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Case Report

Adenomatoid odontogenic tumor in mandible — A rare case report

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ABSTRACT

Adenomatoid Odontogenic tumor (AOT) is an uncommon histologic type of Odontogenic tumor which is characterized by formation of duct-like structures by epithelial component of lesion. It is a non-invasive type of lesion with slow but progressive growth. A 15-year old patient reported to facio-maxillary and dental health center with chief complain of crowding in lower anterior segment. Radiographic examination revealed a radiolucent lesion in relation to lower left unerupted canine. The boundary of lesion was well defined. Enucleation of lesion was done along with removal of canine and defect was filled with bone graft material, R.T.R (Septodont) which consists of Beta-Tricalcium phosphate. Excisional biopsy was performed for excised tissue. Histopathology was suggestive of AOT. The effective treatment of AOT is conservative surgical enucleation guided by tissue regeneration with membrane technique.

1. Introduction

Adenomatoid odontogenic tumour is a benign (hamartomatous), noninvasive lesion with slow but progressive growth.[1] It is classified as an uncommon odontogenic tumor that is generally misinterpreted as odontogenic cyst. It was first described by Steensland in 1905.[2] Numerous terminologies were used to describe the lesion for example; Adenoameloblastoma, adenoameloblastic odontoma, epithelial tumour associated with developmental cysts, ameloblastic Adenomatoid tumour and Adenomatoid or pseudoadenomatous ameloblastoma.[1] In 1969, Philipsen and Birn suggested the name 'Adenomatoid odontogenic tumour'. Later in 1971, World Health Organization (WHO) adopted the term and explained it as 'a tumor of odontogenic epithelium with duct-like structures and with varying degrees of inductive change in the connective tissue. The tumor may be presented as partly cystic and in some cases as solid lesion with masses present in the wall of a large cyst. It is usually considered that the lesion is not a neoplasm'.[1–3]

AOT has three clinical variants: (a) "Follicular" (70% of cases); intra-osseous (associated with impacted teeth); (b) "Extra-follicular" (25% of cases); intra-osseous (present between erupted teeth) and (c) "Peripheral" (5% of cases); on the gingiva (extra-osseous).[3] AOT comprises of about 1% until 9% of all Odontogenic tumors. The commonest variant of all AOTs is follicular type, with a central intraosseous lesion associated with an impacted tooth.[4] 70% of AOT is diagnosed in the second decade of life (age range of 10-19).[3] Women are approximately twice more often affected than men with female: male ratio of 2 .3: 1. The AOT is over two times more located in the maxilla in comparison to the mandible along with the anterior jaw being more affected as compared with the posterior area with an unerupted permanent tooth.[2]

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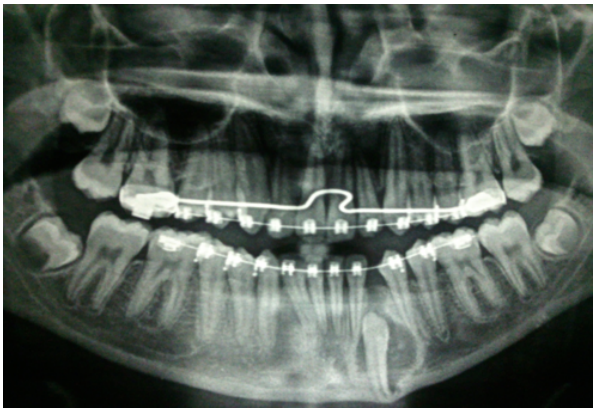

Fig. 1: Panoramic radiograph before therapy. Unicystic radiolucent lesion in the lower left jaw with a comparatively clear demarcation. The tooth 33 is located on the floor of this process. There is no resorption of the root apices.

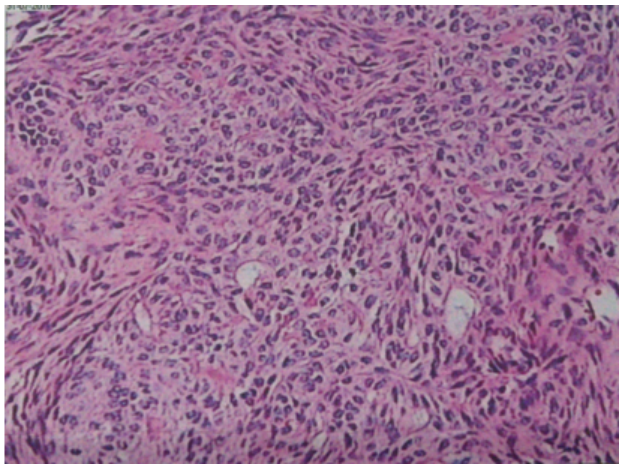

Fig. 2: Tumor with fibrous connective tissue capsule. Nodular aggregates of cells. Duct-like structures .Gland-like spaces are surrounded by cuboidal to columnar cells

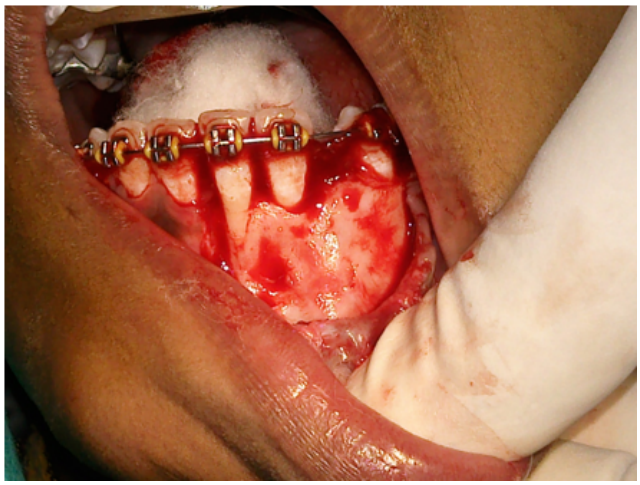

Fig. 3: Underlying bony surface after flap reflection.

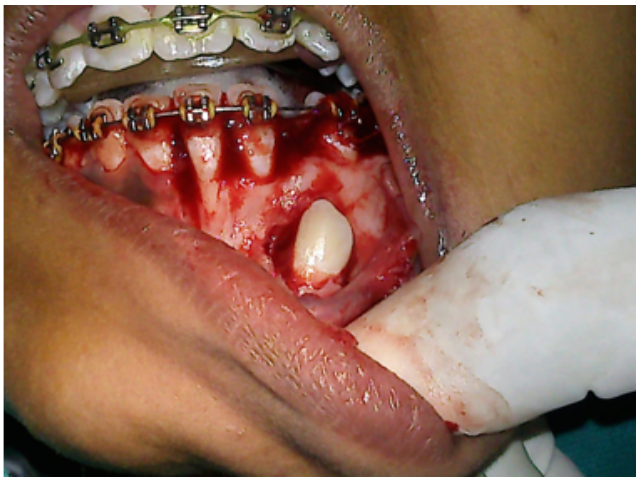

Fig. 4: Impacted canine associated with AOT.

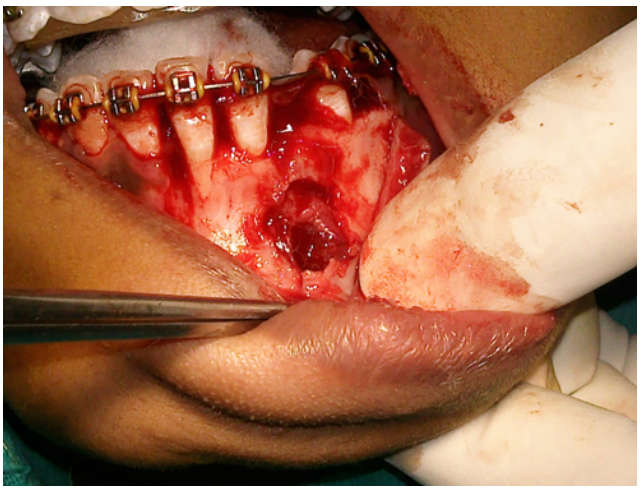

Fig. 5: After extraction of canine underlying calcified tissue.

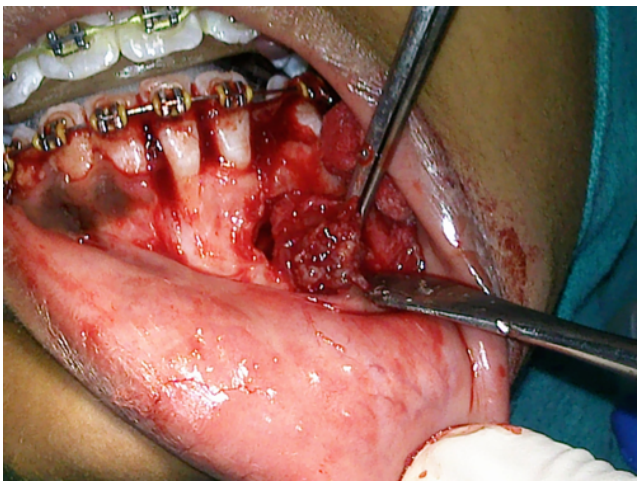

Fig. 6: Enucleation of the cystic lesion.

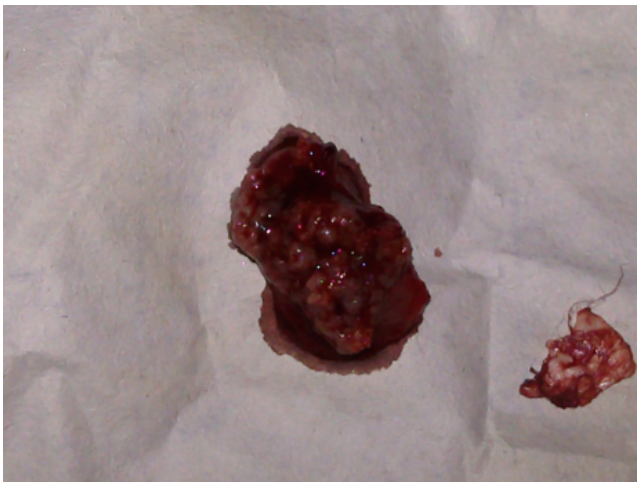

Fig. 7: Excised lesion from the site.

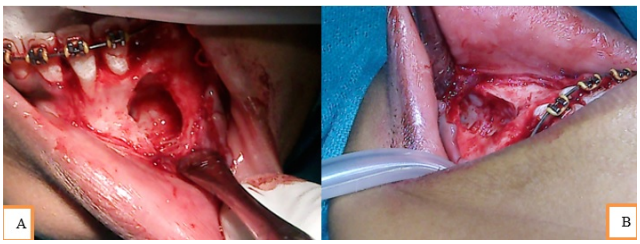

Fig. 8: (a): Bony cavity after removal of the lesion. (b): Bony cavity after removal of the lesion.

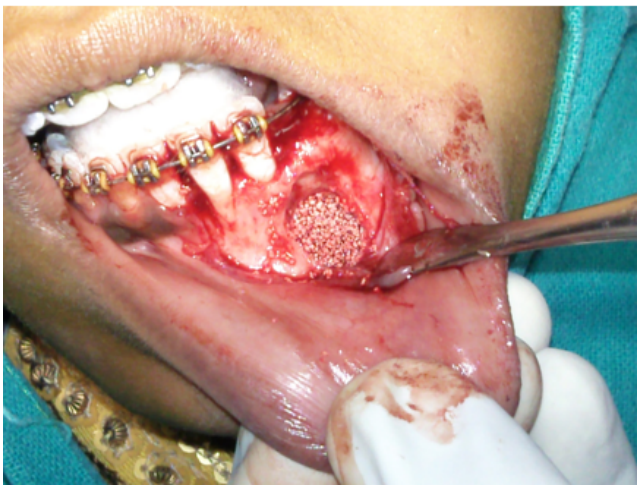

Fig. 9: Bone graft material (R.T. R) filled in the bony defect.

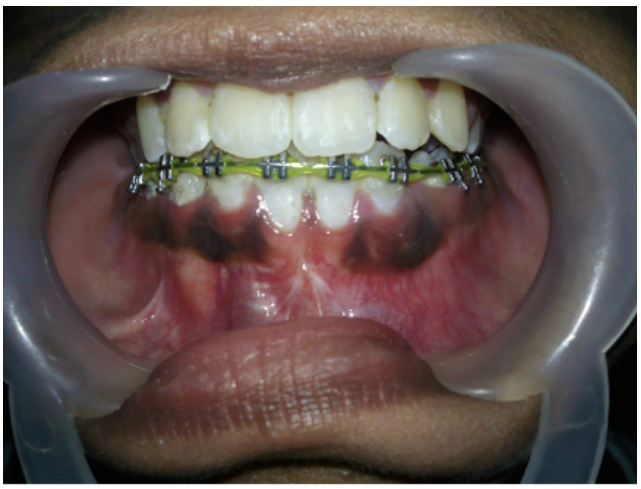

Fig. 10: Follow up after 6 months of surgery.

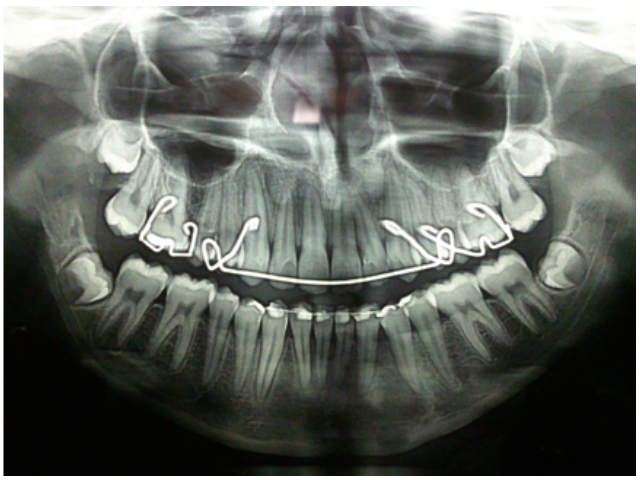

Fig. 11: Panoramic view after 6 month of surgery.

1.1. Clinical features

The lesion usually presents as a well circumscribed and slowly growing asymptomatic swelling which is often associated with an unerupted or missing tooth. But growth of the types comprising of central lesion results in cortical expansion.[1] However, the rare peripheral variant occurs primarily in the gingival tissue of tooth-bearing areas. The majority of lesions are measured between 1.5 and 3.0 cm however, sizes larger than 3.0 cm are described which can exceed upto 7.0 cm.[1,2]

In general, AOT cases are usually mildly expansile.[3] The anterior maxilla (65%) and mandible (35%) are the commonest site of occurrence, with maxilla being almost twice as likely as mandible in association with 76% developing anterior to the cuspid.[1–4] Unerupted permanent canine are the most common teeth involved in follicular variety of AOTs which account for 60% of cases.[4] AOT cases have also been reported in posterior mandible and

maxilla, but rarely beyond premolars. Amongst all AOT cases, 53% are seen in anterior maxilla, whereas 9% are found in maxillary premolar region. About 2% of AOT cases occur in the molar region.[3]

1.2. Roentgenographic features

The radiographic findings of AOT frequently resemble other Odontogenic lesions such as dentigerous cysts, calcifying Odontogenic cysts, calcifying Odontogenic tumors, globule-maxillary cysts, ameloblastoma, Odontogenic keratocysts and periapical disease.[4] The dental roentgenogram of follicular variant usually presents as well-defined to corticated unilocular radiolucent lesions associated with crown and often part of root of an unerupted tooth; about 10% demonstrate some degree of calcifications.[3] In extra follicular type, radiolucency is either located between, above or superimposed upon roots of erupted permanent teeth. It extends apically further than the CEJ. Displacement of neighboring teeth due to tumor expansion is much more common than root resorption. The peripheral variant may show some erosions of the adjacent cortical bone.[4]

Radiolucent lesions are often displayed with presence or absence of calcifications (radio-opacities) within the lesion. The opacities were described as flecks or snowflakes, patchy areas of calcification, spicules of calcification, scattered radio-opacities, irregular radio-opacities, amorphous radio-opacities, fine radio-opacities and faint radio-opacities.[2] Philipsen reported approximately two-thirds of the intrabony AOTs possessed scattered radiopacities in the radiolucent lesions. This intra-lesional calcification with characteristic pattern was suggested as a distinctive radiographic feature of AOT.[5] The influence of age also accounts on the radiological characterizations of AOTs. It is significantly associated with features such as increased root resorption, ill-defined borders, expansion, perforation of cortex and lesions crossing midline. These changes reflects a longer duration of lesion due to a late diagnosis.[2,5]

1.3. Patho-histological features

Histological appearance of all variants is found to be identical.[1,4] The remarkable pattern noticed is that of various sizes of solid nodules of columnar or cuboidal epithelial cells forming nests or rosette-like structures with minimal stromal connective tissue.[1] The epithelial cells are often polyhedral or even spindle shaped exhibiting well-defined cell boundaries and prominent intracellular bridges. Interconnecting strands and ribbons with two or more cells are present throughout the lesion.[3,6] Intra-lesional calcification are found as, eosinophilic, uncalcified, amorphous material and is called 'tumor droplets'.[5] Droplets of unusual eosinophilic material are found between epithelial cells arranged in solid nests, in duct-like structure and sometimes in the midst of cells arranged in a rosette pattern.[1,5] Interestingly, there are a few reports about pigmented cells in AOT. However, all of these reported lesions did not show macroscopically visible pigmentation. Racial pigmentation probably plays an important role in such cases.[4]

1.4. Immunhistological features

Immunohistochemical studies of the lesion recommend expression of keratin and vimentin in the tumour cells at the periphery of the ductal, tubular or whorled structures. Amelogenin and enamel in small mineralized foci are seen in the tumour cells and in hyaline droplets.[1] The classical AOT phenotype is characterized by a cytokeratin (CK) profile similar to follicular cyst and/or oral or gingival epithelium.[4] It is positive for CK5, CK17 and CK19 while negative for CK4, 10, 13 and 18. Positive reactions for amelogenin in limited areas in AOT are also reported as well as in ameloblasts and in the immature enamel matrix.[7]

Interestingly, a positive staining was observed by Takahashi et al. for iron-binding proteins (transferring, ferritin) and proteinase inhibitor (alpha-one-antitrypsin) in various cells of AOT showing their function in the pathogenesis of AOT.[8] Finally, the expression of bone morphogenic protein (BMP) was studied by GAO et al.[9] A positive reaction was shown by cementifying fibromas, dentinomas and compound odontoma, whereas all AOTs along with ameloblastoma and calcifying epithelial Odontogenic tumors were demonstrated negative.[4]

2. Treatment and Prognosis

Conservative surgical enucleation and curettage is treatment modality of choice. It is usually done when tumour is not locally invasive; it is well encapsulated and is easily separated from the bone.[10] For periodontal intrabony defects caused by AOT guided tissue regeneration with membrane technique is suggested after complete removal of tumor. Recurrence of AOT is exceptionally rare. Therefore, prognosis is excellent.[4]

3. Case Report

A 15-year-old girl was referred by her orthodontist to facio-maxillary and dental health centre for evaluation of a mandibular radiolucent lesion noticed on a panoramic radiograph. (Figure 1). The medical history was insignificant and patient was in good general health. Intraoral examination showed that labial vestibule was obliterated by expansion of buccal cortical plate from lower left canine and premolar region (Figure 1). Permanent mandibular left canine was missing clinically. A panoramic radiograph revealed a, well-circumscribed radiolucency around impacted lower left canine (Figure 2). The lesion

produced an expansion of bone and extended into the alveolar processes, disrupting the usual alignment of anterior teeth. On the basis of clinical and radiographic findings, differential diagnosis was Dentigerous Cyst, Unicystic Ameloblastoma, Adenomatoid Odontogenic Tumor, Ameloblastic Fibrous Odontoma, Calcifying Odontogenic Cyst, and Calcifying Epithelial Odontogenic Tumor.

The patient underwent surgery under local anesthesia. A trapezoidal mucoperiosteal flap in left canine-premolar region was reflected to expose labial aspect of lesion. The labial cortex was very thin and had several areas of complete resorption. The lesion was enucleated along with impacted left lower permanent canine. The areas between roots of involved teeth were curetted well, the space was filled with resorbable tissue replacement (R.T. R granules) and flap was sutured in place (Figures 3, 4, 5, 6, 7, 8 and 9). Healing was uneventful, and there was no evidence of recurrence 6 months after the surgery (Figures 10 and 11).

Histopathological examination showed sheets of polygonal cells throughout the fibrous connective tissue stroma (Figure 2) The Ductal Lumina were surrounded by columnar epithelial cells and filled in some areas with eosinophilic material. In other places amorphous calcified material was present. The Histopathological report confirmed the diagnosis of Adenomatoid Odontogenic tumor.

Two years after surgery, a clinical and radiographic follow-up examination was performed. There was no evidence of recurrence and no apical resorption of the adjacent teeth could be observed.

4. Conclusion

Because of being the extra follicular variant of AOT, and with respect to the localization of the lesion in the mandible, the case is a rare case of AOTs. Additionally, it supports the above mentioned general description of AOT in the previous studies. We conclude that the rarity of AOT may be associated with its slowly growing pattern and symptom less behavior. Therefore, it should be distinguished from more common lesions of Odontogenic origin in routine dental examination.

5. Source of Funding

None.

6. Conflict of Interest

None.

References

1. Batra P, Prasad S, Parkash H. Adenomatoid odontogenic tumour: Review and case report. *J Can Dent Assoc*. 2005;71(4):250–3.
2. Becker T, Buchner A, Kaffe I. Critical evaluation of the radiological and clinical features of adenomatoid odontogenic tumour. *Dentomaxillofac Radiol*. 2012;41(7):533–40. doi:10.1259/dmfr/19253953.
3. Al-Shimari F, Chandra S, Oda D. Adenomatoid odontogenic tumor: Case series of 14 with wide range of clinical presentation. *J Clin Exp Dent*. 2017;9(11):1315–21. doi:10.4317/jced.54216.
4. Handschel JG, Depprich RA, Zimmermann AC, Braunstein S, Kübler NR. Adenomatoid odontogenic tumor of the mandible: review of the literature and report of a rare case. *Head Face Med*. 2005;1(1):1–3. doi:10.1186/1746-160X-1-3.
5. Jiang M, You M, Wang H, Xu L. Characteristic features of the adenomatoid odontogenic tumour on cone beam CT. *Dentomaxillofac Radiol*. 2014;43(6):1–9. doi:10.1259/dmfr.20140016.
6. Gomez RS, Castro WH, Gomes CC, Loyola AM. Adenomatoid odontogenic tumor associated with odontoma: a case report and critical review of the literature. *Head Face Med*. 2013;9(1):20. doi:10.1186/1746-160X-9-20.
7. Larson A, Swartz K, Heikinheimo K. A case of multiple AOT-like jawbone lesions in a young patient-a new odontogenic entity? *J Oral Pathol Med*. 2003;32(1):55–62. doi:10.1034/j.1600-0714.2003.00046.x.
8. Takahashi H, Fujita S, Shibata Y, Yamaguchi A. Adenomatoid odontogenic tumour: immunohistochemical demonstration of transferring, ferritin and alpha-one-antitrypsin. *J Oral Pathol Med*. 2001;30(4):237–44. doi:10.1034/j.1600-0714.2001.300408.x.
9. Gao YH, Yang LJ, Yamaguchi A. Immunohistochemical demonstration of bone morphogenic protein in odontogenic tumors. *J Oral Pathol Med*. 1997;26(6):273–7. doi:10.1111/j.1600-0714.1997.tb01236.x.
10. Shivali V, Pandey A, Khanna VD, Khanna P, Singh A, Ahuja T. A Rare Case of Extrafollicular Adenomatoid Odontogenic Tumour in the Posterior Region of the Mandible : Misdiagnosed as Residual Cyst. *J Int Oral Heal*. 2013;5(5):123–8.

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