Regional odontodysplasia in early childhood: A clinical and histological study

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Abstract

Regional odontodysplasia (RO) is a rare disorder of dental development. The affected teeth are clinically hypoplastic and hypocalcified, presenting a ghost-like appearance radiographically. The aim of this work was to report a clinical case of a child with both primary and permanent dentition affected by RO. The conducted therapy was based on a conservative approach, which consisted of follow-up clinical evaluations of the anomalous teeth. However, the endodontic treatment of the primary incisors failed. Then, the chosen option for patient rehabilitation became extraction followed by removable of prosthesis confection. The extracted teeth were processed for histological analysis. In spite of the uncertain prognosis, but taking into account the psychological aspects of the patient, a conservative approach in an attempt to maintain those viable teeth in the oral cavity should be established.

Key words

Ghost teeth, odontodysplasia, permanent dentition, primary dentition

DOI: 10.4103/0970-4388.57099

Introduction

Regional odontodysplasia (RO) is a rare developmental, nonhereditary dental anomaly that involves ectoderm and mesoderm-derived tissues[1–3] with unknown etiology[3–5] and which can affect primary and/or permanent dentition.[1–8] Clinically, the affected teeth appear hypoplastic and hypocalcified.[3,5–8] Radiographically, the teeth present a wider pulp chamber and root canal, due to reduced enamel and dentin thickness and radiodensity.[3–5,8]

The available literature on RO is sparse; thus, it is important to discuss clinical cases of this anomaly to help professionals to better approach the diagnosis and the treatment. This study consisted of an RO case report in which an early diagnosis was possible, allowing a thorough discussion of the diagnosis as well as pathogenic and treatment characteristics of this dental anomaly.

Case Report

A 1.5-year-old male patient sought treatment at the Pediatric Dentistry Department of the School of Dentistry of Araraquara, UNESP. The main complaint was the absence of eruption of the upper anterior primary teeth on the right side of the mouth. The patient's mother had a normal pregnancy, and delivery, medical, and family histories were unremarkable.

The incisors of the upper left side, the lower incisors, and the first primary molars were present. Absence of the right central and lateral incisors was observed in the intra-oral examination. Radiographically, a delay in the development of these teeth was observed compared to its homologous [Figure 1]. The radiographic image determined that the coronal portion of these primary teeth was in the initial phase of mineralization. Regarding the permanent teeth, only the incisors and canines on the left side were undergoing the mineralization process.

The person responsible was informed that this developmental anomaly had affected the teeth and was advised to return every 2 months for the periodical control examinations, but the patient only returned after 1 year. At that time, the patient was 2.5-years old and had all primary teeth present in the oral cavity. The patient's mother indicated that the child was complaining of pain, specifically at the right central incisor. The crowns of the right central and lateral incisors were widely damaged with a fistula in the corresponding region of the central incisor apex. The right canine was healthy, hypoplastic, and partially erupted. The radiographic analysis revealed wide pulp chambers associated with the roots of narrow dentinal walls and opened apices [Figure 2].

Aiming for a conservative approach, the endodontic treatment of the upper right incisors was initially performed. However, considering the absence of regression of the infectious process, after topical and infiltrating anesthesia, the radicular remnants of the upper right incisors were removed. The chosen treatment for the canine was by the means of prophylaxis and topical application of professional fluoride during the monthly assessment visits.

After healing of the alveolar ridge, an aesthetic and functional removable acrylic space maintainer was installed. After 32 months of follow-up visits, continuity in the development of the RO permanent successors was noted [Figure 3]. The patient has been kept under observation and periodical control to monitor the development of dentition and follow-up of craniofacial growth.

The extracted teeth were processed for histological analyses, cut in 6-µm thick sections in the buccolingual direction with a rotatory microtome, and finally stained with Hematoxylin and Eosin (H and E). Morphological analyses were performed under light microscopy (DM2000, Leica, Reichert, Germany), and images were captured with a digital camera (DFC-300 FX, Leica Microsystems, Germany). Histological analysis evidenced the presence of an aprismatic enamel matrix [Figure 4]. Near the residues of necrotic pulp tissue in the radicular portion, the presence of approximately 50-µm thick disorganized, amorphous predentin with the absence of dentin tubules was observed. An amorphous dysplastic tissue with globular characteristics was characterized in the dentin, presenting disorganization and rare dentin tubules that were of reduced and irregular diameter [Figure 5]. Portions of radicular dentin covered by cells from Hertwig's epithelial root sheath with residues of dental follicle represented by fibrous connective tissue were also observed [Figure 6].

Discussion

Since RO is a nonhereditary and rare anomaly, there is no available scientific evidence concerning its prevalence in the population. RO may attack primary and/or permanent teeth and when it affects the primary dentition, the permanent successors are also committed.[6,8–10] In general, this dental anomaly involves groups of teeth,[4,5] but its occurrence has also been reported crossing the midline[3] or all of them.[5]

The main complaints of patients affected by RO are the failure of tooth eruption and the presence of gingival edema.[6,8–10] From a clinical diagnosis, teeth in the oral cavity are hypoplastic, presenting brown or yellowish discoloration, gingival swelling, gingivitis, periapical infection, and abscess formation.[1,4–7,9] Radiographically, the teeth have a ghost-like appearance with narrowed dentinal walls and wide pulp chamber due to the decreased thickness of enamel and dentin and defective mineralization.[5–9] In the present work, the responsible main complaint was the absence of eruption of a group of teeth. Radiographically, the "ghost teeth" aspect was noted as well as the lack of synchronicity in the development of primary germ teeth and of their successors when compared with healthy homologous teeth.

Histologically, all dental tissues were altered.[2,3,5,6,9] The enamel was characterized as hypoplastic and hypocalcified, with variable thickness and irregular surfaces. The enamel prisms were irregularly distributed and could include aprismatic regions with degenerated globular calcifications. The dentin layer was reduced and presented a decreased quantity of tubules that were also irregularly distributed. Other observations included extensive interglobular and amorphous dentin areas, predentin layer enlargement, and clefts that could establish communication between the pulp and oral cavity. It is due to the existence of these clefts between the oral cavity and the pulp that the RO teeth often present pulp necrosis. The dental follicle was composed of dense fibrous connective tissue and could present calcification areas.[2–6] In this work, the histological analysis of the RO teeth has shown similar characteristics to those reported in the literature.[2,3,5,6,9]

Although some authors have suggested hypotheses for

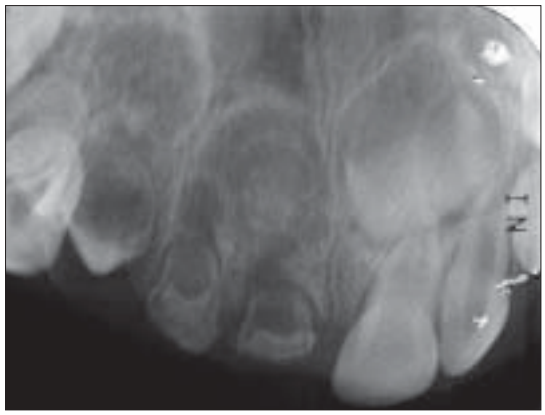

Figure 1: Radiographic image demonstrating developmental delay of the primary and permanent upper right incisors and canines when compared to homologous teeth

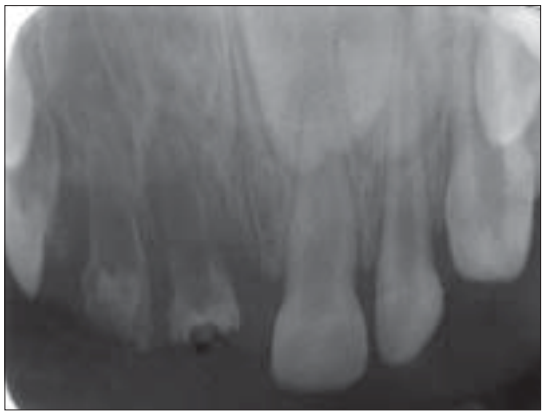

Figure 2: Radiographic aspect suggesting ghost-like characteristic of RO teeth and the presence of wide pulp chambers associated with roots containing narrow dentinal walls

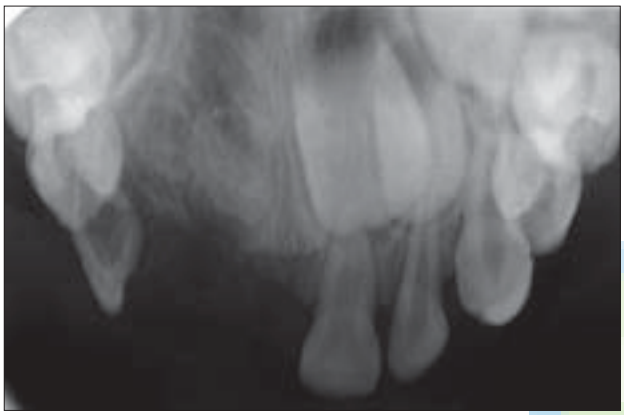

Figure 3: Modified occlusal radiography after 32 months' follow-up, evidencing the continuity of the permanent successors affected by RO

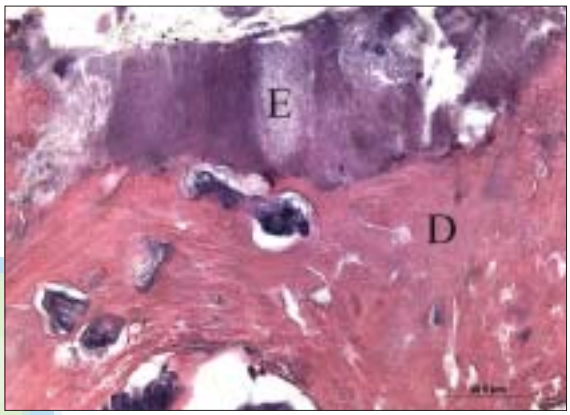

Figure 4: Ghost tooth with the presence of a slender enamel layer lacking mineralization (E). Irregular dysplasia dentin layer (D) characterized by the absence of dentin tubules and with cell inclusions and capillaries vessels. (HE) 400×

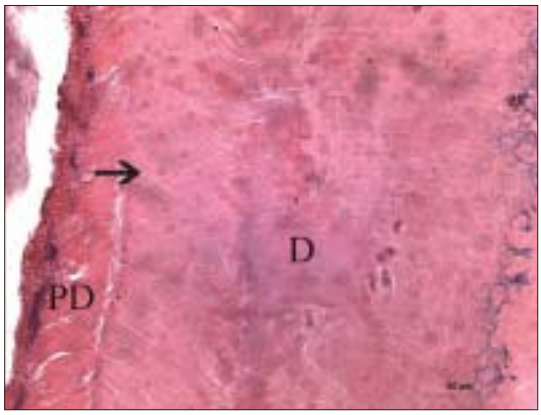

Figure 5: Portion of radicular dentin in which a thick band of amorphous and disorganized predentin can be visualized (PD). The dentin (D) shows rare dentin tubules with reduced and irregular diameter (arrow). Presence of mineralization globules all over dentin extension. (H&E, 400×)

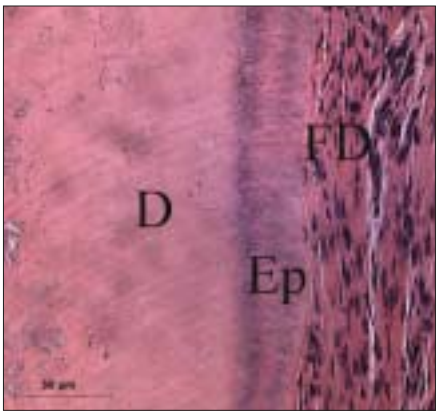

Figure 6: Portion of radicular dentin (D) covered by epithelial cells (Ep), residues of Hertwig sheath associated with connective fibrous tissue, dental follicle (FD). (H&E, 400×)

the development of RO, its etiology has not yet been determined.[4,5] It has been proposed that an imbalance of necessary proteins might lead to the structural disorganization seen in this anomaly,[9] such as the metalloproteinases (MMPs), which are enzymes that play a key role in dental development. In the present case, a possible cause for the development of this anomaly could not be identified.

Removal of the RO teeth has been proposed by several authors, even when this anomaly is diagnosed in an early development stage, since these teeth may result in the delay or failure of the process of eruption in addition to structural defects, and the presence of bacterial infection is frequent.[3–5,10] Less invasive therapeutic approaches have also been suggested due to the potential for psychological problems of the patient and the deleterious effects that the early loss of teeth could cause to the occlusion.[7,8] The treatment depends on the degree of dental involvement and is performed in accordance with the functional and aesthetic needs of each case. The affected teeth should be part of a regular follow-up program in order to observe development and stability in the oral cavity.[7,8]

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Source of Support: Nil, **Conflict of Interest:** Nil