Adenomatoid Odontogenic Tumor Associated with Impacted Mandibular Canine - A Case Report

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Abstract

Adenomatoid odontogenic tumor (AOT) is a relatively rare, benign, hamartomatous, and cystic odontogenic neoplasm that was first described more than a century ago. It accounts for 2-7% of all odontogenic tumors. The lesion still continues to intrigue experts with its varied histomorphology and controversies regarding its development. The present article describes a case of cystic AOT with an unusual histomorphology associated with an impacted 43 in a 15-year-old male.

Keywords: Adenomatoid odontogenic tumor, dentigerous cyst, AOT of mandible

Introduction

Adenomatoid odontogenic tumor (AOT) was first described by Steensland in 1905. In 1907, Dreibladt called this lesion a pseudo-adenoameloblastoma.[1] Since then, clinical and histological features of AOT have been well-studied and documented. The World Health Organization (WHO) currently defines AOT as composed of odontogenic epithelium in various histoarchitectural patterns embedded in mature connective tissue stroma, characterized by slow but progressive growth.[2]

AOT is a rare, noninvasive, benign (hamartomatous) epithelial lesion of odontogenic origin and accounts for 2-7% of all odontogenic tumors. AOT usually affects young patients, mostly during their second to third decades of life. Women are affected more frequently than men, and the lesions tend to occur in the anterior maxillary region.[3]

Based on the clinical and radiologic findings, AOT can be mainly divided into two variants:[4]

A. Central (or intra-osseous) variant
    1. Follicular (dentigerous) type: Tumor is associated with the crown of an embedded tooth (accounting for 73% of total cases)[5]
    2. Extra-follicular type: Tumor has no association with the crown of an embedded tooth (accounting 24% of total cases).[5]
B. Peripheral (or extra-osseous) variant

This type can be often misdiagnosed as a gingival fibroma or fibrous epulis (3% of total cases).

Case Report

A 15 year old male patient presented with a chief complaint of swelling in the mandibular anterior region for 20 days. There was moderate swelling in the chin area, which was nontender and had firm consistency with ambiguous margins. There was no history of trauma and he also had no complaint of fluid, blood, or pus discharge from the swelling. There was no relevant medical history of any disease, especially endocrinal disturbances. He was physically and mentally healthy and not under any medication.

On clinical (extraoral) examination, the swelling existed in the lower third of the face extending from the right to the left parasymphyseal region. Superiorly, the swelling was obliterating the mentolabial sulcus and up to the lower border

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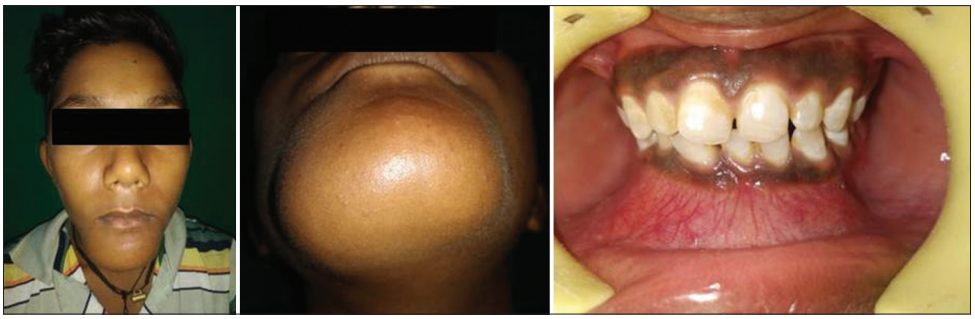

Figure 1: Preoperative clinical photographs

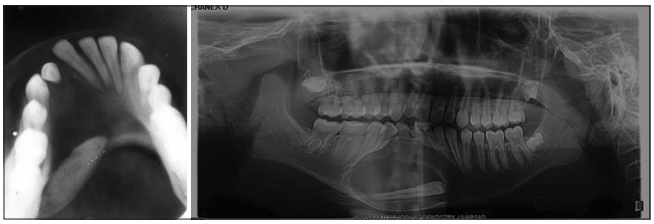

Figure 2: Preoperative radiographs

of the mandible. The overlying skin was normal in color and texture but was stretched. The margins of the swelling were diffuse. On a profile view, there was a slight prognathic appearance [Figure 1]. On palpation, the swelling was firm, nontender, and with a normal temperature.

Intraoral examination revealed an oval, nontender, and firm swelling, measuring 7 cm × 5 cm in size, which was extending from the right second premolar to the left first premolar with buccal and lingual cortical plates expansion. Superiorly, the swelling was at the level of the gingival margins of the anterior teeth. There was no restriction in tongue movements or difficulty in swallowing. The overlying mucosa was nonadherent and with normal color and texture. Severe mesial tilting of all four incisors and mild mesial displacement of left canine and first premolar along with the two right premolars was noticed. The patient had retained his right deciduous canine. No caries was detected. All four anterior teeth were nontender on percussion with Grade I mobility. All findings of inspection were confirmed by palpation.

Mandibular occlusal radiograph revealed a well-circumscribed unilocular radiolucency with radio-opaque border extending from the right second molar to the left first premolar, measuring 7 cm × 5 cm in size with tilting and displacement of some teeth without root resorption. Expansion of the buccal and lingual cortical plates was also observed. There was no perforation of cortical plates [Figure 2].

A digital panoramic view revealed a well-defined unilocular radiolucency of the anterior mandibular region, which contained an impacted right mandibular canine [Figure 2].

The differential diagnosis based on clinical and radiographic findings included a dentigerous cyst, unicystic ameloblastoma, and AOT.

After obtaining written consent, the patient was prepared for surgery under general anesthesia.

Enucleation of cystic lesion was done by giving incision intraorally. Extraction of right deciduous canine, right lower incisors, and left lower incisors was done [Figure 3].

On gross examination, the gross specimen showed one hard tissue (canine) attached with ovoid soft-tissue cystic specimen. Specimen was brownish black in color, measuring from 3.5 cm × 4.0 cm × 3.0 cm. with intraluminal growth, irregular surface, and firm consistency.

Microscopic examination

Hematoxylin and eosin-stained section showed nonkeratinized thin stratified squamous epithelium cell lining showing intraluminal proliferation in the form of sheet strands and rosettes of odontogenic cells with cuboidal-to-columnar shape and hyperchromatic nuclei. Duct-like arrangement of these cells with a central eosinophilic coagulation and basement membrane reduplication was observed. Few foci of calcification and dentinoid were observed. Few areas showed mural proliferations. Underlying connective tissue capsule showed loose collagen fiber bundles with dialed and engorged blood vessels and extravasated RBCs.

Microscopic examination was suggestive of/ compatible with: AOT arising from dentigerous cyst. Figure 4 shows post-operative orthopantomogram after 3 months.

Discussion

The AOT is considered a benign, non-neoplastic (hamartomatous) lesion with slow but progressive growth. It occurs in intraosseous as well as in peripheral forms.[4]

The WHO has described histological features of AOT as follows: "A tumor of odontogenic epithelium with duct like structures and with varying degree of inductive changes in the connective tissue. The tumor may be partly cystic and in some cases solid lesion may be present only as masses in the wall of a large cyst. It is generally believed that lesion is not a neoplasm."

The follicular type of AOT shows a well-defined unilocular radiolucency associated with the crown and often part of the

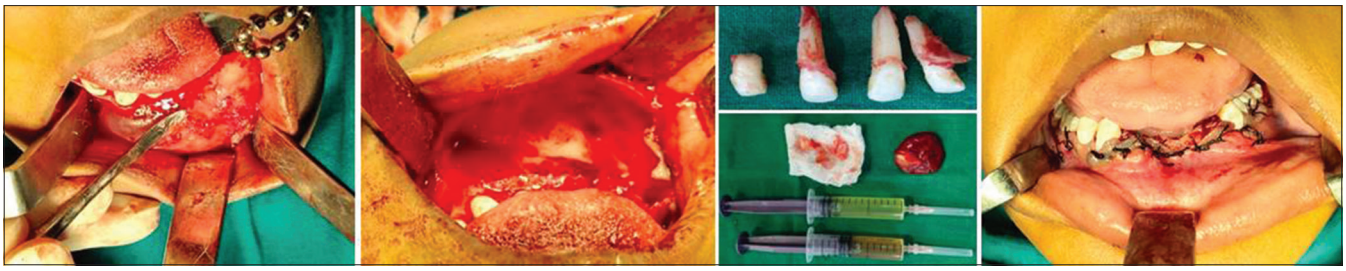

Figure 3: Intraoperative photographs

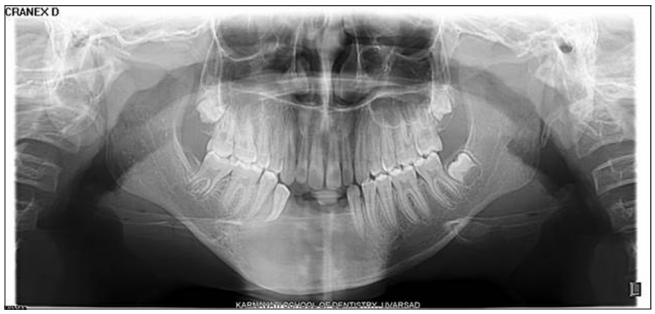

Figure 4: Postoperative radiographs after 3 months

root of an unerupted tooth, thus mimicking a dentigerous or follicular cyst. In fact, 77% of follicular type AOTs are initially diagnosed as dentigerous cysts.[6] Many present as cystic lesions with only mural nodules of AOT lesional tissue, and in some instances, the origin of lesional tissue from the reduced enamel epithelium can be demonstrated histologically.[7] Whether the origin of the follicular variant occurs before or after the cystic expansion has taken place is open to conjecture. If it occurs after cystic expansion, then this effectively means origin from a dentigerous cyst and several such case reports have been published.[8]

The extrafollicular type commonly presents as a well-defined, unilocular radiolucency found between, above, or superimposed on the roots of erupted, permanent teeth, which often leads to a preoperative, tentative diagnosis of a residual, radicular, globulomaxillary, or lateral periodontal cyst depending on actual intraosseous site of lesion.[4,5]

The origin of AOT is controversial. Different hypotheses for the pathogenesis of AOT have been proposed. It could theoretically arise from the enamel organ, epithelial lining of dentigerous cyst, epithelial rests of Malassez of deciduous or permanent teeth, or remnants of the dental lamina and may show an ameloblastic phenotype.[9-18] Some believe that they originate from the odontogenic epithelium of a dentigerous cyst.[7] Although there are arguments both in favor and against origin from a preexisting dentigerous cyst, but no particular conclusion has been deduced.[5,9,12,16,17] Cases like ours may shed more light on the pathogenesis of AOT from a dentigerous cyst or their association with it.

With all possible explanations regarding the development of AOT, more research is required to come to any conclusion regarding their cystic association.

Conclusion

The article presents a rare case of AOT arising from a dentigerous cyst. The case is unusual due to its clinical presentation and important due to its cystic nature and varied histomorphology. On account of the controversies surrounding the lesion, a detailed study of its molecular origin and behavior is suggested.

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Conflicts of interest

There are no conflicts of interest.

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