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Case Report

Mandibular adenomatoid odontogenic tumor: case report

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is a rare tumor of epithelial origin which accounts for 3% of odontogenic tumors. The most common area of involvement is the maxilla, about 65% of cases, followed by the mandibular region accounting to 35% of cases. This case report is of a 16 year old girl operated and diagnosed with AOT at the anterior aspect of the mandible. This case report also discussed about the necessity for a detailed descriptive, differentiating features of AOT from other odontogenic tumors, the most common misdiagnosis being dentigerous cyst.

Keywords: Odontogenic tumor, Mandible, Hamartoma, Epitheloid tumor, Unerupted tooth, Impacted lateral incisor

INTRODUCTION

Adenomatoid odontogenic tumor (AOT) is a rare odontogenic tumor of epithelial origin that arises mostly from the enamel, enamel epithelium, dental lamina or the malleses rests, this is currently the most accepted theory.[1] Most of the times it is misdiagnosed as odontogenic cyst or the dentigerous cyst. Earlier known along a wide range of nomenclature such as adenomeloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantinoma or teratomatous odontoma. It was in 1905 that Steensland described AOT for the first time.[2] Later in 1999 Philipsen et al presented reports and review based literature gathered upto 1997. This showed interesting and distinguished features of AOT.[3]

Now to start with the descriptive features of AOT, firstly histopathologically WHO described AOT as an epitheloid tumor with duct like structures with connective tissue stroma. Tumor shows partial cystic component in few cases, mostly solid lesions are noted. Eosinophilic, uncalcified, amorphous material called tumor droplets are present. This may be in the form of plaques or homogenous material. Highly useful during diagnosis.[3]

Immunohistologicaly, AOT phenotype characterised by cytokeratin (CK): CK5, CK17, CK 19. Criveline detected CK 14 which shows origin from reduced dental epithelium. Tekatrashi et al observed positive staining for iron binding proteins (transferring/ferritin) and proteinase inhibitors (alpha one anti-trypsin).[4]

Radiographically, intra-oral periapical radiograph seems to be more useful than panoramic view as discrete foci with flocculant pattern with minimal small calcifications are visible. The classical picture is of a well circumscribed, radiolucent lesion, causing no cortical bone erosion or thinning having a sclerosing border. Most cases have calcifications within them, which may not be visible in radiographs. It may present as a single lesion, large cystic lesion or multiple cystic masses. This leads to common misdiagnosis of dentigerous cyst, calcifying odontogenic cyst, calcifying epithelial odontogenic tumor, ameloblastoma and odontogenic keratocyst. MRI scans helps to distinguish AOT from other odontogenic tumors.[5] Clinically, 2:1 cases are seen in female ratio,

predominantly in Asian population. Delayed eruption off a permanent tooth, slowly disfiguring facial appearance at the lower jaw, painless swelling is the common presentations. This case report shows the distinct characteristic features of AOT in a 16 year old girl.

CASE REPORT

A 16 year old patient was referred to the department of ENT and Head and Neck Surgery in Shri Satya Sai Sarala Memorial Hospital, Muddenahali with chief complaints of a swelling, progressively increasing in size at the left lower jaw, approximately near the parasymphysis of the mandible noticed for a period of 5 months. Patient also complaints of a mobile accessory tooth on the left side. An orthopantomogram was requested, the report showed a unilocular, radiolucent swelling with sclerotic border at the parasymphysis of the mandible, attached with an unerupted tooth. The OPG also shows a deciduous tooth at the carina. OPG showing unilocular cystic swelling with the unerupted tooth and deciduous tooth at carina. With the clinical and radiological findings, an initial diagnosis of dentigerous cyst was made. Decision to carry out enucleation of the cyst was made and therefore operated through intra-oral approach. Cyst along with the unerupted tooth and the deciduous teeth at the carina was extracted and sent for pathological analysis.

Histopathology report revealed as AOT measuring 2.5×1.8×1.5 cm with the crown of the tooth projecting into the lumen and filled with keratinous debris.

Microscopic examination showed a well circumscribed solid to cystic tumor composed of proliferation of duct like structures lined by columnar type of cells with basal nuclei and clear cytoplasm. The nuclei are polarized away from the lumen. These duct like structures are dispersed in spindly cells arranged in sheet like fashion. Eosinophilic fibrillary material is present within duct like structures. There were foci of calcification.

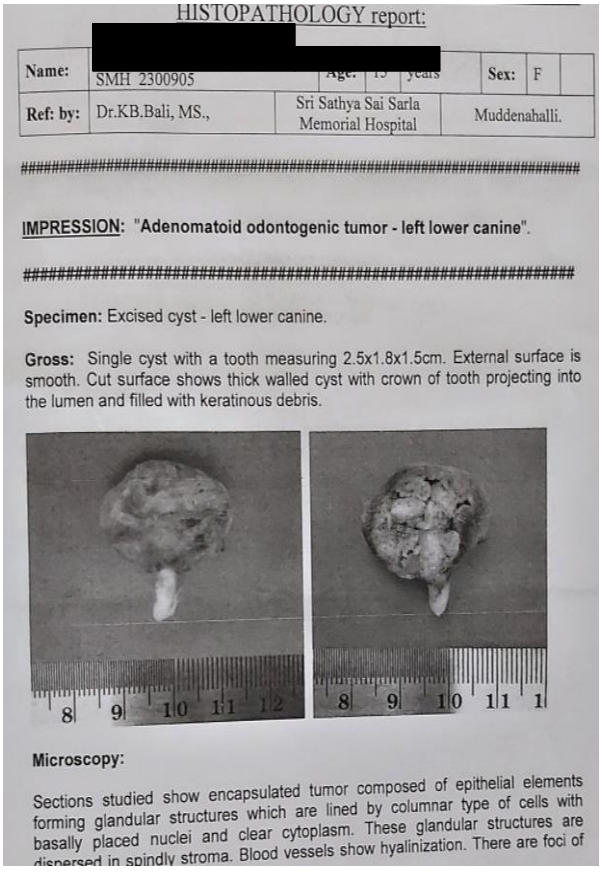
Patient significantly improved post-operatively with no complications. Antimicrobials and anti-inflammatory medications were given for a week and patient was discharged.

Routine radiograph taken during follow-up shows adequate bony regeneration with no remnants.

Figure 1: Pre-operative clinical picture of a swelling noted near the left mandibular parasymphysis.

Figure 2: OPG pre-op.

Figure 3: Post-operative excised lesion with impacted tooth.



HISTOPATHOLOGY report:

Name:	[redacted] SMH 2300905	Age: 15 years		Sex:	F
Ref: by:	Dr.KB.Bali, MS.,	Sri Sathya Sai Sarla Memorial Hospital		Muddenahalli.	

IMPRESSION: "Adenomatoid odontogenic tumor - left lower canine".

Specimen: Excised cyst - left lower canine.

Gross: Single cyst with a tooth measuring 2.5x1.8x1.5cm. External surface is smooth. Cut surface shows thick walled cyst with crown of tooth projecting into the lumen and filled with keratinous debris.

Microscopy:

Sections studied show encapsulated tumor composed of epithelial elements forming glandular structures which are lined by columnar type of cells with basally placed nuclei and clear cytoplasm. These glandular structures are dispersed in spindly stroma. Blood vessels show hyalinization. There are foci of

Figure 4: Histopathology report.

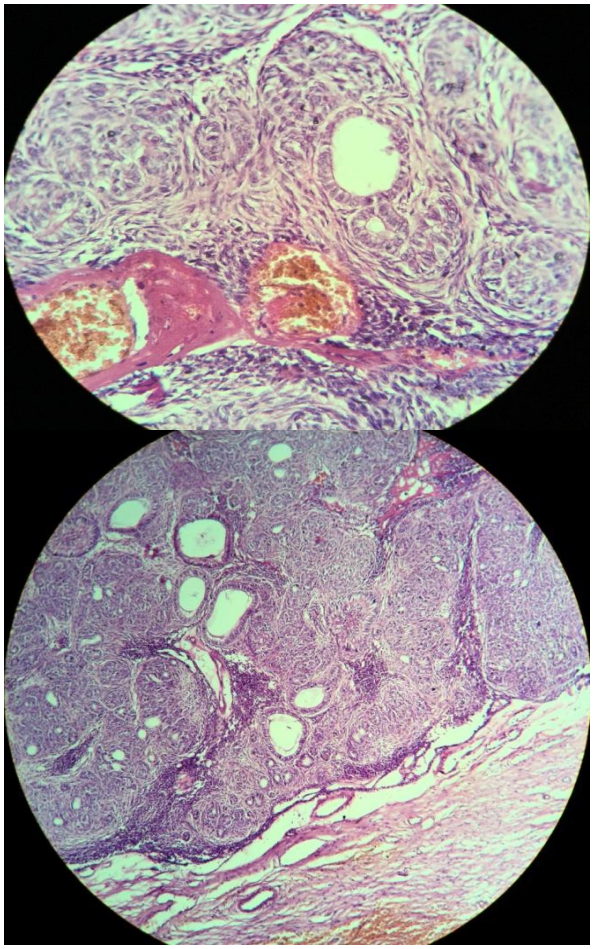


Figure 5: Histopathology slides.

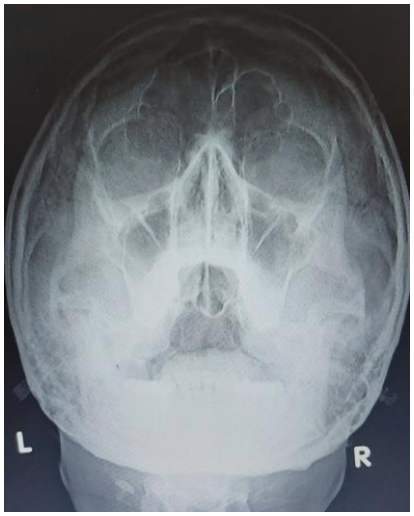


Figure 6: Post-operative ragiograph.

DISCUSSION

Histopathologically AOT is characterised by odontogenic epithelium in the form of sheets or rods and few odontogenic cells in a duct like pattern with eosinophilic material within. A well-defined fibrous tissue capsule is present at the margins.[6]

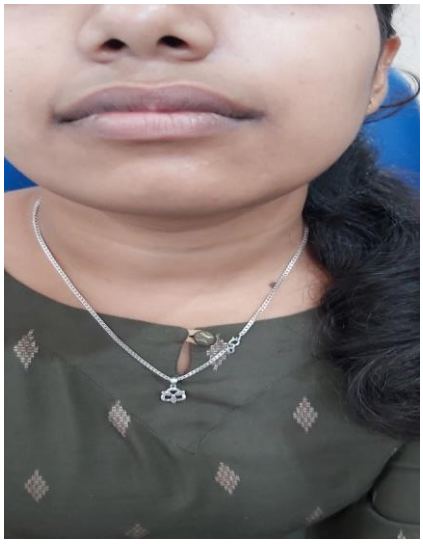


Figure 7: Post-operative follow-up was done for four months and showed no recurrence.

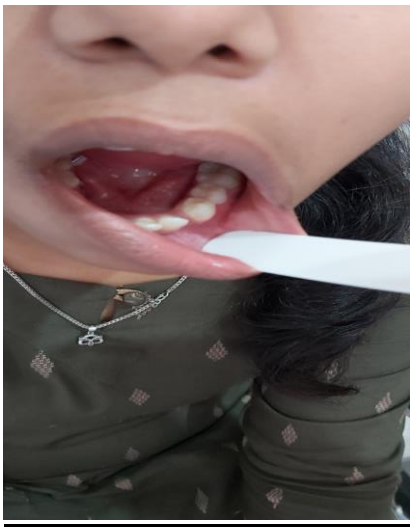


Figure 8: Post-operative intra-oral examination.

Our histopathological report showed spindle shaped stroma with glandular structures lined by columnar epithelium with an unerupted tooth suggestive of follicular type of AOT, which accounts for 73% of all cases. AOT most commonly misdiagnosed as the dentigerous cyst due to their common areas of development and similar radiographic picture. In our case, anterior portion of mandible is affected, in association with an unerupted tooth and a displaced permanent tooth. Radiographically in a panoramic view, it appears as a circumferential, volumetric, radiolucent and unilocular lesion with multiple calcifications. Symptomatically, it is a non-tender, progressive lesion causing cosmetic deformity with no involvement of adjacent structures.[7] AOT is classified under hamartoma, meaning it has limited potential of growth and is a developmental anomaly of tissue differentiation. AOT is called a two-third tumor as two-third cases occur in females, two-third cases with an

unerupted tooth, two-third cases in maxilla. Two-third cases. Canine is the affected tooth.[8]

Our 16 year old patient fits into the characteristic features. Swelling was noted at the parasymphysis of the mandible with an unerupted tooth. The tooth involved was the left canine. The cyst measured around 2.8×1.8×1.5 cm. Generally AOT measures around 1-3 cm.[8] Therefore fitting into the criteria. Since AOT does not infilterate the adjacent bones and treatment mainly is enucleation, therefore most cases it does not recur.[8]Radiopacities and calcifications are present within the lesion which is particularly seen only in AOT. A dentigerous cyst does not have calcifications within them. Also radiographically AOT is shown to involve coronal and radical aspects of the tooth but dentigerous cyst only surrounds the coronal aspect of the involved tooth.[9] AOT does not cause resorption of the root of the tooth involved.

Most cases it causes displacement of the tooth and the adjacent tooth. Canines are most commonly involved followed by premolars and rarely molars. The variants extafollicular type has an intra-bony lesion and no connection with the tooth. Peripheral variant presents as a gingival swelling, small sessile masses on the buccal gingival.[9] Both the variants are only diagnosed histologically as they involve gingival pathologies.

CONCLUSION

The differential diagnosis with other pathologies such as ameloblastoma and keratocyst or a dentigerous cyst is important due to the aggressive treatment they require. AOT requires surgical conservative treatment as they do not involve the bone. Aim is to provide detailed descriptive features of AOT for accurate diagnosis and treatment. Delay in diagnosis leads to increase in size of lesion and therefore larger area of clearance.

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