Regional Odontodysplasia: Report of case.

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A case of regional odontodysplasia in an 8 year-old male whose chief complaint was the absence of eruption of permanent teeth is presented. The regional odontodysplasia is a rare development anomaly of the dental tissues that affects the primary and permanent dentitions involving several adjacent teeth without crossing the midline. The clinical, radiographic and histological aspects are described as well as the treatment accomplished through surgery and prosthetic rehabilitation.

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INTRODUCTION

The regional odontodysplasia is a rare developmental anomaly that affects all the dental tissues, of ectodermal (enamel) and mesodermal origin (dental pulp, dentin, cementum), of the primary and permanent dentitions. These alterations are detected radiographically.[1,2] This pathology tends to involve several adjacent teeth of the same quadrant, usually without crossing the midline.[1,3] When it affects two or three segments or multiquadrants then it is called generalized odontodysplasia.[1]

The regional odontodysplasia was first described by McCall and Wald[4-7] and the denomination odontodysplasia was proposed by Zegarelli *et al*. in 1963,[7] while the term regional was proposed later by Pindborg.[2]

Different terms have been used in the description of this pathology: odontogenesis imperfecta, odontogenic dysplasia, amelogenesis imperfecta non-hereditary segmentalis, localized hypoplasia (Turner teeth) or ghost teeth.[2,3,5]

The regional odontodysplasia is more prevalent in female gender and the maxillary teeth are twice more affected than the mandibular ones.[1,2,6,8] It generally affects the anterior region and does not present an ethnic tendency.[2,4]

When the odontodysplasia is observed in the primary dentition, teeth can be erupted, hypoplastic, hypocalcified, with change in color and form. Gingival tissue can be hyperemic and, usually presents a fistula.[2] In the permanent dentition, teeth usually are not erupted[9] or can be partially erupted with fibrous gingival tissue and swelling.[10] Roots are short with open apices, with large pulp chambers. The main radiographic characteristic is the ghost aspect due to the reduced thickness of the hard tissues.[11,12] Tissue areas relatively radiopaque inside the crown and dental follicle have been described.[6,11]

Due to their defective structure, the dysplastic teeth become more susceptible to caries and are extremely friable, and subjected to fracture.[10] Enamel fractures associated to thin dentin thickness cause the communication between the pulp and the oral cavity, resulting in pulpitis without caries presence. Because of the softened dentin, when these teeth are curetted, they may be mistaken with deep caries lesions.[1,2,6] Patients with regional odontodysplasia can present fever, pain, gingival inflammation, absence of teeth and facial asymmetry.[2]

The regional odontodysplasia is usually seen in adjacent teeth, without the interference of a healthy tooth. However in some cases, teeth occasionally not affected or minimally affected can happen in the permanent dentition.[13]

Microscopic findings include enamel and dentin structures severely hypoplastic and hypocalcified[7], coronal dentin with few tubules and irregularly distributed. The structure and calcification of the radicular dentin is less abnormal than the coronal dentin.[12,14] Predentin is enlarged, with presence of great areas of interglobular dentin formation, calcified bodies within the pulp.[6]

Scanning electron microscopy (SEM) observations displays thin enamel, susceptible at different points to fracture; Enamel can be normal in some areas with irregular enamel prisms presenting a laminated aspect, irregular crystalline aggregates with different sizes and shape.[14]

This paper reports a case of regional odontodysplasia, presenting the clinical, histopatological, radiographic aspects, and the patient's rehabilitation.

CASE REPORT

An 8 year old boy, was refered due to the absence of eruption of permanent teeth.[21,22,23]

Clinically, we observed root stumps of primary teeth 61 and 63 with gingival fibrosis and larger volume as observed in figure 1.

Teeth 64, 65 were radiographically normal. However, 65 presented a lingual caries lesion and tooth 26 was hypoplasic in one of the

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cusps. The periapical X-ray the teeth involved showed a fine line of the radiopaque structure, short roots, open apices, increased radiolucency living "ghost" aspect (figure 2). The panoramic x-ray showed the involvement of the left maxillary quadrant and the presence of an upper right lateral supernumerary (figure 3).

Medical history was not contributory, althought in the clinical exam, parents reported a fall history when he was two years-old.

The treatment suggested to the patient's mother was the extraction of primary root stumps, restoring 65 and 26, clinical and radiographic follow-up of the eruption of the dysplastics teeth. After 1 year, the patient became anxious due to the lack of the anterior teeth in the affected area. Gingival tissue was swollen and teeth were radiographically in the same position (figure 4). Extraction of the dysplastic permanent teeth was proposed. Surgery was accomplished with local anesthesia with a vestibular approach through the mucoperiosteal flap technique. Fibrous gingival tissue was observed in the area affected by the regional odontodysplasia. Extraction of the dysplastic permanent teeth, curettage, irrigation, flap replacement and suture was done (figure 5-7).

No complication in the postoperative phase was seen and 15 days later, a prosthetic appliance was made in order to regain his esthetic, functional and psychological needs (figure 8).

During surgery, three dental structures, a gingival fragment and a bone fragment in the area of central incisor were removed. These pieces were immersed in 10% formaldehyde for histopathological studies .

Biopsy of the gingival fragment showed the presence of islets of odontogenic epithelium in the conective tissue, as well as some fibrosis (figure 9).

One of the extracted incisors was prepared by the decalcification technique. In this technique the enamel is lost, but the pulp and the dentin can be observed. In the analysis of the lamina tinted with H&E, it was possible to observe the largest volume of the pulp in relation to the dentin, pulp nodules, the enlargement of the predentin layer, presence of interglobular dentin and small amount of dentinal tubules, these last ones defective and smaller in thickness (figure 10 -11).

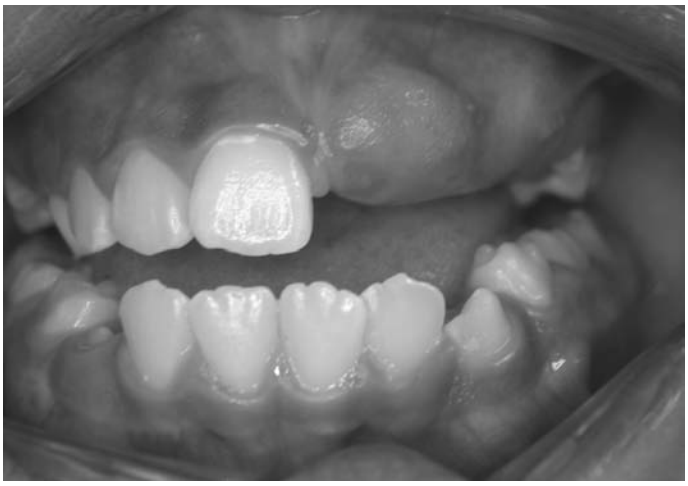

Figure 2. Frontal view of the patient.

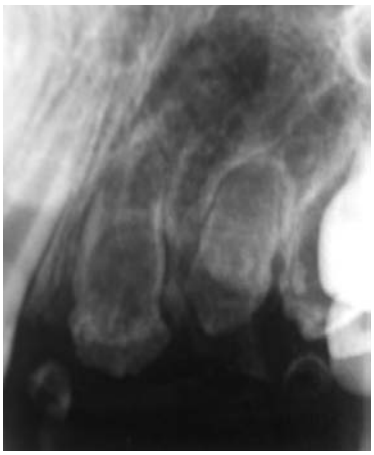

Figure 2. Radiographic aspect 61, 21, 22, 63, 23.

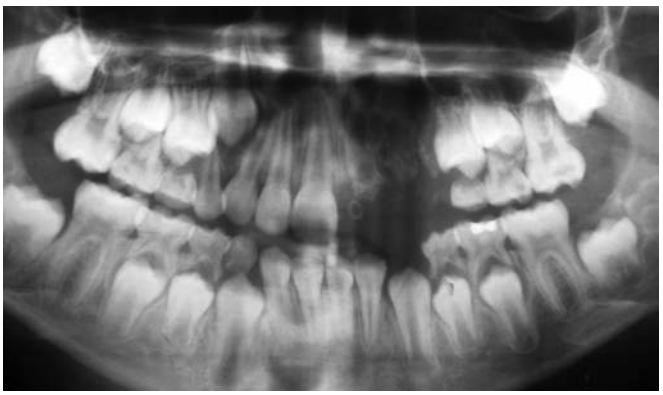

Figure 3. Panoramic x-ray.

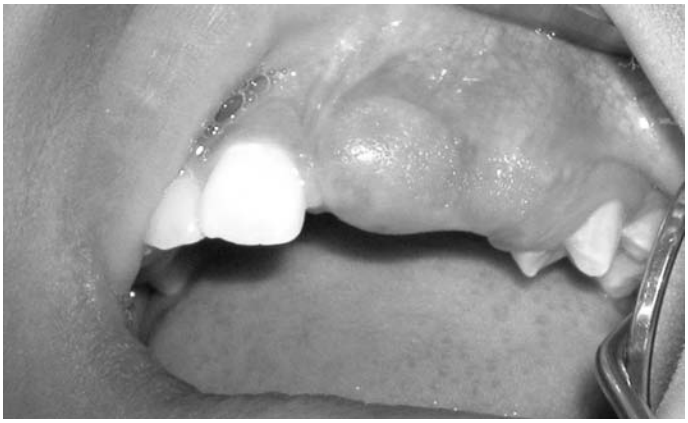

Figure 4. One year-old follow-up.

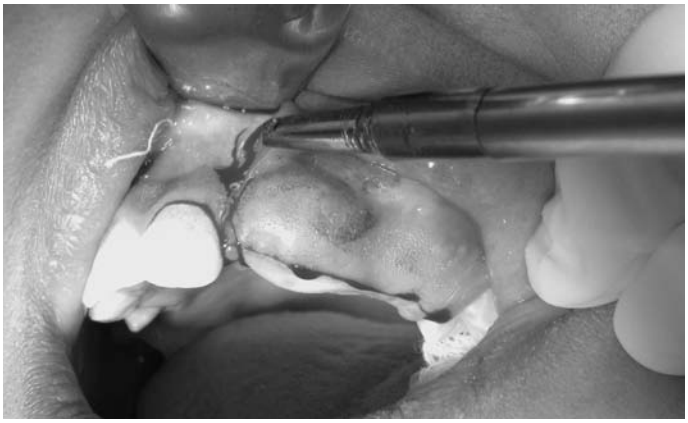

Figure 5. Vestibular approach.

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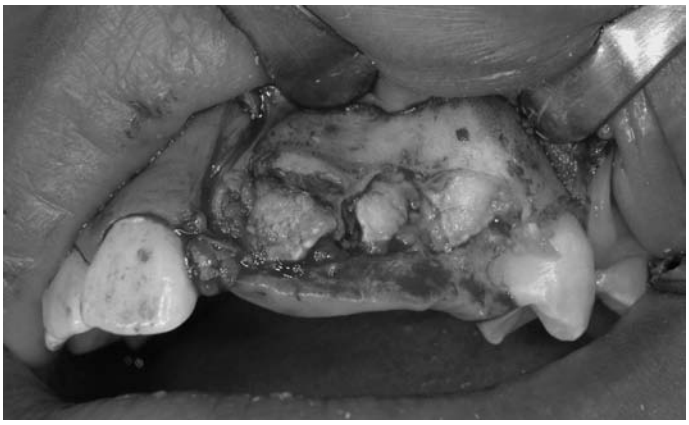

Figure 6. Mucoperiosteal flap and the three dysplastic teeth.

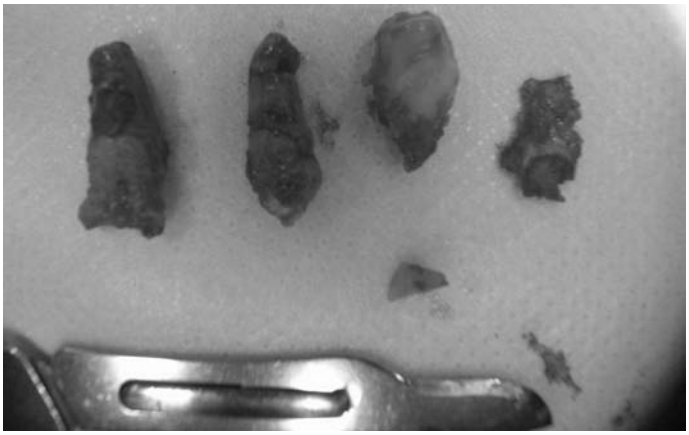

Figure 7. Three dental pieces, bone and gingival fragments.

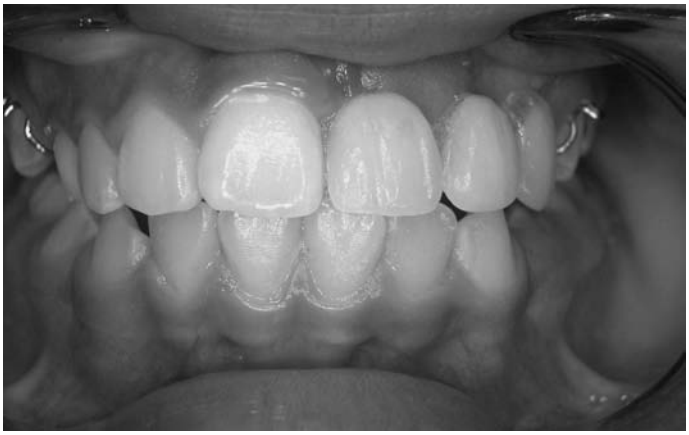

Figure 8. Rehabilitated patient

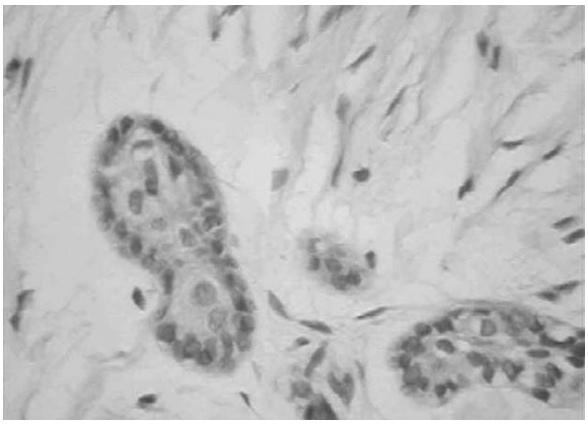

Figure 9. Islets of odontogenic epithelium in conjunctive tissue. Increase of 400X

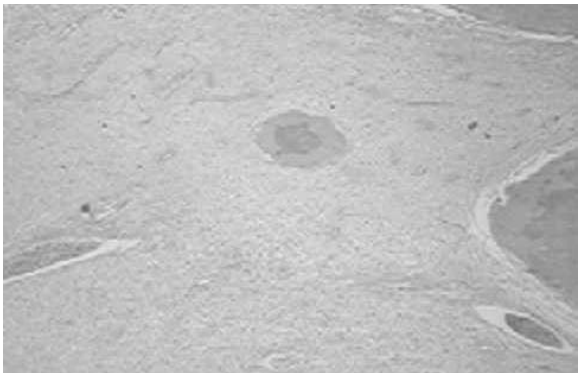

Figure 10. Pulp tissue presenting nodules.

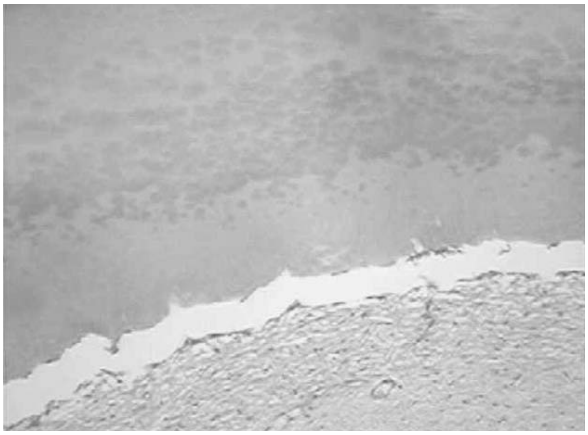

Figure 11. Enlargement of the predentin layer and interglobular dentin.

DISCUSSION:

The odontodysplasia etiology is uncertain; numerous factors have been suggested and considered as local trauma, irradiation, hypophosfatasia, hypocalcemia, hyperpyrexia.[4,7,8,11] The regional odontodysplasia has also been related to the activation of latent viruses in the odontogenic epithelium, to the presence of nevus, hemangiomas and hydrocefalia, but most of the cases present no relevant information.[2,4,11]

The odontodysplasia has been observed without any evident cause or familiar characteristic.[12]

The viral activation theory in the odontogenic epithelium has been accepted, but no viral particle was identified in the affected oral tissue.[4,16]

The vascular theory, in many cases suggests that a disturbance in the circulation happened during tooth development.[2,13,14] The blood overflow from vessels can be responsible for local variations of Mg and Na in the fluid around the crystals, through their connection to blood proteins, like albumin. These factors can affect ameloblastic function, apatite crystal formation and cause enamel hypoplasia.

Aguas de Galli and Carracino[16] studied the evolution of regional odontodysplasia for three years in a patient. They found endocrine and thyroid alterations, an angioma on the same side of the lesion (face skin, ocular sclerotic, tongue mucous membrane and uvula). In the present case it was not possible to determine an etiologic fac-

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tor. So, the patient was directed to the endocrinology department for further testing.

The clinical observation, the X-rays and the histological findings confirmed the diagnosis of regional odontodysplasia. Differential diagnosis should be made between regional odontodysplasia, amelogenesis and dentinogenesis imperfecta, type I and II, dentin dysplasias, Turner teeth and odontogenic calcifying fibroma.[2,6,10]

The treatment of the odontodysplasia is discussed. Surgery and extractions have been suggested.[2,3,7-10,13] Most of the time in the permanent dentition affected teeth are extracted and a prosthetic appliances made.[16] Pulpotomy or apexification has also been suggested depending on pulp vitality to stimulate root formation.[15] In the primary dentition, pulpotomy and restorations with steel crowns can be recommended as form of treatment for dysplastics teeth.[13]

The goal is to recover the masticatory and esthetic function, reducing the psychological effects caused by the absence of teeth, maintaining the vertical dimension and preventing facial asymmetry. Though, due to the growth of the alveolar bone and eruption of the permanent teeth the patient should return every 4-6 months for periodical control [13] and prosthesis should be changed according to the growth and development. The patient was also told about the possibility of implants in the affected area in the future.

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