Evaluation of dental and maxillary development in patients with cleft lip alveolus

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Abstract
Our purpose was to determine in 20 unilateral cleft lip and alveolus subjects characteristics of the maxillary permanent teeth and maxillary arch development. Clinical and radiographic examinations have been carried out to identify congenitally missing teeth and sagittal skeletal patterns. Study cast assessment was undertaken to evaluate mesiodistal widths of individual teeth, intercanine and intermolar distances for the maxillary arch, as well as dental relationships. We observed that the congenital absence of the upper incisors was higher on the cleft side than on the non-cleft side. There was a statistically significant difference between the mesiodistal widths of cleft-side permanent upper lateral incisors, and their antimeres (p<0.05). Most of the patients presented a class II dental relationship on the affected side comparing to class I/III Angle on the unaffected side. We concluded that patients with isolated cleft lip and alveolus present perturbations in dental and maxillary arch development, a more severe disruption being recorded on the cleft side.

Keywords: cleft lip and alveolus, tooth agenesis, maxillary arch.

Introduction
The congenital lack of one or more permanent teeth is the most common congenital abnormality found in humans, affecting about 20% of the population worldwide [1]. Clefting of the lip and palate is one of the most frequent congenital malformations. All degrees of clefting may occur ranging from the benign submucous cleft to major incapacitating forms of combined cleft lip and palate. Oral clefts in humans are often associated with delayed development of dentition on the affected side comparing to the non-cleft side, as well as anomalies of number, size and shape of teeth on both sides [2–4]. Since most oral clefts are unilateral, it is of interest whether maxillary teeth characteristics are systematically different on the cleft side when compared with the non-cleft side. The aim of this study was to evaluate the development of upper permanent teeth and maxillary arch by using different parameters, in a Romanian sample of cleft lip alveolus (CLA) patients.

Material and Methods
Subjects
Twenty patients with isolated CLA, aged between 13–20 years old, mean age 15.6 ± 2.58 years old, and of which 60% were girls, were recruited for the study among the patients of the Department of Orthodontics and Dento-Facial Orthopedics, Faculty of Dental Medicine, “Carol Davila” University of Medicine and Pharmacy, Bucharest. Participation was based on informed consent.

Methods
The assessment of the cleft phenotype was based on the description present in the clinical records and confirmed through examination of study models and radiographs. The occurrence of dental anomalies was determined by oral investigation and examination of dental and radiographic records. Third molars were excluded from the study. The following dental anomalies were evaluated:

Tooth agenesis
Tooth agenesis was taken into consideration when at least one developmentally missing tooth was observed.

Microdontia
As it was defined by D’Souza RN et al. [5], microdontia was considered when a tooth was smaller than its contralateral homologous, a tooth that did not fill its space in the dental arch, or a tooth that appeared small because of the absence of the expected shape.
Atypical teeth position

Malpositions, rotations and transpositions were investigated.

Supernumerary and impacted teeth

The diagnosis of supernumerary teeth was based on the presence of an additional tooth to the normal series, found in any region of the dental arch [6]. Impacted tooth was considered the tooth that failed to emerge through the gums.

Delayed eruption

Delayed eruption was considered whenever it occurred beyond the time limits.

A study cast assessment was undertaken to evaluate the mesiodistal crown diameters of maxillary permanent teeth, intercanine and intermolar distances for the maxillary arch, as well as dental relationships. The intercanine distance was measured between the cusps of upper canines and the intermolar distance was measured between two left-right landmarks, located at the intersection of the lingual groove of the first upper molar with the gingival margin [7].

Sagittal skeletal and dental relationships were also investigated, based on oral examination, panoramic radiographs, lateral cephalograms and medical records. The sagittal skeletal relationships were assessed based on correlation between ANB angle and Wits A0B0 measurement. Dental relationships were considered as follows:

- Class I Angle: the mesiobuccal cusp of the upper first molar occludes in the mesiobuccal groove of the lower first molar.
- Class II Angle: the mesiobuccal groove of the lower first molar is placed behind the mesiobuccal cusp of the upper first molar.
- Class III Angle: the mesiobuccal groove of the lower first molar is placed forward to the mesiobuccal cusp of the upper first molar.

The measurements were made in duplicate and the mean value was calculated.

A single examiner (M.E.) did all measurements and oral examinations.

Data analysis

Data were expressed as appropriate ranges, means, standard deviations (SD) and proportions (%).

The t-Student test was used for group comparisons. Microsoft Office Excel 2003 was used for data analyses. A p value less than 0.05 was considered statistically significant.

Results

Fourteen out of 20 patients had the cleft located on the left side (Table 1, Figure 1).

Table 1 – Distribution of cases studied by cleft location and gender

<table>
<thead>
<tr>
<th>Cleft location</th>
<th>Male</th>
<th>Female</th>
</tr>
</thead>
<tbody>
<tr>
<td>CLA left side</td>
<td>6</td>
<td>8</td>
</tr>
<tr>
<td>CLA right side</td>
<td>2</td>
<td>4</td>
</tr>
</tbody>
</table>

Dental anomalies

Patients with CLA presented most frequently two symptoms of disturbed dental development.

Tooth agenesis

The front teeth were affected in various degrees. Thus, the congenital absence of the lateral incisors on the cleft-side was recorded in five (25%) patients, while its antimere was absent in two cases (10%). The upper central incisor was congenitally missing on the cleft-side in three (15%) cases. The second premolar was absent on the cleft-side in four (20%) patients, while in two (10%) cases the congenital absence was bilateral.

Microdontia

Microdontia was the most prevalent symptom. Mesiodistal crown diameters of central and lateral incisors on the affected side were smaller than the same parameters on the unaffected side. However, a statistical significant difference was recorded only for the lateral incisors (p<0.05) (Figures 2 and 3). There were no statistical differences between the mesiodistal widths of lateral teeth on the cleft side and the same measurements done on the unaffected side (Figure 4).

Atypical teeth position

The maxillary front teeth were also affected by malpositions. The lateral incisors and canines on the cleft side were the most affected teeth. Among patients who presented lateral incisor on the affected side, the tooth was frequently found in palatal position (50%). The canine was often observed in buccal position. Less often, the central incisor was found in malposition, changes in mesiodistal positions and rotations of variable amplitude being recorded, too.

Supernumerary and impacted teeth

No impacted or supernumerary teeth were observed in the study group.

Delayed eruption

Delayed eruption of teeth, was recorded in four patients (20%) and the most affected teeth were those situated at the cleft margins.

Maxillary arch development assessment

The transversal development of the maxillary arch is evaluated in Table 2.

Table 2 – The transversal development of the maxillary arch, assessed by intercanine and intermolar distances

<table>
<thead>
<tr>
<th></th>
<th>Cleft patients</th>
<th>Noncleft patients*</th>
</tr>
</thead>
<tbody>
<tr>
<td>Intercanine distance</td>
<td>27.95 ± 4.84</td>
<td>26.4 ± 1.38</td>
</tr>
<tr>
<td>Intermolar distance</td>
<td>34.13 ± 3.27</td>
<td>37.4 ± 1.7</td>
</tr>
</tbody>
</table>

*According to McNamara J Jr and Brudon WL [7].

The mean value and standard deviation for intercanine distance were 27.95 ± 4.84 mm. However, a malposition of canine on the cleft side (buccal...
position) was recorded in 35% of the cases. The mean value and standard deviation for intermolar distance were 34.13 ± 3.27 mm.

Sagittal skeletodental relationships

In 80% of the patients, there were class II dental relationships on the affected side, although the patients had either class I or class III Angle skeletal malocclusion. Dental malocclusion was either class I or class III Angle on the unaffected side, in agreement with the skeletal relationship (Figure 5).

Figure 1 – CLA located on the left side.

Figure 2 – Mesiodistal widths of upper front teeth in CLA patients (means ± SD).

Figure 3 – The mesiodistal width of lateral incisor on the affected side is smaller than the same parameter on the unaffected side.

Figure 4 – Mesiodistal widths of upper lateral teeth in CLA patients (means ± SD).

Discussion

Orofacial clefts have various distributions in different populations: 1/500 births in Asian population, 1/1100 births in Caucasians and 1/2500 births in African population [8]. The majority of these types of malformations are multifactor and a variety of environmental factors such as maternal smoking, maternal ingestion of anticonvulsivants or pesticides, have been implicated [9, 10]. It has been suggested that mutations in genes described as being important in palate and teeth formation, IRF6, MSX1 (both encode transcription factors) and FGFR1 (encodes a receptor for growth factor), result in occurrence of cleft palate and lip, and dental anomalies [11–13]. A large variety of dental anomalies can be found in patients with oral clefts. Smaller tooth sizes usually occur in these cases [14, 15]. The variability of the lateral incisor is probably
affected in large part by disruption at the cleft site. The central incisor may be diminished because of compromised vascularity [16]. Occurrence of bilateral asymmetries was described in subjects with oral clefts. Van Valen L [17] classified bilateral asymmetry into three kinds: (1) directional asymmetry, (2) antisymmetry, and (3) fluctuating asymmetry. Directional asymmetry occurs when a structure on one side of the body is systematically larger than its antimere, or an unpaired structure characteristically is located to one side of the midline. Oral clefts are an example of directional asymmetry [3]. The pattern of cell movements is directed by the pattern of gene expression, which determines cell surface properties and motility. The left-right anatomical asymmetry of the vertebrate body is foreshadowed by left-right asymmetry in the pattern of gene expression in the early embryo [18]. On the other hand, a left-right asymmetry in mesiodistal dimensions in subjects with unilateral clefts suggests a fluctuating asymmetry. The concept is that the same genetic and environmental factors control the growth of the left and right structures of the body. Moreover, reduced homeostasis causes differences in size and shape of the bilateral structures [3].

To our knowledge, the present study is the first report on cleft lip alveolus in Romanian patients. The primary target was to assess the characteristics of upper permanent teeth on the cleft side and to compare the results with those obtained from the unaffected side. The most commonly affected teeth were the lateral incisors, which were significantly smaller on the cleft side. This result is in agreement with other studies [19, 20]. However, the second premolars were also affected, a congenitally absence being recorded on the cleft side, as well as on the non-cleft side.

Large-scale studies have found lip-palate clefts to be twice as common on the left side as the right [21, 22]. Almost the same rate was found in our study (14 clefts located on the left side vs. six located on the right side).

In the present study, the intercanine distance was higher than the intercanine distance measured at the patients without cleft [7] due to the malposition of canine on the cleft side (buccal position) in 35% of the cases. In 65% cases, the intercanine distance had lower values than the normal ones; therefore, the mean value for the intercanine distance in cleft patients is not representative for the assessment of the transversal development of the maxillary arch in the anterior (canine) region.

The mean value of intermolar distance was lower than the mean value previously reported in a group of patients without clefts [7]. A movement of the maxillary lateral segment to the midline could explain this result.

In most cases (80%), there were class II dental relationships on the cleft side, which do not agree to sagittal skeletal relationships and sagittal development of the maxilla, probably due to the shift of the maxillary lateral segment to the midline. Dental agenesis and dental malpositions on the affected side also represent an important cause. On the other hand, the side without cleft presented dental class I, or III relationships, in agreement with sagittal skeletal malocclusion.

**Conclusions**

Since both upper permanent teeth and maxillary arch are affected, it might be concluded that isolated CLA is not just a single anatomically localized disruption in development. Association of clefts with perturbations in dental development is therefore caused not only by the genetic factor, but also by environmental factors.

Further studies need to be conducted on larger population samples in order to determine the tooth size, shape and structure in cleft lip palate patients, as these markers play an important role in evaluating child’s health.

**References**


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