Adenomatoid Odontogenic Tumor- A Rare Case Report of “Two Third Tumor”

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Authors’ contributions

This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic origin tumor that manifests as a slow-growing cystic neoplasm in the anterior maxilla, often in conjunction with an impacted tooth. AOTs are divided into three kinds based on their histology: follicular, extrafollicular, and peripheral. Because the source of the AOT is unknown, it's impossible to say whether the lining of an associated cyst reflects the cause i.e. a real dentigerous cyst, a cystic alteration within an AOT, or a separate entity. The diagnosis and treatment should be determined following a thorough clinical, radiographic, and histological investigation. The presented case is a rare occurrence of its sort due to the favorable patient’s age and the AOT's site in the lower jaw. The current study reports on a case with follicular AOT in the anterior mandibular region (a rare location), with unusual histomorphology (snow flake and calcified areas) associated with impacted 43 and retained 83.

Keywords: Adenomatoid; dentigerous; adenomatoid odontogenic tumor; impacted; odontogenic cyst.

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ABBREVIATIONS

AOT : Adenomatoidodontogenic tumor
H&E : Hematoxylin and eosin stain
CGCG : Central giant cell granuloma
KOT : Keratocystic odontogenic cyst
CBCT : Cone Bean Computed Tomography
COC : Calcifying odontogenic cyst

1. INTRODUCTION

The most alluring and extremely puzzling hamartomatous malformations, which have been described under a variety of terminologies till the recent years is Adenomatoid Odontogenic Tumor (AOT). Various attempts have been made to give a nomenclature to this entity, but being uncommon distinct odontogenic neoplasm, it was first described by Steensland in 1905 as "epitheliomaadamantinum"[1]. Year after year various terminologies have been discovered like adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, or teratomatous odontoma.

Philipsen and Birn in 1971 proposed a terminology "Adenomatoid Odontogenic Tumour" (AOT) which was widely accepted and later on adopted by World Health Organization (WHO). It was defined as. "A tumour of odontogenic epithelium with duct –like structures and with varying degrees of inductive changes in the connective tissue. The tumour may be partly cystic, and in some cases, the solid lesion may be present only as masses in the wall of a large cyst. According to the most experts, lesion is not thought to be a tumour [2].

It is an uncommon odontogenic tumor with frequency 2.2 to 7.1% with female predilection (F:M = 2.3:1)[3]. It is more commonly seen in anterior maxilla (maxilla: mandible = 2.6:1) in association with an unerupted tooth within second decade of life[3].

There are very few reports of odontogenic tumors either arising from or associated with odontogenic cysts. Such unusual lesions must be carefully diagnosed by an oral physician in order that absolute best treatment needs of the patient are often met. The purpose of this case report is to present a rare case of AOT that originated in the wall of a dentigerous cyst in mandible with an emphasis on radiographic findings and with pathologic correlation.

2. PRESENTATION OF THE CASE

A 20-year-old female reported to the Department of Oral Medicine Diagnosis and Radiology of reputed dental college in North India with chief complaint of swelling on the lower front tooth region from past 4 months. Patient was relatively asymptomatic four months back, when she noticed swelling in front tooth region, which was initially small in size and gradually increased to attain its present size over a period of 4 months. It was associated with dull, intermittent, localized pain without any history of pus discharge. Pain subsided on taking medicines, though the patient was not aware with the details of the same. Medical and personal history of the patient was non-contributory.

On extra oral examination, mild facial asymmetry was evident, with the solitary diffused swelling measuring around 2.5x5cm at right parasympysis region extending supero-inferiorly from the corner of lip till the inferior border of mandible and antero-posteriorly from right mid-pupillary region till the 2 cm ahead of midline. On palpation all inspector findings were confirmed. Swelling was tender with soft to firm consistency, non-fluctuant and non compressible.

On intra oral examination, a well-defined solitary swelling was evident, with the solitary diffused swelling measuring approximately 2.0 x 1.3 cm in size at right mandibular vestibular region extending antero-posteriorly from midline to themesial aspect of 44 and supero-inferiorly from approx. 0.5 cm below cervical margin of 31 41 42 83 44 involving attached gingiva, with obliteration of labial vestibule. Overlying mucosa was bluish in color. On palpation all the inspector findings were confirmed.

On hard tissue examination, grade I mobility of 41, 42 with retained 83 having grade II mobility and 43 missing was evident. Pulp vitality test was performed and it was observed that 31 32 were vital and 41 42 83 were non vital tooth.

So, after the clinical examination and based on the younger age, being well defined, slow growing swelling and association with tooth the clinical provisional diagnosis was given as benign odontogenic cyst /tumor of right mandibular anterior region and the clinical differential diagnosis was given as Dentigerous cyst, Adenomatoid odontogenic tumor, Central giant cell granuloma (CGCG) and Radicular cyst.
Radiographic investigations were advised to the patient and on Intraoral periapical radiograph a well-defined radiolucency with impacted tooth resembling morphology of canine was appreciated. Further mandibular occlusal radiograph and OPG was advised. On mandibular occlusal radiograph, a well-defined circumscribed, corticated expansile radiolucent lesion extending from 31 to 44 teeth with marked labial and mild lingual cortical plate expansion with no discontinuity was appreciated.

OPG revealed an osteolytic well defined radiolucent lesion in the right mandibular region with corticated margins extending antero-posteriorly from mesial aspect of 31 tooth region till periapex of 44 and S1 from the alveolar crest till 1.5 cm above the inferior border of mandible. The internal structure is radiolucent with radiopaque structure resembling impacted tooth i.e., 43. There was evidence of retained tooth with respect to 83 along with the displacement of the roots with respect to 41 42 31 44.

After initial 2-D radiographic investigation, patient was advised CBCT to rule out the exact extent of the lesion and its proximity with the adjacent structures. On CBCT scan, a well-defined hypodensity was evident in sagittal section extending from distal aspect of 31 till the periapex of 44 teeth with the impacted 43. No root resorption was appreciated with adjacent tooth. On this axial section, there was evidence of thinning of labial-lingual cortices with marked labial cortical expansion with mild thinning and mild expansion of lingual cortical plates.

The internal structure of lesion is mixed type with corticated margins, with the area of multiple, discrete radio opaque flecks (snowflakes) which were dispersed in mid apical alveolus region.

On this sagittal section, the hyperdense flecks are noticed throughout the lesion but were more profound at the level of mid third root of the impacted tooth. On further localization, there was evidence of attachment of cystic lining apical to CEJ. So, based upon all the clinical and radiographic findings, the final clinicoradiographic diagnosis was given as Adenomatoid Odontogenic Tumor with respect to right mandibular anterior region.

After clinical examination and radiographic investigations, aspiration was performed with 18-gauge needle which revealed clear straw-colored fluid.

The patient was referred to Department of Oral and maxillofacial surgery for biopsy and further treatment. Incisional biopsy was performed with decompression under local anesthesia and multiple tissue bits, creamish brown in color and firm in consistency was sent to department of oral pathology and microbiology for histopathological correlation.

On histopathological examination a cystic cavity lined by thin nonkeratinized stratified squamous epithelium resembling decreased enamel epithelium and a tumor in the capsule was identified. The connective tissue showed distinct tumor tissue arranged in different patterns like ductlike spaces, rosettelike pattern ducts, cribriform lace like areas. Areas of calcification were seen within the epithelium. Hence, histopathological examination of the lesion confirmed the diagnosis of AOT. So, after the confirmation further treatment was planned with the enucleation of the lesion along with extraction of 83 and orthodontic extrusions of 43.

3. DISCUSSION
AOT is a slow growing lesion, constituting only 3% of all odontogenic tumors with a predilection for the anterior maxilla usually associated with impacted canine among young females in the second decade of life [4].

AOT is also known as “tumor of two-thirds” as its incidence is two-thirds cases in the maxilla, of all cases 2/3 females are involved and in 2/3 cases it is associated with the unerupted tooth[5]. In the present case, the patient was a female in the second decade with an impacted canine. The reported case is an unusual occurrence of this tumour entity due to the patient's age and the location of the AOT in the lower jaw (anterior mandible). However, some investigators like, Lang MJ [6], and Fernandez et al.[7] had reported a slight predilection for the mandible in their subjects which was concurrence with our case.

The lesion is asymptomatic but may result in pain due to cortical expansion which is contributory to the diagnosis. Usually, AOT is associated with an impacted tooth which is enclosed by the lesion with the displacement of adjacent teeth which is similar to the case discussed above with no root resorption [5,8].

This lesion appears as a corticated radiolucency with tiny radiopacities on radiographs and frequently surrounds an unerupted tooth. Unlike
Table 1. The clinical and radiographic differential diagnosis of AOT i.e. dentigerous cyst, central giant cell granuloma, radicular cyst, calcifying epithelial odontogenic tumor and calcifying odontogenic cyst has been described

| S. No. | Lesion                                | Clinical Features                                                                 | Radiological Features                                                                 |
|--------|---------------------------------------|----------------------------------------------------------------------------------|--------------------------------------------------------------------------------------|
| 1      | Central giant cell granuloma          | Seen in people <20 years of age as a slow growing painless swelling. Commonly seen anterior to 1st molar in mandible and anterior to canine in maxilla. In mandible, it usually crosses the midline. | Multilocular lesion with thin wispy septa. There is no evidence of cortication in most of the cases. |
| 2      | Dentigerous cyst                      | Mostly in 2nd decade of life and common in mandibular molar followed by maxillary canine. It is usually associated with missing/unerupted tooth. | Well-defined unilocular radiolucent lesion with corticated or sclerotic margins and the radiolucency is attached to the cementoenamel junction of impacted tooth. |
| 3      | Keratocystic odontogenic tumor (KOT)  | Mostly in 2nd and 3rd decade with slight male predominance, mild swelling and usually posterior to canine in mandible (90% of the cases). Lesions involving anterior mandible crosses the midline. | Well-defined unilocular or multilocular radiolucent lesion with scalloped and corticated margins. Septa are coarse and curved. |
| 4      | Adenomatoid odontogenic tumor         | Common in 2nd decade, slowly enlarging painless swelling in canine premolar region of both the jaws, usually associated with impacted tooth. | Well-defined unilocular radiolucent lesion with corticated or sclerotic border. Internal structure may vary from completely radiolucent to faint radiopaque foci. |
| 5      | Calcifying odontogenic cyst (COC)     | Mostly seen in 2nd to 4th decade, equal propensity for male and female. Seen in both Maxilla & Mandible | Well-defined margins. Internal structure appears as mixed lesion with radio opaque flecks. |

Fig. 1. Intraoral photograph showing a swelling in the right side
Fig. 2. Orthopantomogram reveals a well-defined radiolucency with an impacted tooth.

Fig. 3. CBCT images in sagittal & axial section. (a) Sagittal section reveals a hypodensity from distal aspect of 31 till the periapex of 44 tooth and impacted 43. (b) Axial section depicts marked labial-cortical expansion along with thinning of labial-lingual cortices with internal structure as a mixed type, with the area of multiple discrete radioopaque flecks (snowflakes). (c) Sagittal section reveals radioopaque flecks at the level of mid-apical root of impacted tooth i.e. 43 along with the cystic attachment apical to CEJ.

Fig. 4. Operative pictures (a) During Incision (b) Removal of the lesion and cystic lining (c) Drainage tube in situ.

the dentigerous cyst, which does not encircle the roots, an AOT frequently appears to encircle both the crown and the root [4].

In the present case, the lesion was seen as well-defined unicocular radiolucency with corticated margins and the lesion was enveloping the crown and root of impacted canine with no root resorption. Usually, AOT is associated with the impacted tooth which is enclosed by the lesion along with the displacement of the adjacent tooth which was present in above case.

The diagnosis of AOT should be considered in differential diagnosis of corticated radiolucency with small radiopaque foci, especially in...
teenagers and young adults associated with an impacted tooth. If there are no evidence of radiopaque flecks it may be considered as dentigerous cyst [2].

In the present case the internal structure of the lesion on CBCT sections, showed mixed lesion with the areas of multiple radio-opaque flecks in mid apical alveolus region.

There are three variants of AOT. They are: (i) follicular type (73%); (ii) extrafollicular type (24%); and (iii) peripheral type[2,4]. AOTs are usually solid, but occasionally cystic. Because neoplastic and hamartomatous lesions can occur at any stage of odontogenesis, odontogenic tumors with combined features of epithelial and mesenchymal components may arise within the odontogenic cyst. It is unclear whether the associated cyst's lining is a real dentigerous cyst, a cystic alteration within an AOT, or a separate entity. It is also unclear whether this entity has the ability to be more aggressive[3].

Follicular intraosseous AOT is a central intraosseous lesion associated with an impacted tooth, but extrafollicular intraosseous AOT has no connection to an unerupted tooth [9].

The follicular type is characterized by a smooth, spherical radiolucent lesion with an impacted tooth. It is possible that it will be misdiagnosed as a dentigerous cyst at first. Approximately 77% of the follicular variant is provisionally seen to be diagnosed as dentigerous cysts [5].

The follicular variety of AOT is characterised by a well-defined unilocular radiolucency associated with the crown and often a portion of the root of an unerupted tooth, which might be mistaken for a dentigerous or follicular cyst. In fact, follicular type AOT is initially identified as dentigerous cysts in 77% of cases. It is unclear if the follicular variation appears before or after the cystic growth. If it occurs following cystic enlargement, it is almost certainly the result of a dentigerous cyst, as evidenced by multiple case reports [10].

Fig. 5. Tumor cells are arranged in varied histopathological patterns under hematoxylin and eosin stain (H&E)

Fig. 6. Eosinophilic globules/ tumor droplets showing small foci of calcification under hematoxylin and eosin stain (H&E)
The extrafollicular type commonly manifests as a well-defined, unilocular radiolucency between, above, or superimposed upon the roots of erupted, permanent teeth, leading to the preoperative, tentative diagnosis of a residual, radicular, globulo-maxillary, or lateral periodontal cyst, depending on the lesion's intraosseous location. A solid tumour mass or one big or several tiny cystic cavities containing a yellowish, semi-solid substance with an unerupted tooth implanted in the tumour mass or extending into a cystic cavity may be visible on the cut surface [10].

The histopathology of the present case was very interesting and unique with epithelial lining resembling reduced enamel-like epithelium. Majority of tumor areas shows proliferating epithelial lining as nodular masses. It has a lobular architecture with varied appearance: Pseudo-ductal areas, Rosette formation and cribriform lace like patterns. Tubular or pseudo-ductal spaces lined by single palisaded layer of cubical to low columnar cells with ovoid nuclei polarized away from luminal surface.

AOT usually appears as a well-encapsulated mass with a solid mass in the center or a tiny cyst-like area. The lesion has been explored for marsupialization in an attempt to allow an impacted tooth to erupt, but the tooth is totally encapsulated in the lumen of the lesion since it arises from Hertwig's epithelial root sheath. As a result, surgery becomes impossible since the impacted tooth has no or little radicular bone support. Low recurrence rate of 0.2% has been associated with AOTs in the literature [11]. Enucleation with peripheral ostectomy is effective in treating these lesions because of their low recurrence rate, fibrous encapsulation, and benign nature [5,12].

Similarly in the present case, lesion is encapsulated and hence recurrence is very rare and enucleation is sufficient to treat the case. However, regular follow-up is necessary. For this patient enucleation of the lesion along with extraction of 83rd and orthodontic extrusion of 43 was performed. Postoperative follow up after 2 weeks till 12 months revealed no sign of recurrence.

4. CONCLUSION

As illustrated in the present case, AOT is often mistaken as dentigerous cyst radiologically as well as histopathologically. This case report described a particular case of AOT occurring in an unusual location on the anterior mandibular region which tried to give insight on a variety of clinical and radiological AOT presentations. It emphasizes the overlapping radiographic appearances that a diagnostician should be aware of while identifying more prevalent odontogenic lesions in routine dental examination.

CONSENT AND ETHICAL APPROVAL

As per international standard or university standard guideline Patient's consent and ethical approval has been collected and preserved by the authors.

COMPETING INTERESTS

Authors have declared that no competing interests exist.

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