

Regional Odontodysplasia: An Esoteric Rarity- A Case Report

ABSTRACT

Abstract- regional odontodysplasia is a rare developmental disorder affecting both mesoderm and ectoderm dental components and affecting a localized dentition area. The estimated prevalence of this disease is less than 1 in 1000,000 in the general population. It affects both the primary and permanent dentition in either the maxilla, mandible, or both arches. The etiology of ROD is still unknown. This case presented classical clinical and radiographic features of regional odontodysplasia in a 9-year-old boy with an atypical involvement of both the jaws, crossing the midline. An intricate multidisciplinary team is required to fully rehabilitate a patient affected by ROD. This case was managed conservatively with temporary prosthetic restoration to restore the function and esthetics of the patient.

Keywords: ghost teeth, Odontodysplasia, primary dentition, permanent dentition

1. INTRODUCTION

Introduction- Regional Odontodysplasia (ROD) is a rare developmental anomaly in tooth formation generally observed in young patients. This disease is characterized by the malformation and hypo calcification of the enamel and dentin, both asymmetrically and locally.^[1] ROD affects both primary and permanent dentitions. It is usually limited to one arch and crosses the midline rarely but can be seen more commonly in the anterior region.^[2] The maxilla is affected twice as often as the mandible, with the left quadrant of the maxilla being most commonly affected. Among the teeth, the lateral incisors and central incisors are more likely to be affected than the posterior teeth.^[3] This anomaly has no reported association with any specific racial group; however, females are slightly more affected than males at the ratio of 1.4:1.^[4] The etiology remains unknown of many possible causes that have been proposed in the literature. This includes local circulatory disorders, viral infections, pharmacotherapy during pregnancy, facial-asymmetry, local trauma, metabolic disturbances, somatic and neural mutations, and syndromal involvement. They also suggested that a combination of factors might be involved.^[5]

This paper reports a rare case of regional odontodysplasia in which both maxillary and mandibular jaws were involved including primary and permanent dentition and crossing the midline in both the arches.

2. CASE DISCUSSION

A 9-year-old healthy boy was reported to the department of pediatric and preventive dentistry Mathura, Uttar Pradesh, with a chief complaint of missing teeth with associated pain and swelling in the lower left back teeth region. He had an unremarkable prenatal, birth, medical, and family history. Extraoral examination revealed no pathological abnormalities. The boy was at the mixed dentition stage. Intraoral examination presented a complete set of deciduous teeth except in the upper front and lower front and back teeth region of the mouth. All permanent 1st molars were present except 36. Left primary 2nd mandibular molar was grossly decayed and was present in the soft friable spicules, the crown structure was destroyed because of its hypoplastic nature. (Figures 1,2) According to his parents, his teeth were yellowish-brown in color at the time of the eruption and gradually destroyed after the eruption.



Fig. 1,2- preoperative Maxillary and mandibular occlusal view showing missing teeth

On panoramic examination, a reduced radiodensity was found in the affected quadrant of all primary and permanent teeth. In the affected permanent teeth, the line separating enamel and dentin was not cleared. The involved teeth had enlarged pulp chambers and short, incompletely formed roots with "ghost-like" appearances (Figure 3).



Fig-3 Panoramic view showing reduced radio-density of enamel and dentin in affected teeth of both maxillary upper front and lower left quadrant involving the midline and affected teeth showing ghost-like appearance.

Based on clinical and radiographic findings, a diagnosis of regional odontodysplasia was made. Radicular remnant of the mandibular left 2nd primary molar was extracted under local anesthesia. After healing, rehabilitation with a functional aesthetic space-maintainer was done in the maxillary arch with banding on both, the right and left maxillary 1st permanent molar (Figure-4). Secondly, an alginate impression was taken for the mandibular arch, followed by a special tray fabrication, border-moulding was done, followed by a pickup impression, then a distal extended acrylic partial denture was made with heat cure acrylic. (Figure-5).



Figure 4- showing Prosthetic rehabilitation with Functional aesthetic space maintainer in the maxillary arch



Figure 5 – showing Prosthetic rehabilitation with removal partial denture in the mandibular arch

Oral hygiene instructions were given to the patient and have been followed up periodically to assess the growth and development of both arches. Written consent was taken from parents for the child's photographs and other clinical information to be reported in the journal.

3. DISCUSSION

ROD is a rare dental anomaly that is normally confined to one tooth arch ^[6,7] In this case report, the patient presented with the majority of common clinical and radiographic signs associated with the diagnosis of this condition., but this case was notable due to the uncommon involvement of both the maxillary front and mandibular left quadrants. In 2010, Quinderé ^[6]

described a case of regional odontodysplasia involving three quadrants of the jaws. To date, only six cases of generalized odontodysplasia have been described in the literature. In this case, ROD affected both primary and permanent dentition and crosses the midline in both the arches, which is uncommon. In spite of the fact this patient reported in this case report is a boy, ROD is more common in girls.^[7]

In this case, the affected teeth demonstrated most of the characteristics of ghost teeth described in the literature, both clinically and radiographically. The clinically affected tooth is usually smaller than normal, has a brown or yellowish discoloration, and has an extensive pitting and grooved surface. The enamel is hypoplastic or hypocalcified, and therefore soft on probing, with a delayed eruption or no eruption at all. Teeth are more susceptible to decay due to defective mineralization and extremely fragile fractures at even the slightest trauma.^[8]

Radiographically, enamel and dentin of affected teeth lack contrast, both of which are less radio-opaque than their unaffected counterparts. Also, the enamel and dentin layers are very thin, giving the teeth a “ghostly” appearance.^[2] An enlarged pulp chamber is noticeable, with an open apice and enlarged follicles due to thin walls with little dentine formation and the amelodentinal limit is usually absent. Histologically, the enamel's thickness varies, resulting in an irregular surface, it may lack prismatic structure, hypoplastic in nature, and contains degenerated globular calcification. Interglobular dentin and globular masses interrupting the dentinal tubules are commonly observed. Differential diagnosis includes hereditary conditions like dentinal dysplasia, dentinogenesis imperfecta, shell teeth, or amelogenesis imperfecta.^[9]

The best treatment option for a child with ROD depends on several factors, including the age of the patient, any relevant medical history, and previous dental experience., the child's and parents' attitude toward dental treatment, and the number of affected teeth. There is considerable debate regarding whether to extract or save affected teeth (with or without abscesses). In this case, the residual root of the left primary 2nd mandibular molar was indicated for extraction because it could not be restored. In this critical period, the partial denture was fabricated to provide function, phonation, and esthetics. Currently, it is not possible to place posterior teeth in this denture because there is a lack of space. Early primary molars loss during the growth phase results in a reduction of vertical occlusal dimension. Cahuana, et al.¹⁰ (2005) described auto-transplantation as an alternative treatment option for patients with ROD. The availability of suitable donor teeth, however, limits its use.^[3] Despite the growing use of Osseo-integrated implants in patients with missing teeth, these implants are not recommended for growing individuals and are therefore placed when the patient has reached pubertal maturity.^[2]

Although the child and his parents are happy with the treatment, the rehabilitation can't be considered ideal since the patient's oral esthetic and function aren't completely restored. To monitor the patient's development of the mandible and maxilla, a control visit is being conducted at present. During the appointments, the appliance has been adjusted or replaced. Permanent teeth may not erupt or have a modified eruption pattern. Primary and Permanent teeth that are not infected will not be extracted before eruption since these teeth help maintain the alveolar bone. The presence of teeth is important for skeletal growth. The prognosis of the affected permanent teeth is, however, poor. In the future, it may be necessary to evaluate the extraction of affected permanent teeth and the rehabilitation with dental implants.

4. CONCLUSION

In this case, we describe a 9-year-old boy with unique clinical, radiographic, and conservative management features of ROD. As it involves both dentitions, this case is valuable to pediatric dentists in terms of early diagnosis and treatment. In order to fully rehabilitate a patient affected by ROD, a complex multidisciplinary team is needed.

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DEFINITIONS, ACRONYMS, ABBREVIATIONS

ROD: REGIONAL ODONTODYSPLASIA