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REGIONAL ODONTODYSPLASIA OF MANDIBLE- REPORT OF A RARE CASE AND LITERATUE REVIEW

Dental Science

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ABSTRACT

Regional odontodysplasia (ROD) is a developmental anomaly of teeth involving both mesodermal and ectodermal components in primary or permanent dentition with distinctive clinical, radiographic and histological findings.They have characteristic radiographic findings of ghost teeth appearance. Numerous factors have been suggested to explain the etiopathogenesis of ROD.It shows a slight female predominence.Maxilla is most commonly affected (2.5:1) with a predilection for the anterior teeth.There will be delayed eruption of the affected teeth or sometime it does not occur at all. Here we are reporting a case of regional odontodysplasia affecting the entire left quadrant of mandible in a two year old female child.Ultrastructural findings of affected tooth are also presented here.

KEYWORDS

Regional Odontodysplaia,Ghost Teeth,Delayed Eruption.

Introduction:

Regional odontodysplasia(ROD) is a developmental anomaly of teeth affecting primary or permanent dentition.[1]It has distinctive clinical, radiographic and histological findings.[1] It occurs as a the result of developmental disturbance that locally affects the odontogenic ectodermal and mesodermal tissues.[1]

Regional odontodysplasia is a rare development anomaly affecting the teeth with an unknown etiology[2]. This dental abnormality involves the hard tissues of the teeth that are derived from both epithelial (enamel) and mesenchymal (dentine & cementum) components of the tooth forming apparatus. The affected teeth are described as "ghost teeth," due to malformation of dentin, enamel, and cementum.[3]

Teeth in a region or quadrant of maxilla or mandible are affected to the extent that they exhibit short roots, wide open apical foramen and large pulp chamber, the thinness and poor mineralisation qualities of the enamel and dentin. layers have given rise to a faint radiolucent image, hence the term "Ghost teeth".[4]

Females show a slight predominance than males. It most commonly occurs in maxilla(2.5:1) with a predilection for the anterior teeth[4].There will be delayed eruption of affected teeth or sometimes does not erupt at all. Regional odontodysplasia showed widespread globular dentin, enlarged pulp chambers with calcifications inside it and hypoplastic and hypomineralized enamel.[5] However, necrosis and facial cellulitis appear to be a complication if these teeth are retained.[6]

The first report of this condition was published by Mccall and Wald in 1947, but the term "odontodysplasia" was introduced by Zegarelli *et al* in 1963.[7]Before that a number of cases have been described under a variety of names; such as localized arrested tooth development, familial amelodentinal dysplasia, unilateral dental malformation, regional odontodysplasia, ghost teeth, odontogenesis imperfecta, and amelogenesis imperfecta .[7]The diagnosis of ROD are mainly based on clinical and radiographic findings but sometimes supplemented by histological findings.[8]The etiological features include local trauma, irradiation, hypophosphatasia, hypocalcemia, hyperpyrexia.[7]

Clinical Manifestations :

Regional odontodysplaia shows a localized occurrence within a particular segment of either or both dentition[2].There will be hypoplasia and hypocalcification of enamel and dentin[2].Affected teeth will be small,distorted and show yellowish or brownish discolourations with various surface markings with surface pits and grooves and are soft on probing[2].Due to arrested root formation there will be delayed or failed eruption.[9]The condition is usually unilateral, although exceptions can be found.[8]The anomaly is usually localized in one arch, with incidence higher in the maxilla.All teeth of the same arch are not affected simultaneously. If it affects both arches the presentation is usually unilateral. The affected teeth most often occur as a continuous series, although occasionally the anomaly will "skip" a tooth or group of teeth.[10] Eruption of the affected teeth is often delayed or failed.

Radiographic findings:

Radiographically, the affected teeth show an abnormal morphology and hypoplastic crowns with thin and defective layers of enamel and dentin. resulting in a faint, fuzzy outline, creating a ghost-like appearance.[2]

Histopatholgiacl findings:

Histologically in ground sections enamel shows irregular thickness with a laminated appearance.[11] The dentin contains clefts scattered throughout a mixture of interglobular dentin and amorphous material.[11]Globular areas of poorly organized tubular dentin and scattered cellular inclusions often are seen.[11] Denticles and amorphous calcified materials are seen inside the pulp chamber. The follicular tissue surrounding the crown may be enlarged and typically exhibits focal collection of basophilic enamel like calcifications called enameloid conglomerates.[11]This pattern of calcification is not specific for regional odontodysplasia.[11] But in other developmental disturbances affecting teeth scattered island as of odontogenic epithelium and other pattern of intramural calcifications are also seen.[11]

Case report:

A 2 year old female child complaining of swelling on left lower jaw.vascular nevus is present on left chin near lower lip.maxillary teeth are unaffected. Her prenatal, birth, medical, and family history was unremarkable.

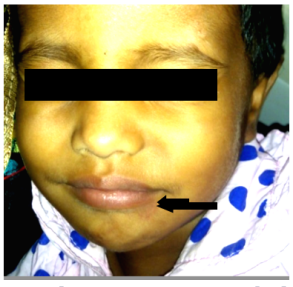

Figure.1 showing vascular nevus present on the left chin near the lower jaw.

Intraoral clinical examination revealed 71 and 72 are hypoplastic.74 is grossly decayed,75 not erupted.Gingival enlargement seen on left lower quadrant. The tooth was extremely tender on percussion and pain on palpation of the surrounding gingiva was also noted

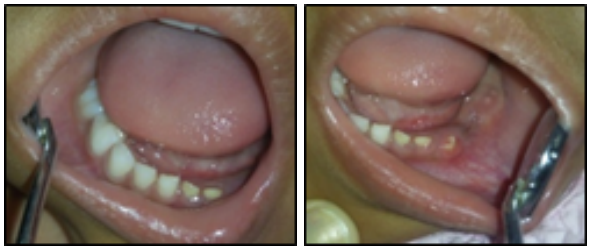

Figure.2 showing affected left mandibular quadrant and unaffected right quadrant.

RADIOGRAPHIC FINDINGS: Ghost tooth appearance on mandibular left quadrant. Absence of succedeneous tooth buds

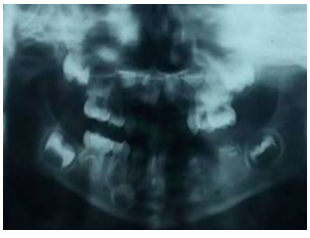

Figure.3 showing ghost teeth appearance on mandibular left quadrant.

GROUND SECTION: Ground section shows irregular dentinal tubules with indistinct dentinoenamel junction.Numerous amounts of interglobular dentin is also seen.

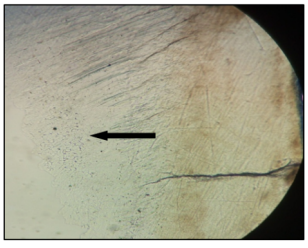

Figure.4 showing ground section of affected tooth with numerous amounts of interglobular dentine.

The case was diagnosed as Regional odontodysplasia of left lower quadrant on the basis of clinical and radiological findings.

Treatment

Since the involved teeth could not be restored effectively, the extraction of the dysplastic teeth was recommended.

Discussion:

Regional odontodysplasia is an uncommon developmental anomaly, and it rarely crosses the midline.,ROD quite rarely cross the midline in mandible frequently when compared with maxilla. Moreover, these patients are usually females Both the permanent teeth and the deciduous teeth are affected. Females are more commonly affected than males. There is a maxillary predominance (2.5:1) with a predilection for the anterior teeth. Eruption of the affected teeth is delayed or does not occur .

Regional odontodysplasia is a rare developmental dental anomaly and the prevalence of this condition is still not definitely clear since the studies have mainly been based on the case reports.[12]Its prevalence is reported to be less than 1/1000000 and only about 140 cases have been reported in the literature up to the time of this study[12]

The affected teeth are clinically small, grooved, hypoplastic and hypocalcified .[2] The involved enamels are soft on probing and the implicated teeth are susceptible to caries .[13] These teeth often display a brown or yellowish discoloration. The most frequent clinical symptoms accompanied by this anomaly are failure of eruption, swelling or abscess of gingiva and periapical infection.[13] Moreover, nine cases of hemangioma and six cases with facial hemi-atrophy were reported to be present with regional odontodysplasia .[14]

The ROD etiology is uncertain; numerous factors have been suggested and considered as local trauma, irradiation, hypophosphatasia, hypocalcemia, hyperpyrexia.[7] It has been suggested the disturbance may derive from a disorderly proliferation of the dental epithelium at an early stage of tooth development.[7] several factors such as, infection, ischemia or vascular defects, neural damage, , nutritional deficiencies, teratogenic drugs, Rh incompatibility, activation of latent viruses residing in odontogenic epithelium, somatic mutations and disorders of neural crest cell migration have been advocated.[15] It reported that regional odontodysplasia is probably a non-hereditary condition.Some patients may also present with systemic anomalies, such as facial asymmetry and other abnormalities. In the present case, parents were unaware/could not recollect any birth injury or medication or any other disease related to etiology of tooth abnormality.

One theory suggests RO could be caused by a vascular disturbance creating a local ischemia affecting odontogenesis.[16] This theory is supported by the occurrence of associated hemangiomas or vascular nevi in areas adjacent to affected teeth in many of the reported cases.[16] In this case also there was an associated vascular nevus present on the left chin near the lower lip .The affected teeth were also present over the left lower quadrant region confirming the etiology behind as some local vascular defects . The etiology of RO remains unknown and further detailed investigations of cases of RO are needed to confirm possible etiologic factors.

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