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CASE REPORT

REGIONAL ODONTODYSPLASIA OF MAXILLARY PERMANENT ANTERIOR TEETH – A CASE REPORT

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ABSTRACT

Odontodysplasia is a rare nonhereditary developmental anomaly of dental hard tissue with unknown etiology, arising from both ectodermal and mesodermal components. Hitchin in 1934 first described it as a localized arrest of tooth development generally affecting one quadrant of the jaw. When it just affects one quadrant, it is referred to as "Regional Odontodysplasia," but when it crosses the midline and affects more than one quadrant, it is referred to as "Generalised Odontodysplasia." In this case, 10 year old female patient was diagnosed with Regional Odontodysplasia affecting the Maxillary Anterior Teeth on left side confirmed with Clinical and Radiographic Features.

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INTRODUCTION

Regional odontodysplasia (RO) is a relatively rare localized developmental anomaly of the dental tissues with specific clinical and radiographic features. The word "odontodysplasia" was coined by Zegarelli, et al. (1963). It is defined as a localized non hereditary developmental anomaly with adverse effects on the formation of dental tissues. (1) McCall and Wald (1947) was the first to report this condition, based only on radiographic findings (2,3). The same condition has been described under other denominations, such as "Odontogenic Dysplasia", "Localized Arrest Tooth Development", "Ghost Teeth", "Odontogenisis Imperfecta", "Unilateral Dental Malformation" and "Familial Amelodentinal Dysplasia". "Regional Odontodysplasia" became the most accepted term to define it. This is a rare developmental dental anomaly with an unknown etiology. It is more often seen in girls than boys. The primary diagnostic criteria for Regional odontodysplasia involves both clinical and radiograph findings. Treatment of RO depends on the individual case. The eruption of affected teeth is delayed. Eruption may not occur in severe cases. (2) We present a rare case of the developmental anomaly called RO or "ghost teeth" in a 10 year old female patient.

CASE REPORT

A 10-year-old female patient visited the department of oral medicine and radiology with chief complaint of delayed eruption and discolouration of tooth in upper left front teeth region. Patients gives history of trauma to the upper front teeth when she was 4-year-old. The past medical history was non-contributory. No congenital or acquired diseases were reported. There was no history of tooth or genetic anomalies in the family. Extraoral examination revealed no facial asymmetry. (Figure 1) Intraoral clinical examination revealed normal developing dentition except for the maxillary left quadrant with Class I molar relation. A solitary sessile swelling was present on the left maxillary gingiva on labial aspect irt left maxillary central incisor, roughly oval in shape measuring

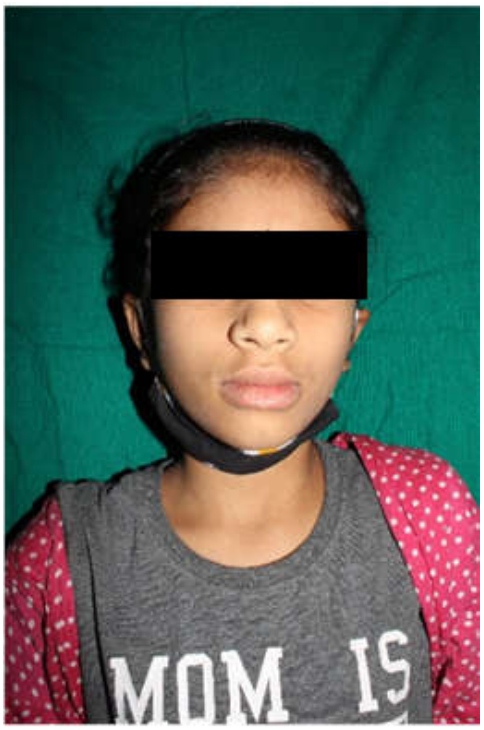

FIGURE 1. Extraoral picture of the child with RO

1x1cm in greatest diameter, extending medio-laterally from Maxillary left central incisor to left lateral incisor and superior-inferiorly from marginal gingiva to vestibule. The borders are ill defined and mucosa over the swelling was normal and there was no visible pulsation noted. On palpation, it was tender, soft in consistency, non compressible, non-fluctuant. There was partially erupted maxillary left central and lateral incisor which were hypoplastic with missing left maxillary canine. (Figure 2)

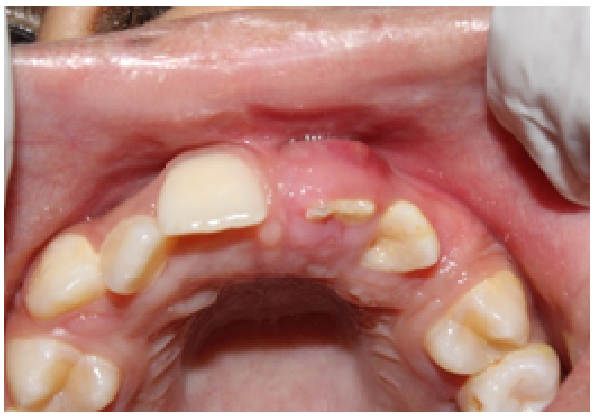

Figure 2. Occlusal view of maxillary arch with left maxillary central and lateral incisors affected

Intra oral (Digital) periapical radiographs in relation to 21,22 and 23 revealed the presence of thin radiopaque contour with no distinction between enamel and dentin and wide pulp chamber giving ghost like appearance irt 21. There was altered morphology of 22 with wide pulp chamber noted. The 23 was rudimentary in its size. (Figure 3). Bases on the history, clinical findings and radiographic examination, the diagnosis of RO was made.

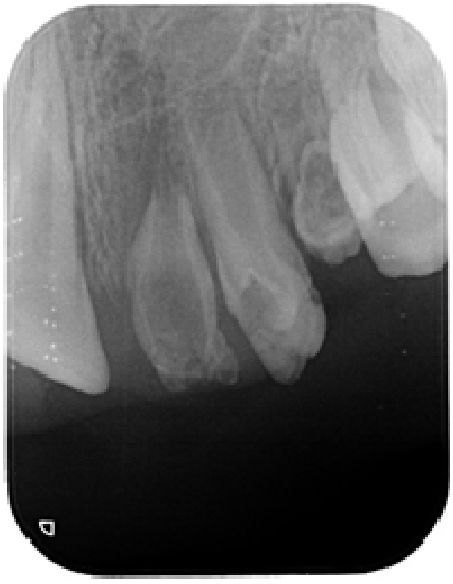

Figure 3. IOPA radiograph showing the teeth with "ghostlike" appearance in maxillary left quadrant and the right permanent central incisor

DISCUSSION

Developmental anomalies are interesting findings, which raise the curiosity of clinicians to seek more knowledge about their origin and nature. Regional odontodysplasia (RO) is a sporadic developmental anomaly that is still ambiguous yet it provokes questions regarding its pathogenesis. The etiology of this dental anomaly is uncertain, although several factors, such as local trauma or infection, teratogenic drugs, local circulatory disorders, Rh incompatibility, irradiation, neural damage, hyperpyrexia, metabolic and nutritional disorders and vitamin.(2) In this case, there was history of local trauma which could be related to RO. The dental follicle calcification is affected in RO. The enamel prisms are generally hypoplastic or hypocalcified with irregular appearance of enamel prisms. The dentin is also poorly mineralized. Coronal part of the dentin is fibrous and contains clefts which could lead to communication between the oral cavity and pulp. Radicular dentin is relatively normal in structure and calcification.(3)

The criteria for diagnosis of RO are generally clinical and radiographic features. The condition affects the dentin and enamel of a group of adjacent teeth. This case presents several common clinical and radiological features related to the regional odontodysplasia. According to the Lustmann et al., teeth in the maxillary arch are affected more frequently by regional odontodysplasia. The patient in this report had "ghost teeth" in the maxillary left region that ended exactly in the midline. Based on the literature, RO has strong female predominance, usually unilateral, rarely crosses the midline and more commonly affects anterior teeth. In most cases, the central and lateral incisors are involved. All these features are seen in our case supporting the diagnosis of RO. In our case there was a swelling in the maxillary left anterior gingiva and delayed eruption of other teeth in accordance with other report**.** In addition, teeth may be affected in different degrees even in the same arch.(4,5) Clinically affected teeth have an abnormal morphology with an irregular contour. The teeth are hypoplastic and hypocalcified, discolored to a yellowish or yellowish-brown. The thin enamel is comparatively soft and susceptible to caries, and is highly susceptible to even mild trauma. Tooth eruption is delayed or does not occur. The most

frequent clinical symptoms after eruption of teeth with RO are gingival swelling, periapical infection, or abscess formation without caries (6). RO-related teeth are often small, discolored, hypocalcified, and hypoplastic, which reflect their abnormal development regarding dental hard tissues originated from the embryonic layers ectoderm and mesoderm (7). The radiographic features have consistently demonstrated thin and defective layers of enamel and dentin, resulting in a faint, fuzzy outline, creating a ghost-like appearance. The pulp chambers and canals are enlarged, and the roots appear short and stubby with open apices. True and false denticles may be seen in the pulp of affected teeth as well as adjacent, clinically normal teeth (8). Treatment of RO has given rise to controversy, the main concern being whether to remove the affected teeth. The treatment depends on the severity of the disorder. Some patients can be successfully treated by conservative restorations or root canal treatment, but the most common method is the extraction of the affected teeth performed in 78.6% of the patients. It is worth noting that in some cases, other possible treatment methods were applied, and in the event of no success, the extraction turned out to be necessary. There are examples of patients who were treated with implantoprosthetics (7.5%). Therefore preserving the teeth is important to promote the development of the bone to make it possible when they reach adulthood.(8)

CONCLUSION

Regional odontodysplasia is a rare developmental anomaly affecting deciduous and permanent teeth. The presentation of this case helps pediatric dentists to review special clinical and radiographic features of RO. Nevertheless, the radiological picture of ghost teeth accompanying clinical discolouration of tooth crowns due to enamel hypoplasia can be considered as a pathognomonic symptom. It should be treated with a multidisciplinary approach to improve the esthetic, psychological, and functional quality of life

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