

Case Report

Adenomatoid Odontogenic Tumour: Immunohistochemical Study: A Rare Case Report and Review of Literature

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ABSTRACT

Adenomatoid odontogenic tumour (AOT) is a rare odontogenic tumour which is often misdiagnosed as odontogenic cyst. To acquire additional information about AOT, all reports regarding AOT and articles cited in "Pubmed" since 1990 onwards were reviewed. AOT accounts for about 1% to 9% of the odontogenic tumours. It is predominantly found in young female patients and located more often in the maxilla. In most cases it is associated with an unerupted permanent tooth. AOT is characterised by positive stain with cytokeratin (CK). Treatment is conservative and prognosis is excellent. Here, we report a case of AOT-follicular variant in the anterior region of maxilla in a 40-year-old woman. The present case was histopathologically proven and tumour exhibited positivity with per iodidic Acid stain (PAS) and Cytokeratin.

Key words: Adenomatoid Odontogenic Tumour (AOT), histopathology, cytokeratin (CK) and per iodidic Acid Stain (PAS).

INTRODUCTION

Adenomatoid Odontogenic Tumour (AOT) is a relatively uncommon distinct Odontogenic neoplasm that was first described by Steensland in 1905. ⁽¹⁾ However, a variety of terminologies like adenoameloblastoma, adenoameloblastoma odontoma, and epithelial tumour associated with ameloblastic adenomatoid tumour, developmental cysts, and Adenomatoid or pseudo Adenomatous ameloblastoma have been used to designate this extremely fascinating entity. ⁽²⁾ There are 3 variants of Adenomatoid odontogenic tumour - follicular type; (accounting for 73% of cases which has a central lesion associated with an embedded tooth), the extra follicular type (24% of cases which has a central lesion and no

connection with the tooth and the peripheral variety (3% of cases). ⁽³⁾ The WHO histological typing of odontogenic tumours, jaw cyst and allied lesions (2005) has defined AOT as a tumour of odontogenic epithelium with duct-like structures and with varying degree of inductive changes in the connective tissue. ⁽⁴⁾ The tumour is readily enucleated without risk of recurrence. ⁽⁵⁾

CASE REPORT

A 40-year-old female, native of Madurai, Tamil Nadu, reported to the department of ortho odontology with complaints of swelling in the right upper anterior part of maxilla for four months duration. Initially the swelling was small and gradually increased in size. It was not

associated with any pain or discharge. Extra oral examination was unremarkable. On intra oral examination, a solitary diffuse cystic swelling was present on the right anterior part of maxilla in relation to 12 and 13. The swelling was 2X1 cm in size. It was soft in consistency and not tender. Intraoral periapical radiograph showed well defined radiolucency with an unerupted tooth (figure 1)



Figure 1: Intra oral periapical radio graph showing unilocular radiolucency.

Periapical cyst in relation to 12 was the provisional diagnosis. Surgical excision was done and the specimen was received in formalin fixative in our histopathology lab for histopathological examination. Grossly, external appearance showed cystic mass measuring 2.5cm X 1 cm and on cut surface showed an encapsulated cyst, unilocular containing straw coloured fluid. (Figure 2A).

On histopathological examination, the tumour was well encapsulated. The thickened wall showed proliferation of epithelium forming solid masses (figure 2B) composed of whorled nodules of spindles cells and columnar cells with polarized nuclei forming rosettes and duct like tubular structures (figure 2C). Eosinophilic amorphous material was also found within and between the tumour cells as well as in duct like structures. (figure 2D). At the intercellular level there were areas of irregular hyaline and amorphous deposits, PAS +ve diastase resistant. (Figure 3 A & B)

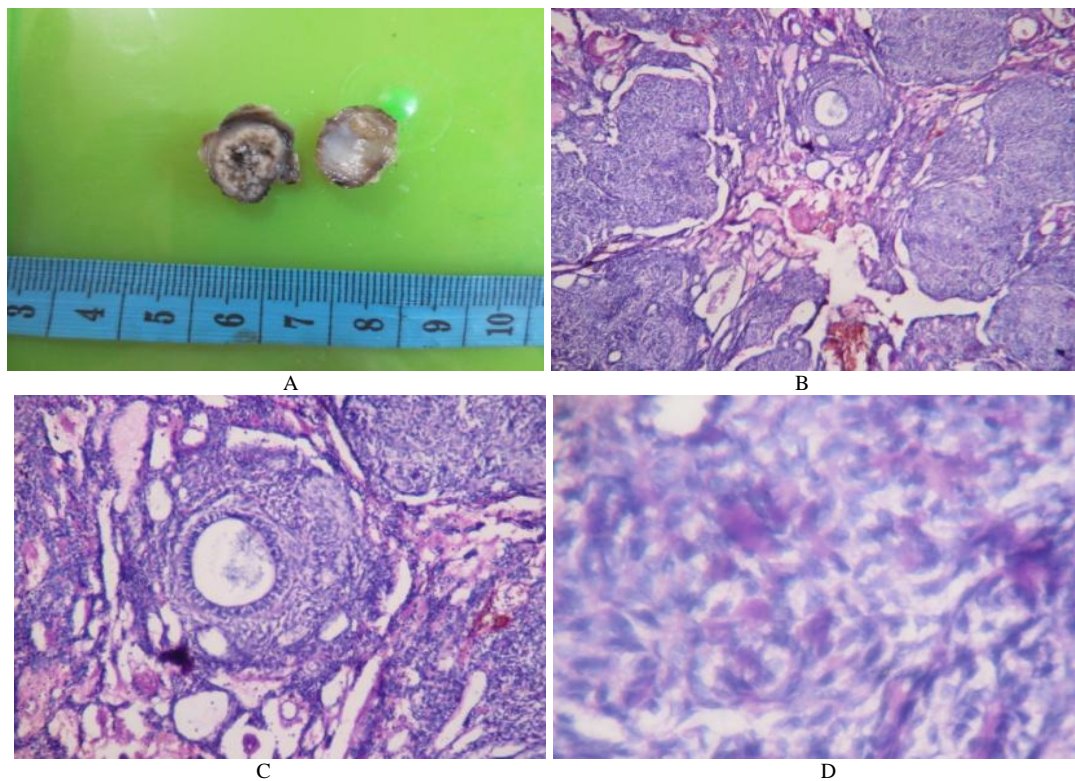


Figure 2 : A.Cut surface shows an encapsulated ,unilocular cystic mass.

B. 4 X –Picture shows tumor cells arranged in solid nodules. (H & E stain)

C. 10X Picture shows cyst like structures lined by columnar epithelium. (H & E stain)

D. 40X Picture shows cuboidal to spindle shaped odontogenic epithelial cells containing eosinophilic material. (H & E stain)

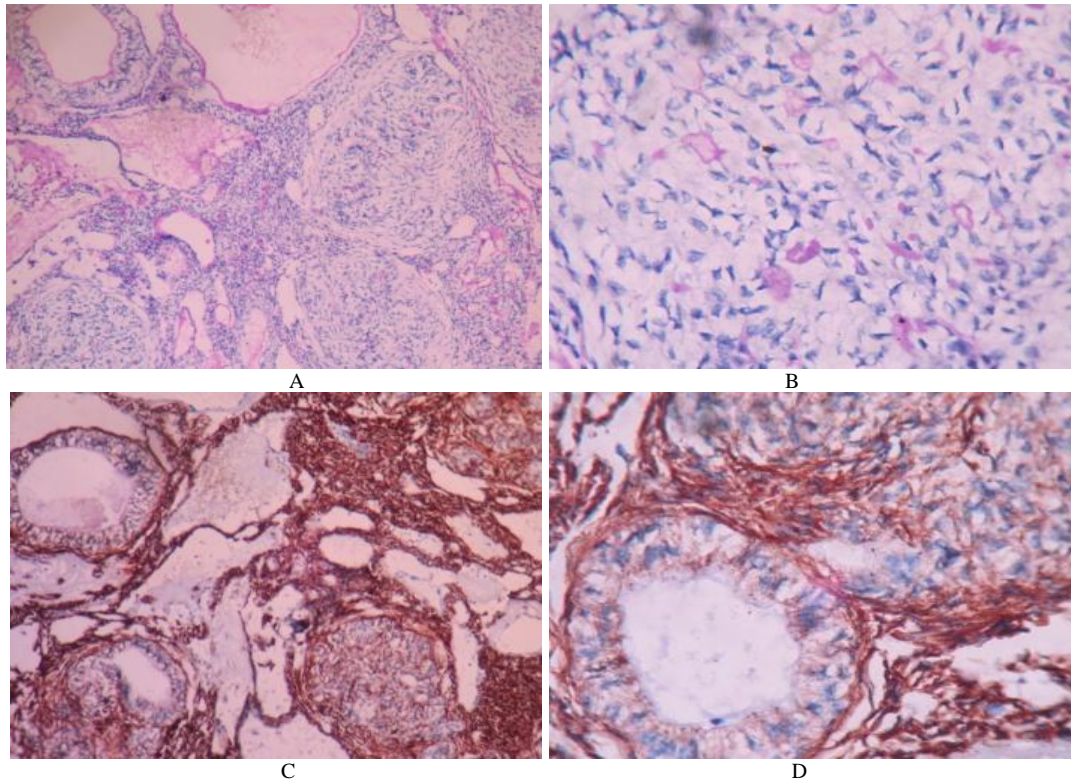


Figure 3: A & B 10x,40x res.] Magnification show columnar epithelial cells separated by a zone of PAS positive material and at the interior of the proliferative nodules. (Periodic Acid –Schiff stain)
C &D 10X,40X resp.] Cytokeratin antibodies study shows a positive stain in epithelial tumor cells in nodular areas and in cells of the duct-like microcyst.(IHC stain)

Broad spectrum CK antibodies revealed a positive stain in epithelial tumour cells, primarily in nodular areas and in cells of the duct-like microcysts. (figure 3C&D) The resulting diagnosis was that of follicular adenomatoid odontogenic tumour associated with unerupted tooth.

DISCUSSION

By definition, Adenomatoid Odontogenic Tumour (AOT) is composed of odontogenic epithelium in a variety of histoarchitectural patterns, embedded in mature connective tissue stromatolites and characterised by slow but progressive growth. ⁽⁶⁾ AOT accounts for 2-7% of all odontogenic tumours. The age range varies between 3 and 82 years .More than two thirds are diagnosed in the second decade of life and 90% are found before the age of 30. More than half of the cases occur among the teenagers. The male: female

ratio is 1:1.9. In some Asian countries, the ratio may reach 1:3.2. ⁽⁶⁾

A peripheral (extra –osseous) counterpart exists; Buchner et al. reported six cases; a total of about 20 cases have been published and almost all have occurred in the anterior part of the maxilla. They are very rare in the mandible. Peripheral AOT presents as a pink gingival swelling. ⁽⁷⁾ In about 75% of cases, the tumour appears as a circumscribed, unilocular radiolucency. The follicular type of AOT (small) may be impossible to differentiate radiographically from the more common dentigerous cyst. The radiolucency associated with follicular type of AOT sometimes extends apically along the root, the part that cements enamel junction. This feature may help to distinguish AOT from a dentigerous cyst. ⁽⁸⁾ The radiolucency located between the roots of erupted teeth (extra follicular type).The lesion often contains fine (snow flakes) calcification. This feature may be helpful in

differentiating AOT from dentigerous cyst.⁽⁸⁾

The pathogenesis of AOT is unknown. The most likely source of tumour development is residues of the dental lamina and proliferation of odontogenic epithelium adjacent to the reduced enamel/epithelium of an unerupted tooth.⁽⁹⁾ Macroscopically, the tumour is circumscribed and usually encapsulated. Some present as a solid mass, those situated around the crown of a tooth.⁽⁹⁾ A peripheral type may occasionally show erosion (“saucerization”) of the alveolar bone crest.⁽⁹⁾ On histopathology, there is an intra cystic epithelial proliferation composed of polyhedral to spindle cells. The pattern typically consists of lobules, although some areas may show syncytial arrangement of cells. Rosettes and duct like structures of columnar epithelial cells give the lesion its characteristic microscopic feature.⁽⁷⁾ There may be scattered foci of polyhedral squamous cells in the tumour. Another conspicuous cellular pattern is characterised by long, narrow epithelial strands consisting of two layers or single layer of cuboidal cells. They may form large loops, which are connected to each other in a plexiform pattern.⁽⁹⁾ In this tumour, a wispy PAS - positive, diastase-resistant eosinophilic material is often seen.⁽⁹⁾ Our case exhibited positivity with PAS stain.

By immunohistochemistry, all epithelial cells in AOT react to pan keratin antibodies. Monoclonal antibodies against individual CK's were used by Leon et al. The tumour cells reacted to the following type II CK s, CK-5, CK-7, CK-8. CK-7 and CK-8 were mostly found in the loop forming narrow epithelial strands. Among the type I CK s, CK-10 and CK-13 are not expressed.⁽⁹⁾

Other markers, Involucrin could not be detected. Nestin, Integrins, surprisingly vimentin, some growth factors (HGF), Extra cellular matrix proteins like

versican, chondroitin sulfate proteoglycan, and tenascin are the other markers expressed. Basement membrane-associated macromolecules, enamelysin, sheathln, amelogenin, enamelin, p 53 and proliferative marker (Ki-67) are useful markers for AOT by various studies⁽⁹⁾ in our case; expression of CK s in AOT was studied.

Mutation of the AMBN, which in human maps to chromosome 4q 21 has been detected in one case of AOT and was considered a tumour-specific mutation. The AOT had 334G>T transversion, causing a R 90 W amino acid change.⁽⁹⁾

Ultra structurally, there is clear cut evidence of glandular differentiation present.⁽¹⁰⁾

Other lesions that might be included in a differential diagnosis of AOT are dentigerous cyst (because of frequent association with impacted teeth) and lateral root cyst (because of this occasionally being located adjacent to roots of anterior teeth). If opacities are evident, calcifying epithelial odontogenic tumour (CEOT) should receive consideration.⁽⁷⁾ AOT is probably best classified as a Hamartoma rather than a true neoplasm; it's not related to ameloblastoma.⁽¹¹⁾ Enucleation curettage is the treatment of choice, and the prognosis is considered to be excellent. Recurrence is very rare.⁽¹²⁾

CONCLUSION

AOT is an epithelial odontogenic hamartoma containing pseudo ducts and enameloid. It is described as tumour of two thirds - maxilla, females, anterior jaws and crown of impacted tooth. Teenagers are most commonly affected and tumour is rarely seen over the age of 30 years. Radiographically, it shows lucent and lucent-opaque patterns. Immunohistochemistry (CK) and histochemical stains (PAS) are expressed by AOT. Treatment is by enucleation and there is usually no recurrence.

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