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Regional Odontodysplasia: Report of a Case in the Mandible Crossing the Midline

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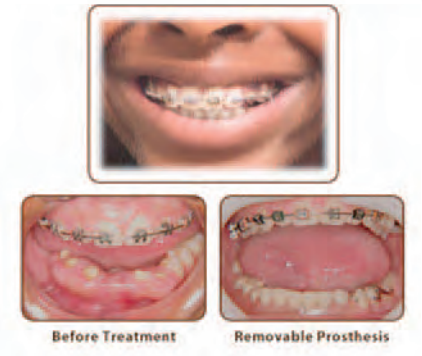

Abstract

Aim: This report presents an unusual case of regional odontodysplasia (RO) in the mandible which crosses the midline along with its clinical management.

Background: RO is an uncommon, nonhereditary unilateral developmental anomaly involving the dental hard tissues, most frequently affecting the maxillary teeth.

Report: RO was diagnosed in a 12-year-old. The dysplastic teeth were the mandibular canines, lateral incisors, central incisors, and the right premolars. The treatment was extraction of the affected teeth followed by provisory prosthetic rehabilitation.

Summary: The etiology of RO remains obscure. In this case a possible association with pre-natal trauma is suspected. The literature is not unanimous as to the management of RO. Nevertheless, the presence of a dental abscess is an indication for extraction. Since general bone quality is not affected, a provisory prosthesis was fabricated as the patient waits for future implant rehabilitation. The executed treatment met the expectations of the patient and her mother as it improved the patient's masticatory function, esthetics, and quality of life.

Clinical Significance: The general practitioner must be prepared to deal with different situations in the dental office. The diagnosis of RO relies mainly on clinical and radiographic findings, and its treatment depends on the affected teeth, the severity of the dysplasia, the presence of infection, and the patient's age.

Keywords: Regional odontodysplasia, RO, dental anomalies

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Introduction

Regional odontodysplasia (RO) is an uncommon, nonhereditary developmental anomaly involving the dental hard tissues derived from both epithelial and mesechymal components of the tooth-forming apparatus.[1] The prevalence of this condition is not known, as the literature largely consists of case reports.[2,3] RO tends to affect several adjacent teeth within a particular segment of the jaw and generally does not cross midline. Frequently, it is located only on one arch, affecting the structures of both primary and permanent teeth. The maxillary teeth are affected more frequently than the mandibular teeth with the central and lateral incisors and canines are more frequently affected than posterior teeth.[1] Some authors have reported RO to be more common in females than males.[1,4] However, Lopes et al.[5] state further studies with a greater number of cases are needed to confirm this tendency. Data on a specific ethnic distribution are not available.[4]

The criteria for diagnosis of RO are mainly based on clinical and radiographic findings but sometimes supplemented by histological findings.[2,3] Clinical examination reveals affected yellowish or brownish discolored teeth that are atypically shaped with surface pits and grooves and soft on probing. The condition is usually unilateral, although exceptions can be found.[4] The anomaly is usually localized in one arch, with incidence higher in the maxilla.[3,4] It is rare to find almost all teeth of the same arch affected. In cases where both arches are involved the presentation is usually unilateral. The affected teeth most often occur as a continuous series, although occasionally the anomaly will "skip" a tooth or group of teeth. Eruption of the affected teeth is often delayed or failed.[3]

Radiographically, the affected teeth show an abnormal morphology and hypoplastic crowns. The enamel and dentin are less radioopaque than unaffected counterparts, and there is little demarcation between enamel and dentin.[4,6] The pulp chambers and root canals are wide, giving the appearance of "ghost teeth."[3,4]

Histologically, all structures of the dental germ are affected.[6] A typical finding is hypoplastic, hypomineralised, and matrix-enriched enamel of variable thickness. Often globular calcifications or apatic needles have been found. The prism structure appears atypical.[4] The greater density of the enamel is not evident in conventional radiographs, probably because of the thinness of the enamel layer in affected teeth.[3] The dentin is reduced in thickness and expanded areas of irregular interglobular dentin are found. A layer of extended predentin is frequently observed. In severe cases the tubules are reduced in number and their processes are disturbed. In less severe cases the dentin and, in particular, the mantle dentin seems to be normally structured.[4] Gingival enlargement, which accompanies the affected teeth in some cases, usually shows a parakeratinized surface epithelium with acanthosis and very hyperplastic rete ridges.[6]

No factor has been positively identified as the single cause of the condition, although many factors have been linked to it such as: medication; viral infection;[2,7] vascular disturbance (localized ischemia); somatic mutation; failure of migration of neural crest cells; localized trauma; and idiopathic factors.[2] The reason the vast majority of cases, particular contiguous teeth, are affected with no involvement of others remains elusive.[6]

Although the histolgical features are distinctive, the diagnosis is based on clinical and radiographic findings and coincides with the eruptive periods of the primary and permanent teeth.[6]

Case Report

A 12-year-old girl was referred to the Pediatric Clinic of the School of Dentistry at the Universidade de Cuiabá - UNIC in Cuiabá, Brazil for evaluation of the absence of eruption of some permanent teeth.

Diagnosis

During a review of the medical history, the mother reported a fall during the third trimester of pregnancy. At birth the baby girl presented with swollen and reddish skin on the right side of the face along with a deviation of the chin to the right side that eventually disappeared over time.

The extraoral examination revealed nothing of significance. The patient had no facial asymmetry or other extraoral abnormality. She was in the permanent dentition stage, with all maxillary teeth

being normal and under orthodontic treatment. The mandibular canines, lateral incisors, central incisors, and the right first premolar had short crowns and were grossly hypoplastic and discolored (Figure 1).

The patient complained of fibrous gingival tissue observed in the area affected by the regional odontodysplasia.

The panoramic (Figure 2) and periapical radiographs (Figures 3 and 4) revealed enlarged pulp chambers and short roots in the mandibular canines, lateral and central incisors, and the right side premolars. Dental abscesses could be seen clinically and in the periapical radiograph in the incisors region (Figure 3).

Development of these teeth was delayed, and they had thin coronal enamel and dentin. The periapical radiographs of the involved teeth also showed indistinct, irregular periapical radiolucency. A diagnosis of RO was made based on these findings.

Treatment

Since the involved teeth could not be restored effectively, the extraction of the dysplastic teeth followed by provisional prosthetic rehabilitation was recommended to the patient's mother.

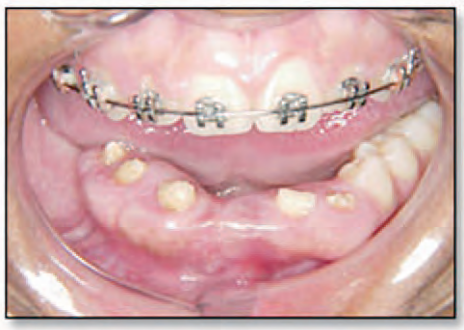

Figure 1. Clinical aspect of the intraoral examination.

Surgery was accomplished under local anesthesia using the mucoperiosteal flap technique via a vestibular approach. Extraction of the dysplasic permanent teeth, alveolar curettage, irrigation of the surgical site, and flap replacement were performed. Fibrous gingival tissue was removed and before sutures were placed, a section of fibrous gingiva and a bone fragment in the area of the regional odontodysplasia were immersed in 10% formaldehyde preservative. The tissue biopsies were sent to an oral pathologist for a histopathological analysis to confirm the clinical and radiographic diagnosis of RO.

Biopsy of the gingival fragment showed the presence of islets of odontogenic epithelium in the connective tissue as well as some fibrosis.

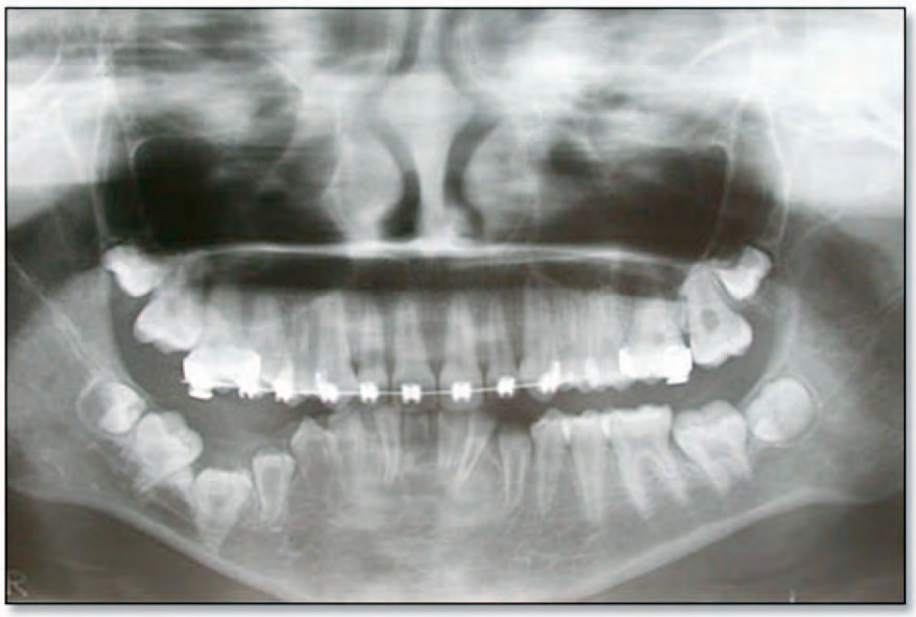

Figure 2. Panoramic view showing affected teeth.

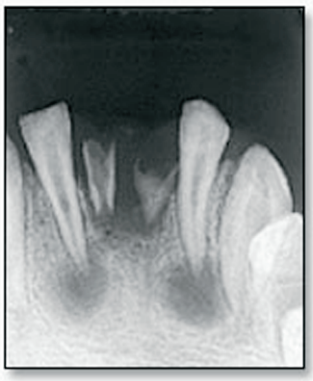

Figure 3. Periapical radiograph showing abscesses in the lower incisors region.

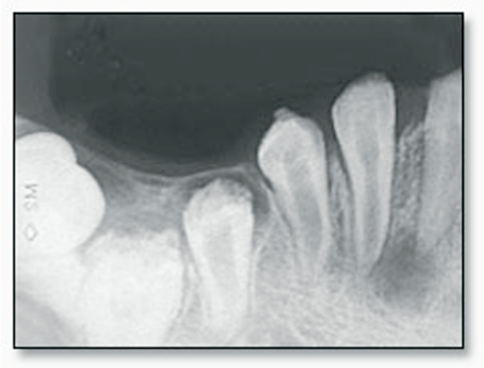

Figure 4. Periapical radiograph showing enlarged pulp chambers and short roots in the lateral incisors, canine, and premolars.

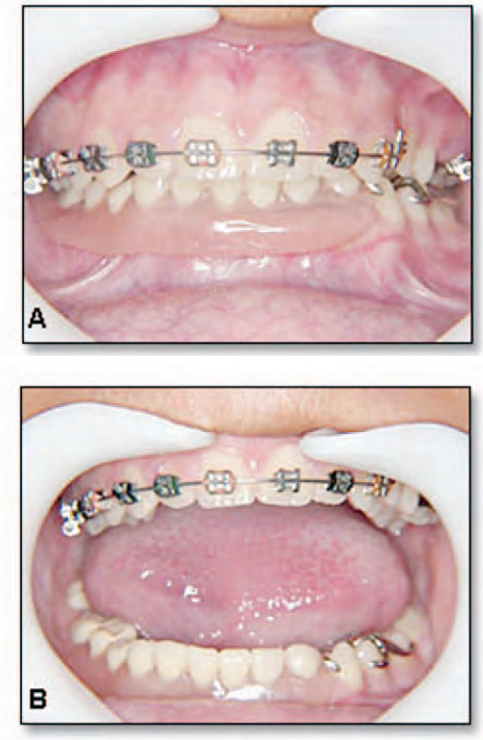

Figure 5. Insertion of the removable prosthesis replacing the extracted teeth to regain esthetics and masticatory function. **A.** In occlusion. **B.** Open mouth.

No complication during the postoperative phase was observed. After three months, the patient returned for a second surgical intervention involving an alveoloplasty of the region where the teeth were extracted.

After a month, a temporary acrylic removable prosthesis was fabricated in order to preserve the alveolar ridge during the period of skeletal growth and to meet the patient's esthetic, functional, and psychological needs (Figure 5).

Oral hygiene instructions, dietary analysis and advice were given, and regular appointments were planned in order to prevent caries and periodontal disease. The patient was placed on periodic recall to monitor the development of the mandibular arch and the prosthesis adaptation.

Discussion

Although there are several hypotheses, the etiology of RO is still obscure. It has been suggested the disturbance may derive from a disorderly proliferation of the dental epithelium at an early stage of tooth development. Other causal factors suggested include viral infection, localized trauma, ischemia, irradiation, metabolic and nutritional disturbances, vitamin deficiencies, local somatic mutation, genetic transmission, medications, failure or migration of neural crest cells, and local vascular defects.[1] Some patients may also present with systemic anomalies, such as facial asymmetry and other abnormalities.[8]

After an extensive literature review, Tervonen et al.[4] concluded the etiology of RO remains unknown and further detailed investigations of cases of RO are needed to confirm possible etiologic factors. One theory suggests RO could be caused by a vascular disturbance creating a local ischemia affecting odontogenesis. This theory is supported by the occurrence of associated hemangiomas or vascular nevi in areas adjacent to affected teeth in many of the reported cases.[7]

In the current case, the reported fall of the mother during pregnancy and the facial alteration at birth were identified as a feasible cause for the RO

because of the possible association with local ischemia that could affect odontogenesis.

Gingival swelling and delayed eruption were the chief complaints of the case, which had also a history of abscesses in the affected area, in accordance with Fanibunda and Soames.[9]

RO affects the enamel and dentin, both of which are hypoplastic and hypocalcified. Because these teeth are structurally defective, they are more susceptible to caries and fractures resulting from trauma. Failed, delayed, or partial eruption is a common finding as well as non-inflammatory gingival swellings, recurrent infections, and abscess formation even in the absence of gross decay.[1]

The radiographic features of RO have consistently demonstrated thin and defective layers of enamel and dentin, resulting in a faint and fuzzy outline. This reduced radiopacity with loss of distinction between enamel and dentin have given rise to the term "ghost teeth." The pulp chambers and canals also appear enlarged with the roots being shortened and stubby with open apices.[1]

The present case of RO is rare since the majority of the reported cases affects only one quadrant of the maxilla and does not cross the mid-line. The appropriate treatment for RO varies. One approach is to protect the erupting affected permanent teeth from fracture and infection by restoring them with preformed crowns without any tooth preparation. Pulpotomy or apexification, depending on the tooth vitality, should also be considered in order to stimulate root formation. However, if the affected permanent teeth erupt with very short roots and wide-open apices, extraction may become necessary due to pulp necrosis and/or tooth instability. In cases of recurrent infection, teeth with RO should be surgically removed.[1] Despite the importance of maintaining the dysplastic teeth in position in the arch to prevent undesirable drifting of unaffected teeth the presence of abscesses contraindicates the retention of these teeth.[1] This is particularly significant for children whose teeth can not be replaced by implants within a short period following surgery.

The goal of treatment is to recover the masticatory and esthetic function, reduce the psychological effects caused by the absence of teeth, maintain the vertical dimension, and prevent the development of facial asymmetry.[10]

Due to the growth of the alveolar bone and eruption of the permanent teeth, the patient should return every four to six months for periodic evaluation and to modify the prosthesis to accommodate growth and development.[10] The temporary prosthesis can be maintained until the age of 17 or 18 years when the gingival margin is stable and restoration with a fixed prosthesis can be considered.[6]

Since the general quality of bone is not affected in RO,[6] the patient and her mother were informed about the possible use of implants in the affected area in the future. The patient is being monitored until she reaches adulthood. At that time, a final rehabilitation for loss of the incisors may be accomplished after the facial bones have stopped growing.

Treatment of RO is controversial and no consensus has yet been reached. The dentist should take into consideration factors such as the age of the patient, the medical history, previous dental experience, the number of affected teeth, the presence or absence of any associated pathology, as well as the attitude and wishes of the child and parents. The aims of the treatment should include aiding mastication and speech, improving aesthetics, reducing the psychological impact, allowing normal jaw growth and development, and, if possible, protection of any affected erupted teeth.[6] Although many clinicians prefer to extract the anomalous teeth as soon as a diagnosis of RO is made, some prefer to retain them in order to facilitate normal jaw development and to lessen the psychological effects caused by premature loss of teeth as long as clinical signs and radiographic aspects indicate no infection.[3,5] Von Arx[2] suggests autotransplantation of suitable donor teeth to the area were dysplastic teeth were extracted as a good treatment alternative.

Summary

The etiology of RO remains obscure. In this case a possible association with pre-natal trauma is pointed. The literature is not unanimous as the

management of RO; nevertheless, the presence of dental abscess is an indication for teeth removal. Since general bone quality is not affected, a provisory prosthesis was fabricated as the patient waits for future implant rehabilitation. The executed treatment met the patient and her mother's expectations, improving the patient's masticatory function, esthetics, and quality of life.

The general practitioner must be prepared to deal with different situations in the dental office. The diagnosis of RO is mainly clinical and radiographic, and its treatment will depend on the affected teeth, the degree of severity of the dysplasia, the presence of infection, and the patient's age.

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