



## Dentigerous cyst associated with Adenomatoid Odontogenic tumor (AOT)- A Rare Case Report and Review of Literature

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### Abstract

Adenomatoid Odontogenic tumor (AOT) is a benign (hamartomatous) lesion of Odontogenic origin, which is relatively uncommon and affects young individuals with a female predominance, mainly in the second decade. This lesion is most commonly located in the anterior maxilla and is usually associated with an impacted canine tooth. The present case report is a 15 year old female patient, who presented with a large AOT in the anterior mandible associated with an impacted canine – a very rare situation.

**Keywords:** Adenomatoid Odontogenic Tumor, Dentigerous cyst, Impacted tooth

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### Background

Adenomatoid Odontogenic tumor (AOT), an uncommon benign epithelial lesion of odontogenic origin was first described by Dreivaldt in 1907 as a pseudoameloblastoma [1, 2]. Different terminologies such as adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinum or teratoma odontoma have been used before to define the lesion which is currently known as AOT, accepted in the first WHO classification of odontogenic tumors in 1971 [3]. The term AOT seems to be the most appropriate, as these tumors unlike the ameloblastoma, is benign and present in a very low recurrence, making it unnecessary to carry out

extensive and aggressive surgery. [4]. The surgical management of this lesion would be enucleation along with removal of the associated impacted tooth. There are three variants of AOT, the

follicular type (73%), extra follicular type (24%) and the peripheral type (3%) [4-6]. AOT is a tumor of odontogenic epithelium having duct like structures which may be partly cystic, and in some cases the solid lesion may be present only as masses in the wall of a large cyst [7]. Dentigerous cysts are odontogenic and arising in the jaws [8]. This is a case report of dentigerous cyst associated with AOT which is a rare occurrence.

### Case report

A 15 year old female patient reported to the dental clinic with a chief complaint of swelling in the lower left front region of the jaw since 2 months. History of intermittent pain was present in that region. No significant medical and dental history. A dental examination revealed, all vital signs were within normal limits. Patient was afebrile. Extra orally, no gross asymmetry was seen and no lymph nodes were palpable (Fig 1).

Intraorally, Grade I mobility with 42,41,31,32,33,34,35 and Grade II mobility with 73 was present. On inspection, swelling

was extending from 42 to 35 region. On palpation, swelling was firm in consistency, tender and was obliterating the labial vestibule. All hematological investigations were within normal limits.



**Figure-1:** A case of 15years female patient with swelling in the lower left front region of the jaw.

Radiographically, radiolucency involving the anterior region of the lower jaw with impacted canine was seen extending upto the lower border of the mandible(Fig 2).



**Figure-2:** Orthopantomogram shows radiolucency involving the anterior region of the lower jaw with impacted canine.

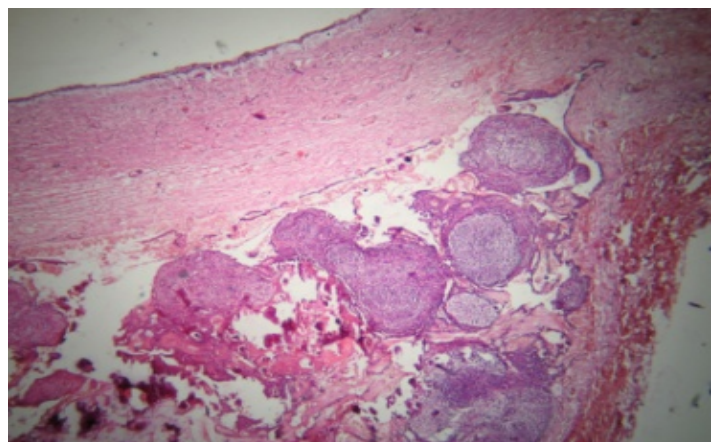
Provisional diagnosis was given as Dentigerous cyst. Excisional biopsy of the lesion was taken. Grossly, two bottles of specimen were received, one containing single soft tissue specimen, soft in consistency, bluish in colour, measuring about 3x2x2cm<sup>3</sup> and

another bottle containing surrounding hard tissue measuring about 3.5x1.5cm<sup>2</sup>(Fig 3).



**Figure-3:** Gross specimen is soft tissue attached to the tooth, which is soft in consistency, blackish blue in colour, measuring about 3x2x2cm<sup>3</sup>

Histopathological examination revealed, a cystic cavity lined by thin nonkeratinized stratified squamous epithelium. Connective tissue capsule showed presence of encapsulated tumor mass. Odontogenic cells were arranged in the form of sheets, follicles, ducts and rosette pattern. Odontogenic follicles lined by tall columnar ameloblast- like cells and centrally placed stellate reticulum- like cells were seen. Tumor mass consisted of odontogenic islands and calcified bodies. Blood vessels and chronic inflammatory cells were seen (Figure 4).



**Figure-4:** Histological section under low power objective(10X) showing a cystic cavity lined by thin non-keratinized stratified squamous epithelium and connective tissue capsule

encapsulating the tumor mass containing odontogenic follicles and calcified bodies.

Overall features were suggestive of “Dentigerous Cyst With Adenomatoid Odontogenic tumor”.

Patient was followed up after 3 months, during which the intraoral healing was normal & advised for longer follow-up periods for prosthetic & orthodontic rehabilitation.

## Discussion

The specialty of a dentigerous cyst is encasement of the crown of an unerupted tooth by expansion of its follicle, and is attached to its neck. Mandibular posterior region is affected more and occurrence is in 2nd or 3rd decade of life. Males are more prone compared with females with a ratio of 1.6:1. Radiographically or as clinical expansion this condition can be detected with unilocular radiolucency associated with the crown of an unerupted tooth [8]. Histologically, it is usually composed of a thin connective tissue wall with a thin layer of stratified squamous epithelium lining the lumen. If there is any secondary infection, Rete peg formation takes place. The connective tissue wall is frequently quite thickened which is composed of a very loose fibrous connective tissue or of a sparsely collagenized myxomatous tissue. Varying numbers of islands of odontogenic epithelium is histological characteristics [6].

Adenomatoid odontogenic tumor is an uncommon tumor (3%), and 0.1% of tumors of jaws, occurs mostly in females (64%) in association with unerupted maxillary cuspid, with a predilection for the anterior maxilla (ratio 2:1 relative to mandible) developing anterior to the cuspid. Mean age of occurrence of the tumor is 20 years. Usually, the tumors do not exceed 1-3 cm in greatest diameter [6, 9-11].

Earlier studies mentioning this clinical condition asymptomatic, but in this case we found associated with cortical expansion. The involved teeth are commonly impacted and adjacent teeth may be slightly displaced, root resorption is not a usual feature. Most commonly canine is impacted 40% [12, 13].

This uncommon tumor which occurs even less frequently than the comparing with odontoma, cementoma, myxoma and ameloblastoma this clinical condition occurs rarely and considered a hamartoma rather than a neoplasm [5]. Histologically, the tumor is made up of polyhedral or even spindle-shaped epithelial cells, usually with only a scanty stroma of connective tissue. However, an AOT often appears to envelop the crown as well as the root; unlike the dentigerous cyst which does not envelop the roots [9, 14, 15].

Pathogenesis of AOT is controversial. Santos et al, reported a case of AOT being developed in the fibrous capsule of the dentigerous cyst. Garcia Pola et al, described the proliferation of an AOT in the epithelial border of a dentigerous cyst where as Cassiano Francisco Weege et al, reported a case of AOT associated with dentigerous cyst [4, 16, 17]. The tumor in association with a dentigerous cyst is reported to occur in the anterior maxilla as in our case and other areas of the jaw such as the angle of the mandible. Rick et al have reviewed cases of AOT in association with dentigerous cysts and stated that although most central AOTs occur in a pericoronal relationship with an associated tooth, there is no way to be certain whether the lining of an associated cyst represents a true dentigerous cyst or a secondary cystic change within the AOT [18].

In this case, AOT and dentigerous cyst are found in the same lesion. All the earlier cases of AOT associated dentigerous cyst showed radiolucency. Maxilla and maxillary sinus are the common area of occurrence and mainly affecting age group between 8-25 years. Our case also supporting this truth [19,20]. However, our patient is a female and most of the above lesions have occurred in male and it completely differs as none of the above have manifested in the mandible, making this case truly rare. Some other authors proposed that the epithelial lining of the odontogenic cyst may transform into an odontogenic neoplasm-like ameloblastoma or AOT [17, 21, 22 ].

## Conclusion

In summary AOT in the anterior mandible associated with an impacted canine is relatively rare condition. There is slow growth and clear delimitation, low recurrence rate, the treatment option is enucleation and simple curettage or in exceptional cases of large tumors or risk of bone fracture, partial resection, en bloc of the mandible or maxilla was recommended. In the present case, the cyst was associated with adenomatoid odontogenic tumor of the 15 years old female patient. These findings are not common and hence, this case is reported.

## Competing interests

Authors do not have any competing interests.

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