Adenomatoid odontogenic tumor mimicking a lateral periodontal cyst – A rare case report in the mandible

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Abstract:
Adenomatoid odontogenic tumor (AOT) is benign tumor of the oral cavity characterized by its slow growth accounting to 3%–7% of the odontogenic tumors. AOTs mostly occur in the maxillary anterior region and most often than not associated with impacted anterior teeth. It affects the younger age group, especially below 20 years and is commonly seen in females. It occurs as two main variants – (a) central/intraosseous which is more common and (b) peripheral which is rare. The location of the lesion, its association with the impacted tooth, is the basis for the classification of AOT. The intraosseous type may be related to unerupted tooth (follicular variant) or may not be related to unerupted tooth (extra-follicular variant). Radiologically, AOT presents predominantly as a unilocular cystic lesion enclosing the unerupted tooth. The lesion presents rarely with a cystic component. Radiopacities on the intraoral periapical radiograph are seen as discrete foci having a flocculent pattern within radiolucency even with the presence of minimal calcified deposits. AOT being benign in nature is enucleated with least chances of recurrence. Here, we present a rare case of extra-follicular variant of adenomatoid odontogenic tumor in a 19-year-old female patient.

Key words:
Adenomatoid odontogenic tumor, benign ameloblastoma, extrafollicular, odontogenic tumor, unilocular

INTRODUCTION

Steenland described Adenomatoid odontogenic tumor (AOT) for the first time in 1905.[1] The lesion is also called as adenoameloblastoma, ameloblastic adenomatoid tumor, adamantinoma, epithelioma adamantium, and teratomatous odontoma. Currently, it is known as AOT. It is a well-established epithelial lesion of benign odontogenic origin. The tumor may be partly cystic, and sometimes, it may present as a solid lesion in the wall of a large cyst. It is believed to be a nonneoplastic lesion.[2] The definition states it to be of cystic nature, but very minimal literature is available describing the cystic lining. Harbitz in 1915, reported cystic presentation of AOT as “cystic adamantoma.”[3] Histopathologically, AOT is a pure epithelial odontogenic tumor and has an overall distinctive histomorphology which exhibits a remarkable consistency making its diagnosis easy and nonambiguous.[2,4,5]

Here, we present a case of AOT with the aim of providing an insight into the clinical and radiographic manifestation of the lesion.

CASE REPORT

A 19-year-old female patient reported with a chief complaint of swelling in the lower left back tooth region since 1 month. The swelling was slow growing in nature and gradually increased in size leading to marginal extraoral asymmetry of the face. She presented with no pathological or systemic disorder. A 1.5 cm × 2 cm diffuse swelling was palpable on the left side of the face. Superio-inferiorly, the swelling was seen to extend from the corner of the mouth to the inferior border of the mandible, antero-posteriorly from the left corner of the mouth up-to the middle third of the mandible. The swelling on palpation was not tender, hard, and fixed to the bone. The body of the mandible on the left side presented a solitary swelling extending from the gingival margin up to the vestibule from lateral incisor to second premolar was seen intraorally. The mucosa over the lesion was normal. The margins...
were diffused and ill-defined, the consistency varied from rubbery to hard in nature. The premolars were vital and exhibited Grade I mobility. Orthopantomograph revealed a pear shaped unilocular radiolucency of size 1.5 cm × 2 cm with well-defined sclerotic border was observed in the left body of the mandible. An intraoral periapical radiograph revealed a well circumscribed unilocular translucency surrounded by a distinct radiopaque lining. The roots of mandibular left canine and 1st premolar were seen to have drifted away from each other due to the pressure exerted by the growing lesion. Expansion of the buccal cortical plate was observed on the occlusal radiograph. Cone-beam computed tomography presented a radiolucent area in relation to the mandibular left premolar region and also showed mandibular nerve involvement in the first premolar region [Figure 1a-e]. The differential diagnosis included dentigerous cyst, lateral periodontal cyst, calcifying odontogenic cyst, and keratocystic odontogenic tumor.

Enucleation of the lesion was planned. After anaesthetizing, the area with 2% lignocaine hydrochloride with adrenaline 1:80,000 dilution, a crevicular incision was placed extending from the distal surface of the mandibular left lateral incisor up to the distal surface of 2nd premolar. Distal to mandibular lateral incisor a vertical releasing incision was placed. A full thickness flap was raised. The thin osseous lining of the lesion was carefully removed for clear visibility of the lesion. The cyst measuring 1.5 cm × 2 cm was enucleated carefully. The hollow cystic cavity was curetted to remove the granulation tissue and the cystic remnants. The enucleated cyst was sent for histopathological examination. Platelet-rich fibrin was placed in the hollow cystic cavity and 3.0 nonabsorbable silk sutures were placed [Figure 2a-e]. Nine months postoperatively on radiographic examination, root resorption was observed in the first premolar which was treated endodontically and mineral trioxide aggregate was placed. Histopathological examination under the scanner view showed one bit of tissue with cystic cavity and connective tissue stroma. A multinodular proliferation of spindle, cuboidal, and columnar cells in the variety of patterns comprising duct like structures and eosinophilic material delineated by a fibrous capsule of varying thickness was seen. Anastomosing strands of epithelial cells arranged in a cribriform, plexiform pattern between cell-rich nodules, and the presence of a whorled mass of columnar cells with a thin layer of homogenous eosinophilic material was also observed. Cystic component was also evident. Connective tissue stroma was highly vascular composed of numerous areas of hemorrhage, and extravasated red blood cells. Mild-to-moderate inflammatory infiltrate was also evident. Immunohistochemistry revealed the tumor to be immunopositive for CK 5 and 6, CK 14, CK 19, CK 34, and Beta catenin. The Ki67 proliferative index was observed to be approximately 3%. There was no evidence of any malignancy reported. The overall features were suggestive of AOT [Figure 3a-h].

The sutures were removed after 2 weeks and the healing was uneventful. The patient was under observation for more than 9 months postoperatively. No clinical as well as radiographic signs of recurrence was observed in this period. The orthopantomogram showed resolution of the lesion. The intraoral periapical radiography showed the tooth roots that had drifted away preoperatively to be returning back to their original position. The occlusal view showed remission of cortical expansion. Cone beam computed tomography confirmed the bone fill in the cystic cavity [Figure 4a-e].

**DISCUSSION**

AOT is a slow growing epithelial tumor, but of a nonaggressive nature; is benign and also rare. It originates from the epithelial component of the tooth forming tissues, but no morphological defects were observed in the associated teeth. Thus, the disturbance must occur after odontogenesis is complete.[6] AOT is mainly of central/intra-osseous nature, has two variants, the follicular type (73%) and extra-follicular type (24%) and other being peripheral in nature, is rare (3%). The extra-follicular type is subdivided into E1-where the tumor has no relation to the tooth structure either erupted or unerupted, E2 – where the tumor causes the intra-radicular adjacent roots to diverge apically due to expansion, E3 – where the tumor is superimposed on root apex and E4 – where the tumor is superimposed on the mid root level.[7]

In the present case, there was divergence of adjacent roots seen as seen in the E2 variant of extra-follicular AOT. Although typically AOT is seen in the maxilla, in the present case, it was seen to be occurring in the mandible. The tumor commonly is called as “two third tumor,” as (a) two-third of the cases are seen in maxilla, (b) two-third of the affected individuals are young females, (c) two-third of the cases are seen to occur in association with un-erupted tooth, and (d) two third to be affected are the canines.[8] However, this lesion may appear as other odontogenic tumor/ cyst.[9] However, here, the presentation was in a young female and in the mandibular canine and premolar region. Hence, only 2 out of the 4 features mentioned for the two-thirds tumor matched in the case presented.

Advanced stage of AOT shows swelling. As it is painless, patients delay the treatment that increases the risk of complications which includes the facial asymmetry and/or functional disorders commonly.[10] In the case presented, a painless well-defined lesion was seen with marginal facial disfigurement. Radiograph showed unilocular lesion with root divergence. Histologically, all the three variants of AOT have common features indicating common origin; derived from the complex system of dental lamina or its remnants. Gubernaculum dentis (GuDe) is considered to be of importance in the development of AOT. GuDe comprises two structures, the gubernacular cord (GuCo) and the gubernacular canal (GuCa). Philipsen et al. state that AOT is derived from odontogenic epithelium of the dental lamina complex or its cellular remnants that is located in the GuCo.[11]

The lesion is considered controversial; it is considered as a hamartoma or a neoplasm of benign nature or sometimes even as a developmental anomaly or a cyst. Clinically, its limited size, inability to recur and metaplastic mineralization warrants it to be considered as a hamartoma.[12] A study of Ki-67 and B-cell lymphoma 2 expression in ameloblastoma and AOTs suggested the latter to be less proliferative in comparison to the former, pointing toward its hamartomatous nature.[13] As it is slow growing and can be detected early
before reaching a clinically appreciable size, it has also been described as a neoplasm. Immunohistochemistry protein expression in AOT has been in the last 10 years widely described in the literature. Reduced dental epithelium has been attributed as the probable origin of AOT due to the CK14 expression. As the staining is seen to be positive with CK5, CK17, and CK19, it confirms the classical AOT phenotype characterized by a CK profile that is very similar to dentigerous cyst and/or oral or gingival epithelium. 

The mild nature of AOT has been attributed to the low proliferation marker Ki67. The observation of cases by Neha S, Santosh M, Sachin MG, Poonam SR, Simranjit S, Abdul KA showed no root resorption, but displacement of the adjoining teeth and nonassociation of the tumor with impacted tooth in two cases. Whereas; the case reported here presented with resorption of the root, 

Figure 1: (a) A well-circumscribed swelling extending from mandibular left lateral incisor to the 1st premolar; (b and c); Orthopantomograms and Intraoral periapical reveal a well-circumscribed pear-shaped radiolucent lesion in relation to mandibular left canine and 1st premolar; (d) Occlusal radiograph showing expansion of buccal cortical plate and a thin sclerotic lining; (e) Cone-beam computed tomography showing a pear-shaped bone defect involving both buccal and lingual bony plate

Figure 2: (a) Incision placed; (b) Cyst being enucleated; (c) Hollow cystic cavity; (d) PRF prepared to be placed in the cystic cavity; (e) Sutures placed

Figure 3: Immunohistochemistry analysis revealing tumor to be immunopositive for; (a) H and E stain showing duct like structures arranged in plexiform pattern; (b) H and E stain showing rosette like structure; (c) CK 5 and 6; (d) CK 14; (e) CK 19; (f) CK 34 Beta E2; (g) Beta catenin; (h) Ki67

Figure 4: 9-month postoperative pictures; (a) Clinically healing was satisfactory; (b) Orthopantomograms showing resolution in the size of lesion; (c) Intraoral periapical view showing resolution of cortical expansion discrepancies; (d) Cone-beam computed tomography view showing bone formation in canine premolar region
displacement of the adjoining teeth and also non-association with impacted tooth.

CONCLUSION

Over the past years, many researchers have provided vast literature on the description of AOT and its treatment option. An unusual extrafollicular type of AOT, with its characteristic feature of occurrence in female in second decade of life but in the canine – premolar region on the left side of the mandible is presented here. Surgical excision is the treatment of choice. Careful follow-up should be considered as recurrence of the lesion has been reported. However, over a period of 9 months, recurrence of the lesion was not observed.

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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Conflicts of interest

There are no conflicts of interest.

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