher's exact test; P<.05 considered statistically significant.*

patients; 15%). In 6 cases (6%) no side could be assigned (ie, cleft palate, submucous). Eighty-two cases (80%) involved the primary and secondary palate, 14 (14%) involved only the primary palate, 3 (3%) involved only the secondary palate, and 3 (3%) were submucous clefts. After excluding all submucous and isolated cleft palate cases, the study sample consisted of 67 UCLP and 29 BCLP cases.

There was a high prevalence of dental anomalies in our sample, with 93% of UCLP cases and 96% of BCLP cases having at least 1 dental anomaly. The distribution of the 9 dental anomalies is shown in Table 1. Of the 9 anomalies assessed, only 3 presented statistically significant differences by cleft type: (1) anodontia of a single tooth; (2) anodontia of multiple teeth; and (3) anterior malocclusion. The most prevalent dental anomalies were: anodontia multiple (54%), anterior malocclusion (41%; ie, crossbite involving ≥1 tooth), and supernumerary teeth (34%) in BCLP; and single tooth anodontia (39%) and supernumerary teeth (37%) in UCLP. Hypoplasia and other enamel defects, posterior crossbite, rotated/ectopic teeth, and discolored teeth were present in less than 20% of the cases in both groups.

DISCUSSION

The presence of dental anomalies complicates the functional and esthetic rehabilitation of CLP sequelae. To the best of our knowledge, no longitudinal study to date has been conducted comparing the prevalence of dental anomalies and malocclusion of the primary and permanent dentition in individuals with unilateral or bilateral CLP. In this study, the prevalence of 9 dental phenotypes, including malocclusion and dental anomalies of number, morphology, and position, was assessed in the primary and permanent dentitions of nonsyndromic UCLP and BCLP patients. These anomalies were investigated because they often affect the treatment of patients with CLP in both the primary and permanent dentition.[1]

More than two thirds of patients in our randomly selected sample had UCLP. Left-sided UCLP was the most prevalent, 3 times more prevalent than right-sided UCLP, followed by BCLP, isolated cleft palate, and submucous cleft. The latter two were excluded from analysis because the primary objective of this study was to compare UCLP and BCLP cases that affected the maxillary alveolar ridge. Although our sample was obtained at a tertiary cleft lip and palate center, the cleft type ratio found agrees with other studies.[3,11,21,22,35]

Sixty-seven UCLP and 29 BCLP cases were included in the study, a sample size comparable to previous studies.[21] The sample size, however, limits the generalization of our results. Males and females, similarly represented in the sample, were not studied separately since similar studies have identified no significant differences between genders regarding dental anomalies associated with clefts.[12,27,32,33,35-37]

Several studies have reported that the prevalence of the most common dental anomalies is higher in CLP patients than in the general population.[9,19,20,33,37] The results have varied depending on the specific anomaly included in the analysis, dentition and population studied, specific data collected, and type and severity of orofacial cleft.[2-7,11,15,27,35,37]

The prevalence of dental anomalies found in this study was 96% in UCLP and 93% in BCLP cases. This is similar to prior reports regarding the permanent dentition.[1,35] Some studies including both primary and permanent dentition, however, have reported, in general, a lower prevalence of dental anomalies.[20,21,37] The high prevalence of dental anomalies in this study is likely due to the inclusion of both the primary and permanent dentition in cleft and noncleft areas. Assessment of the malocclusion, as part of dental anomalies of number and structure, also contributed to the high prevalence found. Of the 9 anomalies studied, however, only 3 of the 4 most prevalent showed a statistically significant difference between UCLP and BCLP: missing teeth; supernumerary teeth; and anterior malocclusion. Interestingly, a single missing tooth was more prevalent in UCLP patients, whereas multiple missing teeth were more prevalent in BCLP. Of note, UCLP and BCLP cases were not subdivided into complete and incomplete clefts for analysis. Although all cases included in this study had involvement of the alveolar ridge, the combination of incomplete clefts (which may minimally affect the alveolus) and complete clefts (which, in general, produce more severe alveolar clefts) may have mischaracterized the prevalence of dental anomalies in ULCP and BCLP. Neither the primary nor permanent dentition were independently evaluated; hence, the results cannot specifically be applied to either dentition.

Anterior teeth on the cleft side, especially the lateral incisors, are reported to be the most frequently absent, malformed, or associated with supernumerary teeth in CLP.[12,18,21,27,33,35] This seems to be related to a disrupted tooth formation caused by the lack of coalescence of the maxillary and medial nasal process and consequent deficiency of the mesenchymal mass in the incisor area.[9,33,35] Therefore, it may be expected that fewer malformed or absent teeth would be found in UCLP cases because, generally, maxillary tissue deficiency is not as severe as in BCLP cases.[33]

Trauma from early surgical lip and palate repair and related scar tissue formation has also been suggested to contribute to the presence of hypoplastic and missing incisors on the cleft side.[15,16,34] The high frequency of missing teeth in areas far from the cleft, such as maxillary or mandibular premolars and incisors in the noncleft area, are more likely related to common etiological genetic factors involved in clefts and tooth anomalies. Genes MSX1, IRF6, TGFB3, TGFA, PAX9, and FGFR1

have been reported as potentially shared etiologic factors.[11,13,21,27,36]

Deficient midfacial growth as a consequence of the cleft severity and the surgical repair of the cleft also contributes to the high prevalence of anterior crossbite, especially in BCLP cases.[23-26] The lack of alveolar continuity in complete UCLP and BCLP cases is also responsible for the collapse of the premaxilla and posterior maxillary alveolar segments, which may contribute to the presence of tooth crowding, ectopic eruption, and anterior and posterior crossbite.

CLINICAL RELEVANCE

Dental anomalies are highly associated with both UCLP and BCLP and often need to be addressed early in the primary dentition to prevent further long-term functional and esthetic complications. For instance, the decision of keeping primary teeth in areas where permanent teeth are missing, or extracting supernumerary teeth that are disrupting the normal permanent teeth eruption, needs to be made before the establishment of the mixed dentition. On the other hand, esthetic concerns associated with the presence of discolored or hypoplastic teeth may be reserved for permanent teeth. When the patient's self esteem is compromised, however, consideration should be given to addressing the problem in the primary dentition.

The absence of a single tooth, such as a permanent lateral incisor or a premolar, or multiple missing teeth should be considered in space maintenance for future implant and/or prosthodontic rehabilitation. Closing the space of 1 or 2 missing teeth by orthodontic means may be a possible treatment option in some CLP cases; however, it is more difficult to accomplish when multiple teeth are missing.

Anterior and posterior crossbite, which are also common findings in CLP, particularly in BCLP patients, are initially corrected during the first transitional period of the mixed dentition. Orthodontic expansion is performed at this stage to improve the arch form and correct the anterior and posterior collapse of the alveolar segments before any bone graft procedure is performed. When the permanent dentition is complete, the treatment of the malocclusion, by orthodontic means or in combination with orthognathic surgery, is dependent on the severity of the occlusal discrepancies and the degree of facial involvement and severity of the maxillary deficiency.

Finally, it is important to discuss with expecting parents (in cases of prenatal diagnosis of CLP) or a newborn's parents the dental development and anomalies associated with CLP. This will better educate parents about the specific home oral care needs these children have as well as the importance of a sequential dental evaluation and treatment of these anomalies to achieve adequate oral function and a balanced facial and dental appearance.

CONCLUSIONS

Based on this study's results, the following conclusions can be made:

1. At least 1 dental anomaly is present in more than 90% of cleft lip with/without palate cases involving the alveolar ridge, regardless of being unilateral (UCLP) or bilateral (BCLP).
2. Missing a single tooth is a frequent finding in UCLP patients, whereas anterior malocclusion in the anterior area and multiple missing teeth are more prevalent in BCLP cases.
3. A high prevalence of dental anomalies and their variability should be realized by clinicians and parents, since they pose additional challenges for accurate, timely, and effective comprehensive dental treatment.

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