# Regional odontodysplasia: Report of an unusual case involving mandibular arch

N. S. Venkatesh Babu, R. Jha Smriti, D. Bang Pratima

## Abstract

Regional odontodysplasia (RO) is a rare developmental anomaly involving both mesodermal and ectodermal components in primary or permanent dentition. It affects the maxilla and the mandible or both; however, maxilla is more commonly involved. This article reports the case of 33-month-old boy who came with the chief complaint of delayed eruption of mandibular teeth. Findings of clinical and radiographic examination were consistent with those of RO. Maxillary dentition was unaffected. Clinical and radiographic features and treatment options are discussed.

**Keywords:** Mandibular arch, primary teeth, regional odontodysplasia

## Introduction

Regional odontodysplasia (RO) is a rare developmental dental anomaly that involves ectoderm and mesoderm derived tissues.[1] It can affect either primary or permanent dentition.[2] This condition was first described by Hitchin in 1934. The prevalence of this condition is still not clear since the studies reported till date have mainly been based on case reports.

Although several factors such as local trauma, infection, ischemia, neural damage, and somatic mutations of neural crest cell migration have been advocated, the underlying pathophysiology of the condition remains unclear.[3,4] It is reported that RO is probably a nonhereditary condition.[4] It affects females more than males (ratio female: male = 1.7:1).[3]

Regional odontodysplasia is usually unilateral, with no tendency to cross the midline.[2,5-7] However, cases have been reported with bilateral or multiquadrant involvement.[3] The maxillary teeth are more commonly affected, and only four cases of mandibular involvement have been reported so far.[5,8,9]

The teeth with RO often display a brownish or yellowish discoloration and most frequent clinical symptoms accompanied by this anomaly are failure of eruption and gingival enlargement. Radiologically, the affected teeth illustrate hypoplastic crowns and lack of contrast between enamel and dentin is usually apparent. Enamel and the dentin are very thin, displaying a "ghost-like appearance."[5] The other pathognomic radiological characteristics are enlarged pulp chambers, short roots, and open apices.[5]

## Case Report

A 33-month-old boy reported to the Department of Pediatric and Preventive Dentistry of our college with the complaint of delayed eruption of teeth in the lower jaw. The patient was a healthy boy with no relevant medical history. The prenatal and natal history of the patient was unremarkable. There was no history of abnormal tooth anomalies on both maternal and paternal part of the family. No congenital or acquired diseases were reported.

Extra-oral examination revealed normal symmetrical face and normal skin, hair, and nails. The intra-oral examination revealed an adequate maxillary arch with fully erupted primary teeth. All the maxillary teeth appeared normal except for some carious involvements in molars [Figure 1]. Although all the primary teeth were visible in the mandibular arch, they were not fully erupted and seemed to be embedded in abnormal, slightly hyperplastic alveolar mucosa. Furthermore, these teeth revealed abnormal crown morphology with yellowish discoloration and hypoplastic enamel [Figure 2]. Generalized gingival enlargement in the mandibular arch was evident [Figure 2]. However, none of the teeth showed any abnormal mobility.

The orthopantomogram (OPG) [Figure 3] which was taken after detailed clinical examination, showed a full complement

*Department of Pedaitric and Preventive Dentistry, V. S. Dental College and Hospital, Bengaluru, Karnataka, India*

**Correspondence:** Dr. N. S. Venkatesh Babu,
V. S. Dental College and Hospital, K. R. Road, V. V. Puram,
Bengaluru, Karnataka, India.
E-mail: drnsvbabu@gmail.com

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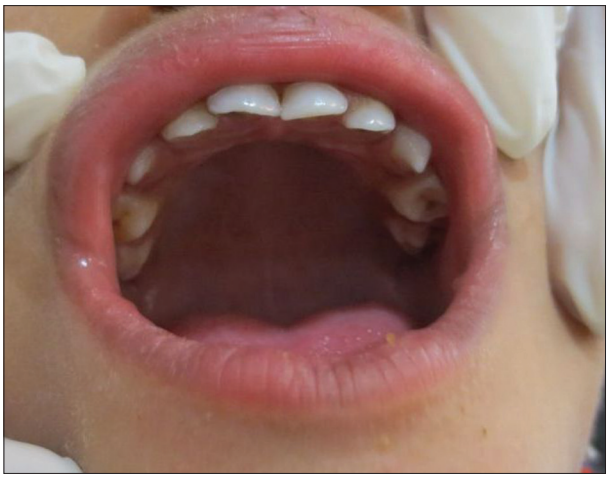

**Figure 1:** Normal maxillary arch with a full complement of teeth

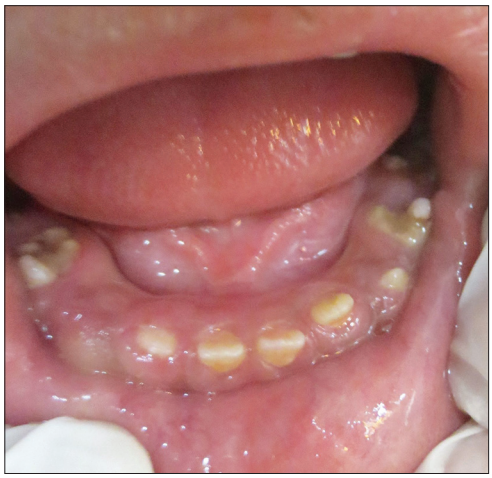

**Figure 2:** Hypoplastic crowns and gingival hyperplasia in entire mandibular arch

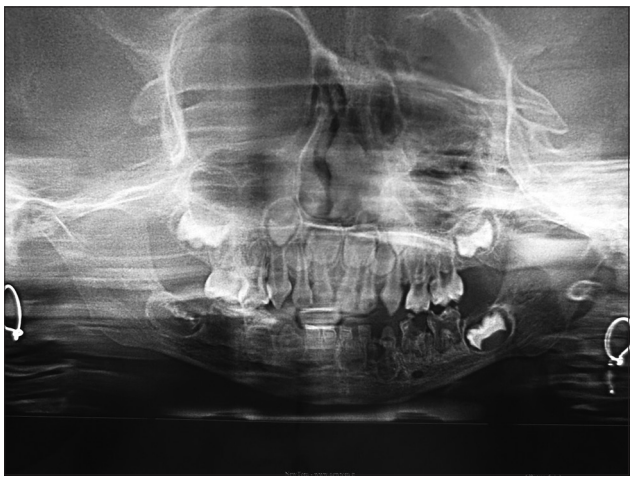

**Figure 3:** Orthopantomogram showing "ghost teeth" in entire mandibular arch

of maxillary deciduous teeth and follicles of permanent teeth except for tooth germs of premolars, which was normal for his age. However, in the mandibular arch, all the primary teeth showed a striking "ghost-like" appearance [Figure 3]. All the teeth had abnormal crown structure with very thin enamel and dentin layers. The demarcation line between enamel and dentin in these teeth was not clear, and enamel was hypoplastic. All the teeth exhibited enlarged pulp chambers with short incompletely formed roots. None of the permanent tooth germs were seen except for the follicles of first molars.

The laboratory investigations showed that the serum calcium, phosphorus, sodium, and potassium levels were in normal range. Since the child was only 33 months old, and none of the teeth showed any abnormal mobility, extraction was not carried out. Therefore, histopathologic examination of the tooth was not done. However, gingival biopsy examination revealed the odontogenic tissue in the epithelium and intramesenchymal calcifications.

On the basis of clinical and radiographic findings, a diagnosis of RO was proposed. The treatment plan for the child was mainly conservative. Maintenance of proper oral hygiene and regular follow-up examinations for monitoring the developing dentition was advised. Considering the age of the child, conservative treatment was preferred over extractions because it preserves oral functionality and esthetics until the end of growth period after which a more definite treatment could be planned. Restorations with glass ionomer cement for cariously involved maxillary teeth were done.

## Discussion

Regional odontodysplasia is a rare tooth anomaly that affects maxillary teeth more often with very less tendency to cross the midline.[7,8] However, cases have been reported with bilateral or multiquadrant involvement.[7,8] In the present case, clinical and radiographic findings were consistent with that of RO, but this case was notable due to involvement of the entire mandibular arch. Till date, only four cases of mandibular involvement have been reported.[5] All these cases had, however, reported a segmental involvement of mandible.

The etiology of RO is not well-known.[3,5] The cause of the present case also remains unknown because the patient's past medical history and family history was noncontributory, and no congenital or acquired diseases were reported.

Regional odontodysplasia has shown to affect both primary and permanent dentition.[7] In the present case, the primary dentition was affected. Delayed development of permanent tooth germs in the mandibular arch was also evident which was in agreement with some cases reported previously.[10]

Typical clinical features in this case included gingival hyperplasia and teeth embedded in the gingival overgrowth;

both of these features were consistent with the majority of cases reported previously.[7]

A differential diagnosis of RO includes hereditary conditions, such as dentin dysplasia, dentinogenesis imperfecta, and amelogenesis imperfecta as these conditions may present similar features of enamel hypoplasia, abnormal pulp morphology, and calcification. Hereditary developmental anomalies of enamel and dentin, however, usually affect the entire dentition rather than segments.[8] Dentinogenesis imperfecta type III bears close resemblance to RO but can be excluded if opalescent dentine, bell-shaped crowns, and family history are absent. Failure of eruption or delayed eruption and gingival enlargement occur only in RO.[11] Both of these features were seen in the present case.

Treatment of RO is controversial and requires a multidisciplinary approach. Consultations between pediatric, orthodontic, prosthodontic, and surgical specialties are often necessary. Factors such as the patient's age, extent of the lesion, and eruption of the teeth, medical history, and esthetics need to be carefully considered. For the present case, conservative approach was considered. This was because the child reported at a very early age. Furthermore, he did not present with any abnormal mobility or difficulty in chewing. In addition, since the OPG showed absence of developing permanent tooth germs, it was thought appropriate to preserve the primary teeth. Follow-up examinations were planned to observe the development of mandibular permanent tooth germs. The carious teeth in the maxillary arch were restored with glass ionomer cement.

## Conclusion

The case presents unique clinical and radiographic features of RO involving the entire mandibular arch in a child aged 33 months. A dental professional must be prepared to manage cases of dental anomalies, providing an opportunity for early diagnosis, adequate monitoring and treatment plans that minimize the after-effects on the patient's development. In cases of RO, treatment depends on the age of the patient, extent of the involvement of the teeth, and the individual functional and esthetic needs.

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