10.5005/jp-journals-10037-1085

CASE REPORT

Bilateral Regional Odontodysplasia: A Rare Case Report

[1]Sumit Majumdar, [2]Divya Uppala, [3]K Padma Priyanka, [4]S Praveen Kumar

ABSTRACT

Context: Regional odontodysplasia (RO) is an uncommon developmental anomaly affecting a localized area of dentition, with distinctive clinical and radiographic findings. This article reviews a case of a 10-year-old female who reported with RO in partially erupted mandibular anterior teeth.

Aims: To highlight the likelihood of occurrence of bilateral mandibular RO as a rare developmental anomaly. It commonly affects a localized area of dentition.

Settings and designs: The representative tissue received was 10% formalin fixed hard tissue measuring about 1×0.5 cm in diameter.

Materials and methods: Ground section was done for surgically removed teeth and observed under optical microscope.

Conclusion: In the ground section, the enamel and dentin were of variable thickness producing an irregular surface with central enlarged pulp cavity. Interglobular dentin and globular masses interrupting the dentinal tubules were also seen. All the features are suggestive of regional odontogenic dysplasia.

Keywords: Delayed eruption, Ghost teeth, Regional odontodysplasia.

How to cite this article: Majumdar S, Uppala D, Priyanka KP, Kumar SP. Bilateral Regional Odontodysplasia: A Rare Case Report. Oral Maxillofac Pathol J 2016;7(2):755-757.

Source of support: Nil

Conflict of interest: None

INTRODUCTION

Regional odontodysplasia (RO) is a rare developmental anomaly of the tooth apparatus involving enamel, dentin, and cementum.[1] The affected teeth show thin enamel and dentin with large pulp chambers, enlarged apical foramen, and short roots. This overall appearance gives a faint radiolucent image, hence commonly termed as "ghost teeth." The other terminologies for RO are odontogenic dysplasia, localized arrested tooth development, unilateral dental malformation, odontogenesis imperfecta, and familial amelodentinal dysplasia.[2] The etiology of RO remains unclear. However, numerous factors like local trauma, teratogenic drugs, Rh incompatibility, neural damage, nutritional, and vitamin deficiency, etc. have been considered. The maxilla is affected twice as often as the mandible. It is usually unilateral but rarely bilateral and rarely crosses the midline. Females are affected more often than males (1.4:1).[3] This is an interesting case report of bilateral mandibular involvement, a feature that is rarely reported in the literature.

CASE REPORT

A 10-year-old girl reported to the Department of Pedodontics and Preventive Dentistry, GITAM Dental College & Hospital, Visakhapatnam, Andhra Pradesh, India, with a chief complaint of discoloration of partially erupted lower front teeth. Patient history revealed trauma at the age of 2 months. Family history was irrelevant. Intraoral examination revealed a firm swelling on the mandibular alveolar mucosa extending from distal surface of right lateral incisor to the mesial surface of left first molar along with pus discharge irt 31, 41 (Fig. 1). Panoramic radiograph revealed malformed mandibular anterior teeth, left first and second premolars. The teeth had malformed crown structure with no distinction between enamel and dentin with wide pulp chambers giving the "ghost-like appearance" (Fig. 2). Based on clinical and radiographic findings, we made the provisional diagnosis of RO. Under local anesthesia, the right mandibular canine and lateral incisors were surgically removed. The teeth were of

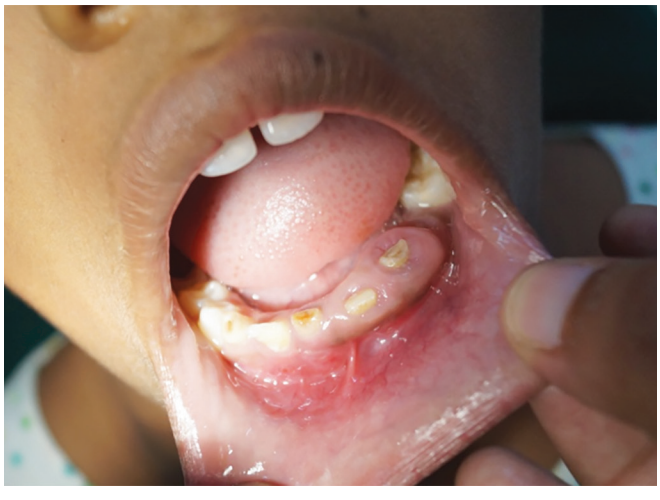

Fig. 1: Swelling on the alveolar mucosa extending from distal surface of right lateral incisor to the mesial surface of left first molar

[1]Professor, [2]Reader, [3,4]Postgraduate Student

[1-4]Department of Oral and Maxillofacial Pathology, GITAM Dental College & Hospital, Visakhapatnam, Andhra Pradesh, India

Corresponding Author: K Padma Priyanka, Postgraduate Student, Department of Oral and Maxillofacial Pathology, GITAM Dental College & Hospital, Visakhapatnam, Andhra Pradesh India, e-mail: vindhyakassetty@gmail.com

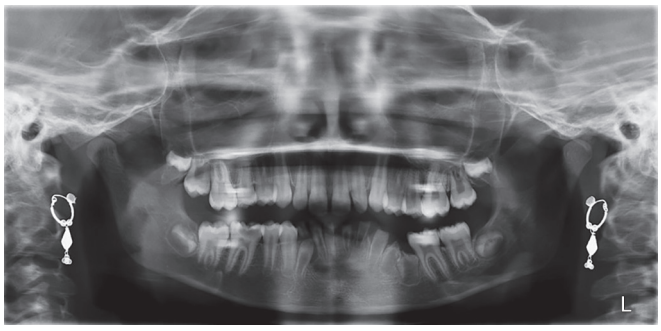

Fig. 2: Panoramic radiograph showing the teeth with "ghost-like" appearance of mandibular anterior teeth with the absence of distinction between enamel and dentin and wide pulp chambers

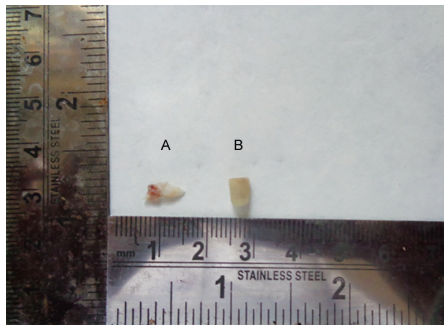

Fig. 3: Surgically removed teeth with altered morphology, yellowish in color, very short, or unformed roots

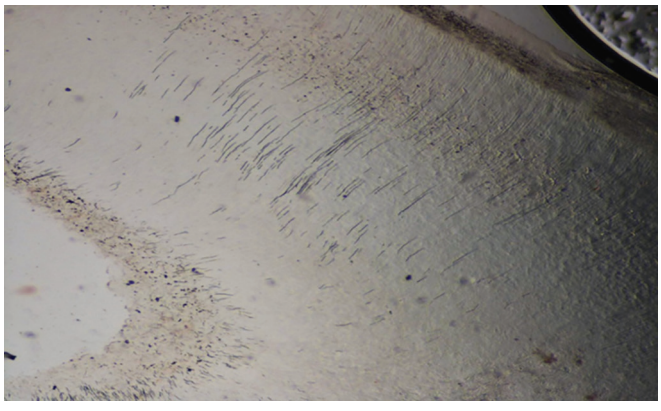

Fig. 4: Enamel and dentin with variable thickness, producing an irregular surface and central enlarged pulp cavity

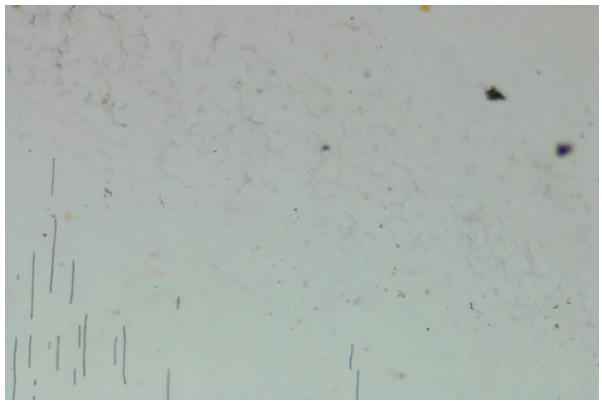

Fig. 5: Interglobular dentin and globular masses interrupting the dentinal tubules

altered morphology, yellowish in color, soft and rubbery in consistency, and had short or unformed roots (Fig. 3). Surgically removed teeth were histologically examined under an optical microscope. In the ground section the enamel and dentin were of variable thickness and producing an irregular surface with central enlarged pulp cavity (Fig. 4). Interglobular dentin and globular masses interrupting the dentinal tubules were also seen (Fig. 5). All the features were suggestive of regional odontogenic dysplasia.

DISCUSSION

Regional odontodysplasia is a relatively rare, nonhereditary, localized developmental anomaly that involves the hard tissue derived from both epithelial (enamel) and mesenchymal (dentin, cementum) components.[4] Many cases are misdiagnosed as malformed teeth or odontomas.[5,6]

This condition was first reported by Hitchin in the year 1934.[7] McCall et al in 1947 named this lesion as arrested tooth development. Rushton in 1954 introduced the term "shell teeth" based on radiological appearance. The word "odontodysplasia" was coined by Zegarelli et al in 1963. As the condition affects a group of several adjacent teeth in a particular segment of the jaw, Pindborg added the prefix "regional" in 1970, thus the name "regional odontodysplasia" was derived[7] (Table 1).

Table 1: Historical perspectives of regional odontodysplasia

First described by Hitchin	1934
McCall et al – named "arrested tooth development"	1947
Rushton – introduced the term "shell teeth" based on radiological appearance	1954
Zegarelli et al – proposed the term "odontodysplasia"	1963
Pindborg – gave the prefix as "regional"	1970

Prevalence of RO is less than 1/1,000,000. Most cases are reported under 23 years of age, but have bimodal age distribution at 4 and 10 years of age. Although this condition mostly affects one quadrant, seven cases of generalized odontodysplasia have also been reported in the literature till now. A special feature of regional odontodysplasia is that it does not cross the midline.[8]

ETIOLOGY AND PATHOGENESIS

Though many theories have been put forth, the etiology of odontodysplasia is still unclear. Some authors suggested a correlation with infection and trauma but this correlation was not proved, because that was based on patient history alone. According to Witkop,[9] distribution of affected teeth in all four quadrants suggests that somatic mutation may be a causative factor. They also suggested that a somatic mutation in the early development could cause alterations in the odontogenesis of the teeth involved. Sibley and

Zimmerman proposed that an imbalance of important proteins, such as metalloproteinases, might lead to the structural disorganization seen in this anomaly.[10] All the reports agree that this is a noninherited condition, because no cases were reported in other family members. The newer reports revolve around idiopathic etiology, as most of them failed to identify any local or systemic etiological factors.[11] Maxilla is affected twice than the mandible. Regarding the teeth, the central and lateral incisors are more frequently affected than the posterior teeth. It can occasionally cross the midline.[4]

Clinically, the affected teeth have an abnormal morphology with an irregular, rough surface with brown or yellowish discoloration, swelling or abscess of gingival and periapical infection.[5] The affected tooth enamel is soft on probing and more susceptible to caries and extremely friable, fracturing at the slightest trauma.[4] Radiographic view shows a ghost-like appearance, which is due to reduced radiodensity of the thin enamel and dentin, enlarged radiolucent pulp, and also due to lack of contrast between enamel and dentin, the roots are short with open apices. Thus the criteria for diagnosis of RO is primarily clinical and radiographic.[11]

To evaluate the mineral content of affected teeth, a microradiograph is indicated. As in conventional radiography, the density of enamel may not be evident due to the thinness of enamel layer.[2] Under microscopic examination the findings are reduction in the thickness of dentin layer and the expanded areas of interglobular dentin and predentin. In milder cases the mantle dentin appears normal, whereas in severe cases the dentinal tubules are reduced in number. The pulp often contains denticles and amorphous calcified materials.[5]

TREATMENT

The management of RO is often challenging and it requires a multidisciplinary approach and an organized team effort.[5] The objective of the management is to maintain good oral health and function, improve esthetics, and facilitate normal jaw growth.[12,13] Some authors recommend for the removal of the affected teeth as these are considered to be a potential source of infection.[11] Others suggest a conservative approach that saves the alveolar bone and enhances normal jaw development.[12]

CONCLUSION

- The presentation of this case helps pediatric dentists to review special clinical and radiographic features of RO.
- The therapeutic considerations of RO should be based on the degree of the anomaly and the functional and esthetical needs of each case.

CONSENT

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

AUTHORS' CONTRIBUTIONS

The oral surgeon excised the tissue and Dr. Divya Uppala, Dr. KP Priyanka, and Dr. S Praveen Kumar have contributed in analyzing, reading, and writing this article. Dr. KP Priyanka has done the research for the article.

REFERENCES

1. Cho S. Conservative management of regional odontodysplasia: case report. J Can Dent Assoc 2006 Oct;72(8):735-738.
2. Magalhaes AC, Pessan JP, Cunha RF, Delbem ACB. Regional odontodysplasia: case report. J Appl Oral Sci 2007 Dec;15(6): 465-469.
3. Crawford PJM, Aldred MJ. Regional odontodysplasia: a bibliography. J Oral Pathol Med 1989 May;18(5):251-263.
4. Naik LK, Shetty P, Girish Babu KL. Regional odontodysplasia crossing the midline: a unique case associated with bilateral impacted supernumerary teeth. J Pediatr Dent 2014;2(3):96-100.
5. Kappadi D, Ramasetty PA, Rai KK, Rahim AMB. Regional odontodysplasia: an unusual case report. J Oral Maxillofac Pathol 2009 Jul-Dec;13(2):62-66.
6. Venkatesh Babu NS, Smriti RJ, Pratima DB. Regional odontodysplasia: report of an unusual case involving mandibular arch. Contemp Clin Dent 2015 Apr-Jun;6(2):237-239.
7. Hitchin AD. Unerupted deciduous teeth in a youth aged 15 1/2. Br Dent J 1934;56(12):631-633.
8. Rashidian A, Zadeh MA, Azarshab M, Zarrabian T. Regional odontodysplasia: report of a case. J Dent Shiraz Univ Med Sci 2013 Dec;14(4):197-200.
9. Witkop CJ. Hereditary defects of dentin. Dent Clin North Am 1975 Jan;19(1):25-45.
10. Sibley LC, Zimmerman ER. Odontogenic dysplasia: report of a case. Oral Surg 1962;15:1370-1373.
11. Alotaibi ON. Regional odontodysplasia: an overview. Pakistan Oral Dent J 2015;35:175-178.
12. Cahuana A, Gonzalez Y, Palma C. Clinical management of regional odontodysplasia. Pediatr Dent 2005 Jan-Feb:27(1): 34-39.
13. Hamdan MA, Sawair FA, Rajab LD, Hamdan AM, Al-Omari IK. Regional odontodysplasia: a review of the literature and report of a case. Int J Paediatr Dent 2004 Sep;14(5):363-370.