Dentigerous cyst associated with multiple complex composite odontomas

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Abstract

Odontomas are considered to be hamartomatous malformations rather than true neoplasms. This most common odontogenic lesion results from the growth of completely differentiated epithelial and mesenchymal cells that give rise to ameloblasts and odontoblasts. Dentigerous cyst is an epithelium-lined sac enclosing the crown of an unerupted tooth. Apparently, the dentigerous cyst arises by the accumulation of fluid between reduced enamel epithelium and the tooth crown. Occasionally it is associated with supernumerary tooth or odontoma. We report a case of dentigerous cyst associated with complex composite odontoma and an impacted lateral incisor in a 30-year-old male patient.

Keywords: Complex composite odontoma, dentigerous cyst, impacted lateral incisor

Introduction

Odontoma, complex type (OC) is a tumor-like malformation (hamartoma) in which enamel, dentin, and pulp, and sometimes cementum, is present in a disorganized form.[1] The etiology of odontoma is unknown but genetic factors and environmental causes such as trauma and infection have been proposed.[2] This developmental anomaly results from the growth of completely differentiated epithelial and mesenchymal cells that give rise to ameloblasts and odontoblasts. Complex odontoma is one of the most common odontogenic lesion. It occurs before the age of 30 with the peak in the second decade of life and - male: female ratio being 1.5:1.[3] Majority of complex odontomas are located in the posterior mandible followed by anterior maxilla.[3] Unerrupted teeth are associated with 10% to 44% of complex odontomas.[3] Overall 17% of impacted permanent maxillary lateral incisors are associated with complex odontomas.[4]

Dentigerous cyst is a developmental odontogenic cyst seen around the crown of unerupted tooth and attached to its neck. Apparently, the dentigerous cyst arises by the accumulation of fluid between reduced enamel epithelium and the tooth crown. Amongst all the jaw cysts its prevalence is 16.6% and the most common site is mandibular third molar region followed by maxillary permanent canine, mandibular premolars, and maxillary third molars. Male: female ratio is 1.84:1. Occasionally, they are associated with supernumerary teeth or odontomas.[5] Dentigerous cysts are seen in conjunction with odontomas in 27.6% of cases.[6]

In the present case, complex composite odontoma associated with an impacted maxillary lateral incisor was an incidental radiographic finding. The lesion was surgically excised along with the soft tissue attached to it. Histopathological examination revealed that the soft tissue was a dentigerous cyst associated with an odontoma. The association of a cyst arising from odontoma and an impacted permanent lateral incisor makes this case interesting.

Case Report

A 30-year-old male patient presented with the chief complaint of missing anterior teeth to the Department of Oral and Maxillofacial Pathology Modern Dental College and Research Centre, Indore. Patient gave history of meeting with an accident one month back in which his upper left central incisor was fractured and the retained part of the tooth was mobile, which was extracted by the dentist on the same day. Intra oral examination revealed missing 21, 22, and lacerated wound, lateral to the extraction socket of 21. Single, intrabony, hard, and tender swelling was present in the anterior part of the hard palate. The swelling was extending from the mesial aspect of 23 to the mesial aspect of 11 with the dimension of 1.5 × 1 × 0.5cm. Small white globular structure was seen within the lacerated wound in 22 region, which was of 0.3 × 0.2 × 0.3cm in size, hard in consistency, smooth textured, and shiny in appearance. IOPA, occlusal view of anterior maxilla, and orthopantomogram were taken and radiographs revealed multiple, small globular to irregular radiopaque structures in 21 and 22 region and was surrounded by a thin radiolucent rim around it. On the

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occlusal side and adjacent to the radiopacities, a small well defined radiolucency was noted of approximately 1 × 0.5cm. The 22 was impacted and placed palatally and horizontally superior to the radiopacities [Figure 1].

Multiple hard tissues were surgically excised with two soft tissues. One bigger piece of hard tissue showed an association of cystic lining. The impacted permanent lateral incisor was also removed as it was horizontally placed [Figure 2].

On histopathological examination, H and E stained decalcified section showed distantly placed small areas of pulpal tissue of variable volume and few disorderly placed enamel spaces surrounded by dentin. Some enamel spaces showed homogenous eosinophilic areas adjacent to them, suggestive of immature enamel. At places, cementum like tissue was admixed with the dentin. One of the hard tissues showed an association of cystic lining of considerable length. The cystic lumen was lined by 3−4 cell layers thick, flat, non keratinized stratified squamous epithelium. Other soft tissue excised from the same area showed similar epithelial features as those seen in the lining attached to the odontoma. Underlying connective tissue capsule was moderately dense fibrocellular, moderately vascular, and showed diffuse mild to moderate chronic inflammatory cell infiltration. Few odontogenic islands were interspersed in the connective tissue capsule [Figures 3 and 4].

Discussion

Complex odontoma is most prevalent in the dental field superceded in frequency only by compound odontoma. These non-aggressive hamartomatous malformations consist of mixed lesions with epithelial and mesenchymal tissues, which show differentiation into odontogenic tissues such as enamel, dentin, cementum and pulp.[2] Two types of odontomas recognized are compound and complex composite. Compound odontomas consist of encapsulated, discrete, small tooth-like structures, or denticles in a fibrous connective tissue stroma,[2] mostly located in anterior maxilla. Complex odontomas on the other hand, consist of an irregular mixture or mass of mature hard and soft dental tissues, which has no resemblance to teeth.[2]

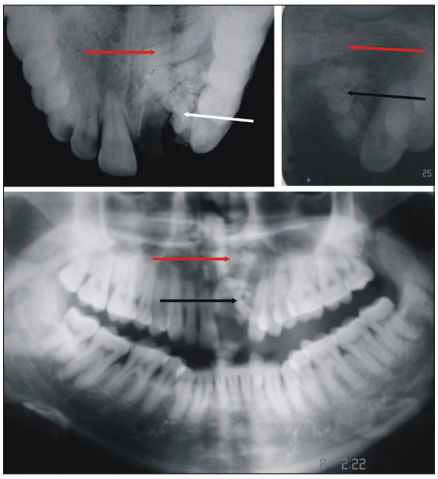

Figure 1: Occlusal radiograph, IOPA, and orthopantomogram show missing left central incisor, horizontally impacted left lateral incisor(red arrow) and multiple radiopaque masses with a thin radiolucent rim(white arrow). IOPA shows radiolucency around and in association with radio-opaque mass.

Despite their unknown etiology, odontomas are usually discovered during the second and third decades of life as an incidental finding as they are asymptomatic.[3] Sometimes it may present as bone expansion, pain, and tooth displacement or show association of unerupted normal teeth. Intra-oral radiographs usually show well-defined borders of a similar density of calcified dental tissue, having a ground-glass appearance, and a radiopaque mass occupying the affected region,[7] surrounded by a thin radiolucent halo. Although they are usually tooth-sized or smaller, the complex variant can occasionally exhibit considerable size and can attain diameter upto 6 cm.[1]

The frequency of complex variant constitutes between 5% to 30 % of all odontogenic tumors.[3] Usually they are located in the posterior mandible and the second most common site is the anterior maxilla.[3]

The present case shows a complex odontoma in anterior maxilla, which is second common site for it and was associated with impacted lateral incisor. When the lesion was exposed surgically the soft tissue lining was attached to the odontoma and not the impacted tooth, which confirms the association of cyst with the odontoma only.

Large odontomas are associated with local disturbances such as the eruption delay of permanent teeth and the development of cystic lesions as dentigerous cysts. These

Figure 2: Multiple hard and soft tissues with impacted lateral incisor. The tissue circled is showing a cystic soft tissue attached to the hard tissue.

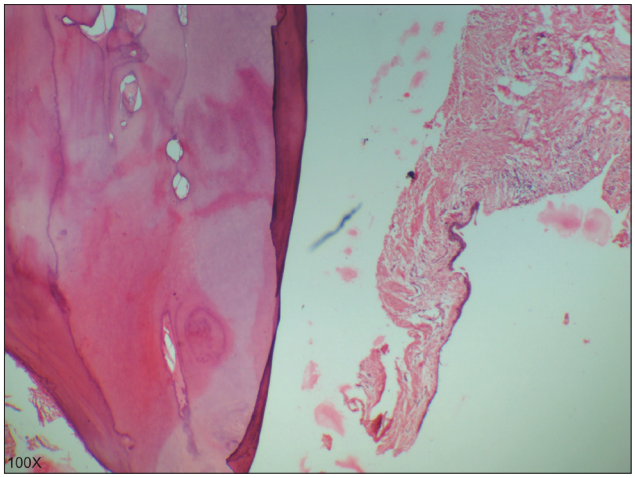

Figure 3: Decalcified H and E-stained section shows haphazard arrangement of enamel space, dentin, pulp, and cementum. A soft tissue adjacent to it showed cystic lining

cases of simultaneous pathologies are uncommon and diagnosis based on the radiographic appearance of such lesions is a challenge to dental professionals because huge lesions with solid features can mimic pathologies such as fibro-osseous lesions, for e.g., fibrous dysplasia, ossifying fibroma. However, wrong diagnosis can lead to serious damage to maxillofacial structures.[8]

In addition to their potential for attaining large size and destroying the jaw bone, these cysts are capable of causing resorption of roots of adjacent teeth with neoplastic changes such as ameloblastoma, mucoepidermoid, and epidermoid carcinomas within isolated segments of the cyst wall. The potential for neoplastic change and invasion beyond the cyst wall justifies complete enucleation of the dentigerous cyst and its histopathological examination.[5]

Odontoma is a common incidental finding when radiographic evaluation is performed and should be carefully analyzed. Kaugars *et al* found that roughly half of all odontomas block the eruption of a tooth. This study demonstrated that 77% (62/81) of odontomas were located on the eruption pathway of a permanent or deciduous tooth. This, in turn, prevented the normal eruption of the involved impacted tooth.[6]

The association of this lesion with the unerupted tooth is high and three-quarters of impacted teeth related to odontomas can erupt after removal of the odontoma. This finding indicates

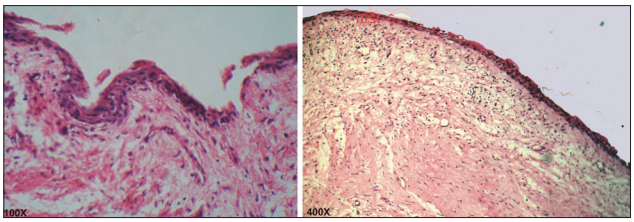

Figure 4: H and E-stained section shows cystic lumen lined by 3–4 layers thick nonkeratinized stratified squamous epithelium

that a careful consideration has to be made before removing the impacted tooth associated with the odontoma.[9]

Another reason for removing odontomas is their association with the dentigerous cysts in significant number of cases (27.6%). Looking at the association of these two lesions and the potential complications associated with the cyst, odontomas should be properly evaluated and removed in case of any suspicion of cystic transformation.

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