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Prevalence of dental anomalies in children with cleft lip and unilateral and bilateral cleft lip and palate

ABSTRACT

Aim To examine the prevalence of different types of dental anomalies in children with nonsyndromic cleft lip, unilateral cleft lip-palate, and bilateral cleft lip-palate.

Materials and methods A sample of 90 patients (aged 4-20 years) affected by isolated cleft lip, unilateral and bilateral cleft lip and palate was examined. Cleft patients were classified into one of three groups according to cleft type: (1) Unilateral Cleft Lip-Palate, (2) Bilateral Cleft Lip-Palate, and (3) Cleft Lip. Intraoral exams, panoramic radiographs and dental casts, were used to analyse the prevalence of the various dental anomalies included in this study.

Results There were no statistically significant differences between patients with cleft lip, unilateral cleft lip and palate and bilateral cleft lip and palate. The congenital absence of the cleft-side lateral incisor was observed in 40% of the sample, and a total of 30% patients showed supernumerary teeth at the incisors region. Second premolar agenesis was found in 4.4% of patients, whereas in 18.9% of the sample there was an ectopic dental eruption. Lateral or central incisors rotation was noted in 31.1% of the sample, while

shape anomaly, lateral incisor microdontia, and enamel hypoplasia were detected respectively in 25.6%, 5.6% and 18.9% of cleft patients.

Conclusion High prevalence of different dental anomalies in children with cleft lip and unilateral and bilateral cleft lip and palate has been confirmed. This study, in particular, shows the presence of ectopic and rotated teeth in the cleft area.

Keywords Cleft lip and palate; Dental anomalies; Prevalence.

Introduction

The most critical events affecting lip and/or palate formation occur between the 4th and the 10th week of embryogenesis, almost concurrently to the odontogenic process; indeed, it has been proposed that the aetiological factors disturbing face development, and leading to cleft formation, may even affect the odontogenesis [Stahl et al., 2006; Jordan et al., 1966; Böhn, 1963]. Many studies have revealed the higher prevalence of dental anomalies in children with isolated cleft lip or cleft palate or unilateral and bilateral cleft lip and palate than in general population [Akcam et al., 2010; Ribeiro et al., 2003; Ranta, 1986; Vichi and Franchi, 1995]. According to these studies, clefting should be considered as a part of a complex spectrum of malformations, caused by disturbed face and tooth development, also involving dental anomalies [Stahl et al., 2006; Perillo et al., 2011a; Perillo et al., 2011b; Edwards et al., 2014; Johal et al., 2013]. Several authors have demonstrated that the congenital absence of the cleft-side lateral incisor is the most common finding in children with cleft lip, cleft palate or both, while supernumerary tooth in the cleft area is the second most common dental alteration in these patients [Ranta, 1986; Ribeiro et al., 2003]. Tooth number anomalies, shape and structure alterations, timing of teeth formation, and dental eruption were frequently investigated in cleft patients but there are few studies [Tortora et al., 2008] that analysed extensively the presence of ectopic and rotated teeth inside the cleft area. The aim of this study is to determinate the presence of different types of dental anomalies in children with cleft lip, unilateral cleft lip-palate, and bilateral cleft lip-palate.

Materials and methods

A sample of 90 Caucasian children (59 boys and 31 girls) with isolated cleft lip and unilateral or bilateral cleft lip and palate, attending the Regional Centre for Rehabilitation of Cleft Lip and Palate – Second University of Naples, Italy, was investigated. Cleft patients included in this study were born between 1990 – 2008, with a

median age of 10 years (range 4-20), and did not show an associated syndrome or relevant medical diseases, to rule out possible dental anomalies associated with the presence of other illnesses. All subjects received the same surgical treatment. Cleft palate patients and those subjects with insufficient and incomplete records were excluded from investigation. The study was approved by the local ethical committee and the participants signed an informed consent agreement; in case of a minor, the informed consent form was signed by the parents.

Data were obtained by a single examiner to avoid differences due to variations in examination criteria.

Cleft patients were classified according to cleft type into one of three groups:

- 1) Unilateral Cleft Lip-Palate (UCLP);
- 2) Bilateral Cleft Lip-Palate (BCLP);
- 3) Cleft Lip (CL).

Even if male gender was prevalent (65.6%), the authors have not considered separately males and females because there are no statistically significant differences between the two gender and the presence of dental alterations in the cleft area (Table 1) [Böhn, 1963; Ranta, 1986; Vichi and Franchi, 1995]. The following dental anomalies were investigated inside the cleft area.

- 1) Tooth agenesis: congenital absence of a permanent tooth or germ.
- 2) Supernumerary tooth: a tooth additional to the normal series.
- 3) Ectopic eruption: the eruption of a tooth in an abnormal position.
- 4) Tooth rotation: the rotation of a tooth around its longitudinal axis.
- 5) Microdontia: a tooth that is much smaller than its contralateral homolog or a tooth of the same group from the opposing arch, a tooth that does not "fill" its space in the dental arch, or a tooth that appears small because of lack of proper shape.
- 6) Enamel hypoplasia: a hereditary condition in which the dental enamel shows either a break in continuity or surface loss, often because of insufficient calcification.
- 7) Tooth shape anomaly: a tooth with an abnormal anatomy, for example peg-shaped incisor.

The investigation was carried out with clinical exams, panoramic X-rays, and dental casts; past dental treatment records, such as previous tooth extractions, were considered.

Statistical analysis

In this study the prevalence rates for the different dental anomalies in the cleft area were calculated in CL, UCLP, BCLP and in total CLP groups. Continuous variables were reported as median and range, while categorical variables were reported as absolute number and percentage. For a more reliable analysis exact inference methods were adopted which do not make assumption about the null distribution. The overall prevalence of dental anomalies was reported as percentage and 95% exact confidence

	UCLP (n=36)	BCLP (n=17)	CL (n=37)	p-value
Age, years*	10 (4-20)	9 (4-16)	10 (4-20)	0.141
Gender				0.365
Male	23 (63.9%)	9 (52.9%)	27 (73.0%)	
Female	13 (36.1%)	8 (47.1%)	10 (27.0%)	

UCLP: unilateral cleft lip and palate; BCLP: bilateral cleft lip and palate; CL cleft lip; * median (range)

TABLE 1 Demographic characteristics of subjects by cleft group.

intervals (CI). Differences in characteristics of subjects among the three cleft groups were tested by means of the exact Kruskal-Wallis test and exact Pearson χ^2 test for continuous and categorical variables, respectively. Statistical significance was assessed for p-values<0.05. Data were analysed using SAS version 9.2 (SAS, Cary, NC).

Results

Tables 2 and 3, respectively, report the overall distribution of dental anomalies and the distribution of dental anomalies by cleft groups. There were no statistically significant differences between patients with cleft lip, unilateral cleft lip and palate and bilateral cleft lip and palate. The statistical analysis revealed a high prevalence (31.1%) of lateral or central incisor rotation in the sample and the presence of ectopic tooth (18.9%) in the cleft area. The congenital absence of the cleft-side lateral incisor was observed in 40% of the sample, whereas second premolar agenesis in 4.4% of patients. A total of 30% of the patients showed supernumerary teeth at the incisors region while in 18.9% of the sample there was an ectopic dental eruption. Lateral or central incisors rotation was noted in 31.1% of the sample, while shape anomaly, lateral incisor microdontia and enamel hypoplasia were detected respectively in 25.6%, 5.6% and 18.9% of cleft patients. In this study we found that a high proportion of patients have at least one dental anomaly. Median (range) for numbers of anomalies per subjects was 2 (0-4) for UCLP and BCLP subjects and 1(0-5) for UCL patients.

Discussion

Clefting can affect different areas of the maxillary structure, and the current classification includes: Cleft Lip (CL), Cleft Palate (CP), Unilateral Cleft Lip and Palate (UCLP), Bilateral Cleft Lip and Palate (BCLP) [Jiang et al., 2006]. Statistical differences in the prevalence of each dental anomaly for each cleft type have been reported by many authors [Akcarn et al., 2010; Menezes and

	N of anomalies	% (95%CI)
Lateral incisor agenesis	36	40.0% (29.9%-50.1%)
Lateral or central incisor rotation	28	31.1% (21.6%-40.7%)
Supernumerary teeth in the anterior region	27	30.0% (20.5%-39.5%)
Lateral incisor peg shaped	23	25.6% (16.5%-34.6%)
Enamel hypoplasia	17	18.9% (10.8%-27.0%)
Ectopic eruption	17	18.9% (10.8%-27.0%)
Lateral incisor microdontia	5	5.6% (0.8%-10.3%)
Second premolar agenesis	4	4.4% (0.2%-8.7%)

TABLE 2 Overall distribution of dental anomalies.

Vieira, 2008; Lai et al., 2008] and a correct evaluation of teeth anomalies is really important, because in these patients dental alterations worsen the aesthetic and functional problem of these children.

Dental anomalies can be found even in patients that do not show this congenital disease, but their prevalence is much lower than in cleft children [Akcem, 2010; Tortora et al., 2008]. Tooth number anomalies such as tooth agenesis and supernumerary tooth, are the most common dental anomalies found in cleft patients [Böhn, 1963; Vichi and Franchi, 1995]. Tooth agenesis and particularly congenital absence of the lateral incisor may be explained by a severe deficiency of mesenchymal mass, a deficiency in blood supply or by the surgical treatment [Jiroutová and Müllerová, 1994; Lekkas et al., 2000] in the cleft area. A less severe deficiency, instead, may be accompanied by a supernumerary tooth [Brook, 2009]. Anyways, tooth agenesis in individuals with cleft seems to be controlled by homeobox *Msx-1* genes and it is considered almost four times more frequent, compared to the general population [Vastardis et al., 1996]. It has also been postulated that the surgical procedure of palatal cleft closure may itself disrupt the permanent lateral incisor formation [Akcem, 2010]; moreover, Lekkas et al., did not observe congenitally missing teeth outside the cleft area in non-operated adult patients with cleft [Lekkas et al., 2000].

According to previous studies on the prevalence of congenitally missing lateral incisor on the cleft side, our results confirm that the maxillary lateral incisor is the tooth most frequently missing in the cleft area and the most common dental anomaly in all cleft types (CL, UCLP, BCLP) [Ranta, 1986]. In our sample, in CL patients the prevalence rates for congenitally missing maxillary lateral incisors were 40.5% while in UCLP and BCLP patients were respectively of 41.7% and 35.3% (Table

	UCLP (n=36)	BCLP (n=17)	CL (n=37)	p-value
Lateral incisor agenesis	15 (41.7%)	6 (35.3%)	15 (40.5%)	0.922
Lateral or central incisor rotation	9 (25.0%)	9 (52.9%)	10 (27.0%)	0.100
Supernumerary teeth in the anterior region	9 (25.0%)	4 (23.5%)	14 (37.8%)	0.413
Lateral incisor peg shaped	8 (22.2%)	5 (29.4%)	10 (27.0%)	0.857
Enamel hypoplasia	6 (16.7%)	3 (17.7%)	8 (21.6%)	0.938
Ectopic eruption	8 (22.2%)	5 (29.4%)	4 (10.8%)	0.216
Lateral incisor microdontia	2 (5.6%)	2 (11.8%)	1 (2.7%)	0.334
Second premolar agenesis	2 (5.6%)	0 (0.0%)	2 (5.4%)	0.685
N of anomalies per subject*	2 (0-4)	2 (0-4)	1 (0-5)	0.417

UCLP: unilateral cleft lip and palate; BCLP: bilateral cleft lip and palate ;CL unilateral cleft lip;. * median (range)

TABLE 3 Distribution of dental anomalies by cleft group.

3). Previous studies revealed that prevalence rate for lateral incisor agenesis range from 45.3 to 58.6 for UCLP patients and from 45 to 48.1% for BCLP subjects [Suzuki and Takahama, 1992a; Suzuki and Takahama, 1992b].

In our study, the statistical analysis showed no significant differences for lateral incisor agenesis in CL, UCLP and BCLP groups; however, tooth agenesis has been reported even outside the cleft area, in about 24% of clefts [Jiroutová and Müllerová, 1994] with a prevalence of 20.7% on the cleft side, and of 15.7% on the non-cleft side. In the present study, the maxillary second premolar was absent in 5.6% of UCLP patients and in 5.4% of CL subjects, while this anomaly was absent in BCLP children. Our result for UCLP patients is close to the prevalence rate reported by Tortora et al. investigation, while it is different for BCLP patients. Missing second maxillary premolars in BCLP patients according Tortora et al. data was of 25%, while in our study no BCLP patients showed second maxillary premolar agenesis [Tortora et al., 2008].

Supernumerary teeth are the second most common dental anomaly at the cleft area and are more frequent in cleft patients, distal to the cleft region [Ribeiro, 2003; Hellquist et al., 1979]. Our study confirms the high prevalence of this dental anomaly, showing that it is present in 25% of UCLP children, in 23.5% of BCLP patients and in 37.8% of UCL subjects.

Lateral or central incisor rotation was seen in 25% of the UCLP, and in 52.9% and 27% of BCLP and UCL patients [Larson et al, 1998]. According to Meazzini [Meazzini et al., 2007], tooth rotation could be the consequence of the surgery carried out to repair this malformation. Indeed, early secondary gingivoplasty reduces the anterior

alveolar space as well as space availability for tooth alignment [Meazzini et al., 2007]. Even if we support this hypothesis too, it cannot be actually confirmed because there are no patients with non-operated cleft to be used as comparison [Baccetti, 1998].

Anomalies in teeth size and shape, such as malformed and peg-shaped teeth, have been commonly reported in cleft patients [Akcem et al., 2010; Ribeiro et al., 2003]. It is generally accepted that there is a relationship between tooth agenesis and an overall reduction in tooth size, and alterations such as hypodontia and microdontia can be observed in the same patients [Baccetti, 1998; Garn and Louis, 1970]. Previous research has shown the prevalence of microdontia varies from 1.5% to 2% in the general population [Akcem et al., 2010]. In our study, we found that the prevalence rate for lateral incisor microdontia was 5.6% for UCLP patients, 11.8% for BCLP subjects and 2.7% for UCL children. The first result is similar to the prevalence rate for lateral incisor microdontia found by Ackam et al. in their investigation (1.9% - 4.2%) [2010]. Furthermore, in our study the percentage of shape anomaly of lateral incisor was 22.2% for UCLP group, 29.4% for BCLP group and 27% for UCL group.

Enamel hypoplasia, which is a quantitative enamel alteration, clinically observed as fossae, striae, or generalised absence of the enamel surface, has frequently been reported in patients with cleft lip and palate. Dixon attributed to trauma at the time of CLP surgery the high prevalence of enamel discolouration in children with CLP, as before [Dixon, 1968]. Malanczuk et al., instead, related enamel hypoplasia to the pathologic processes that cause the cleft itself [Malanczuk et al., 1999]. The percentage of enamel hypoplasia found in our investigation was 16.7% for UCLP patients, 17.7% in BCLP subjects and 21.6% UCL children.

Ectopic eruption is considered to be due to systemic as well as local factors [Tortora et al., 2008; Ballini et al., 2012]. The general population average for ectopic teeth ranges between 2 and 6% for the maxillary first molars and from 1.5 to 2% for the permanent canines [Ballini et al., 2012; Paolantonio et al., 2009; Bondemark and Tsiopa, 2007]. There are no other studies in literature that show ectopic eruption in cleft patients. Our results strongly indicate that 18.9% of the sample (UCLP 22.2% BCLP 29.4% and UCL 10.8%) showed an ectopic tooth in the anterior region of the cleft area.

Conclusions

Based on our results, it can be concluded that there is a high prevalence of dental anomalies in UCL, UCLP and BCLP patients, and that there are no statistical significances between these three groups. This study has confirmed the high prevalence of dental anomalies, affecting especially teeth of the anterior maxillary area where the cleft is located. Probably there is an interaction

of genetic factors that cause the cleft and environmental factors, as well as the surgical treatment, to modify the normal odontogenic process. This study adds other findings that are not extensively treated in literature, such as the presence of ectopic and rotated teeth in the cleft area. Dental alterations can represent aesthetic and functional problems especially in cleft patients, for this reason their study can be considered a very important step to complete cleft patients rehabilitation.

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