TREATMENT APPROACH FOR THE RARE MULTIPLE DENTAL ANOMALIES IN A SINGLE TOOTH IN PATIENT WITH OROFACIAL CLEFT

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ABSTRACT

Background. Orofacial cleft (OC) is the craniofacial malformation that affects facial structures. It is often associated with a high prevalence of dental anomalies that can be morphological, numerical, and structural. However, a single tooth is rarely affected by two anomalies. This article describes the diagnostic and treatment of a rare case of a permanent maxillary lateral incisor with three dental anomalies in a patient with OC.
Case Description. A male with cleft lip and palate, operated of the lip, at nine years of age, after a radiographic examination, was diagnosed with three dental phenotypes: microdontia, dens invaginatus, and root dilaceration in the right upper permanent lateral incisor. At the age of 11, a secondary alveolar bone grafting (sABG) was performed with the autogenous iliac bone in the cleft area. After the maxillary expansion with Hyrax expander, a traditional fixed appliance was installed for dental alignment, without involving the right upper permanent lateral incisor due to the anomalies in this tooth. At the age of 16 years old, a small extrusion of this tooth was performed with a self-ligated bracket, moving this tooth for better alignment to facilitate the aesthetic procedure of re-anatomization.
Practical Implications and Conclusion. The rehabilitative dental treatment with orthodontic intervention was proposed to restore aesthetics and function. This approach improved the patient's oral health and quality of life. In patients with OC, considering the possibility of multiple dental anomalies, a broad and accurate diagnosis is important to develop the most appropriate rehabilitation treatment plan for each case.

Keywords: Dental anomalies. Microdontia. Dens invaginatus. Root dilaceration. Cleft lip. Cleft palate. Craniofacial abnormalities.

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INTRODUCTION

Orofacial cleft (OC) is among the most common congenital anomalies and is classified as a heterogeneous disorder that can affect different craniofacial anatomical structures. It presents a complex multifactorial etiology when it occurs as an isolated phenotype [1,2]. Dental anomalies are highly prevalent in individuals with OC. These dental characteristics have been attributed to anatomical alteration resulting from the presence of the cleft in the alveolar ridge or even to possible interferences in genetic signalling or gene expression that would have an impact on the odontogenesis process, thus resulting in changes in the number, size, and shape of the teeth, among others [3].

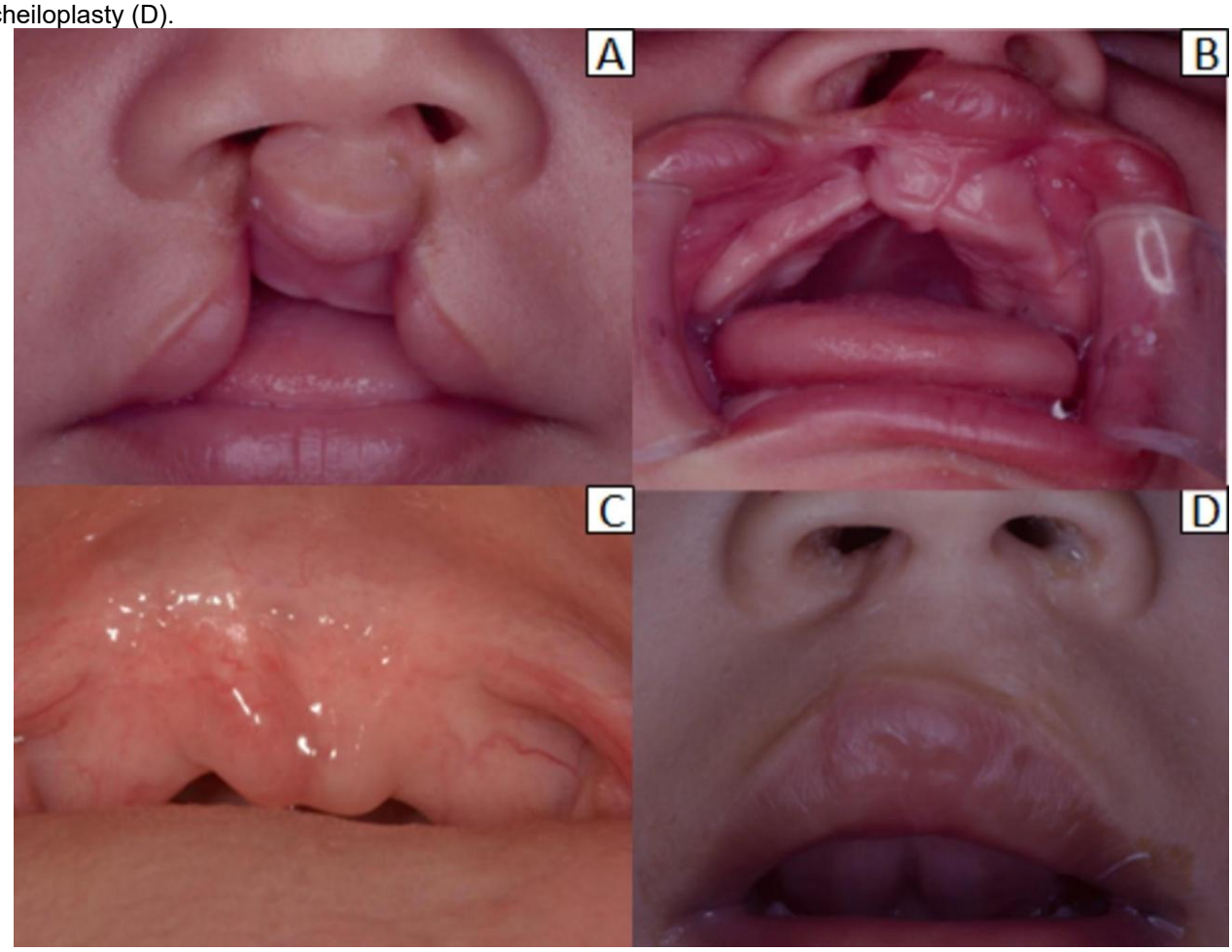
Microdontia, dental agenesis, taurodontism and supernumerary are some of the developmental anomalies that can affect the crown and/or root of tooth described in literature associated with OC [4-11].Although subjects with OC present many dental phenotypes, the concomitant occurrence of the multiple dental phenotypes in a single tooth in an individual with OC is extremely rare and has not yet been reported in the literature. From a clinical point of view, the occurrence of dental anomalies in the extensive dental rehabilitation process of the OC represents an additional challenge for the dentist [5,7].Therefore, this study aimed to report a rare case of a subject with OC with three dental phenotypes (microdontia, dens invaginatus and root dilaceration) in a single tooth. The clinical characterization of the OC and the three dental phenotypes diagnosed of the permanent upper lateral incisor on the right side (tooth 12) are presented through clinical and radiographic images, and the therapeutic proposal adopted for the rehabilitation process.

CASE REPORT

PATIENT INFORMATION, CLINICAL FINDINGS AND DIAGNOSTIC ASSESSMENT

A 2-months-old male was admitted to the Hospital for Rehabilitation of Craniofacial Anomalies of the University of São Paulo (HRAC-USP) for treatment. The patient was diagnosed with two different types of orofacial clefts associated: on the right side a cleft lip and alveolus and on the left side a cleft lip only, associated concomitantly with a submucous cleft palate (Figure 1 A-C). No other major anomalies were detected during the physical examination or family anamnesis. Therefore, the diagnosis was non-syndromic orofacial cleft (OC), as the clinical findings did not indicate any associated syndrome. During the diagnostic examination, the family was advised on the possibilities of breastfeeding and general guidelines on oral hygiene, including the cleft area.

Figure 1. Clinical image in frontal view showing cleft lip and alveolus on the right and cleft lip only on the left (A). Intraoral clinical image showing submucous cleft palate (B-C). Clinical image in frontal view after bilateral cheiloplasty (D).



THERAPEUTIC INTERVENTION

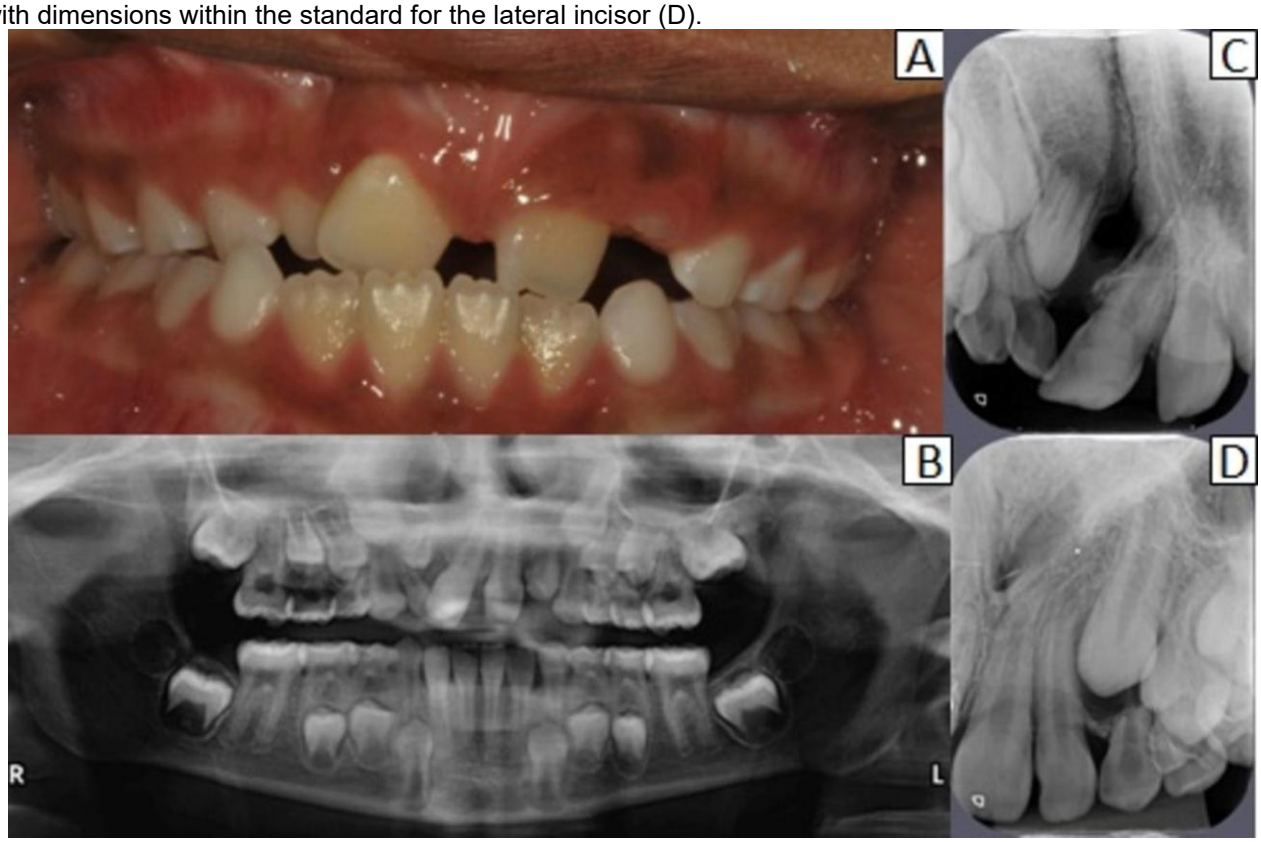
The treatment protocol in the Craniofacial Center consists of two primary reconstructive surgeries. The first is the cheiloplasty, which is the surgical repair of the lip, and after that, the palatoplasty, for the surgical repair of the palate. In this case report, the cheiloplasty was performed bilaterally at one year and six months of age (Figure 1 D).

After the first surgery, the multidisciplinary team evaluated the patient for palatoplasty. As the patient had only an incomplete cleft palate, classified as submucosal, in which there was only the clinical characteristic of bifid uvula and was asymptomatic, the initial option was not to operate and to monitor the functional evolution. Functionally, during follow-ups with the team, muscle diastasis was not observed, and speech was considered adequate and satisfactory by the parents, the patient, and the speech therapist. Thus, because this submucous cleft palate remains asymptomatic, there was no recommendation for palatoplasty.

The patient was continually and periodically monitored by the multidisciplinary team, including pediatric dentistry and orthodontics (Figure 2 A). During dental attendance at age

9, as a complementary examination to the clinical dental examination, a panoramic radiograph was performed. It was possible to diagnose that the right upper lateral incisor (tooth 12) presented three dental anomalies: microdontia, dens invaginatus type II Oehler's classification 12 and root dilaceration (Figure 2 B-C and Figure 3 B). Other radiographic evaluations were conducted throughout the treatment process, including periapical radiographs (Figure 2 C-D). These radiographic studies provided comprehensive insights into the dental structures, roots, and surrounding bones.

Figure 2. Patient aged 9 years in the mixed dentition, clinical image (A) and panoramic radiograph (B). Periapical radiograph of the cleft region where it is possible to observe the bone defect in the alveolar ridge and the intra-osseous right upper lateral incisor (12), in formation, showing dens in dente and microdontia (C), in contrast to the periapical radiograph of the region of the left upper lateral incisor (22) already erupted and with dimensions within the standard for the lateral incisor (D).

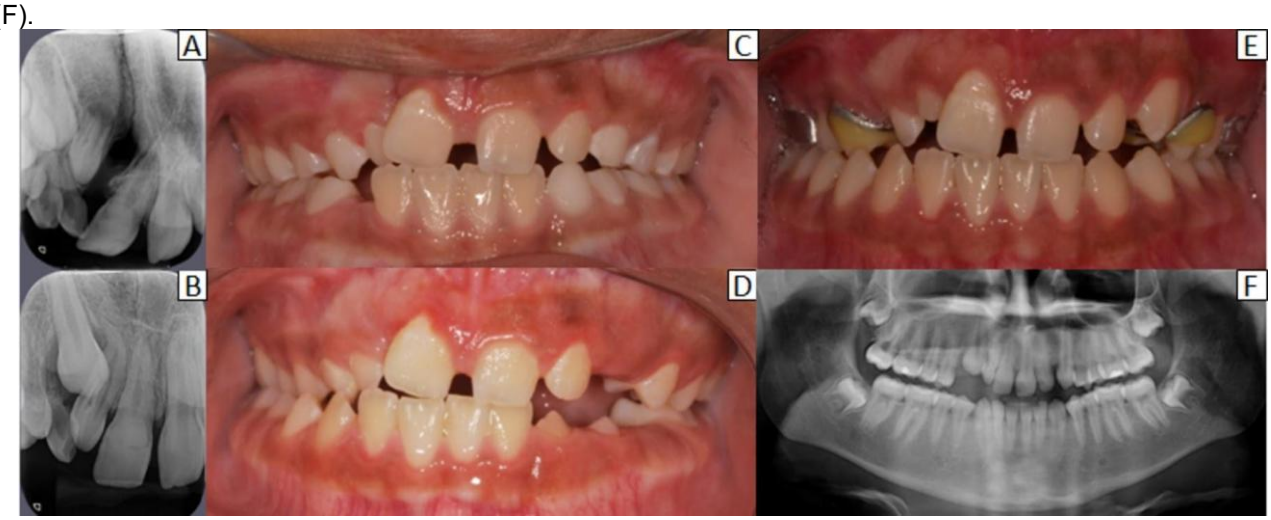


As the patient presented the cleft on the right side involving the alveolar ridge, what determines a bone defect in the region, at the age of 11, following the aesthetic and functional rehabilitation protocol, a secondary alveolar bone grafting (sABG) was performed in the region of the bone defect (Figure 3 A-D). The sABG aims to fill the defect caused by the cleft with the autogenous iliac bone, encouraging gain of bone support and stabilizing the maxillary arch to perform the orthodontic movement mechanics of the teeth in the region. In the postoperative control of 6 months sABG, the confirmation of osseointegration with bone neoformation in the area was confirmed radiographically (Figure 3 B). Clinically, it

is also possible to observe the continuity of the right vestibular alveolar region, which made it possible to continue with great security orthodontic treatment (Figure 3 D).

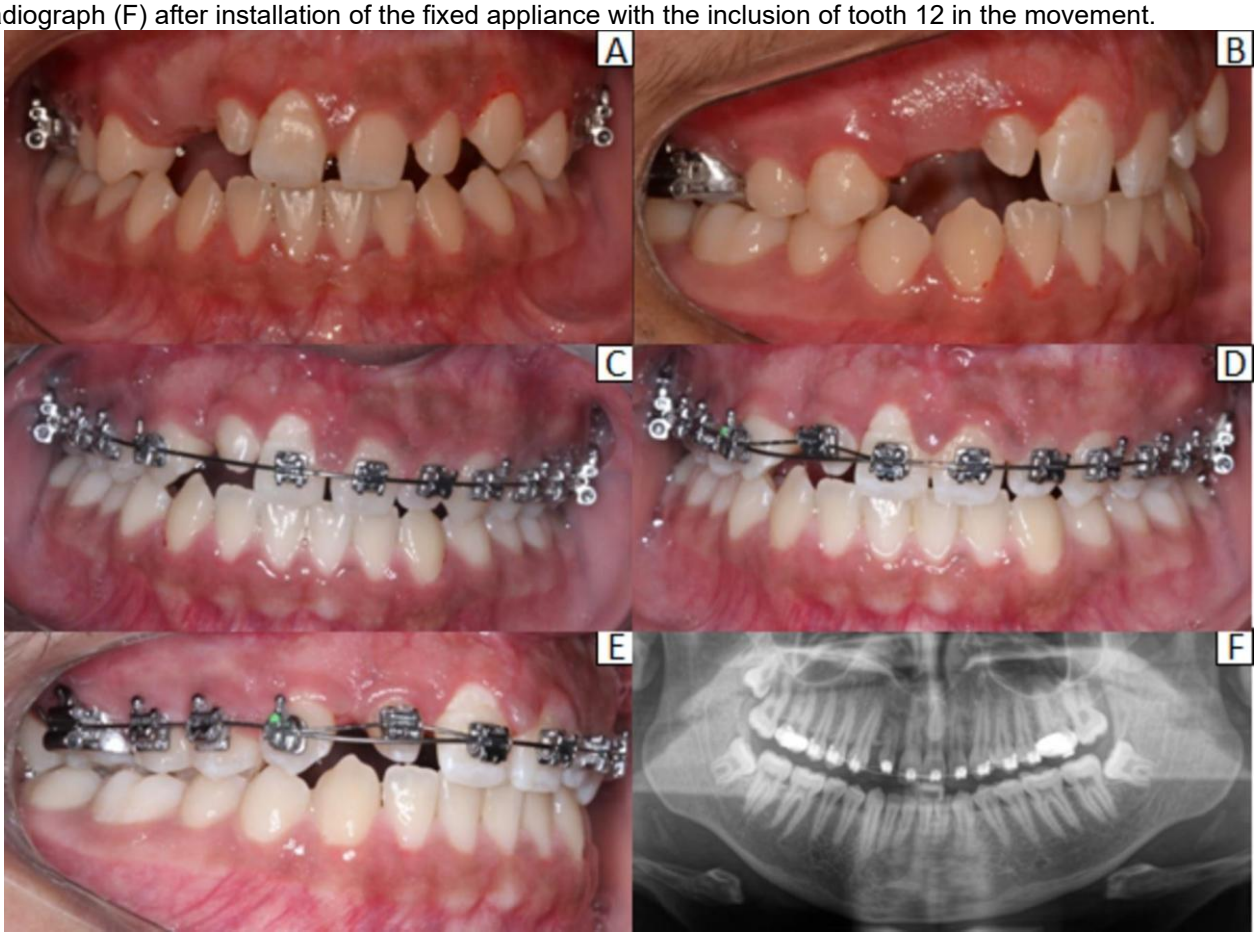
In the context of the dental rehabilitation process, at the age of 14, due to maxillary atresia, there was an indication of maxillary expansion. The Hyrax expander was installed, providing a better conformation of the arches and adequate space for dental repositioning (Figure 3 E-F).

Figure 3. Periapical radiograph on the right side (alveolar cleft region) and clinical image in frontal view in occlusion: Before sABG (A and C) and 6 months after sABG (B and D). In the first radiographic image it is possible to observe dens in dente and microdontia of the right upper lateral incisor (12) with incomplete root development (A). The Periapical radiograph after sABG shows the right upper lateral incisor (tooth 12) with complete root development presenting root dilaceration, in addition to dens in dente and microdontia (B).Clinical image in frontal view in occlusion after installation of the expander (E), in these clinical images it is possible to observe the right upper lateral incisor (12) with microdontia erupted in the grafted area. Panoramic radiograph of in the permanent dentition after expansion, taken prior to the installation of the fixed appliance (F).



After expansion, a traditional fixed appliance was installed in the upper arch for dental alignment, up until this phase, without involving tooth 12 in the movement, especially due to the changes in dimension of this tooth (microdontia) and root (root dilaceration) (Figures 4 A-C). With the eruption of the tooth 12, when the patient was 16 years old, a small extrusion of this tooth (12), with self-ligated bracket, was performed, moving this tooth for better alignment on the edge and to facilitate the aesthetic procedure of re-anatomization (Figure 4 D-F).

Figure 4. Clinical image in occlusion in frontal and right side view, in the preparation phase for installation of the fixed appliance (A-B). Clinical image in frontal view in occlusion after installation of the fixed appliance without including tooth 12 (C). Clinical images in occlusion in frontal view (D) and side view (E) and panoramic radiograph (F) after installation of the fixed appliance with the inclusion of tooth 12 in the movement.



FOLLOW-UP AND OUTCOMES

Throughout the treatment process, interdisciplinary discussions were conducted among professionals, primarily involving specialists in orthodontics, restorative dentistry, and radiology, among others, to assess the path forward, analyzing possibilities and defining the best course of action while considering the patient's desires and expectations. These collaborative efforts ensured a holistic approach to addressing the multiple dental anomalies affecting the single tooth, allowing for informed decision-making and personalized treatment plans. By engaging in these discussions, dental specialists from various fields could pool their expertise, resulting in comprehensive evaluations and optimized outcomes tailored to meet the specific needs of each patient. This interdisciplinary collaboration not only enhanced the quality of care provided but also fostered a patient-centered approach, ultimately leading to greater satisfaction and success in treatment outcomes.

The patient involved in this study has provided their free and informed consent for the publication of information related to their case.

DISCUSSION

Orofacial clefts are considered the most common malformations related to the craniofacial region, involving different oral structures with great morphological diversity[1]. Individuals with OC require extensive dental care. The presence of dental development disorders resulting in a more complex treatment that demands greater attention [6]. In this case report, three developmental anomalies (microdontia, dens invaginatus and root dilaceration) were diagnosed in the right upper lateral incisor (tooth 12). From a dental clinical point of view, these phenotypes have an aesthetic and/or functional impact.

Microdontia consists of the reduction of the standard dimensions of one or more teeth [13]. The hypothesis is that the microdontia would be a developmental defect in the context of the phenotypic spectrum of dental agenesis. In this line, in some teeth, the dental germ does not form, resulting in agenesis, while in others, there could be a "hypo-development" of the germ, resulting in microdontia[14]. The literature points out that the occurrence of this phenotype is related to the anterior teeth, which directly affects the aesthetics and harmony of the smile [13]. Additionally, microdontia can result in diastema and, in terms of occlusion, can cause dental misalignment with malocclusion. The most conservative rehabilitation protocol for a tooth with microdontia involves aesthetic restorations aiming at its re-anatomization. On the other hand, in cases where the root structure is not sufficiently effective for the maintenance of the tooth, extraction may be recommended, with the rehabilitation of the area being carried out by moving the contiguous canine or even with the placement of implants [13].

The dens invaginatus, the other anomaly diagnosed, is considered a rare developmental anomaly [12]. This anomaly occurs due to an invagination of the enamel organ on the palatal face towards the internal region of the tooth, the enamel is thinner and more susceptible to fracture or even demineralization due to infection resulting from the accumulation of bacterial plaque [12,15]. Difficult access to the root can lead to complications such as internal resorption, abscess, or cyst formation when the endodontic intervention is required [16]. Oehlers [12] in 1957 classified this anomaly into three types. Type I is restricted to the coronary region, type II crosses the line of the cement-enamel junction without reaching the apex, and type III extends to the apex region and may result in an independent apical or lateral foramen [12]. In the case report in question, dens in dente was classified as type II, since it is located in the cervical and middle root thirds. There was no consequence to the health of the dental pulp due to the dens invaginatus, and thus, we opted for preservation, with hygiene guidance measures and plaque control [16].

The third anomaly, the root dilaceration, consists of an abnormal change in the curvature of the crown and/or long axis of the root. The etiology is quite varied and may be related to trauma, insufficient space for development, presence of supernumerary teeth, and hereditary factors. The identification of the root dilaceration is especially important for endodontics and orthodontics[17,18]. In endodontics, root dilaceration can complicate instrumentation, cleaning and obturation, increasing the risk of instrument fracture, incomplete disinfection and inadequate sealing. Effective management strategies include thorough diagnostic imaging, careful planning, and the use of appropriate instruments. In orthodontics, root dilaceration can make tooth movement difficult and increase the risk of root resorption. Applying controlled, gentle forces, frequent monitoring with radiographs and interdisciplinary collaboration are crucial for successful outcomes in such cases. Therefore, recognizing and appropriately managing root dilaceration is essential for achieving optimal results in both endodontic and orthodontic treatments[18].

The subject of this case report has not so far required any endodontic but required extensive orthodontic treatment. The involvement of an orthodontist is essential for the decision of maintenance or extraction of a tooth, especially in the area of the cleft, since the occlusion is directly related to complete rehabilitation [19]. In the present case, the choice of orthodontic movement properly positioning the permanent upper lateral incisor adjacent to the cleft after sABG surgery can contribute to the maintenance of the bone graft by mechanical stimulation. In addition, providing a functional, stable occlusion and a more harmonious facial and dental aesthetics [19], since the natural tooth allows more favorable aesthetic results [20].

From a clinical point of view, the phenotypes described in the reported case represent an additional challenge to the rehabilitation process of orofacial clefts. In this context, we believe that ongoing clinical and radiographic care is extremely important, as are interventions at appropriate times, aiming at satisfactory rehabilitative dental treatment to restore aesthetics and function, resulting in improved oral health and quality of life for the patient.

This case report emphasizes the importance of recognizing the potential coexistence of multiple dental anomalies in patients with orofacial clefts. Early and precise diagnosis is crucial for developing an effective treatment plan to ensure optimal oral rehabilitation.

Conflict of Interest Disclosure: The Authors declare that there is no conflict of interest.

Ethics Approval Statement: This case report has been reviewed and approved by the Research Ethics Committee of Hospital for Rehabilitation of Craniofacial Anomalies of the University of São Paulo (HRAC-USP) (No.76333523.3.0000.5441), ensuring compliance with the ethical standards laid down in an appropriate version of the Declaration of Helsinki (as revised in Brazil 2013).

Patient Consent Statement: The patient involved in this study has provided their free and informed consent for the publication of information related to their case. The consent form has been duly signed and filed by the authors.

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