



CASE REPORT

Adenomatoid Odontogenic Cyst: A Rare Case Report

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ABSTRACT

Adenomatoid odontogenic cyst (AOC) is a benign, slow growing, relatively uncommon lesion of odontogenic origin. Histogenesis of AOC is still uncertain; however, it is often considered as a hamartomatous lesion rather than a true neoplasm. It is described as a cyst that has a hamartomatous intraluminal proliferation of epithelial cells derived from Hertwig's epithelial root sheath. It usually presents as an expansile lesion in maxillary anterior region. Adenomatoid odontogenic cyst is characterized histopathologically as well-demarcated cysts that typically appear with intraluminal masses. In the present paper, we report a rare case of AOC, thereby emphasizing the terminology and the histoarchitectural spectrum.

Keywords: Adenomatoid odontogenic cyst, Hamartoma, Hertwig's epithelial root sheath, Odontogenic.

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INTRODUCTION

Adenomatoid odontogenic cyst (AOC) is a benign, slow growing, relatively uncommon lesion of odontogenic origin.¹ Adenomatoid odontogenic cyst accounts for about 1 to 9% of all odontogenic lesions. It is best considered as a hamartomatous proliferation rather than neoplasm.² It is known to arise from Hertwig's epithelial root sheath as a hamartomatous intraluminal proliferation of epithelial cells.³ The lesion is most frequently encountered in the second decade of life (68.6%) and 53.1% of cases occur within 13 to 19 years of age. It has a female predilection in almost 2:1 ratio.¹ The cyst presents as an expansile lesion most commonly in the anterior maxillary region.³ The lesion is asymptomatic but may cause cortical expansion and displacement of the adjacent teeth. Adenomatoid odontogenic cyst is usually associated with an impacted tooth, most often involving unerupted permanent canine.^{1,2}

In the present paper, we report a case of AOC in anterior maxillary region, thereby emphasizing the terminology and the histoarchitectural spectrum.

CASE REPORT

A 19-year-old male patient presented at our institute with a firm, nontender swelling of right maxillary region, since 1 month (Fig. 1). The patient was under medication for epilepsy since past 10 years. The lesion presented as a diffuse swelling extending superoinferiorly from the infraorbital region to alae of right nasal cavity and anteroposteriorly till the zygomatic process. Intraorally, a diffuse swelling was observed extending from 11 to 15 region obliterating the buccal vestibule (Fig. 2). Orthopantomography (OPG) showed radiopacity in relation to maxillary sinus with over retained 53 and impacted 13. Computed tomography showed a well-defined radiolucency with spicules of radiopaque structure and the associated impacted tooth. Obliteration of maxillary



Fig. 1: Extraoral view shows diffuse swelling extending over the right maxillary region



Fig. 2: Intraoral view shows diffuse swelling extending from 11 to 15, obliterating the buccal vestibule

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Fig. 3: Computed tomography shows well-defined radiolucency with spicules of radiopaque structure and the associated impacted tooth. Obliteration of maxillary sinus was also observed

sinus was also observed (Fig. 3). A provisional diagnosis of dentigerous cyst was given.

Incisional biopsy of the lesional tissue microscopically revealed, multinodular proliferation of spindle, cuboidal, and columnar cells in a variety of patterns comprising of rosettes, scattered duct-like structure, hyaline ring, and calcifications with few areas of hemorrhage (Fig. 4). A diagnosis of adenomatoid odontogenic tumor (AOT) was made. Surgical enucleation was done (Fig. 5). The gross excised specimen received was white to black in color measuring $3.0 \times 3.0 \times 2.0$ cm. The lesion was firm in consistency, giving a cystic sac appearance with an irregular surface contour. Microscopically, a cystic space surrounded by thin nonkeratinized stratified squamous epithelial lining of two to three-cell thickness was seen

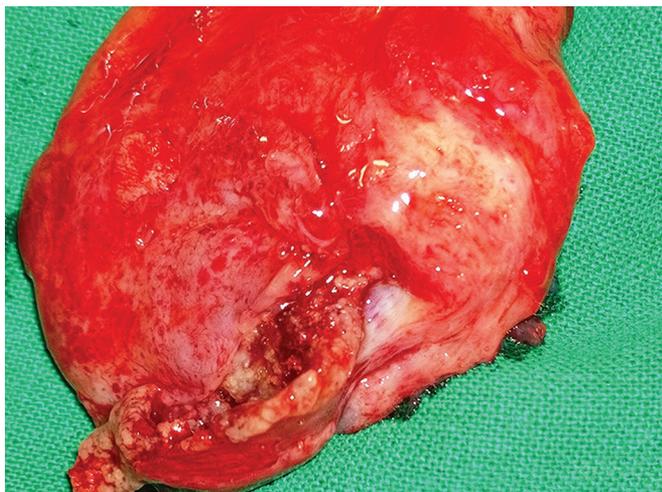


Fig. 5: Surgically enucleated specimen

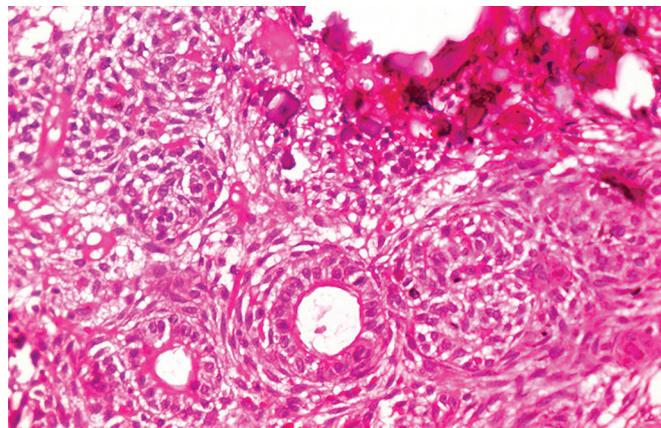


Fig. 4: Hematoxylin and eosin-stained (400 \times) soft tissue section shows duct-like spaces lined by tall columnar cells with polarized nuclei. Few foci of calcifications are also noticed

(Fig. 6). A cribriform pattern of the epithelial lining was evident at places. Lace-like pattern of proliferating epithelium was also observed with juxta-epithelial hyalinization (Fig. 7). A focal area pathognomonic of AOT was also observed (Figs 8 and 9). The histopathological features confirmed the diagnosis of AOC.

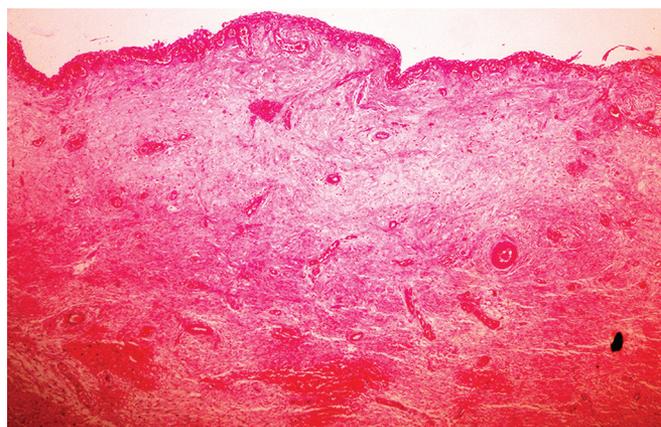


Fig. 6: Hematoxylin and eosin-stained (40 \times) soft tissue section shows cystic space lined by a thin nonkeratinized stratified squamous epithelial lining of two to three-cell thickness

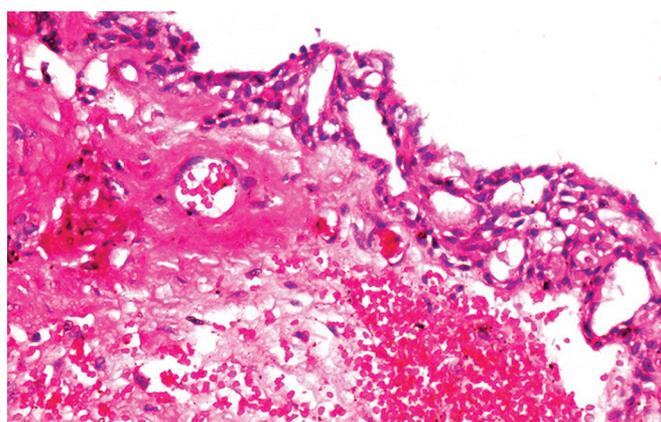


Fig. 7: Hematoxylin and eosin-stained (100 \times) soft tissue section shows lace-like pattern of proliferating epithelium with juxta-epithelial hyalinization

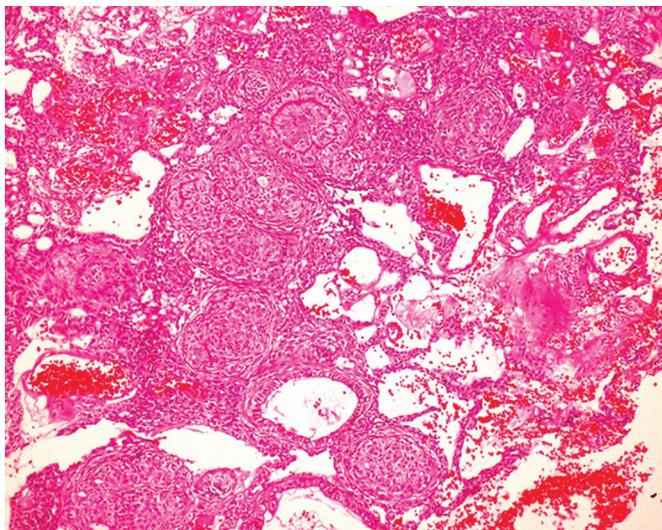


Fig. 8: Hematoxylin and eosin-stained (100×) soft tissue section shows multinodular proliferation of spindle, cuboidal, and columnar cells in a variety of pattern comprising of rosettes, whorls, and scattered duct-like structure

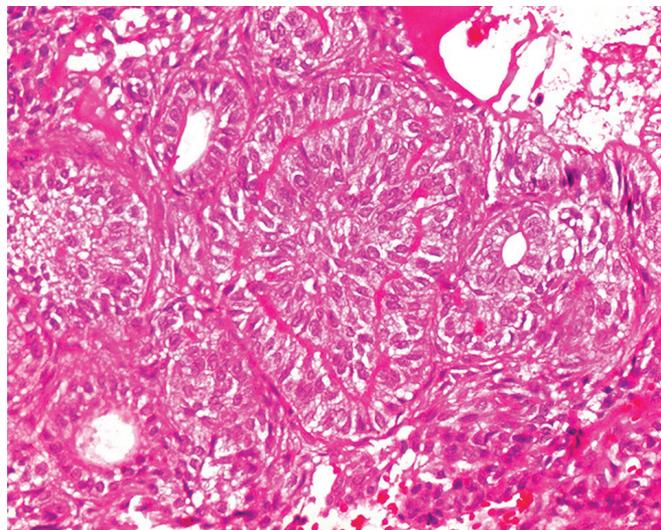


Fig. 9: Hematoxylin and eosin-stained (100×) soft tissue section shows rosette and scattered duct-like structure

DISCUSSION

The term AOC has been proposed by Marx and Stern. They have described the lesion as a hamartomatous intraluminal proliferation of the epithelial cells derived from Hertwig epithelial root sheath (HERS), which fills the cystic space, giving a solid appearance. It has also been referred to as two-thirds tumor as about two-thirds occur in maxilla, two-thirds occur in young women, two-thirds are associated with unerrupted tooth, and two-thirds of those unerrupted teeth are canine.³ Regezi et al⁴ described the lesion as an intracystic epithelial proliferation composed of polygonal and spindle cells. Although cystic presentation of AOT has been reported way back in 1915 by Harbitz who reported the lesion as “cystic adamantoma,” a systematic review of literature, however, reveals very few cases.⁵ Gadewar et al⁶ described a similar cystic presentation and diagnosed it as cystic AOT. Kurra et al⁷ in their case report of AOC designated it as a hamartomatous odontogenic cyst. Uppada et al⁸ reported a case of AOC that was earlier misdiagnosed as a dentigerous cyst on incisal biopsy.

Microscopically, AOCs are well-demarcated cysts that typically appear with intraluminal or even intramural proliferating masses.³ The cystic space is usually lined by 3 to 10-cell thick nonkeratinized stratified squamous epithelium that might exhibit nodule formation. The cystic lining may also show focal areas giving origin to strands of smaller cuboidal cells going into the connective tissue in a lace-like pattern. The juxta-epithelial connective tissue shows hyaline material, surrounded by a vascular stroma.⁶ The epithelial cells may exhibit

a bimorphic appearance with spindle-shaped cells and cuboidal or columnar cells. The spindle-shaped cells may form sheets, strands, or whorled nodules and rosettes. Areas of duct-like spaces lined by a single layer of tall columnar cells with polarized nuclei may also be evident. Calcifications of varying degrees are present and may occur in small droplets. These calcifications may resemble dentin or cementum that would further confirm AOC's histogenesis from Hertwig root sheath.³ Our case was in accordance to similar microscopic features.

Adenomatoid odontogenic cyst should be included in the differential diagnosis of corticated radiolucency with small radiopaque foci.² Radiographically, the cyst appears well-demarcated, unilocular radiolucent lesion and may contain fine calcification. They are usually associated with an impacted tooth. Frequent displacement of roots of adjacent teeth may also be observed but irregular root resorption is rare. Adenomatoid odontogenic cysts that appear without radiographic evidence of calcification will be most suggestive of the more common dentigerous cyst. Adenomatoid odontogenic cyst is known to arise from Hertwig's epithelial root sheath, whereas dentigerous cyst arises from follicle of tooth crown. This explains the attachment of cystic lining of AOC on root surface completely enveloping the tooth, whereas in dentigerous cyst, the attachment is at clinical attachment level (CEJ).³ Gadewar et al⁶ suggested that absence of ameloblast-like cells and ameloblastoma-like proliferation in the lining epithelium could exclude the possibility of unicytic ameloblastoma. Adenomatoid odontogenic cyst in which calcifications can be observed resemble a calcifying odontogenic cyst (COC). The presence of ghost cells, characteristic of COC, is distinctive for the differentiation.⁶

Adenomatoid odontogenic cyst is considered to be a slowly growing benign lesion; hence, a conservative surgical enucleation or curettage is sufficient. The lesion is encapsulated by a thick connective tissue capsule that readily separates from its bony crypt, and hence recurrences are rare.³ However, Xiang and Yan,⁹ in their review of 16 cases, reported one case that recurred twice over a period of 20 years. Gadewar et al⁶ in their review have also reported four cases of recurrences. Although the prognosis is considered excellent, regular follow-up is necessary.² Our case was treated with surgical enucleation with no recurrence reported in 2 years follow-up period.

CONCLUSION

Herein, the goal of this paper is to add on one more case of this rare entity to the literature. The rarity of AOC may be associated with its slowly growing pattern and benign behavior. Therefore, it should be always distinguished from more common lesions of odontogenic origin in routine dental examination. Also, uncommonness of such cases hinders any lasting conclusions regarding the lesions behavior. Thus, it necessitates a periodic review of these cases so as to understand the actual incidence, biological behavior, and outcome associated.

REFERENCES

1. Gupte S, Shetye A, Chhadva S, Malusare P. Adenomatoid odontogenic cyst of the mandible: report of a case. *Int J Sci Res* 2015;4(7):656-658.
2. Kumar R, Hashmi GS, Amjad SM. Adenomatoid odontogenic cyst of mandible: a rare case report. *Int J Health Sci Res* 2015;5(5):490-494.
3. Marx RE, Stern D. *Oral and maxillofacial pathology: a rationale for diagnosis and treatment*. Hanover Park: Quientessence Publishing; 2003. p. 609-612.
4. Regezi JA, Sciubba JJ, Jordan RCK, editors. *Odontogenic tumours. Oral pathology clinical pathologic correlation*, Fourth edition. St Louis, Missouri: Saunders; 1999. p. 276-277.
5. Harbitz F. On cystic tumours of the maxilla, and especially on adamantine cystadenomas (adamantomas). *Dental Cosmos* 1915;57:1081-1093.
6. Gadewar DR, Srikant N. Adenomatoid odontogenic tumour: tumour or a cyst, a histopathological support for the controversy. *Int J Pediatr Otorhinolaryngol* 2010 Apr;74(4):333-337.
7. Kurra S, Gunupati S, Prasad P R, Raju. Y S, Reddy BVR. An adenomatoid odontogenic cyst (aoc) with an assorted histoarchitecture: a unique entity. *J Clin Diagn Res* 2013 Jun;7(6):1232-1235.
8. Uppada UK, Salavadi R, Agarwal A, Paul D. Adenomatoid odontogenic cyst mimicking dentigerous cyst: a case report. *J Cranio Max Dis* 2015;4(1):90-94.
9. Xiang ZC, Yan G. Adenomatoid odontogenic tumour: a report of a rare case with recurrence. *J Oral Pathol Med* 2007 Aug;36(7):440-443.