

“Two third tumor”: A case report and its differential diagnosis

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ABSTRACT

Adenomatoid odontogenic tumor (AOT) is an odontogenic tumor with a prevalence of 2.2–7.1%. AOT is a benign, noninvasive, and progressive lesion which is also known as “a two third tumor.” As the name suggests the tumor occurs in the maxilla in two third of cases. It occurs in young patients in two third of cases and associated with missing or unerupted teeth in two third of cases. Two third cases are associated with the maxillary canine. Characterized by slow growing, gradually enlarging, painless swelling associated with missing teeth. We report a case of a male patient of age 22 years, with characteristic findings. AOT resembles different odontogenic cysts and tumors which may include dentigerous cyst, globulomaxillary cyst, ameloblastoma, and other entities, hence must be well differentiated. Conservative surgical enucleation is the treatment of choice. Recurrence rate for AOT is 0.2%. Prognosis is excellent when completely removed in toto.

Keywords: Adenoameloblastoma, adenomatoid odontogenic tumor, ameloblastic adenomatoid tumor, enamel organ epithelium

Introduction

Adenomatoid odontogenic tumor (AOT) is also known as adenoameloblastoma or ameloblastic adenomatoid tumor, originating from the enamel organ epithelium. Predominant site is the maxillary canine region. Predominant in young females, second decade of life and associated with missing tooth.^[1] There are three variants of AOT, the follicular type 73%; extrafollicular type 24%, and the peripheral variety 3%.^[2] According to World Health Organization, histological typing of odontogenic tumors, jaw cyst, and allied lesions AOT can be defined as a tumor of odontogenic epithelium.^[3]

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Case Report

A 22-year-old male patient reported to the dental OPD with a painless swelling in the upper left anterior region of jaw since 6 months [Figure 1]. He gives a history of trauma in the same region 1 year back. The swelling was initially peanut sized and gradually increased to present size of 2 cm by 2.5 cm. The swelling was presently not associated with pain. History of hot fomentation extraorally and balm application intraorally in the same region was reported at the time of injury. Also, history of sensitivity of tooth was reported in the same region to cold beverages since 2 months. On general examination, gait was normal, built was thin, and all the vital signs were within normal limits. On extraoral examination, there was facial asymmetry present. Swelling was present on the left maxillary canine region which was extending anteroposteriorly from the philtrum region to the level of outer canthus of eye and superoinferiorly from the ala-tragus line in the left side of face to the upper lip region.

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Figure 1: A 22 years old male with a painless extra oral swelling on left side of maxilla

On inspection, size was 3×2 cm approximately, shape was roughly oval, surface was smooth, margins were diffuse, and color was same as that of contralateral skin. On palpation, all the inspektor findings were confirmed and temperature was afebrile, tenderness was absent, consistency was firm to hard in nature. There was obliteration of the philtrum on the left side. On intraoral examination, there was a single diffuse dome shaped swelling present in the left maxillary canine region, extending anteroposteriorly from mesial aspect of left permanent maxillary canine region to distal aspect of left permanent maxillary second premolar region and superoinferiorly from gingival margin to depth of buccal vestibule. On inspection, size was around 3.5×4.2 cm, shape was roughly oval, surface was smooth, margins were diffuse, and color was same as that of the adjacent mucosa [Figure 2]. On palpation, the inspektor findings were confirmed and consistency was soft to firm in nature, tenderness was absent, and fluctuation was present. On examination of the teeth in the vicinity was observed that there were over-retained 62, palatally placed 23, missing 22.

Vitality test

Vitality test were performed with gutta-percha point which showed delayed response with 12, 13, and 25 and no response with 21, 62, 23, and 24.

Radiographic features

On intraoral periapical radiograph it shows missing left permanent maxillary lateral incisor and a single radiolucency seen in apical region of 62, size approximately 4×3 cm, shape roughly oval, margins well defined [Figure 3].

Cross-sectional occlusal radiograph shows missing with 22. A single radiolucency was seen extending from 21 to 26 and mediolaterally from midpalatal raphae to the alveolar crest of 24 region causing expansion of the buccal cortical plate. Size approximately 3×3 cm, shape roughly round, margins well defined, and corticated. Internal structure is radiolucent with



Figure 2: The intra oral swelling for the same patient in the maxillary canine region on the left side

no radiopaque foci. Effect on adjacent structure: displacement with left permanent maxillary central incisor and left deciduous maxillary lateral incisor [Figure 4].

Orthopantomogram shows missing 22, a single well-defined radiolucency seen in the upper left posterior region extending anteroposteriorly from distal of 21 to distal of 26 and superoinferiorly from level of alveolar crest to the maxillary antrum superiorly, root resorption with 23, 24 and 25. Displacement of teeth with 23, 24, and 25. Loss of cortication seen with the inferior border of left maxillary sinus [Figure 5].

Waters's view shows haziness seen in the left maxillary sinus [Figure 6].

Macroscopic Features

The tumor was of size $2.5 \times 1.5 \times 1.5$ cm approximately, shape was oval; surface was rough and irregular, color brownish, and consistency soft to firm [Figure 7].

Microscopic features

Under 10X view, odontogenic epithelial cells arranged in ductal pattern [Figure 8] presence of odontogenic epithelial cells arranged in rosette pattern [Figure 9] and plexiform pattern was appreciated [Figure 10].

Discussion

The first report of AOT in history was made by Steensland's (1905), who defined it as epithelioma adamantinum.^[4] The present term AOT has been first described by Dreyblatt in 1907.^[5] The most common site in maxilla in descending order is canine, central, and lateral incisor and the premolars.^[1] The most widely accepted clinicotopographic variants include (i) follicular, (ii) extrafollicular, and (iii) peripheral.^[6] The follicular type is associated with the crown and often part of the root of an unerupted tooth.^[7] The follicular variants are mostly predominant in the maxilla, whereas

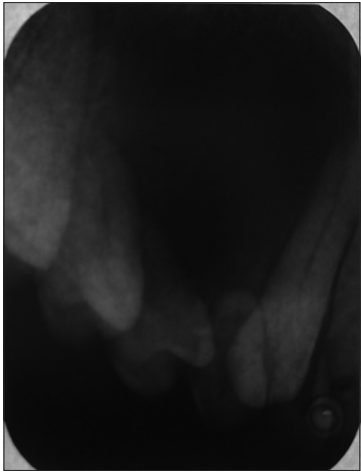


Figure 3: Intra Oral Periapical Radiograph it shows radiolucency in apical region of left deciduous maxillary lateral incisor



Figure 4: Cross Sectional Occlusal Radiograph shows radiolucency



Figure 5: Orthopantomogram shows a single well defined radiolucency seen in the maxillary left posterior region

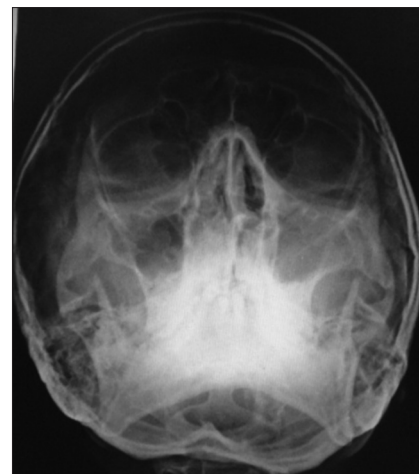


Figure 6: Water's view shows haziness seen in the left maxillary sinus

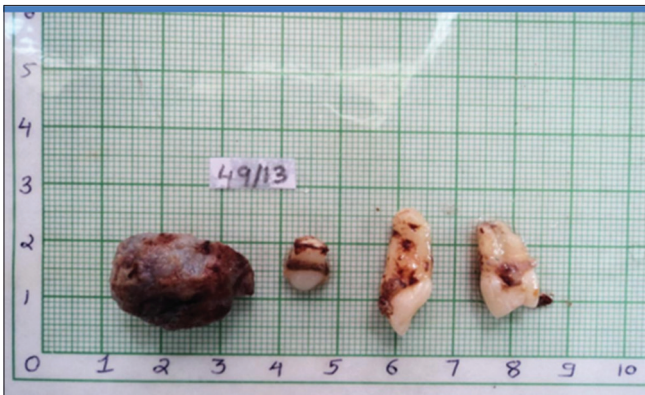


Figure 7: The lesion was oval in shape and surface was rough and irregular. Colour was brownish and consistency was soft to firm in nature

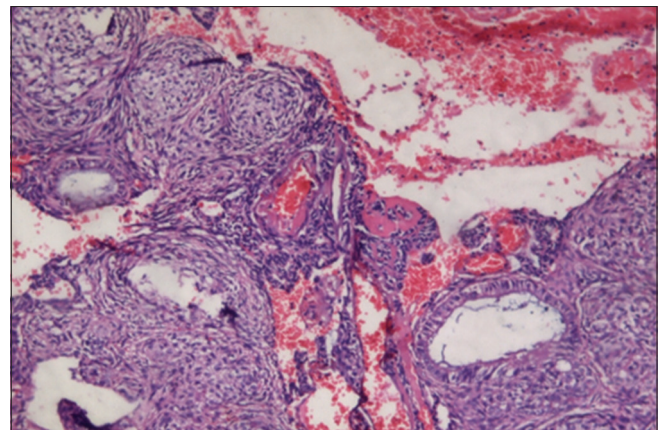


Figure 8: Under 10X view, odontogenic epithelial cells arranged in ductal pattern

the extrafollicular variants are seen mostly in the mandible. The peripheral type is located in the gingival mucosa.^[8] Benign odontogenic cysts and tumors which need to be differentiated from AOT for proper diagnosis and its management. The most common odontogenic cyst to be differentiated from AOT is dentigerous cyst. Predominant site for dentigerous cyst is the mandibular posterior region and AOT in the maxillary anterior region. A lesion associated with dentigerous cyst arises from the

cemento-enamel junction which is characteristic. The next entity to be differentiated is keratocystic odontogenic tumor (KCOT) and a unicystic ameloblastoma (UA) in cases when the lesion is located pericoronally. All of these three lesions are mostly seen in the second and third decade of life. But KCOT and UA are more common in the posterior regions of the mandible

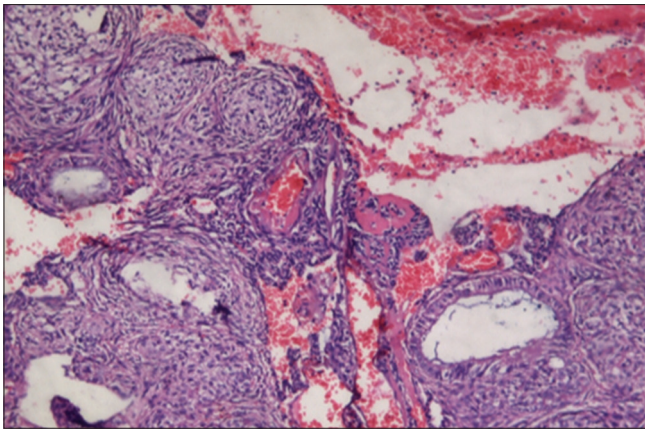


Figure 9: Under 10X view, odontogenic epithelial cells arranged in rosette pattern

and AOT in the maxillary anterior region. The calcifying cystic odontogenic tumor (CCOT) mimics an AOT. CCOT and AOT is observed in the anterior region of jaws and is associated with an unerupted tooth and presence of radioopacities and diagnosed in the second decade of life.^[9] Permanent canines are the most common unerupted teeth associated with these two entities. Opacities are described as flecks, snow-flakes, and patchy areas of calcification.^[8] The extrafollicular variant of AOT is also differentially diagnosed from lateral periodontal cysts located between the roots of erupted teeth in the anterior region of the mandible, diagnosed in old age groups, and AOT in the early age group.^[10] A unilocular radiolucency with opacities and tooth displacement in the anterior region are the characteristic radiological features.

Conclusion

AOT can be considered as a differential diagnosis of the swellings in the maxillary anterior region. The other sites are mandible and maxillary premolar region for this lesion but rarity. Conservative surgical enucleation is the treatment of choice as the lesion is a well-encapsulated lesion and can be separated from the bone easily. AOTs are hamartomas due to the innocuous behavior of the lesion; because the tumor stops developing after the tooth structures completes their growth.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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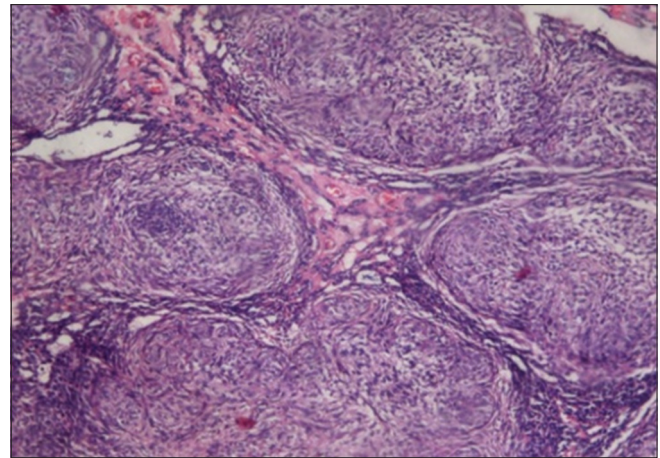


Figure 10: Under 10X view, odontogenic epithelial cells arranged in plexiform pattern

Conflicts of interest

There are no conflicts of interest.

References

1. White SC, Pharaoh MJ. Oral Radiology: Principles and Interpretation. 5th ed. Mosby; 2004.
2. Dare A, Yamaguchi A, Yoshiki S, Okano T. Limitations of panoramic radiography in diagnosing adenomatoid odontogenic tumor. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1994;77:662-8.
3. Mutalik VS, Shreshtha A, Mutalik SS, Radhakrishnan R. Adenomatoid odontogenic tumor: A unique report with histological diversity. *J Oral Maxillofac Pathol* 2012;16:118-21.
4. Almeida RB, Capelari MM, Marzola C, Filho JL, Toledo GL, Zorzetto DL. Adenomatoid odontogenic tumor. Literature review and clinical-surgical case report. *Rev Odontologia* 2012;12:1229-55.
5. Patil NN, Nayyar AS, Wadhwan V. Adenomatoid odontogenic tumor: A series of four clinico pathological variants. *Int J Case Rep Images* 2014;5:1-7.
6. Philipsen HP, Reichart PA, Zhang KH, Nikai H, Yu QX. Adenomatoid odontogenic tumor: Biologic profile based on 499 cases. *J Oral Pathol Med* 1991;20:149-58.
7. Philipsen HP, Birn H. The adenomatoid odontogenic tumor: Ameloblastic adenomatoid tumor or adeno- ameloblastoma. *Acta Pathol Microbiol Scand* 1969;75:375-98.
8. Becker T, Buchner A, Kaffe I. Critical evaluation of the radiological and clinical features of adenomatoid odontogenic tumor. *Dentomaxillofac Radiol* 2012;41:533-40.
9. Buchner A, Merrell PW, Carpenter WM. Relative frequency of central odontogenic tumors: a study of 1,088 cases from Northern California and comparison to studies from other parts of the world. *Int J Oral Maxillofac Surg* 2006;64:1343-52.
10. Neville BW, Dam DD, Allen CM, Bouquot JE. Oral and Maxillofacial Pathology. 3rd ed. St. Louis: Saunders Elsevier; 2009.