REGIONAL ODONTODYSPLASIA: REPORT OF A RARE CASE AND REVIEW OF LITERATURE

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Abstract

Regional odontodysplasia is a rare developmental anomaly involving both mesodermal and ectodermal components in a group of teeth normally in the same area of jaw. It affects the primary and permanent dentition in maxilla or mandible.

The affected teeth are often grossly malformed showing deficient and abnormal formation of dentine and enamel and develop abscesses soon after eruption. Radio graphically the appearance of the teeth is very characteristic and is called as “ghost teeth” due to the thin shell of enamel and dentine enclosing a large pulp chamber. The etiology is uncertain but it is suggested that multiple factors play a role. The condition is not inherited nor related to genetics though many patients of regional odontodysplasia are seen to suffer from vascular nevi. The treatment plan in each case should be based on degree of involvement as well as functional and aesthetic needs in each case. Treatment modalities vary from conservative procedures that will help retain teeth for longer periods of time to auto transplantation or extraction and implants in hopeless cases.

This is a case of a fourteen year old girl with the rare anomaly on the right side of the mandibular arch involving only the first and second permanent molars. The clinical and radiographic features and treatment is discussed. The literature is reviewed in detail.

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Introduction

Regional odontodysplasia is a rare dental anomaly that affects one or multiple teeth from a localized area in an unusual manner. The condition being rare the literature available with respect to regional odontodysplasia is basically in the form of case reports only. It has been more than 60 years since the condition was first reported radio graphically by McCall et al in 1947 [1], but the incidence is so low that even now we find case reports in the international literature where authors state that the case is the first one reported in the country.[2]

A thorough search of literature was conducted where we stumbled upon a single article that has reviewed and analyzed 138 internationally published cases[3]. The understanding about the etiology and ultrastructural findings has not changed much in the past 7 decades either.

In this article we have reported a similar rare case of mild regional odontodysplasia involving only two adjacent permanent molar teeth and no other teeth in the jaw.

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Case Report

A 14 year old female patient belonging to a tribal community in the Central Part of India reported with a chief complaint of recurrent abscesses occurring in lower right molar region for more than six months.

The patient’s prenatal, birth, medical and dental history was unremarkable. The patient’s mother explained that her primary dentition was normal in every aspect and exfoliation and permanent teeth eruption was uneventful. The

patient did not have access to any formal dental care but the recurrent abscesses had made her visit the urban area for treatment.

General examination showed that the patient was afebrile and normal in height for her age but underweight.

On extra oral examination the submandibular lymph nodes on right side were mildly enlarged and slightly tender. Halitosis was noted and was of oral variety.

On intra oral examination the area of chief complaint was noted to be right mandibular first and second permanent molar. The teeth were seen to have an unusual coronal morphology and were much longer occluso-cervically.

The tooth structure was seen to be mildly hypoplastic and yellow to brownish in color but not soft to probing. The teeth were non carious. The gingiva around both the molars had receded mildly and the teeth showed deep periodontal pockets around them.

The gingiva was inflamed and suppurating. The teeth were mobile and depressible in their sockets. The examination of the rest of the teeth showed no other teeth in oral cavity having similar pathology.

The permanent molars and incisors were probed carefully for loss of attachment suggestive of any aggressive periodontitis but the attachment loss was absent.

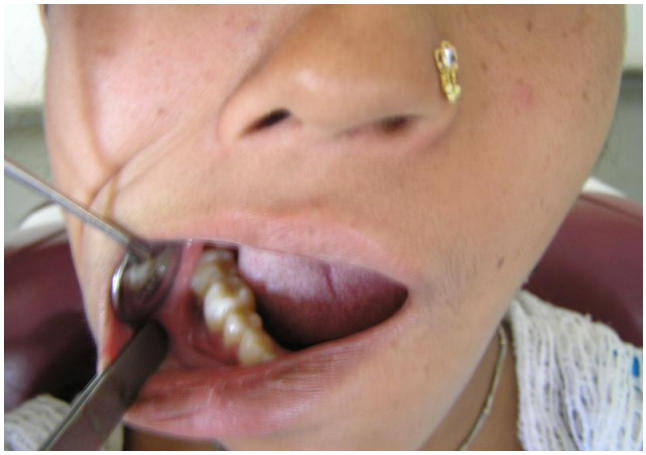

Figure 1. Clinical Photograph Showing the Involved Teeth.

The intra oral periapical radiograph of the area showed a markedly altered morphology and radio density of the structure of teeth. Both molars showed extremely short roots. The roots were shorter even than adjacent premolar root.

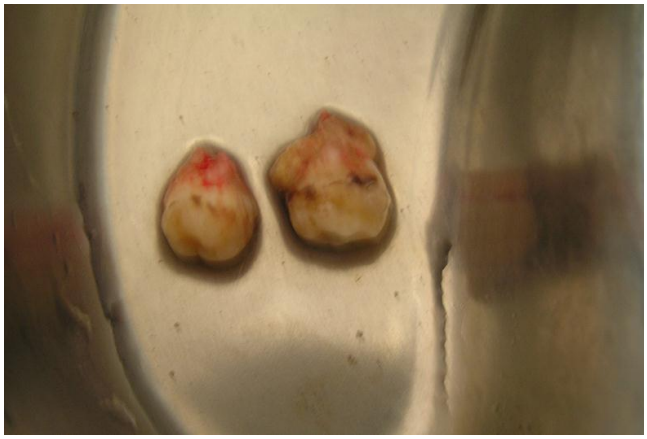

Figure 2. Extracted teeth specimens.

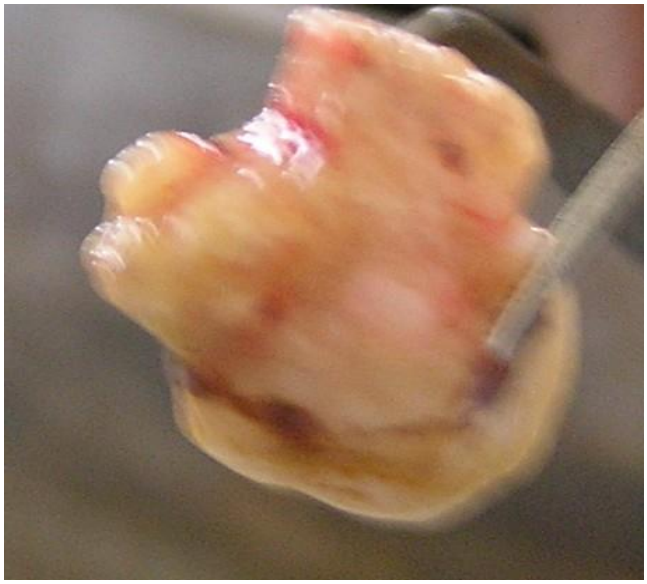

Figure 3. Roots showing irregular anatomy.

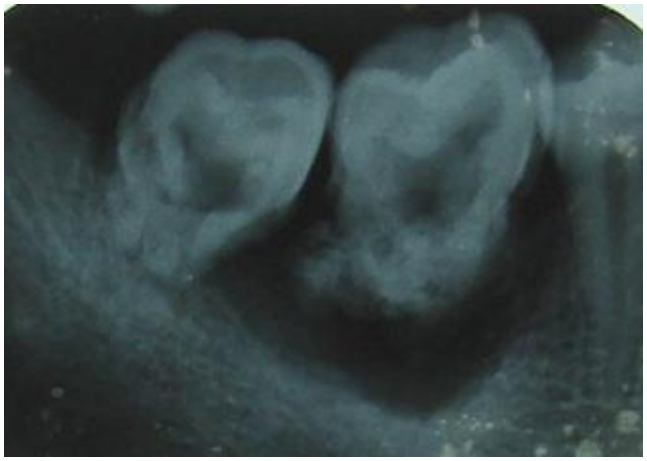

Figure 4. Intra Oral Periapical Radio graph showing altered roots, absence of root canals, periodontal bone loss and large pulp chambers.

The mesial and distal roots of both molars were fused and irregularly formed. The pulpal

morphology was altered. The pulp chamber was much larger and the roots did not have regular root canals. The first molar roots had lost their attachment and alveolar bone completely around them. The second molar had some bone distal to it but none mesially or apically.

The patient had blood sugar levels within normal limits and was mildly anemic.

The patient was advised extraction of the teeth as they were periodontally hopeless and was advised regular follow up with the rehabilitation with implants when she reaches adulthood.

The parent followed the advice about extraction of teeth but did not give consent for further investigations i.e. radiographic or pathological or histopathological. The patient was eventually lost to follow up.

Discussion and Review of Literature

The condition described in this case report is known by various terms in the literature. Odontodysplasia, Odontogenic Dysplasia, Odontogenesis Imperfecta, Ghost teeth to name a few. The term Odontodysplasia was coined by Zegarelli et al in 1963 [4] and later Pindborg added the prefix “regional” to it. The condition is a relatively rare nonhereditary developmental anomaly of hard tissues of mesodermal and ectodermal origin[4].

Clinical feature of teeth involved with regional odontodysplasia is gross malformation. The teeth involved may be single or multiple but are normally localized to a particular region of oral cavity and hence the name. We came across only one case in literature where the condition was affecting three quadrants of the same patient [5]. If anterior teeth are involved the condition may be seen to cross the midline[4].

The cases reported so far show a strong predilection for female sex, for anterior teeth compared to posteriors and for maxilla compared to mandible[3]. At the same time any teeth in the oral cavity may be involved.

The presenting complaints vary and range from failure of eruption to eruption soon followed by gingivitis, gingival swelling, gingival overgrowth, recurrent abscess or fistula formation[3]. Majority cases report both deciduous as well as permanent teeth being affected in the same manner[4]. Even teeth that erupt are shown to be hypoplastic or hypocalcified having grossly malformed crown and roots [6]. The defect in the structure of enamel and dentine results in caries, attrition and resultant esthetic and restorative problems.

Radiographically they are shown to have a very thin layer of enamel and dentine that surround a pulp chamber that is too large. Traditionally this appearance is called as Ghost teeth[7].

Histologically the teeth are shown to have a marked reduction in the amount of dentine, widening of predentine layer, presence of large areas of interglobular dentine and an irregular tubular pattern of dentine[7].

Proposed etiology: All the reports agree that this is a non-hereditary condition and there is no history of any systemic illness or of trauma associated with the condition[7]. It has been suggested that the condition may represent a somatic mutation and some suggest that it could be due to a latent virus residing in the odontogenic epithelium which subsequently becomes active during development of tooth[7].

One of the earliest reports documented that all the three patients of regional odontodysplasia had vascular nevi of the overlying facial skin as infants. They reported similar involvement in three additional cases in literature and these findings suggested to them that local vascular defects are involved in the pathogenesis of the condition [8]. The brown discoloration on the skin of patients has been noted even in one of the latest reported case involving three quadrants of jaws[5].

The newer reports give a more elaborate description insight about etiology and state that the anomaly involves both mesodermal and ectodermal dental components which result in deficient and abnormal function of dentine and enamel[9].

Treatment Suggested: The cases of regional odontodysplasia show a range of symptoms. They vary from a mild form involving altered shape of tooth to a severe form involving shell like teeth that show recurrent abscess formation. Hence a single treatment plan will not suit all. The treatment plan has to be based on degree of involvement as well as functional and esthetic needs of a patient. Older reports have uniformly advised extraction and replacement [7]. But the newer cases in literature especially with milder form of disease have tried various treatment modalities. Conservative treatments

have tried to retain the involved teeth for a long time without extraction[6].

Auto transplantation has been suggested as a good partial treatment option during period of mixed dentition in some cases[7,10].

Implants have been suggested as the definite treatment option when patient reaches adulthood[7,10].

In the case reported here, the grossly altered shape of the teeth along with recurrent abscess formation following eruption in oral cavity and localized appearance of the condition pin pointed to the diagnosis of regional odontodysplasia.[3]

The teeth though altered in morphology and color did not have a very soft consistency and this made the case to be one of the mild ones.[6]

The radiograph too showed the crowns of the teeth to be almost normal in radio density. However the maximum abnormality was seen to be in the roots and this seemed to result in recurrent abscess formation and large amount of bone loss with associated teeth. The mild form of dysplasia in coronal part but severe form of dysplasia in roots and localization of the condition only to permanent molar teeth hence absence of condition in deciduous dentition make the case one of rare ones.

The condition was not associated with any vascular nevi as described often in literature[8]. The condition was non-hereditary, not associated with systemic illness or local trauma as described by almost all the reported cases in literature. Recurrent periodontal abscesses were not a manifestation of undiagnosed diabetes as shown by her normal blood sugar levels. No attachment loss with any other teeth in oral cavity proved that it was not a case of aggressive periodontitis.

SHORTCOMINGS: Though the condition is known to be localized and it was confirmed that clinically no other teeth were involved, an orthopantomograph would have confirmed the findings radiographically. An ultrastructural and histochemical study of the specimen and its comparison with the similar report in literature would have shed more light on the cases of regional odontodysplasia[11].

Certain factors like whether deciduous teeth were unaffected and normal with regards to morphology, eruption and exfoliation could not be verified and we had to rely on the history only. This may be a vital issue as some cases in literature have described eruption disorders in cases of regional odontodysplasia[12].

Also rehabilitation of the young patient was needed. But the remote locality, certain communal beliefs, inaccessibility to basic dental care, lower education and socioeconomic status could be few of the multiple factors for the short comings and also the reasons for why the patient was lost to follow up.

Conclusions

Regional odontodysplasia is a rare non hereditary condition with very specific clinical and radiographic findings. Though known in dental literature for more than six decades the incidence of the condition is so low that almost all the literature consists of only the case reports.

The cases vary from mildly affected to severely affected forms. The teeth affected by regional odontodysplasia are seen to have a hypoplastic or hypocalcified enamel and dentine and hence are very soft in consistency and have a shell like appearance called as Ghost Teeth.

Routinely, the teeth showing the condition show uniformity in the degree in which crown as well as roots are affected. Also both the deciduous as well as permanent dentition are affected.

This case report describes a similar case but rare and unusual because the dysplasia of severe degree was affecting roots though crowns were only mildly affected. Teeth involved were permanent molars and hence the deciduous dentition was not involved.

The recurrent abscess formation had resorbed the alveolar bone severely and the teeth had to be extracted. The age and the clinical features of the condition suggested a differential diagnosis of aggressive periodontitis or undiagnosed diabetes mellitus. But the radiographic features pointed to the correct diagnosis of regional odontodysplasia.

Declaration of Interest

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