

# Regional Odontodysplasia: A Pediatric Case Report

Arifa Bakerywala, DMD, Adrienne Nguyen, DDS, Azita Khanbodaghi, DMD, Cheen Loo, PhD, MPH, DMD ( Tufts University School of Dental Medicine, Boston MA)

## Introduction

- Regional odontodysplasia (ROD) is an uncommon, severe, non-hereditary developmental dental anomaly involving epithelial and mesenchymal derived dental tissues with no known etiology
- ROD affects both dentitions and is commonly seen unilaterally, without crossing the midline
- Clinically, the affected teeth appear hypoplastic, with a yellow-brown discoloration, soft upon probing and have a higher susceptibility to caries
- Symptoms include abscesses, with or without the presence of caries, fever, gingivitis, and delayed or failure of tooth eruption
- Radiographically, the teeth show enlarged pulp chambers, open apices and reduced radiopacity of the enamel and dentin

## Case Report

A 2 year and 9 month-old healthy female presented to the clinic with a referral for treatment for multiple carious lesions. The mother and patient report no history of dental pain. The medical history was non-contributory and the mother reported a normal, full-term pregnancy with no complications. The patient had a 1 week-long hospitalization, where she was intubated due to pneumonia and pneumothorax of the right lung at 2 months old, but has been healthy since. The patient is not currently taking any medications and has no known drug allergies. Clinical examination revealed no extraoral pathology or asymmetry. All primary teeth were fully erupted with normal morphology except the lower left first and primary molars, which were discolored and partially erupted. The gingival in the lower left quadrant appeared inflamed and swollen. Radiographic examination revealed lower left mandibular teeth with thin radiopaque contours and no clear demarcation between the enamel and dentin. The affected primary canine and molars showed wide pulp chambers with the classic ‘ghost-like’ appearance. In the affected quadrant, the coronal aspect of the unerupted permanent teeth were surrounded by larger radiolucent areas representative of the enlarged dental follicle as well as showing wide pulp chamber which suggest that these teeth could be potentially affected as well. A diagnosis of ROD limited to mandibular left region was confirmed by a consultant from the Department of Diagnostic Sciences at Tufts University. Due to limited cooperation and young age, the patient was referred to the operating room for treatment under general anesthesia.



Figure 1: (left) Full mouth radiographs taken in the operating room while the patient was under general anesthesia during full mouth dental rehabilitation. Caries is noted on upper anterior teeth #D, #E, #F, #G. All teeth in the lower left quadrant (both primary and permanent) are affected by ROD.



Figure 2: (below) Clinical photographs taken in the operating room under general anesthesia prior to full mouth dental rehabilitation.

## Management

The patient was seen in the operating room for full mouth dental rehabilitation under general anesthesia. Radiographs and case photographs were obtained with signed consent from the mother. The anterior teeth were extracted and resin restorations were placed on the upper right lateral incisor and first and second lower right primary molars. The maxillary central and left lateral incisors were extracted. The entire lower left quadrant was affected by ROD in both primary and permanent dentition. The affected lower left first and second primary molars were extracted due to non-restorability from severe breakdown from ROD. The lower left incisors and canine, were less affected by ROD and clinically intact, and therefore left untreated to prevent a midline shift to the left side. The unerupted ROD affected lower left permanent first molar was left untreated. The patient's mother was informed that the patient's permanent dentition in the lower left quadrant are all affected by ROD, verified radiographically and will likely need extensive treatment and management in the future.

## Oral Pathology Report

The findings of biopsy of teeth #K and #L reported fragments of variably well-differentiated tooth structure. Fibrovascular tissue with acute and chronic inflammation was present. Microscopically, decalcified sections showed fragments of tooth structure in the form of mature tubular dentin transitioning to amorphous masses of mineralized tissue. Regions of mature tubular dentin surfaced by lightly basophilic linear calcifications consistent with tinned enamel matrix was also noted. The histological findings support a diagnosis of regional odontodysplasia.

## Conclusion

This case demonstrates the classical characteristics of ROD based on clinical and radiographic finding with support of histological review. Early diagnosis of ROD is important given both primary and permanent teeth are affected and likely will require a multidisciplinary approach. Since there is no consensus on the best treatment option, each ROD case should be individually assessed for proper management.

*Acknowledgment:*  
Funding to support these educational activities for our residents was made possible by the Golub Family Advancement in Education Fund in Pediatric Dentistry.

References:

- Hitchin AD. Unerupted deciduous teeth in a youth aged 151/2. Br Dent J 1934; 56:631-3.
- Hamdan MA, Sawair FA, Rajab LD, et al. Regional odontodysplasia: A review of the literature and report of a case. Int J Paediatr Dent 2004; 14:363-70
- Tervonen SA, Stratmann U, Mokrys K, et al. Regional odontodysplasia: A review of the literature and report of four cases. Clin Oral Invest 2004; 8:45-51.
- Al-Tuwirgi A, Lambie D, Seow WK. Regional odontodysplasia: literature review and report of an unusual case located in the mandible." Pediatric Dentistry, vol. 36, no. 1, pp. 62-67, 2014.
- Rushton MA. Odontodysplasia: ghost teeth. Br Dent J 1965; 119: 109-13
- Zegarelli EV, Kuischer AH, Applebaum E, Archard HO: Odontodysplasia. Oral Surg 1963; 16: 187-93
- Pindborg JJ. Pathology of the dental hard tissues. Copenhagen Munksgaard, 1970: 120-3
- Jahanimoghadam F, Pishbin L, Rad M. Clinic, Radiographic, and Histologic Evaluation of Regional Odontodysplasia: a Case Report with 5-year Follow-up. J Dent Shiraz Univ Med Sci., 2016 June, 17(2): 159-63
- Crawford PJ, Aldred MJ. Regional odontodysplasia: a bibliography. Journal of Oral Pathology and Medicine. 1989; 18: 251-263.