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Case Report

Extrafollicular adenomatoid odontogenic tumor: a case report

Sankar Vinod Vichattu, Arun George, Joy R. Das, Atic Thomas, Athira K. Sasi*

Department of Oral and Maxillofacial Surgery, Mar Baselios Dental College, Kothamangalam, Kerala, India

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***Correspondence:**
Dr. Athira K. Sasi,
E-mail: athiraks1996@gmail.com

ABSTRACT

Adenomatoid odontogenic tumor (AOT), a benign epithelial odontogenic tumor, constitutes around 2-7% of all odontogenic tumors that are biopsied. This lesion is unique with regard to its clinical, radiographic, and histopathological features making its diagnosis more predictable. This article emphasizes an extrafollicular variant of AOT leading to maxillary anterior jaw swelling along with cortical plate expansion, perforation, and root resorption within a short duration of time pointing towards its aggressive nature.

Keywords: Adenomatoid odontogenic tumor, Extrafollicular variant, Enucleation, Root resorption

INTRODUCTION

AOT is not a new entity in the literature, World Health Organization (WHO) in 1971 adopted the term 'adenomatoid odontogenic tumor' till then various terminologies were used to describe AOT. The name adenomatoid is given because histologically the tumor shows numerous duct-like structures interposed within the lesion giving rise to a glandular or adenomatoid appearance.

Terminologies to describe AOT evolved from epithelioma adamantium by Steenslandin in 1905, pseudo-adamantium by Dreybladt in 1907, glandular adamantinoma by L'Esperance in 1910, and cystic adamantoma by Harbitz in 1915. Stafne in 1948 coined it as a distinct entity, still others described it as an ameloblastoma variant. Philipsen and Birn suggested 'adenomatoid odontogenic tumor' terminology and WHO later adopted it.[1]

Odontogenic tumors were classified primarily based on whether they are biologically benign or malignant. 2005 classification divided the benign tumors into "Odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation, Odontogenic epithelium without odontogenic ectomesenchyme, and Odontogenic ectomesenchyme with or without odontogenic epithelium." AOT is categorized under odontogenic epithelium with odontogenic ectomesenchyme, with or without hard tissue formation subclass. While accurate, this seemed overly complex and the 2017 version recognized epithelial, mesenchymal, and mixed tumors. Now it is categorized under benign tumor of epithelial origin.[2]

CASE REPORT

A 22-year female patient reported a chief complaint of swelling in the upper right front tooth region since one month. Swelling increased in size within a short duration. No pain, pus discharge, and trauma history were reported. Extra oral examination unveiled a diffuse solitary swelling of size 3.5×4 cm, with a shiny surface extending anteroposteriorly from the lateral aspect of the nose to about 4 cm in front of tragus and superoinferiorly approximately 1 cm below the infraorbital rim to right corner of mouth, also 1cm above with nasolabial fold obliteration and lifting right ala of nose causing gross facial asymmetry. On palpation, firm consistency,

non-tender, non-reducible, non-compressible, and non-fluctuant lesion was found (Figure 1).

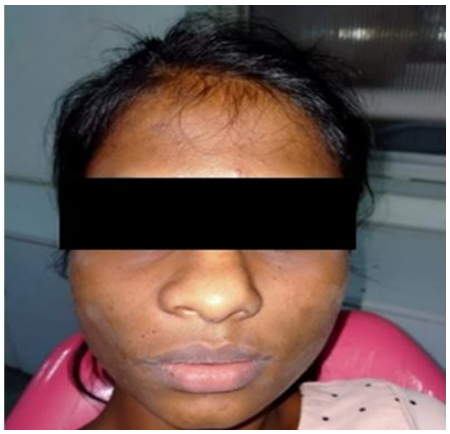

Figure 1: Extraoral image. Note the ala of nose lifted upward due to swelling on right side.

On intra-oral examination a solitary swelling on right labial vestibule, measuring approximately 3x4 cm, oval in shape with a shiny surface extending from the labial frenum to the buccal frenum of right side anteroposteriorly and with vestibular obliteration (Figure 2). On palpation, swelling was soft in consistency, non-tender, non-compressible, and non-reducible. Grade II mobility noted with respect to 11,12,13,14 and 12,13 was found to be deviated. Pulp vitality test showed non vital 13 and delayed response with respect to 11,12,14.

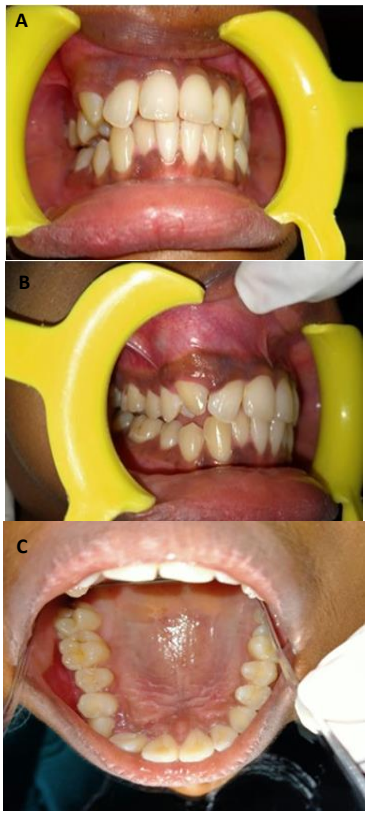

Figure 2 (A-C): Intraoral images of the lesion.

Blood investigations were within normal limits. Panoramic view disclosed a well defined unilocular radiolucent lesion approximately 29.60x26.30 mm in size extending from the apical third of 11 to the apical third of 15 and from the crest of alveolar ridge between 12 and 13 to involve the floor of right maxillary sinus superiorly with sclerotic borders. The long axis of 12 was tilted mesially with respect to 11. The long axis of 13,14 and 15 were tilted distally with respect to 16 (Figure 3 and 4).

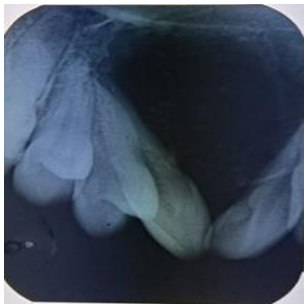

Figure 3: Intraoral periapical radiographic image.

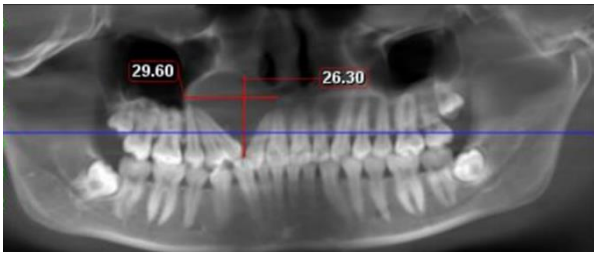

Figure 4: Distinct, radiopaque margins of a well-circumscribed radiolucent lesion seen in panoramic radiograph.

Cone beam computer tomography (CBCT) revealed well-defined non-homogenous area of soft tissue density with multiple pinpoint hyperdense areas distributed within the lesion. Ballooning expansion of the buccal cortical plate was obvious (Figure 5). Discontinuity was noted in the buccal and alveolar cortical plate, floor of right nasal cavity, and anterior border of nasopalatine canal (Figure 6). The contents of the lesion were displaced superiorly elevating floor of right maxillary sinus. Root resorption noted with respect to 11 and 14 (Figure 7).

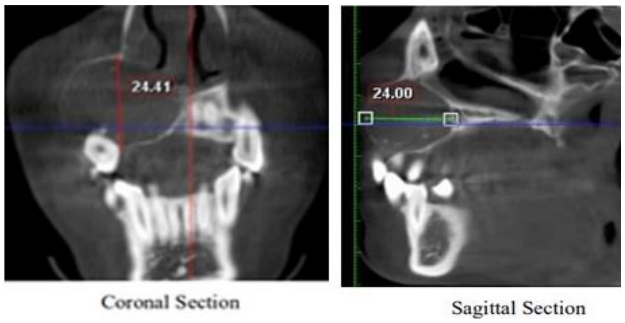

Figure 5: CT images showing coronal and sagittal sections.

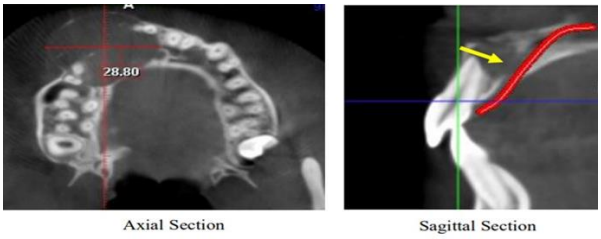

Figure 6: Axial section and sagittal section showing discontinuity of buccal cortex and anterior border of nasopalatine canal (yellow arrow) respectively.

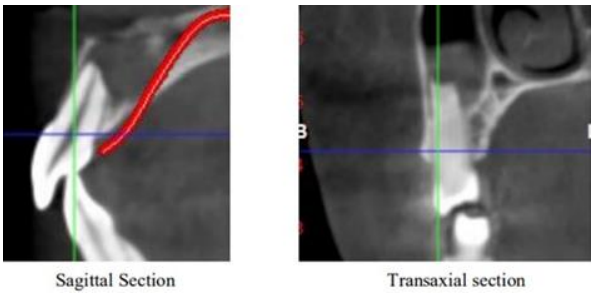

Figure 7: Root resorption noted in 11 and 14.

Based on the above clinical and radiographical presentation and the presence of fine calcifications (snowflakes) provisional diagnosis was given as calcifying epithelial odontogenic cyst (CEOC), and the differential diagnosis was given as AOT, dentigerous cyst, odontogenic keratocyst, unicystic ameloblastoma and calcifying epithelial odontogenic tumor (CEOT). Under general anesthesia surgical excision was done and a histopathological examination of the specimen was taken (Figure 8 and 9).

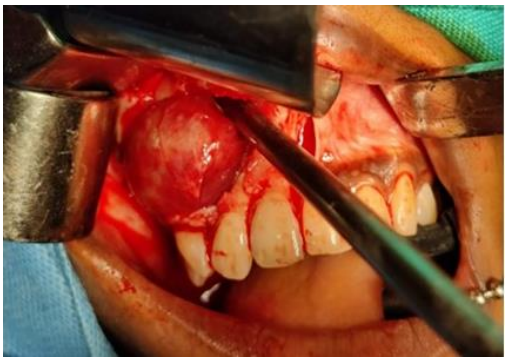

Figure 8: Surgical removal of the lesion.

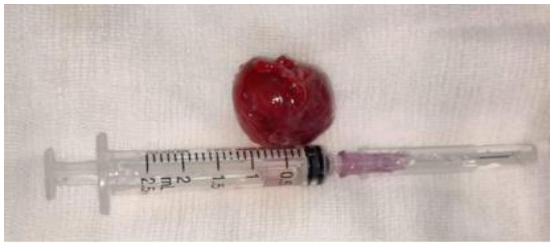

Figure 9: Enucleated specimen.

Results revealed a well-encapsulated tumor mass, made of proliferating odontogenic epithelial cells in the form of ducts, whorled mass, sheets, and rosettes pattern with small foci of calcification, and hence the lesion was confirmed as AOT. Post surgically composite splinting was done on the palatal aspect of 15, 14, 13, 12, 11, and 21 to stabilize the mobile teeth, with a follow-up of 3 months and root canal treatment (RCT) was done (Figure 10).

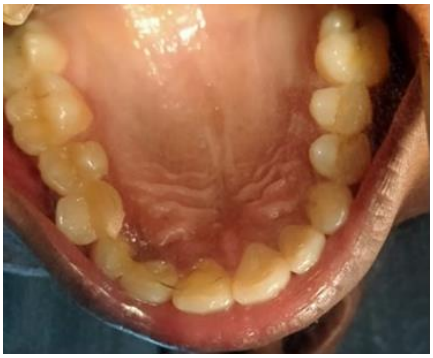

Figure 10: Palatal splinting done after 2 days of procedure.

DISCUSSION

AOT is a neoplasm with locally invasive nature characterised by ameloblastoma like islands of epithelial cells in a mature connective tissue stroma. Keratinization is seen with varying amounts of dysplastic dentin.[3]

AOT is classified under benign odontogenic tumor arising from odontogenic epithelium, indicating its origin from an enamel organ, reduced enamel epithelium (REE), Mallasez rests or dental lamina rests.

Crivelini et al conducted an immunohistochemistry study on the origin and nature of AOT and concluded that those tumors which have origin from REE are likely to have inductive capacity and can be neoplastic in nature, while those arising from rests of dental lamina or gubernacular dentis can result in hamartomatous malformation.[4]

Philpsen et al gave the concept of gubernaculum dentis in the development of AOT. Gubernaculum dentis is a bony crypt that lies between the lingual crypt of primary and permanent teeth. Whenever a permanent tooth follicle develops into gubernaculum dentis, there will be formation of a follicular variety of AOT, and if it bypasses gubernaculum dentis, it gives rise to an extrafollicular variant of AOT. The follicular variant of AOT clinically and radiologically resembles that of a dentigerous cyst, therefore AOT is considered as a master of disguise or a perfect imitator of dentigerous cyst.[5]

This tumor commonly affects young females with a predominance of 1.9:1 ratio, maxilla more than mandible, and anterior region more frequently than posterior region;

as in our case.[6] Unerupted maxillary canine is the commonly associated tooth, but in our case, there was no impacted tooth, making it an extrafollicular variant. Although it is common in maxillary anterior region, it has also been reported in the mandibular angle region.[7]

As mentioned earlier profuse terminologies were put forth to describe AOT, one such is 2/3rd tumor, because: (1) 2/3rd occur in the maxilla, (2) 2/3rd of young females, (3) 2/3rd in unerupted tooth and (4) 2/3rd of affected teeth were canines.

AOT usually doesn't go beyond the limit of 1-3cm in diameter. There were only a few reports of large AOTs in the literature. This can be due to higher growth potential in young individuals or delay in treatment.[8]

AOT was recognized under three variants, i.e., Follicular, Extra follicular, and Peripheral, with identical histology.

Follicular variant: associated with an embedded tooth, mostly a permanent canine, which accounts for 73% of AOT cases. AOT surrounds the entire involved tooth, while a dentigerous cyst surrounds only the coronal part of an impacted tooth.

Extrafollicular variant has no pericoronal or other relationship with an impacted tooth, they resemble periapical cyst radiographically and accounts for about 24% of AOT.[1]

The peripheral variant, was seen on the gingival mucosa as a gingival fibroma or epulis clinically and accounts for about 3% of AOT.[1]

The histopathological features of AOT are very diverse. The tumor is mostly well-encapsulated which accounts for its easy enucleation. AOT can either be completely solid, partially, or majorly cystic, which brings on the argument of considering it as a cyst or a tumor among researchers. AOTs are usually cellular with less amount of connective tissue, epithelial cells may be polygonal, spindle, columnar, cuboidal, or stellate-shaped in solid nodules, rosette patterns, or whorls. The most distinct pattern is a duct-like arrangement, formed by a columnar or cuboidal single layer of cells with nuclei away from the lumen. The lumen can be empty or may have eosinophilic material lined by an eosinophilic rim called a hyaline ring. These duct-like structures resulted in the term AOT. Studies have shown that these structures are not ducts, rather they are closed spherical cysts. Calcification of varying amounts was also found within the tumors.[9]

A gross examination of the tumor revealed a distinct fibrous capsule. Upon sectioning white to tan; solid to crumbly tissue with cystic spaces; yellow-brown fluid to semisolid material and gritty granular materials can also be found with several calcified masses. Intact specimens showed an impacted tooth crown projecting into the cystic cavity in the tumor.[10]

A unilocular radiolucent area with a distinct radiopaque border is a radiographic feature of AOT1. Bartake et al in 2009 reported a case of multilocular radiolucency associated with AOT.[11] This tumor often results in the displacement of the adjacent tooth, but root resorption is uncommon1, and it was noted in our case. Root resorption by AOT is rarely reported in the literature. Faintly detectable radiopaque foci of varying sizes were also found within the radiolucent lesion of AOT. These radiopacities are best diagnosed with an intraoral periapical radiograph when compared to a panoramic radiograph.[12] Extra-osseous variants are seldom detected radiographically.

Multiple AOTs are seen in association with Sebaceous nevus syndrome or Schimmelpenning syndrome (SS), a congenital neurocutaneous disorder identified by linear nevus sebaceous, with neurologic, ophthalmic, and skeletal disabilities. Rarely oral manifestations like bifid uvula, exophytic papillomatous growth in the mucosa, and fibromatous enlargement of the tongue are seen in SS. It is also found to be associated with odontoma, ameloblastic fibro-odontoma, ameloblastoma, and central giant cell lesion.[13]

AOTs are sometimes found to be associated with dentigerous cysts and along with other odontogenic tumors.[10] Conservative management usually produces excellent outcomes without recurrence. Treatment should consist of enucleation and curettage as it is a benign tumor with non-aggressive biologic behavior, absence of invasion, progressive growth, and presence of connective tissue capsule, rarely recurrence has been reported. But in case of unusual findings, a longer follow-up period is mandatory.[14]

CONCLUSION

To consider AOT as a hamartoma or benign neoplasm is still a puzzle. Due to its limited size of the growth, lack of recurrence, and metastatic mineralization it favors the hamartoma concept, while microscopic features show a greater departure from normal odontogenic epithelium, it can also be considered as a neoplasm. Limited size can be because they are detected early because of their anatomical location, age, and sex predilection; hence, they are removed before they grow into their full size.

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