

Ghost Teeth: A Rare Developmental Anomaly of the Dental Tissues

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Abstract - Regional odontodysplasia (RO) is a sporadic (not inherited) developmental defect involving only a few contiguous teeth in a small region of the jaw; developmentally it involves both mesodermal and ectodermal dental components. It affects the primary and permanent dentition in the maxilla and mandible or both jaws. Generally it is localized to only one arch. The etiology of this dental anomaly is uncertain. Clinically, affected teeth have an abnormal morphology, are soft on probing and typically discoloured, yellow or yellowish-brown. Radiographically the affected teeth show a thin layer of dentin and enamel with a huge pulp space producing a "ghost like" appearance. This paper reports a case of a 3-year-old boy who presented with this rare anomaly on the left side of the maxillary arch and the sequelae of the same on the involved primary teeth. The primary maxillary left central, lateral and canine were involved with varying degrees of severity. Management of RO is challenging and long term, the various treatment modalities will be outlined and discussed.

Keywords: Ghost teeth, Regional odontodysplasia, Shell teeth, Developmental anomalies of primary teeth.

Introduction

Developmental anomalies involving the primary and permanent dentition can pose confusing and challenging problems for the clinician in diagnosis and clinical management, both short and long term and require an interdisciplinary approach. Regional odontodysplasia (RO) is a localized disorder of tissues of dental origin resulting in characteristically bizarre clinical and radiographic appearances. It most commonly affects the maxillary anterior teeth of both the permanent and primary dentitions. [1]

Crawford [1] ascribed the first report to Hitchin in 1934. The condition was defined by Gibbard et al. as "a developmental anomaly, principally affecting developing coronal odontogenic tissues, that is non-familial (and ultimately related to the vascular supply)". Enamel, dentin, pulp and follicle are all affected. [1, 2]

Many authors however, give credit to McCall and Wald in 1947 for the first published report of this condition which they termed 'arrested tooth development'. In 1954, the term 'shell teeth' was introduced by Rushton. [2]

The word "odontodysplasia" was coined by Zegarelli, et al. in 1963. Because this abnormality has a tendency to affect only one quadrant, "regional odontodysplasia" became the most accepted term to define it. [3, 4]

No specific racial predilection has been described. It affects both primary and permanent dentition and can occur in the maxilla, mandible or both. The maxilla is twice as involved as mandible and it is usually unilateral with rare cases crossing the midline.[5] It occurs with increased frequency in females as opposed to males with a ratio of 1.4:1. [1, 6]

The RO etiology is uncertain; numerous factors have been suggested such as local trauma, irradiation, hypophosphatasia, hypocalcemia, hyperpyrexia. It seems to be a non-inherited condition because no cases with affliction for family members have been described. The cause is unknown but several theories have been advanced such as vascular defects involving ischaemia, local infections, trauma, Rhesus incompatibility, irradiation, neural damage, hyperpyrexia, metabolic and nutritional disturbances, hereditary and somatic mutation. [1-4]

The Condition affects enamel, dentine and the pulp producing delayed or failure of tooth eruption, early exfoliation, abscess formation, malformed teeth and non-inflammatory swellings of the jaw presenting as a firm painless gingival enlargement and inflammation around the affected teeth.[7, 8]

The clinical and radiographic characteristics of Regional odontodysplasia will be discussed in this article, describing this anomaly with certain unique findings and treatment options that need to be considered with specific reference to pediatric patients especially those with primary teeth. The management of regional odontodysplasia is somewhat controversial and revolves around the question of whether or not to remove the affected teeth. Although many clinicians prefer to extract the anomalous teeth as soon the diagnosis is made, some would prefer to retain them as long as they are free of infection until the skeletal growth is complete. [5]

Case Report

A 3-year-old healthy boy was referred to the Pediatric dentistry clinic at our esteemed dental College, for detailed evaluation of his oral condition. According to his mothers report, the primary maxillary left teeth had erupted differently from the other teeth. The mother also reported that the affected teeth had a yellowish color and were getting rapidly destroyed and appeared to be broken.

On recording the case history from the mother it was found that the pregnancy and the birth occurred uneventfully. There was no history of tooth or genetic anomalies in the family. The child's general health was good and no congenital or acquired disease was found while recording the history. Extra oral examination revealed no facial asymmetry.

Intraoral clinical examination revealed a relatively caries-free mouth with normal occlusion, soft tissues and developing dentition except for the maxillary left quadrant (**Figures 1 and 2**). On the left side of the maxilla, the primary central and lateral incisors and primary canine were affected. The panoramic radiograph and IOPAR (**Figures 3 and 4**) which were taken revealed the presence of primary teeth and the germs of permanent teeth, except in the maxillary left quadrant. In the affected area the root of primary left lateral incisor was also present. In addition, the primary left canine presented reduced radio density and showed wide open apex and abnormally wide pulp chambers and canals in comparison to unaffected teeth.

Germes of permanent teeth from the maxillary left quadrant and also presented a "ghost like" appearance. Dental development appeared age-appropriate. Normal thickness of enamel and dentin in primary and permanent dentitions was observed in the other quadrants.

The affected three teeth were restored with a resin modified glass ionomer and kept under observation to limit further breakdown. The boy was followed up periodically (once a month) to observe if eruption of the "affected teeth" occurred and to monitor the growth and development of the maxillary and mandibular arches.

Treatment options for a child with RO are based on factors such as age of the patient, any relevant medical history, previous dental experience, child's and parental attitude regarding dental treatment and number of affected teeth.

There has been much debate as to whether affected teeth (with or without abscesses) should be extracted or saved. In this case, at a six month review, the maxillary left primary lateral incisor (62) was extracted as it had developed an abscess and fistula (**figure 5**). In contrast, the maxillary left primary central incisor (61) and canine (63) were restored with resin modified glass ionomer as they were less affected by the anomaly.

Discussion

The purpose of this case report is to increase the recognition of regional odontodysplasia. Diagnosis of this developmental anomaly is primarily based on clinical and radiographic criteria so clinicians should not only clarify the pattern of clinical features that may be regarded as being within a normal range and be aware of the possible clinical sequelae of this rare condition but also lead to an improvement in both the short and long term management of patients with the condition.

Although many theories have been proposed [¹⁻⁴], in this particular case it was not possible to determine a precise etiological factor.

Other conditions, such as, dentinal dysplasia, shell teeth, hypophosphatasia, dentinogenesis imperfecta, or amelogenesis imperfecta can mimic some features of regional odontodysplasia. However, these disorders tend to affect the entire dentition.^[2]

The criteria for diagnosis of RO are primarily clinical and radiographic. Clinically, affected teeth have an abnormal morphology and an irregular surface contour, with pitting and grooved surfaces. The teeth appear to be discoloured yellow, yellowish or brown, are hypoplastic and hypocalcified. The thin enamel is soft on probing and the affected teeth are more susceptible to caries and are extremely friable.

Regarding the teeth, the central and lateral incisors are more frequently affected than the posterior teeth [⁴] as was seen in this case. When RO is observed in the primary dentition, teeth can be erupted, hypo plastic, hypo calcified, with changes in color and form. Affected teeth are likely to be small, brown, grooved, and hypo plastic. Gingival tissue can be hyperemic and usually presents a fistula. In the permanent dentition, teeth usually do not erupt or can be partially erupted with fibrous gingival tissue and swelling. [^{3, 4, 6}]

This case confirms the clinical presentation of regional odontodysplasia. In the condition, affected teeth are hypoplastic, mobile and radiolucent, giving rise to the term 'ghost teeth'. Radiographically, there is a lack of contrast between the enamel and dentin, both of which are less radiopaque than unaffected counterparts. Additionally, enamel and dentin layers are thin, giving the teeth a 'ghost-like' appearance. [^{3, 6, 9}]

The patient in this case report exhibited many of the common clinical and radiographic features consistent with the diagnosis of RO. The clinical and radiographic characteristics involving the permanent dentition in the maxillary left quadrant (and also including the right permanent central incisor) strongly supported the diagnosis of this condition. RO seems to in both dentitions primary and permanent, as found in this case. The permanent teeth will probably not erupt and will have an altered eruption pattern. The non infected permanent teeth will not be extracted before eruption because these teeth help maintaining the alveolar bone. [⁹]

This case showed some features that were not in accordance with the literature, in that the condition seemed to affect females more than males but in the current case the patient in question was a male even though cases of RO involving males have been ascribed. Therefore, further studies with a greater number of cases are necessary to confirm this tendency. [^{1, 6, 9}]

According to the literature, abscess formation is the main reason for extraction of affected teeth as was seen in our patient six months after first visit. [^{1, 3, 4, 7, 9}]

The treatment of RO has given rise to controversy. These cases require a continuous and multidisciplinary approach. Most clinicians advocate extracting the affected teeth as soon as possible and inserting a prosthetic replacement. Other clinicians prefer restorative procedures, if possible, to protect the affected erupted teeth. However, selection of method

and timing appear to be critical factors in the treatment of RO.

Although in very young children, teeth in the arch should be retained, teeth involved with abscesses can not be restored, and need to be extracted. In contrast, in older children, abscessed permanent teeth should be extracted with others retained until final rehabilitation with implants and/or fixed prosthesis.

As mentioned by Cahuana A, Gonzalez Y, Palma C in their article in 2005 they stated that in a child with RO, conservative treatment should be applied to preserve the affected teeth for as long as possible to provide normal jaw development. Several reports state that if abscessed teeth are present, they should be extracted and edentulous areas should be restored with acrylic removable appliances to:

1. maintain aesthetic and masticatory functions;
2. avoid over eruption of opposing teeth;
3. achieve space preservation and normal vertical dimension;
4. lessen the psychological effects of premature tooth loss

Despite the increasing use of osseointegrated implants in patients with missing teeth, their use is contraindicated in growing patients. Implants are preferably placed after pubertal growth. [9]

Taking these goals into consideration, we examined the affected teeth clinically and radiographically and decided to initially conservatively manage the case and follow up the child regularly. On his sixth month visit an intraoral abscess with a fistula was noted in relation to left primary lateral incisor which was subsequently extracted. The affected maxillary left primary central incisor and canine were not extracted and restored conservatively and are currently under observation.

Conclusion

The presentation of this case helps clinicians to review special clinical and radiographic features of Regional Odontodysplasia

or Ghost teeth as the condition is referred to commonly. The therapeutic considerations of RO should be based on the degree of the anomaly, the functional and esthetical needs of each case. Individual management is required until the patient reaches the age for prosthetic rehabilitation.

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Figure 1: Extraoral View of the Child with RO



Figure 2: Intraoral View of the Child with RO



Figure 3: Periapical Radiograph Showing the Characteristically Ghost-like Appearance of Regional Odontodysplasia



Figure 4: Panoramic Radiograph Showing the Teeth with “Ghostlike” Appearance in Maxillary Left Anterior Segment



Figure 5: Intraoral View of the Child with RO after 6 Months Showing 62 with Intraoral Abscess

