ABSTRACT
Asymptomatic, slow growing lesions are common in maxillofacial region especially in the mandible. These lesions are of odontogenic origin and become evident on routine radiological or clinical investigations. Proper treatment planning is very important to save associated teeth and related structures. The purpose of this article is to present a case of a well defined unilocular lesion in canine-premolar region of left mandible and its management.

Key Words: Unilocular, Asymptomatic, AOT, Enucleation, Mandible

INTRODUCTION
Non tender, painless, expansile lesions are common in the maxillofacial region.[1] Cysts and tumours of odontogenic and non odontogenic origin are early diagnosed due to noticeable facial asymmetry. Adenomatoid Odontogenic Tumour (AOT) is an uncommon odontogenic tumour, accounting for 3 to 7% of all odontogenic tumours. The lesion was once believed to be a variant of ameloblastoma and was previously designated as Adenoameloblastoma.[2] In about 75% of cases, the lesion appears as a well-circumscribed, unilocular radiolucency involving the crown of an unerupted tooth, frequently a canine. Being an encapsulated tumour, it separates easily from the surrounding bone. Enucleation is the treatment of choice. Rare cases of recurrence are reported in previous literature.[3]

This article reports a case of extrafollicular variety of Adenomatoid Odontogenic Tumour (AOT) in an unusual location of mandible (canine-premolar region) and its management.

CASE REPORT
A 19 year old female was referred from a private practitioner with the chief complaint of asymptomatic swelling on left side of her lower face since three months. Patient did not give any history of trauma and toothache. Her medical history was non contributory.
On palpation of the buccal vestibule, a diffuse, round, bony, non-tender swelling was present in relation to the left mandibular canine-premolar region. The periodontium was normal with no visible sinus or fistula. There was absence of any carious, missing or impacted tooth. Aspiration was negative of any cystic fluid or blood. Panoramic radiograph was advised which revealed a well-defined radiolucent lesion of 2cm x 1cm located between the left mandibular canine and 1st premolar (Figure 1) with displaced roots without any sign of resorption.

After thorough clinical and radiological examination, the lesion was diagnosed as being benign without any vascular malformations. Surgery was planned under local anesthesia for complete removal of the lesion. After ensuring complete anesthesia, crevicular incision and releasing incision anteriorly and posterior to mental foramen was given. Flap was reflected exposing thin egg shell like bone. Bone was removed and the whole tumour mass (size 2x1 cm) was enucleated (Figure 2 & 3). On gross examination, the lesion was round, firm in consistency, covered with a thick fibrous capsule. When bisected centrally it was solid.

The bony cavity was irrigated with normal saline, curettaged and then sutured. (Figure 3)

The specimen (Figure 4) was put into 10% formalin solution and sent for histopathological examination. On seventh post operative day, sutures were removed. The post-operative healing was satisfactory.

**HISTOPATHOLOGY**

Microscopically, ovoid to spindle shaped epithelial cells forming sheets and whorled masses in fibrous stroma were seen. Dystrophic calcification was also present.

**DIAGNOSIS**

From clinical, radiological and histopathological report the lesion was diagnosed as extra-Follicular Adenomatoid Odontogenic Tumour.
DISCUSSION

AOT is an uncommon benign, slow growing lesion of odontogenic origin characterised by the presence of an unusual duct or gland like structure. In 1907, Dreibaldt called it as 'Pseudo-adenoameloblastoma'. Later in 1909 a case of “Epithelial Odontome” was described by James and Fordes in England. Herbitz called it as 'Adamentoma'. Stafne in 1948 first reported a case series of AOT like lesions in the name of 'Epithelial tumour associated with cyst of maxilla'. Bernier and Tiecke reported a case series under the name of “Ameloblastic adenomatoid tumour”. The term AOT for the first time was described by Phillipson and Birm in 1969. In 2005, Marx and Stern considered it as a cyst with hamartomatous intraluminal proliferation of epithelial cells derived from Hertwig epithelial root sheath but this terminology is not accepted in the literature till date. 

It comprises 3-7% of all odontogenic tumours and its predilection for young age is well established. It is also called as “two third tumour” because about two third of it occurs in maxilla, two third of it occur in young women, two third occurred with unerupted tooth and mostly the canine. 

Its clinical emergence may be subtle and can be discovered in routine radiological investigation as an incident finding. Mobile teeth, asymmetry of face and swelling are less frequent but can be seen if lesion is large. Peripherally, these are mostly seen in maxilla as soft, gingival coloured, asymptomatic painless lesions. 95 % of these lesions are centrally located. Grossly, the tumour is soft covered with a fibrous capsule. On sectioning, it reveals solid mass with some cystic spaces, calcified masses, yellowish to brown fluid and semisolid material. Crown of the tooth seems to be embedded in to solid tumour or projecting into cystic cavity. 

Phillipsen described the three varieties of AOT according to radiological and clinical finding as Follicular, associated with the crown of an unerupted tooth; Extrafollicular, never associated with tooth but sometimes superimpose the roots of teeth and Peripheral which occurs in gingival tissue. Follicular and extrafollicular are osseous tumours formed approximately in 97 % of cases and Follicular alone account for 73% cases. 

Radiologically – These lesions are mostly pericoronal or juxtracoronal and are seen as well demarcated unilocular radiolucency with smooth cortical borders but sometimes radiopaque foci can be seen. Rare multilocular cases have been reported in literature.

Microscopic examination show a cellular multinodular proliferation of spindle, cuboidal and columnar cells in a variety of patterns (Figure 5). It has droplets of eosinophilic material composed of heterogenous fibrils and calcified material. This material is positive for Periodic Acid Schiff and appears to replicate basal cell lamina. Varying numbers of duct like striations are distinctive feature of AOT and are mostly lined by an eosinophillc rim of varying thickness. Liesegang rings may be present and calcification resembles CEOT. 

These lesions are benign unilocular, mostly capsulated and small in size. The typical treatment option is enucleation of whole lesion and curettage of the cavity. Recurrence is rarely seen.

CONCLUSION

AOT is a benign, slow growing, painless tumour with rare reoccurrence rate. Enucleation of whole lesion is the treatment of choice because of its benign nature.

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