Case Report

Regional Odontodysplasia: A Case with Radiographic Evidence of Advancing Development

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Abstract

Regional odontodysplasia, also commonly known as ghost teeth, is a rare dental anomaly affecting the teeth. It is a nonhereditary disorder and affects the maxillary teeth of females. This condition is localized to one or more teeth of one quadrant; it hardly crosses the midline to affect the teeth of the next quadrant. Controversy regarding the management of this condition is still prevalent, and some believe in extraction while others in retaining them. A case of ghost teeth in an 11-year-old boy, crossing the midline with radiographic evidence of advancing development, is being presented, which supports the "wait and watch" policy in the management of this condition.

Keywords: Ghost teeth, odontodysplasia, regional odontodysplasia, tooth abnormality

Introduction

Regional odontodysplasia (RO) is a rare developmental anomaly of the tooth which is nonhereditary.[1,2] Mc Call and Wald are credited for their first published report on RO, although it was Hitchin who identified this condition.[2] This entity is christened differently by different investigators. Ghost teeth, nonhereditary segmental amelogenesis imperfecta, unilateral dental malformation, arrested tooth formation, familial amelodental dysplasia, odontodysplasia, and odontogenesis imperfecta are the synonyms used to describe this entity.[2-5]

Case Report

An 11-year-old boy was brought for evaluation of unerupted maxillary anterior teeth. The deciduous counterparts had been exfoliated following mobility at the age of 8 years. There was no history of preceding trauma or toothache, and there were no associated symptoms as well. His medical and surgical histories were noncontributory. The child was born as a first child to parents who were not blood relatives. The parents had consulted a dentist previously about 1 year back with the same complaint and radiograph had been made. The parents were not cognizant about the diagnosis. They expressed their inability to follow-up with the clinician due to lack of accessibility of clinic. Extraoral examination revealed no abnormalities, whereas intraoral examination revealed missing deciduous maxillary right and left central incisors and left lateral incisor and unerupted permanent counterparts [Figure 1]. The alveolar mucosa in the region appeared normal. There was no evidence of eruption bulge, while the adjacent and other teeth in the oral cavity appeared normal. With simple delayed eruption in mind, an intraoral periapical radiograph and maxillary anterior occlusal radiograph were ordered [Figure 2]. The radiograph showed improperly formed maxillary right central incisor and left central and lateral incisors, while normally developing right lateral incisor. The teeth lacked root development, and there was indistinct demarcation between enamel and dentin. The pulp chambers and follicular spaces appeared wide. The previous radiograph made a year ago [Figure 3] showed similar findings, but upon careful observation, the right central incisor appeared to be less developed and mineralized than presently. The radiographic appearance was consistent with RO. A panoramic radiograph was made to rule out more generalized involvement which proved to be negative. The patient and the parents were informed about the condition and

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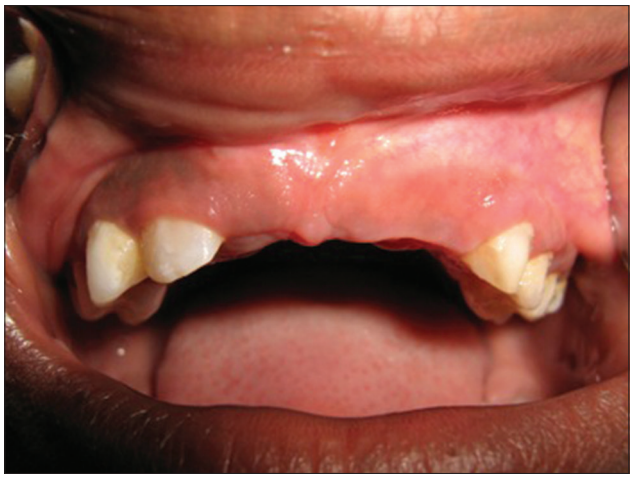

Figure 1: Intraoral photograph showing unerupted maxillary anterior teeth

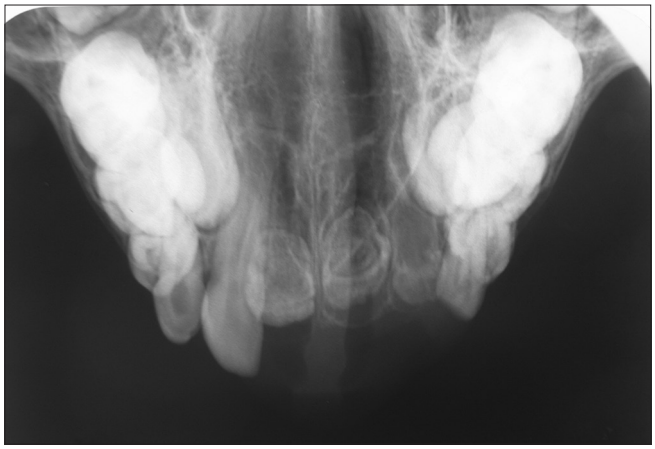

Figure 2: Maxillary anterior occlusal radiograph showing the ghost teeth

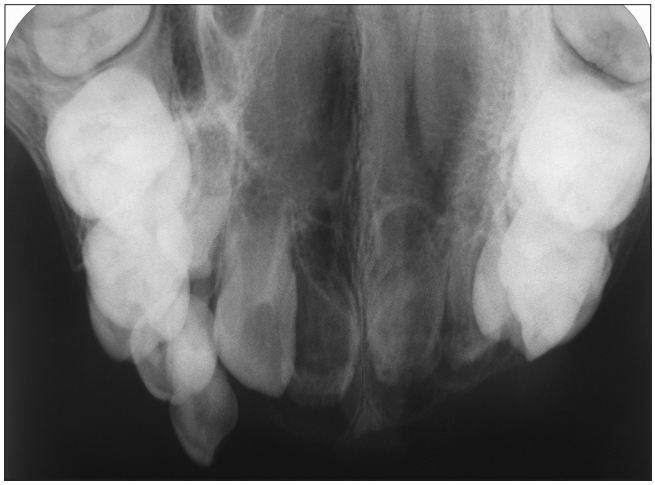

Figure 3: Maxillary anterior occlusal radiograph made 1 year back showing the ghost teeth

motivated for close follow-up. Further, they were educated about the "wait and watch" policy as there were more chances of the teeth erupting and normalizing in due course, the evidence of which was already seen through radiographs. Furthermore, a removable denture was fabricated for esthetics. The patient reported for a follow-up after 2 months with no further development of the ghost teeth. Despite motivation, the parents failed to bring the child for additional follow-ups.

Discussion

Etiology

Etiology of this rare entity is incompletely understood, and many theories have been proposed and much of them have been either disproven or negated.[1] It is one of the anomaly teeth where all the three structures, the enamel, dentin, and pulp, are affected.[5] It is now clear that it is a nonhereditary condition.[3,4]

Epidemiology

Prevalence remains largely undetermined as there are no planned epidemiological studies. Literature states that it is lower than 1 in 10 lakhs.[1,6] Interestingly, close to 150 cases, from worldwide, of this entity are reported so far.[6] There is no predilection for any race or gender, while a few reports point toward a female preponderance with the female-to-male ratio being 1.4:1.[1,3]

As the terminology implies, this condition is regional meaning solely affecting teeth in one quadrant which rarely crosses the midline.[1-3] The left side of the maxilla is more common than the other sides or the other jaw.[1] Rarely, this rule of unilateralism is broken, and teeth in the adjacent jaw as well as being a generalized condition is also reported.[2,3] Teeth commonly affected are the left lateral incisor and right 1st and 2nd molars.[7,8]

Clinical features

The affected teeth may fail to erupt, and if at all they erupt, the teeth may appear to be misshapen, discolored, and smaller in the dimensions. The color may vary from yellow to brown, may show grooves, and may be hypoplastic.[4] Gingival enlargement is not an uncommon finding.[6,7] Erupted teeth being caries prone may also develop periapical abscesses.[7]

Radiographic appearance

The classic radiographic feature is the one that contributes this disease its name. The radiographs of the affected teeth show overall reduced density of the mineralized structures, and hence, they lack a distinct demarcation between them resulting in a "ghost-" like appearance. There is generally a wide pulp chamber, while the enamel and dentin are thin.[3,4,6,8]

The teeth may have short roots with open apices, and rarely, those teeth which fail to erupt remain in the bone impacted with pericoronal radiolucencies.[2] These radiolucencies occasionally can be so large to be confused with a cyst or a tumor.[9]

Computed tomography has been recently utilized to study the ghost teeth as well as their pericoronal radiolucencies. The Hounsfield Unit for the radiolucencies has been determined to be equal to that of fibrous connective tissue, while of

enamel, it was found to be lower than normal teeth indicating hypomineralization. It was additionally found that the hypomineralization was more severe in enamel than dentin.[9]

Histopathology

Those teeth which failed to erupt and subsequently extracted have served as excellent candidates for histopathology which has shown that the enamel is hypocalcified and the prisms are irregular, dentin is fibrous, composed of clefts, while the dentinal tubules are fewer. The pulp may additionally show calcifications.[2,3,5]

Management

Much controversy surrounds the management of this unusual entity.[1-3,5,7] Some advocate extraction of the affected teeth while others advocate retention.[2,4,5] It is argued that extraction of affected teeth would prevent development of the alveolar bone and may also lead to psychological disturbances. Further, anecdotal cases of affected teeth eventually erupting and normalizing after a delay have also been reported.[2] Auto-transplantation of unaffected teeth also has been suggested, provided, normal donor teeth are available.[3]

Prognosis

Some clinicians believe that affected erupted teeth will, in due course, develop periapical infection owing to hypomineralization and need extraction. Conversely, affected teeth that have survived for more than 25 years, without intervention, are also available.[2]

Conclusion

As much as the etiology of this rare entity needs a closer research, so does its management. As there are fewer reports worldwide, sufficient evidence regarding management is lacking. Anecdotal reports play a vital role in detailing the clinical and radiographic features of this rare anomaly as does the management. This paper illustrates the fact that affected teeth may still have the eruption potential, may normalize, and may refute the belief of extraction.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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