ODONTOGENESIS IMPERFECTA- ANUNUSUAL CASE

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ABSTRACT

Regional odontodysplasia (RO) is an unusual, non-hereditary anomaly of the dental hard tissues with typical clinical and radiographic features. RO is a rare developmental anomaly involving both mesodermal and ectodermal dental components of teeth. The primary and permanent dentitions of maxilla, mandible or both the jaws are involved. Most of the cases seen in females involving single quadrant of maxillary arch. Here we report a unique case of RO involving maxillary arch crossing midline in a 9-year old boy.

Key words: *Odontodysplasia, Ghost teeth, Development anomaly.*

INTRODUCTION

Regional odontodysplasia (RO) affecting the dental hard tissues is a rare developmental anomaly of unknown etiology, which was first reported by McCall and Wald in 1947.(1) The term 'odontodysplasia' was introduced by Zegarelli et al, in 1963,(2) due to the tendency of this anomaly to occur in a single quadrant, Pindborg added the prefix "Regional" to it.(3) Since then various authors described the anomaly as localized arrested tooth development, ghost-teeth, Odontogenesis Imperfecta, unilateral dental malformation, amelogenesis-imperfecta non-hereditary segmentalis and familialamelodentinal dysplasia.(4) Clinically, RO involves both the dentitions in the maxilla, the mandible or both, most often affecting only one quadrant, but cases with bilateral or multi-quadrant involvement also been reported.(2) Radio graphically the teeth have abnormal morphology and hypo plastic with thin enamel and dentin giving "ghost-like" appearance.(3) Histologically, thin dentin with decreased dentinal tubules in severe cases, wide areas of interglobar and predentin is observed and Pulp shows calcifications.(5) Management of RO is controversial and requires a continuous Multi displinimary approach. The affected teeth are preserved as long as possible to maintain normal jaw development, and affected teeth with abscess should be extracted and replaced with removable appliances.(6) Here we report this rare anomaly affecting the right maxillary arch in a 9-year old boy.

CASE REPORT

A 9-year old boy reported to our department with a chief complain of missing teeth in the right upper back teeth region and yellowish discoloration of upper front teeth. The patient revealed difficulty in speech, mastication and unpleasant appearance due to missing teeth and yellowish discoloration of erupted teeth. There is no history of trauma. History of present illness revealed right maxillary deciduous teeth were yellowish in color, which were lost due to carious lesion. His prenatal, birth, medical and family history was non-contributory. On general physical examination patient was apparently normal. On intra oral clinical examination (Fig.1) revealed caries in relation to 75 and 85, and the overlying mucosa in the edentulous area was normal. Teeth in relation to 21 presented with intrinsic yellowish discoloration. Radiographic evaluation by maxillary anterior occlusal view (Fig.2) and panoramic radiography (Fig.3) revealed maxillary right permanent tooth germs of 11 12 13 14 and 15 with incomplete root formation, had low radio density, thin enamel and dentin with less demarcation between them, open apex in relation to 21 and 22 and with large pulp chambers giving them a typical "Ghost-teeth" like appearance. Based on clinical and radiolographical findings a diagnosis of Regional Odontodysplasia was made with a differential diagnosis of dentin dysplasia, dentinogenesis imperfect, and amelogenesis imperfect. Patient was not willing for any prosthesis and currently under follow up.

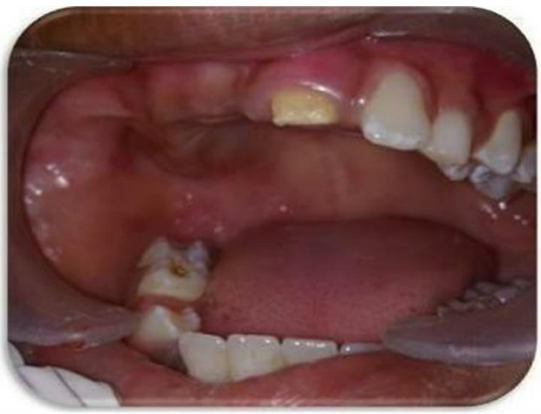

Fig. 1: Intraoral photograph showing the edentulous region on the right side maxilla.

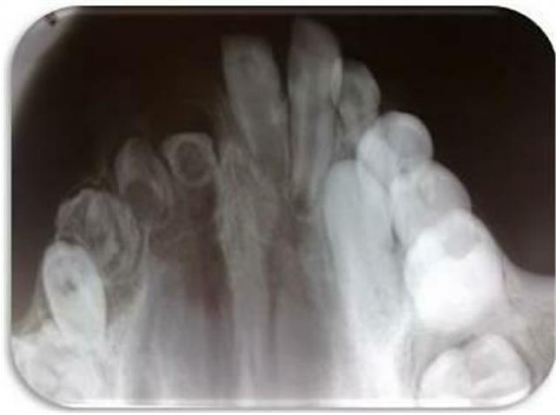

Fig. 2: Maxillary topographic occlusal radiograph view showing the unerupted teeth irt 11 12 13 14 and 15 with thin enamel, dentin and wide pulp chambers.

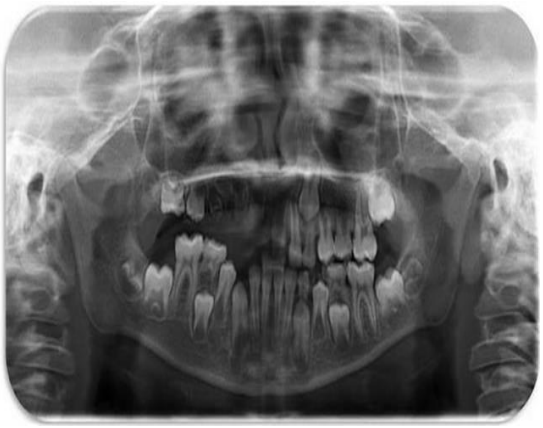

Fig. 3: Orthopantomogram revealed typical "Ghost-like" appearance teeth.

DISCUSSION

RO is a rare, localized developmental anomaly of dental hard tissues. According to International Statistical Classification of Diseases and Related Health Problems (10th Revision Version for 2007), RO is classified as K00.4.[3] Though the etiology of this anomaly is uncertain, multi factorial etiologies like, infections, trauma, vascular disorders, Rh incompatibility, irradiation, nerve damage, hyperpyrexia, metabolic and nutritional disturbances, hereditary.[7,8] It affects maxilla and mandible with a ratio of 2:1, predominantly maxillary left quadrant, more commonly involving the anterior region,[4,9] but in the present case maxillary right quadrant was involved affecting the incisors crossing midline. RO is more prevalent in females than males at a ratio 1.4:1,[7] but in our case the patient was a male. The diagnosis of the condition is primarily based on the clinical and radio graphical findings. Clinically the teeth are morphologically altered with pits and grooves, giving irregular surface contour.[10] The teeth appear to be hypo plastic, hypo calcified, and show yellowish or brownish discoloration with more susceptibility to caries and readily fracture with minute trauma.[4] The affected teeth either erupt late or do not erupt at all, as in the present case few of the affected teeth were not erupted. The radiographic features are uniquely characteristic, with decreased radio density of hard tissue, thickness of enamel and dentin with large pulp chamber giving a ghost-teeth like appearance.[7,11] The diagnosis was confirmed as Regional odontodysplasia, as the present case exhibited many typical clinical and radiographic features of this condition. Similar features can be exhibited in conditions like dentin dysplasia, hypophosphotasia, dentinogenesis imperfect, amelogenesis imperfect an affect the entire dentition.[7, 12] Treatment of RO is controversial and requires a continuous Multi displinimary approach and conservative approach like preserving the affected teeth without infection and pain by giving oral hygiene instructions and antibiotic therapy for a week. If missing teeth are present they should be replaced with acrylic removable appliances to maintain space for normal jaw development, masticatory function, esthetics, prevention of over eruption of opposing teeth, and less the psychological effects of premature tooth loss.[6] According to Cahuana et al in 2005 auto transplantation can be considered as an alternative treatment for RO if suitable donor teeth available.[13]

CONCLUSION

Dentist should be aware of the characteristic clinical and radiographic features of RO in pediatric patients, and it should be considered as one of the differential diagnosis in morphologically altered teeth and anodontia. The treatment of RO should be done depending on the severity and considering the functional and esthetic needs. Maintenance of oral hygiene and periodic checkups are necessary in such cases.

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