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.

INTRODUCTION

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. Adenomatoid odontogenic cyst accounts for about 1 to 9% of all odontogenic lesions. It is best considered as a hamartomatous proliferation rather than neoplasm. It is known to arise from Hertwig’s epithelial root sheath as a hamartomatous intraluminal proliferation of epithelial cells. 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. The cyst presents as an expansile lesion most commonly in the anterior maxillary region. 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.

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 sinus and displacement of teeth were noticed. The case was planned to be operated and treated by extraction of impacted tooth.

In the present paper, we report a case of AOC in anterior maxillary region, thereby emphasizing the terminology and the histoarchitectural spectrum.
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 × 3.0 × 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. 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.
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 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. However, Xiang and Yan, in their review of 16 cases, reported one case that recurred twice over a period of 20 years. Gadewar et al in their review have also reported four cases of recurrences. Although the prognosis is considered excellent, regular follow-up is necessary. 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