CASE REPORT

Adenoid ameloblastoma with dentinoid: A rare hybrid odontogenic tumor

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ABSTRACT

Background: Adenoid ameloblastoma with dentinoid (AAD) is a hybrid odontogenic tumor comprising histopathological presentation of ameloblastoma (AM) and adenomatoid odontogenic tumor (AOT) along with extracellular dentinoid material.
Case Presentation: A 35-year-old female reported an asymptomatic swelling in the left mandibular posterior region. Histopathological examination revealed composite features of AM with AOT along with dentinoid material, which stained positively with Van Gieson and trichrome stains.
Conclusion: The present case report serves to add further to the modicum of literature reports pertaining to AAD, which may gain recognition as a distinct entity in future World Health Organization (WHO) classification of odontogenic tumors.

KEY WORDS: Adenoid odontogenic tumor, hybrid tumors, immunohistochemistry

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INTRODUCTION

Two or more different but exactly defined tumor entities co-existing within the same lesion have been described as hybrid tumors. These have been defined as "a lesion showing the combined histopathological characteristics of two or more previously recognized tumors and/or cysts of different categories" by Ide *et al.*[1]

One such peculiar hybrid variant exhibits a histological picture which is a composite of features of ameloblastoma (AM) and adenomatoid odontogenic tumor (AOT). The lesion was first reported by Slabbert *et al.* in 1992 as "dentinoameloblastoma," whereas the term "adenoid ameloblastoma with dentinoid" (AAD) was first suggested by the Armed Forces Institute of Pathology (AFIP) in 1994.[2] Over time, the lesion has been ascribed with various terminologies such as "atypical plexiform ameloblastoma with dentinoid," "ameloblastoma with features of dentinoid," "hybrid ameloblastoma and adenomatoid odontogenic tumor," and "atypical adenoid ameloblastoma."[3]

The lesion is extremely rare with less than 30 cases reported in the literature till date.[4-6] Here, we report a case of AAD, emphasizing its aggressive clinical nature and histopathologic features with an objective to add further to the modicum of literature reports pertaining to this unusual hybrid odontogenic tumor.

CASE REPORT

A 35-year-old female patient reported an asymptomatic swelling in the left posterior mandibular region which had gradually increased in size over the past 6 months. No history of medical illness or trauma was elicited; however, a history of spontaneous exfoliation of the mandibular left second molar 3 months ago was alluded by the patient. A hard, non-tender, diffuse swelling was noted of size 8 cm × 5.5 cm on the left side of the lower jaw extending from the corner of the mouth until 1 cm in front of the lower border of the tragus anteroposteriorly

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was notable extra orally [Figure 1a]. Intraoral examination revealed a well-defined, firm, and nontender swelling extending from the distal of the second premolar to the left pterygomandibular raphe region posteriorly, obliterating the buccal vestibule [Figure 1b]. Buccal and lingual cortical plate expansion was noted without sinus tract or pus discharge. The differential diagnosis for the lesion after clinical examination included AM, dentigerous cyst, odontogenic keratocyst (OK), and central giant cell granuloma.

An orthopantomogram revealed a well-defined multilocular radiolucency extending anteroposteriorly from the mesial root of the left mandibular first molar up to the ramus of the mandible [Figure 1c]. Superoinferiorly, the lesion extended from the alveolar ridge until the mandibular lower border displacing the mandibular third molar inferiorly. Resorption involving more than two-thirds of the distal root and apical third of the mesial root of the first molar was noted.

Cone-beam computed tomography illustrated a large, expansile multilocular hypodense lesion along the left mandibular posterior region with impacted left mandibular third molar within the interior of the lesion, which was displaced towards the inferior border of the mandible. The margins of the lesion appeared to be partially corticated with thinning and perforation of the buccal and lingual cortical plates. The inferior alveolar nerve canal appeared to be displaced to the lower border of the mandible. Root resorption was noted in relation to the distal root of the left mandibular first molar.

On histopathological examination of the tissue following an incisional biopsy, odontogenic epithelium proliferating predominantly in the form of interconnecting plexuses as well as follicles in a background of fibrous connective tissue stroma was observed [Figure 2a]. The odontogenic follicles and plexuses were lined by cuboidal to tall columnar ameloblast-like cells with hyperchromatic nuclei exhibiting reversal of polarity and stellate reticulum-like cells were present centrally. Confluent with these areas resembling a classical AM, areas of spindle-shaped to low cuboidal epithelial cells proliferating in the form of sheets, cords, and whorls were noted [Figure 2b and d].

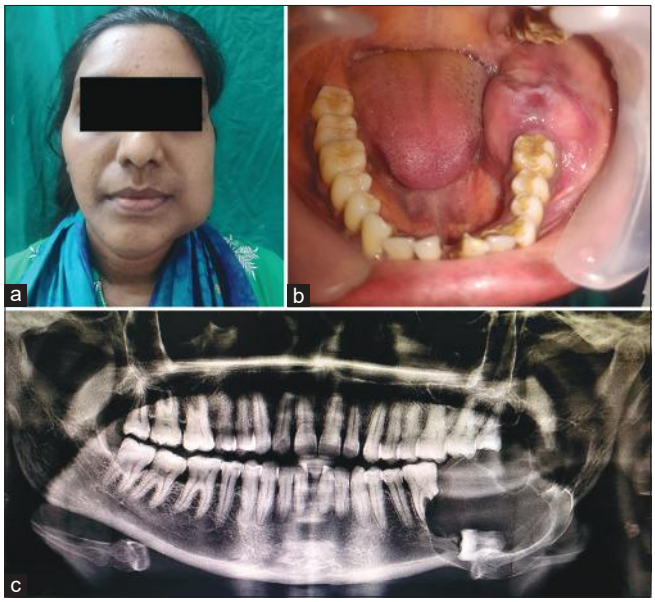

Figure 1: Well-defined swelling noted on clinical examination (a) Extraorally and (b) Intraorally; (c) Orthopantomogram showing a well-defined multilocular radiolucent lesion in the left posterior mandibular region

Some areas in the connective tissue stroma exhibited desmoplastic changes comprising hyalinization of collagen fibers with evident compression of odontogenic follicles [Figure 2c]. Duct-like structures lined by cuboidal to low columnar epithelial cells with homogenous eosinophilic material within the lumen were also noted [Figure 2e]. The lumen of some of the duct-like structures also exhibited a ring of hyalinized eosinophilic material. These features resembled a characteristic picture of AOT.

Abundant areas of extracellular homogenous eosinophilic material were present alongside the AM and AOT components. The material was stained red with Van Gieson stain and blue by Masson's Trichrome stain suggesting it to be of collagenous nature [Figure 3a-c]. The material, however, did not exhibit positive uptake of Alcian blue and Congo red stains.

About 15% of the cells exhibited nuclear expression of Ki-67 in the peripheral ameloblast-like as well as central stellate reticulum-like cells only in the AM component of the lesion. The stellate reticulum-like cells and focal areas of peripheral ameloblast-like cells also exhibited intense calretinin immunoexpression in the nuclei as well as cytoplasm [Figure 3d and e].

A final diagnosis of "adenoid ameloblastoma with dentinoid" was made on the basis of histopathological collocative presence of AM as well as AOT components along with dentinoid material in concordance with biochemical stains and immunohistochemistry findings.

Although the lesion is to be considered more aggressive compared to other benign odontogenic tumors, a conservative approach was adopted in the management of the present case because the patient was not willing to undergo an extensive surgical procedure. The lesion was excised along with the extraction of second and third molar teeth while preserving the inferior border of the mandible. Uneventful healing was noted after a 2-year post-surgical follow-up of the patient with no evidence of disease.

DISCUSSION

AAD is a rare hybrid odontogenic tumor with about 30 cases reported to date.[3] Although the cause for such a peculiar tumor to develop is yet unclear, the transformation of one phenotype to another has been cited as a possible reason.[4] The predominance of either of the entity (AM or AOT) in the histopathological picture may lead to the other one being overlooked, and subsequent misdiagnosis.

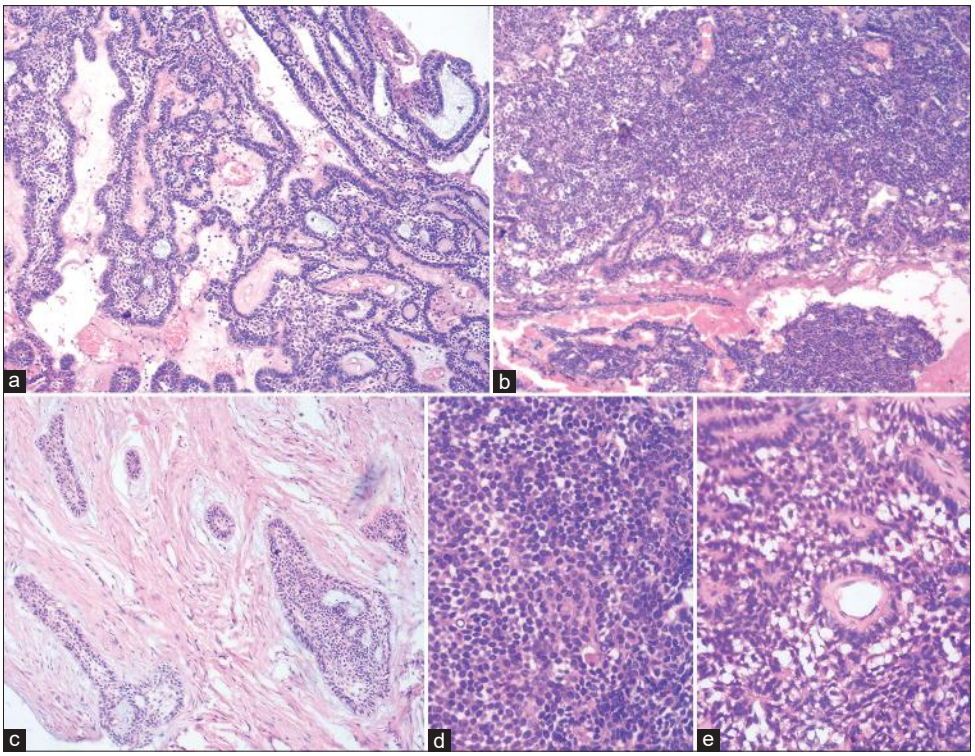

Figure 2: Low power view exhibiting (a) Ameloblastomatous component in the form of interconnecting plexuses, (b) Adenomatoid odontogenic tumor-like component comprising sheets of hyperchromatic odontogenic cells, and (c) areas of compressed follicles with desmoplastic changes in the stroma (H and E, original magnification × 100); High power view of (d) the cuboidal to spindle-shaped epithelial cells and (e) duct-like structure lined by cuboidal epithelium with an intraluminal hyaline ring. (H and E, original magnification × 400)

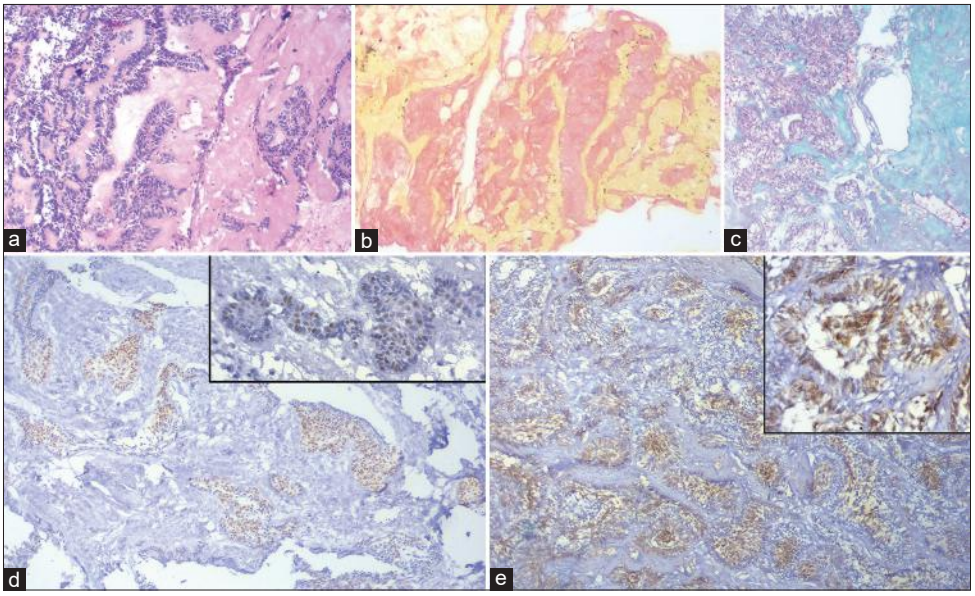

Figure 3: Microscopic examination of "dentinoid" areas by (a) H and E stain (b) Van Gieson stain (c) Masson Trichrome stain (original magnification × 100); Immunohistochemical expression of (d) Ki-67 (inset: original magnification × 400) and (e) Calretinin (inset: original magnification × 400)

In the present case, the lesion presented in a manner similar to a typical AM, which frequently occurs as a multilocular radiolucency involving the posterior mandible, especially the angle-ramus area.[5] The extensive involvement causing the inferior border of the mandible on the verge of pathological fracture along with extreme displacement of the third molar indicated the aggressive clinical course of AAD. It can be surmised that the AM component in AAD could be responsible for its tendency to invade the local structures.

Our interpretation of the extracellular homogenous material as "dentinoid" was in accordance with that of previous reports

wherein the material was found to be positively stained for collagen, but with negative uptake of stains for mucin and amyloid.[2,6] It has been theorized that odontogenic epithelial cells committed to ameloblastic differentiation, comprising the neoplasm, can produce this dentinoid material owing to conversion and coexpression of mesenchymal phenotype.[7]

Calretinin is a calcium-binding protein that has a possible role as a messenger in cell proliferation and differentiation in the neoplastic epithelium of AM but not in odontogenic keratocyst (OKC) or AOT. In corroboration with previous reports of AAD,[3,8] focal but intense positive immunoexpression of calretinin was noted in the present case, that was limited to the cells in AM component of the tumor.

With more than 10% of the cells exhibiting Ki-67 nuclear immunoexpression, an intermediate Ki-67 index was discerned for the case, which reflected the inherent aggressive biological potential of the lesion.[8] Even so, the conservative approach adopted in the present case had yielded positive results in preserving the integrity of the mandible and satisfactory healing after a follow-up of 2 years.

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Conflicts of interest

There are no conflicts of interest.

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