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Dental anomalies in individuals with cleft lip and/or palate

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SUMMARY Significant heterogeneity has previously been reported but with no consensus on the prevalence of dental anomalies in subjects with a cleft lip and palate (CLP), thus, the purpose of this study was to investigate the frequency of various dental anomalies in the upper dental arch in different cleft groups.

Diagnostic records, i.e., panoramic, occlusal and periapical films, dental casts, and intra-oral photographs, of 122 subjects (mean age: 14 ± 5 years; 67 males and 55 females) were grouped as either unilateral left cleft lip and palate (ULCLP), unilateral right cleft lip and palate (URCLP), bilateral cleft lip and palate (BCLP), or cleft palate (CP). Prevalence rates of 15 different dental anomalies were calculated for each group. Wilcoxon's test was used to determine if there was a statistically significant difference in the number of missing teeth between the right and left sides, in each cleft group.

Overall, 96.7 per cent of patients were found to have at least one dental anomaly. The most prevalent was agenesis in the anterior region on the cleft side (70.8–97.1 per cent). There was a statistically significant difference in the prevalence of agenesis by cleft and non-cleft sides but only in the ULCLP group ($P < 0.001$). Significantly higher rates of impaction were observed in the anterior and premolar regions in the CLP groups (2.9–29.2 per cent), with the highest rates in the anterior region on the cleft sides.

A very high proportion of subjects were found to have at least one dental anomaly. Thus, the management of dental anomalies should be central to the treatment planning process of individuals with a cleft.

Introduction

Dental anomalies may occur as a result of genetic and environmental factors. While abnormalities are most commonly caused by defects in specific genes, pre- and post-natal aetiological events have also been implicated in anomalies in tooth dimension, morphology, position, number, and structure (Garn *et al.*, 1963; Sofaer, 1979; Kotsomitis *et al.*, 1996).

When compared with the general population, subjects with a cleft lip and palate (CLP) have been found to have a higher prevalence of dental anomalies, such as variations in tooth number and position, and reduced tooth dimensions, most of which are localized in the area of the cleft defect (Haring, 1976; Ribeiro *et al.*, 2003). Lucas *et al.* (2000) reported a higher prevalence of enamel discolouration in children with a CLP when compared with a control group, and attributed this defect to trauma at the time of CLP surgery. Ribeiro *et al.* (2002, 2003) found a high prevalence of hypodontia of the permanent cleft-side lateral incisor (49.8 per cent) as well as delayed root development in comparison with the contralateral tooth.

Most previous studies investigating dental deformities among individuals with clefts have included different types of cleft cases among their samples and have not differentiated their results according to cleft type. As a result, little information is available in the literature regarding the prevalence of various dental anomalies in different cleft groups. Since cleft formation may be the result of a combination of genetic and environmental factors and may occur at different times during gestation, thus affecting different parts of the craniofacial and dentofacial structure, it is possible that specific patterns of deformities may be related to different cleft types (Ranta and Rintala, 1982; Trotman *et al.*, 1993). It has therefore been suggested that epidemiological studies conducted on cleft individuals require classification by cleft type (Baek and Kim, 2007).

Because 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. In the light of the above, the current retrospective study aimed to investigate the prevalence of dental anomalies in a group of individuals with different types of clefting and to elucidate relevant clinical suggestions from the results.

Materials and methods

Standardized diagnostic records, i.e., panoramic, occlusal and periapical radiographs, dental casts, intra-oral photographs, and dental histories, of 122 Caucasian subjects (67 males and 55 females) with clefts were retrieved from the archives of the Orthodontic Department, Ankara University. The films were taken by the same technician using the same device (Siemens, P10E, Palomex Instrumentarium, Hyrylä, Finland) using the same standardized method. The mean age of the patients was 14 ± 5 years (range: 12–30 years). The subjects, who had

undergone surgical treatment (lip and hard palate closure) before 3 years of age, were in the permanent dentition and had no syndromes, no extraction of permanent teeth, no endodontic/prosthodontic/orthodontic treatment, and no trauma to any tooth before the initiation of orthodontic treatment.

The subjects were classified by cleft type into one of four groups (Table 1): (1) Unilateral left cleft lip and palate (ULCLP), (2) Unilateral right cleft lip and palate (URCLP), (3) Bilateral cleft lip and palate (BCLP), and (4) Cleft palate (CP). Subjects in groups 1, 2, and 3 had a complete cleft of the lip, alveolus, and palate.

In order to eliminate inter-examiner differences, all records were examined by one observer (SE).

The following anomalies were investigated (Figure 1):

1. Agenesis: congenital absence of a permanent tooth or germ.
2. *Dens invaginatus*: developmental malformation resulting from invagination of the crown or root surface before calcification (Hamasha *et al.*, 2002).
3. *Dens evaginatus* (talon cusp): a developmental aberration of a tooth resulting in the formation of a supernumerary tubercle that extends from the occlusal aspect of an otherwise normal tooth (McCulloch *et al.*, 1997, Levitan and Himel, 2006).
4. Impaction: a tooth that is not expected to erupt completely into its normal functional position based on clinical and radiographic assessment (Thilander and Jakobsson, 1968).
5. Taurodontism: the tooth trunk is elongated and the floor of the pulp chamber is displaced apically with proportionately shortened roots (Darwazeh *et al.*, 1998).
6. Pulp stone: a calcified mass in the dental pulp of a healthy, diseased, or unerupted tooth (Hamasha and Darwazeh, 1998).
7. Microdontia: an inherited condition that produces one or more disproportionately smaller teeth (Kocabalkan and Özyemisci, 2005).
8. Dilaceration: a deviation or bend in the linear relationship between tooth crown and root; an angulation or sharp curve of 90 degrees or more in the root or crown of a developed tooth (Hamasha *et al.*, 2002).
9. Enamel hypoplasia: an hereditary condition in which the dental enamel shows either a break in continuity or surface loss, often because of insufficient calcification (Lai and Seow, 1989).
10. Short or blunt roots: defined as developmentally very short blunt dental roots (Apajalahti *et al.*, 2002). Roots as long as or shorter than the crowns of the incisors and visually evaluated as very short blunt roots bilaterally in the posterior teeth were recorded as short or blunt roots.
11. Supernumerary teeth: those that appear in addition to the regular number of teeth.
12. Ectopic eruption: the eruption of a tooth in an abnormal position (Toutountzakis and Kastaris, 1990).

Table 1 Distribution of subjects by cleft group and gender in the unilateral left cleft lip and palate (ULCLP), unilateral right cleft lip and palate (URCLP) and bilateral cleft lip and palate (BCLP), and cleft palate (CP) groups.

	URCLP	ULCLP	BCLP	CP
	n (%)	*n* (%)	*n* (%)	*n* (%)
Male (*n* = 67)	14 (20.9)	25 (37.3)	23 (34.3)	5 (7.5)
Female (*n* = 55)	10 (18.2)	28 (50.9)	12 (21.8)	5 (9.1)
Total (*N* = 122)	24 (19.7)	53 (43.4)	35 (28.7)	10 (8.2)

Statistical analysis

Intra-examiner reliability was tested using Kappa statistics and confirmed in a previous study (Uslu *et al.*, 2009). A chi-square test was used to evaluate differences by gender and a Mann–Whitney *U*-test to determine whether or not significant differences existed in the occurrence of dental anomalies by age.

Figure 1 (From left to right) *dens invaginatus*, *dens evaginatus*, pulp stones, and taurodontism.

The rates of occurrence of each anomaly were calculated as a percentage of the total sample in each group. A dental anomaly was defined as the presence of any type of dental anomaly within a subject's mouth. A detailed explanation has been provided previously (Uslu *et al.*, 2009). Differences in the incidence rates of each dental anomaly by cleft type were analysed using Wilcoxon's test.

Statistical analysis was performed with the Statistical Package for Social Science Version 11.0 for Windows (SPSS, Inc., Chicago, Illinois, USA).

Results

No statistically significant differences were found in the incidence of dental anomalies between males and females. A Mann–Whitney *U*-test revealed no statistically significant differences in dental anomalies by age ($P = 0.779$, Table 1).

Tables 2–4 show the distribution and rates of dental anomalies by cleft group. Overall, the vast majority of patients (96.7 per cent; $n = 118$) were found to have at least one dental anomaly. Agenesis in the anterior region on the cleft side was found to be the most frequently occurring dental anomaly (70.8–97.1 per cent). Wilcoxon's test showed no significant differences between overall agenesis rates by side in the URCLP, BCLP, or CP groups; however, in the ULCLP group, the agenesis rate was higher on the left side compared with the right side ($P < 0.001$; Table 5). A higher rate of agenesis on the cleft side in the ULCLP group compared with the URCLP group ($P < 0.05$) was observed. Agenesis in the CP group was found to occur at lower rates than in the other groups ($P < 0.05$ to $P < 0.001$).

Microdontia was observed in all groups but only on the cleft side, while tooth rotation was noted in the anterior region in all groups. Impaction was observed in the anterior and premolar regions in the three CLP groups with the highest rates in the anterior regions on the cleft sides in the URCLP (29.2 per cent) and ULCLP (18.9 per cent) groups.

Table 2 Distribution of dental anomalies in the unilateral right cleft lip and palate (URCLP, $n = 34$) and unilateral left cleft lip and palate (ULCLP, $n = 53$) groups. *n*, number of teeth.

	Region	URCLP	ULCLP	URCLP	ULCLP
		Right		Left	
		n (%)	*n* (%)	*n* (%)	*n* (%)
Agenesis	Molar	13 (54.1)	28 (52.8)	12 (50)	27 (50.9)
	Premolar	1 (4.2)	9 (16.9)	3 (12.5)	14 (26.4)
	Anterior	17 (70.8)	16 (30.1)	7 (29.2)	43 (81.1)
Microdontia	Molar			1 (4.2)	1 (1.9)
	Anterior	1 (4.2)			2 (3.8)
Ectopic eruption	Premolar				3 (5.7)
	Anterior	1 (4.2)	1 (1.9)	2 (8.3)	3 (5.7)
Rotation	Premolar			2 (8.3)	
	Anterior	5 (20.8)	8 (14.8)	5 (20.8)	12 (22.6)
Impaction	Premolar	1 (4.2)	1 (1.9)		
	Anterior	7 (29.2)	2 (3.8)	1 (4.2)	10 (18.5)
Retained teeth	Molar		2 (3.8)		3 (5.7)
	Anterior	4 (16.7)	3 (5.7)	1 (4.2)	9 (16.9)
Dilaceration	Molar		1 (1.9)		1 (1.9)
Shape anomaly	Premolar	1 (4.2)		1 (4.2)	
	Anterior	1 (4.2)	1 (1.9)		6 (11.3)
Short or blunt roots	Molar		1 (1.9)		
	Premolar	1 (4.2)	3 (5.7)		3 (5.7)
Pulp stones	Molar	2 (8.3)		4 (16.7)	
Supernumerary teeth	Premolar			1 (4.2)	1 (1.9)
	Anterior				1 (1.9)
Dens invaginatus	Anterior	2 (8.3)	4 (7.5)	1 (4.2)	
Dens evaginatus	Anterior		2 (3.8)		1 (1.9)
Taurodontism	Molar		1 (1.9)		1 (1.9)
Enamel hypoplasia	Anterior	1 (4.2)			1 (1.9)

Table 3 Distribution of dental anomalies in the bilateral cleft lip and palate group ($n = 35$). *n*, number of teeth.

	Region	Right	Left
		n (%)	*n* (%)
Agenesis	Molar	18 (51.4)	17 (48.6)
	Premolar	15 (42.8)	17 (48.6)
	Anterior	32 (91.4)	34 (97.1)
Microdontia	Molar	1 (2.9)	1 (2.9)
Ectopic eruption	Premolar	2 (5.7)	1 (2.9)
	Anterior	1 (2.9)	1 (2.9)
Rotation	Premolar		1 (2.9)
	Anterior	1 (2.9)	4 (11.4)
Impaction	Premolar	2 (5.7)	1 (2.9)
	Anterior	7 (20)	9 (25.7)
Retained teeth	Molar	3 (8.6)	4 (11.4)
	Anterior	9 (25.7)	5 (14.3)
Shape anomaly	Premolar	1 (2.9)	1 (2.9)
	Anterior	6 (17.1)	7 (20)
Short or blunt root	Molar	2 (5.7)	2 (5.7)
Pulp stone	Molar	2 (5.7)	4 (11.4)
Enamel hypoplasia	Molar	2 (5.7)	
	Anterior	4 (11.4)	1 (2.9)

Table 4 Distribution of dental anomalies in the cleft palate group ($n = 10$). *n*, number of teeth.

	Region	Right	Left
		n (%)	*n* (%)
Agenesis	Molar	2 (20)	5 (50)
	Premolar	2 (20)	2 (20)
Ectopic eruption	Premolar	1 (10)	1 (10)
Rotation	Anterior	1 (10)	1 (10)
Retained teeth	Anterior		1 (10)
Shape anomaly	Anterior		1 (10)
Pulp stones	Molar	3 (30)	
	Premolar	1 (10)	1 (10)
Supernumerary teeth	Molar		1 (10)

Table 5 Wilcoxon test comparing agenesis between the cleft and non-cleft sides by group.

Agenesis(Number of teeth)			
	Right side (median, maximum, minimum)	Left side (median, maximum, minimum)	*P* value
Unilateral right cleft lip and palate	31 (1,3,0)	22 (1,3,0)	NS
Unilateral left cleft lip and palate	53 (0,8,0)	84 (1,10,0)	***
Bilateral cleft lip and palate	65 (2,6,0)	68 (2,10,0)	NS
Cleft palate	5 (0,1,0)	7 (5,2,0)	NS

NS, not significant. ***$P < 0.001$.

Pulp stones were found in all groups except in the ULCLP group, mainly in the molar region. Supernumerary teeth were observed in all groups except the BCLP group. *Dens invaginatus* was recorded in the anterior region in the URCLP and ULCLP groups, while *dens evaginatus* was observed in the ULCLP group in the anterior region. Taurodontism was present only in the ULCLP group, while enamel hypoplasia was detected in the teeth on the cleft sides in all three CLP groups.

Discussion

The prevalence of dental anomalies has been found to vary among different racial/ethnic groups. Derijcke *et al.* (1996) reported an increased frequency of dental anomalies among Caucasian patients with a UCLP. The present study investigated the frequency of anomalies in a sample of Caucasian subjects and classified them according to cleft type.

No relationship was found between dental anomalies and gender in the present investigation. This is in agreement with the findings of Ribeiro *et al.* (2002, 2003). On the other hand, the number of female subjects was higher than males in the ULCLP group, which was not as expected. Demirjian *et al.* (1973) observed that the mechanisms controlling dental development are independent of somatic and sexual maturity but appear to be highly influenced by aetiological factors such as a cleft. The Mann–Whitney *U*-test revealed no statistically significant difference in dental anomalies with regard to age, although the number of congenital tooth anomalies may increase with age, as they become more recognizable.

The cleft sample in this study was subdivided into four groups, as it has previously been reported that different cleft types could be related to specific patterns of deformities (Ranta and Rintala, 1982; Trotman *et al.*, 1993). Moreover, it has been underlined that epidemiological studies conducted on cleft individuals require classification by cleft type (Baek and Kim, 2007).

Dewinter *et al.* (2003) reported that for patients with a UCLP, the left side is more affected than the right side (ratio 2:1). This ratio is similar to the findings of the current study. It has previously been reported that dental anomalies occur with a higher frequency on the cleft side in patients with a UCLP (Böhn, 1963; Ranta, 1983, 1986). Some authors support the view that the aetiological factors that lead to cleft formation (poly- or monogenetic inheritance and multiple exogenous factors) may also affect development of the dentition (Bhatia, 1972; Eerens *et al.* 2001).

Previous studies have reported congenital absence of the cleft-side permanent lateral incisor to be the most common finding in children with a cleft lip, CP, or both (Böhn, 1950, 1963; Ranta, 1986). Dewinter *et al.* (2003) found agenesis of the lateral incisor on the cleft side in more than 50 per cent of patients with a cleft. In the present study, the rates of anterior agenesis on the cleft side varied between 70.8 and 97.1 per cent, depending on cleft type, with the differences in rates being statistically significant. It has been suggested that the high rate of agenesis near the cleft may be due to a deficiency in blood supply, either congenital or secondary to surgery, or to a deficiency in the mesenchymal mass (Jiroutova and Mullerova, 1994; Vichi and Franchi, 1995; Ribeiro *et al.*, 2003). On the other hand, Dewinter *et al.* (2003) reported agenesis outside the cleft area in 27.2 per cent of patients, and Brattström and McWilliams (1989) a 27.8 per cent rate of agenesis outside the cleft area in UCLP patients, which was markedly higher than that of non-cleft controls (3.6 per cent). In the current study, the rate of non-cleft side agenesis in the anterior, premolar, and molar regions varied between 12.5 and 52.8 per cent. Different rates of agenesis can be related to the severity of the cleft phenotype, which has been shown to have a correlation with the number of affected teeth (Dewinter *et al.*, 2003).

Previous research of the general population has shown the prevalence of microdontia to vary from 1.5 to 2.0 per cent. Teeth in the region of an alveolar cleft have commonly been reported to be malformed, peg-shaped, microformed, or congenitally absent (Böhn, 1963; Ranta, 1986; Vichi and Franchi, 1995; Uslu *et al.*, 2009). In the current study, there was a higher prevalence of microdontia (1.9–4.2 per cent) on the cleft side in the URCLP and ULCLP groups when compared with the general population. Werner and Harris (1989) reported tooth size in UCLP individuals to be significantly smaller (2.3 per cent) than that in non-cleft

controls and commented that since CLP patients show a compromised growth potential, the dentition may be reduced in size.

Ectopic eruption has been reported to depend on systemic or local factors (Bondemark and Tsiopa, 2007) and the population average for ectopic teeth has been reported to range between 2–6 per cent for the maxillary first molars and 1.5–2 per cent for the permanent canines (Fox *et al.*, 1995; Barberia-Leache *et al.*, 2005). In the current study, the rate of ectopic eruption varied by cleft group from 1.9 to 10.0 per cent. In a radiographic study of 225 children (Bjerklin *et al.*, 1993), a higher prevalence of ectopic eruption of the maxillary first permanent molar (15.4 per cent) was found in children with a cleft lip or cleft lip and alveolus. In the current study, all cleft groups showed ectopic eruption on both sides of the dental arch, while the URCLP group showed ectopic eruption only in the anterior region, and the CP group only in the premolar region.

Impacted permanent maxillary canines occur in 1–3 per cent of the general population (Peck *et al.*, 1994). The findings of the current study showed a significantly higher rate of impaction in the anterior and premolar regions in the CLP groups (1.9–29.2 per cent), with the highest rates in the anterior region on the cleft side. It has been reported that impacted maxillary canines often present in conjunction with other genetically linked dental abnormalities (Baccetti, 1998).

The current study revealed dilaceration in the molar region only in the ULCLP group at a rate of 1.9 per cent, which is in the range found in the general population (Thongudomporn and Freer, 1998; Hamasha *et al.*, 2002). Identifying a dilaceration is particularly important prior to root canal treatment, extraction, and orthodontic tooth movement.

Shape anomalies of teeth in patients with clefts have been reported (Ribeiro *et al.*, 2003, Akcam *et al.*, 2008); however, none of the studies distinguish between cleft types. In the current investigation, shape anomalies were found not only on the cleft side but also on the non-cleft side in the three CLP groups, particularly in the anterior region, with the greatest frequency observed in the BCLP group. This could indicate that the clefting not only affect the shape of the teeth on the cleft side but also on the non-cleft side.

The rates of short or blunt roots and pulp stones have been found to be in the range of those in non-cleft subjects (Hamasha *et al.*, 2002). Dewinter *et al.* (2003) reported that 5 out of 75 children (6.7 per cent) with a CLP showed root resorption of one tooth at the cleft site; however, no obvious cause could be demonstrated. Patients with short or blunt roots before orthodontic treatment have been reported to experience significant root shortening during treatment (Grover and Lorton, 1985).

Hamasha and Darwazeh (1998) reported the prevalence of pulp stones in a sample of 814 dental patients to be 22 per cent. In the current study, pulp stones were present in all groups, at rates ranging from 8.3 to 30.0 per cent. Subay *et al.* (2001) found no evidence of any correlation between the presence of pulp stones and the application of orthodontic force; nevertheless, it should be borne in mind that a pulp stone may complicate endodontic therapy.

Following agenesis, the presence of a supernumerary tooth in the cleft region has been stated to be the second most common dental anomaly (Ribeiro *et al.*, 2003). Supernumerary teeth have been reported to be present in various populations at rates ranging from 0.1 to 3.8 per cent (Peck *et al.*, 1994; Baccetti, 1998). The current study found high rates, from 1.9 to 10.0 per cent in the UCLP and CP groups. A higher rate (22.2 per cent) of supernumerary permanent teeth in the cleft area was observed in children with a unilateral cleft lip or palate, or both, and that supernumerary teeth related to a CLP result from fragmentation of the dental lamina during cleft formation (Vichi and Franchi, 1995).

The reported rates of *dens invaginatus* in the general population range between 2.0 and 2.95 per cent (Thongudomporn and Freer, 1998). Significantly higher rates (4.2–8.3 per cent) of *dens invaginatus* were found in the present study in the anterior region of the URCLP and ULCLP groups. Although *dens invaginatus* is not a common anomaly, it may present difficulties during endodontic treatment due to tooth anatomy (Garn *et al.*, 1963; Thongudomporn and Freer, 1998). While invagination is not considered to be a risk factor for apical root resorption during orthodontic tooth movement, invaginated teeth have been reported to have malformed roots more often than non-invaginated teeth (Horowitz, 1966); therefore, the clinician should be aware of this dental anomaly in making decisions regarding extractions. In addition, *dens invaginatus* places the tooth at a higher risk of devitalization from pulpal exposure and precautions should be taken, before any orthodontic treatment is carried out.

Dens evaginatus in the anterior region of the ULCLP group in the current study was found in 1.9 per cent on the cleft side and in 3.8 per cent on the non-cleft side. In order to avoid any unexpected loss of vitality during orthodontic treatment, early diagnosis of *dens evaginatus* is important so that alternative treatments, such as aesthetic restoration or a full crown, with or without root canal therapy, may be considered (Mattheeuws *et al.*, 2004).

Darwazeh *et al.* (1998) found a high rate of taurodontism (8.0 per cent) in non-cleft dental patients, with the maxillary second molar reported to be the most commonly affected tooth. In the current study, taurodontism was found at a lower rate (1.9 per cent) in the maxillary right and left molar regions but only in ULCLP group.

Among the different cleft groups, dilaceration, taurodontism, and *dens evaginatus* was found only in the ULCLP group. This supports the interpretations of Trotman *et al.* (1993) and Ranta and Rintala (1982) that different patterns of deformities may be related to different cleft

types. The rate of overall dental anomalies has generally been found to be higher among cleft patients in comparison with the general population, with anomalies most often located in the area of the cleft (Ribeiro *et al.*, 2003). The varying results reported in the literature can be explained by the application of different criteria in different studies and a lack of standardization of the data. Moreover, previous studies evaluated different types of clefts jointly, whereas the present study evaluated them separately; however, a much greater sample size would be necessary to draw more precise conclusions. The literature also suggests that associations between different tooth anomalies are 'clinically relevant', with individuals diagnosed with one anomaly at a possible increased risk for others (Thongudomporn and Freer, 1998; Hamasha *et al.*, 2002).

Conclusions

1. A significant proportion (96.7 per cent) of individuals with a cleft was found to have at least one dental anomaly.
2. Agenesis was the most common dental anomaly in this overall study sample.
3. The rates of dental anomalies varied among the different cleft groups.
4. The management of dental anomalies, which can easily be detected by careful inspection of routine orthodontic diagnostic records, should be taken into consideration in treatment planning of individuals with a cleft.

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