

## Case report

# An Ambiguous case report: Regional odontodysplasia affecting both maxillary and mandibular arch

### ABSTRACT

Abstract- Regional Odontodysplasia is an unusual 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 maxillary, mandibular, 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 aged boy with an atypical involvement of both the jaws, crossing the midline. In order to fully rehabilitate a patient affected by ROD, a multidisciplinary approach is required. 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. The condition was first reported by McCall and Wald in 1947, but Zegarelli et al. introduced the term "odontodysplasia" in 1963, and the prefix "regional" was added by Pingborg in 1970.<sup>[1]</sup> There is malformation and hypo calcification of the enamel and dentin in an asymmetrical and localized fashion in this disease.<sup>[2]</sup> The condition affects both primary and permanent teeth. This condition is generally limited to one arch, crossing the midline only rarely, but more frequently located in the anterior region of the jaw.<sup>[3]</sup> In contrast to the mandible, the maxilla is twice as likely to be affected (ratio 2.5: 1). The left quadrant of the maxilla is most commonly affected. Among the teeth, the lateral incisors and central incisors are more likely to be affected than the posterior teeth.<sup>[4]</sup> There is no known association between this anomaly and a specific racial group; however, females are little more affected than males (1.41:1).<sup>[5]</sup> According to Lustmann et al., only 11 of the 51 cases reported involved teeth on both sides of the midline.<sup>[1]</sup> The etiology of this disease is unknown despite the number of possible causes reported in the literature. These include circulatory disorders, viral infections, pharmacological treatment during pregnancy, facial asymmetry, trauma, metabolic disorders, somatic and nural mutations, and syndromal disorders. They also suggested that a combination of factors might be involved.<sup>[6]</sup>

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 PRESENTATION

A 9-year-old healthy **male** child 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 1<sup>st</sup> molars were present except 36. Left primary 2<sup>nd</sup> 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.

The diagnosis of regional odontodysplasia has been made based on clinical and radiographic findings. Radicular remnant of the mandibular left 2<sup>nd</sup> primary molar was extracted under local anesthesia. After healing, rehabilitation with a functional aesthetic space-maintainer was accomplished in the maxillary arch with banding (Denext band material) on both, the right and left maxillary 1<sup>st</sup> permanent molar (Figure-4). A Diagnostic alginate impression was made of the mandible arch and was poured with dental stone (NeelKanth -DSP). A self-cure acrylic resin (DPI RR cold cure) custom tray was fabricated on this primary cast which only extended on the left mandibular arch. Border moulding was performed by a low fusing impression compound. Right labial and buccal vestibule movements were recorded also with premylohyoid, mylohyoid, and retro-mylohyoid region by tongue movement on the same side along with the retromolar pad. A wash impression was then made with zinc oxide eugenol impression paste. The impression made was then poured with die stone (NeelKanth - DSP), and then a self-cure acrylic resin denture base was fabricated over the master cast. A wax occlusal rim was formed on the denture base and bite registration was done and the casts were mounted on a mean value articulator. After arranging teeth, a trial procedure was done and occlusal relations were examined. A complete wax-up was then followed by heat cure acrylisation (DPI heat cure) and the partial denture was fabricated, finished, and polished. (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.

### 3. DISCUSSION

ROD is an uncommon dental disease that is usually confined to a single tooth arch<sup>[7,8]</sup> In the present case report, the child displayed the majority of common clinical and radiographic signs associated with the determination of regional odontodysplasia, A unique aspect of this case was the involvement of both the front left quadrant of the maxilla and the left quadrant of the mandible. As reported by Quinderé<sup>[7]</sup> in 2010, three quadrants of the jaws were affected by regional odontodysplasia. Only six cases of generalized odontodysplasia have been documented so far 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.<sup>[8]</sup>

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 prone to decay because of defective mineralization and extremely fragile fractures at even the slightest trauma.<sup>[9]</sup>

Radiographically, the enamel and dentin of affected teeth lack contrast, both of which are less radio-opaque than their non-affected counterparts. Also, the enamel and dentin layers are very fine, giving the teeth a "ghost appearance".<sup>[3]</sup> An enlarged pulp chamber is noticeable, with open apices 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 of ROD includes hereditary conditions like dentinal dysplasia, dentinogenesis imperfecta, shell teeth, or amelogenesis imperfecta.<sup>[10]</sup>

For a child with ROD, the best treatment option will depend on several factors, including the age of the patient, any relevant medical history, and previous dental experience. The attitude of the child and his parents toward dental treatment, as well as the number of affected teeth. It is debatable whether to extract or save affected teeth (with or without abscesses). It was indicated in this case that the left primary 2nd mandibular molar root should be extracted since it was unable to be restored. In this critical time, the partial denture was constructed to offer function, phonation, and aesthetics. Currently, this denture is not able to accommodate posterior teeth due to its limited space. The loss of early primary molars can result in a reduction of the vertical occlusal dimension during the growth phase.

Cahuana, et al.<sup>[6]</sup> (2005) described auto-transplantation as an alternative treatment option for patients with ROD. The availability of suitable donor teeth, however, limits its use.<sup>[3]</sup> Although the utilization of osseointegrated implants in patients with missing teeth is increasing, these implants are not recommended for growing patients and are therefore placed when the patient has reached pubertal maturity.<sup>[3]</sup>

Despite the fact that the child and his parents were pleased with the treatment, the patient's oral esthetic and function were not fully restored. To monitor the patient's development of the maxilla and mandible, a control visit has been scheduled and the appliance will be altered or adjusted during these appointments.

As per the present scenario the permanent teeth may or may not emerge or could have an altered eruption pattern. Non-infected primary and permanent teeth will not be extracted before they emerge, because these teeth help in maintaining the alveolar bone and are crucial for skeletal development. However, the prognosis for the affected permanent teeth is poor. In the future, we may have to determine whether to remove affected permanent teeth and rehabilitate them 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. A pediatric dentist would find this case valuable for early diagnosis and treatment since it involves both dentitions. In order to fully rehabilitate a patient affected by ROD, a complex multidisciplinary team is needed.

#### CONSENT

The parent's written consent was obtained before photographs and other clinical information were included in the journal.

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**ABBREVIATIONS**

ROD: REGIONAL ODONTODYSPLASIA

UNDER PEER REVIEW