Case Report

Adenoid ameloblastoma with dentinoid: A rare hybrid variant

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Abstract

Odontogenic tumors comprise an unusual group of lesions of the jaw and present diverse histological patterns. Derived from the primordial tooth-forming tissues, they represent a heterogeneous group of lesions that range from hamartomas to benign and malignant neoplasms of variable aggressiveness. Sporadic case reports and diverse complex histogenetic source also defy categorization of odontogenic tumors. Many can be diagnosed accurately based on the distinctive clinical, radiological and histopathological presentation. Considerable variations in the clinicopathological presentation of odontogenic tumors can be confusing, increasing the chance of misdiagnosis. An interesting case of adenoid ameloblastoma reported in a 55-year-old male patient in the mandible, presenting with a diverse and intriguing histopathology, is discussed here.

Keywords: Adenomatoid odontogenic tumor, ameloblastoma, architecture, hamartoma, odontogenic tumor

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INTRODUCTION

Odontogenic tumors encompass a large variety of rare lesions that originate from odontogenic tissue and present with variable levels of differentiation. They result from diverse differentiation potential in odontogenic epithelial cells, while their multipotentiality has been represented by the histopathological varieties of odontogenic tumors themselves. Determination of their precise nature is sometimes difficult and inconclusive.[1] Adenoid ameloblastoma with dentinoid is considered a rare phenotype of ameloblastoma with composite histological features of both ameloblastoma and adenomatoid odontogenic tumor (AOT), including calcified tissue.[2] Here, we report a case of adenoid ameloblastoma with dentinoid matrix with focus on its diverse histological features. This article also provides a comprehensive review of the various composite archetypes presented in the literature regarding this rare odontogenic tumor.

CASE REPORT

A 55-year-old male patient reported to our institution with a complaint of pain and swelling in the lower right jaw region for the past 3 months, which was increasing in size. No history of trauma was elicited. Extraoral examination revealed a nontender, diffuse, hard swelling of size 6 cm × 2.5 cm on the right side of the jaw extending 2 cm from the midline till 2 cm anterior to the angle of the mandible. Paresthesia was present in the same region. Intraoral examination revealed a swelling in the vestibular region which was soft, fluctuant and nontender.

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Radiographic examination revealed a well-defined unilocular radiolucency of size 3.5 cm × 4 cm having irregular margin extending from 43 to 46 region. Another well-defined radiolucency of size 2.5 cm × 2.5 cm was seen on the left parasymphysis region extending up to 35 region [Figure 1]. Both the lesions were enucleated and specimens were sent for histopathological examination.

Histopathological examination of the lesion on the left side of the jaw showed multinodular proliferation of spindle-shaped and cuboidal cells. Numerous scattered duct-like structures were also seen which were lined by cuboidal to columnar cells and lumen filled with eosinophilic material exhibiting characteristic histological findings of AOT.

Histopathological examination of the lesion on the right side presented unusual features. The lesion showed a large cystic lumen surrounded by odontogenic epithelium, predominantly cuboidal cells showing ductal or glandular pattern [Figure 2]. Numerous follicles lined by cuboidal and columnar cells with centrally placed stellate reticulum-like cells with thin rim of eosinophilic material were seen [Figure 3]. Anastomosing cords of epithelial cells with peripheral tall columnar cells intermingled with areas of spindle-shaped cells were also seen [Figure 4]. Supporting connective tissue was highly vascular containing large areas of extravasated red blood cells, few chronic inflammatory cells and numerous multinucleated giant cells peripherally. A few areas of the sections showed homogenous eosinophilic material resembling nontubular dentinoid matrix with entrapment of few epithelial cells [Figure 5].

Cytokeratin 19 (CK19), a potentially useful polypeptide for identification of odontogenic epithelial component, was used for immunostaining, which was strong and diffusely positive with tumor cells [Figure 6]. It further substantiated the evidence of dentinoid formation in association with the epithelium.

Based on the clinical, radiological, histopathological, immunohistochemical study and aided by a review of case reports in the literature, a final diagnosis of adenoid ameloblastoma with dentinoid was made.

DISCUSSION

The rarity, unusual behavior and prognosis of odontogenic tumors make its diagnosis an enigma. Ameloblastomas generally do not show any evidence of induction; however, rare cases associated with hard tissue formation are reported similar to our case report. Lesion on the left side was characteristic of AOT, and a definitive diagnosis was made histopathologically. However, the lesion on the right side was indicative of a composite of ameloblastoma and AOT, leading to inconclusiveness in its histopathological diagnosis. Odontogenic lesions which we considered in the microscopic differential diagnosis included unicystic ameloblastoma, AOT and adenoid ameloblastoma.

Unicystic ameloblastoma refers to those cystic lesions that show clinical and radiologic characteristics of an odontogenic cyst; however, on histological examination, it shows a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation.[3] Even though the interlacing strands of odontogenic epithelium in our case were suggestive of plexiform ameloblastoma, no palisading of the nuclei or polarization of nuclei away from the connective tissue was detected. Furthermore, plexiform ameloblastoma does not exhibit an odontogenic hard tissue component.[4]

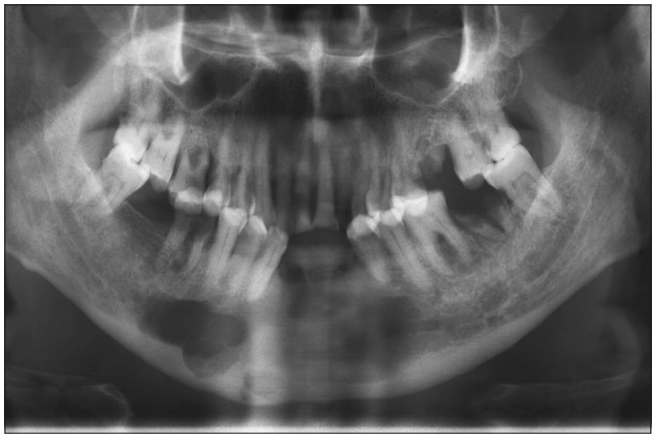

Figure 1: Orthopantomogram showing radiolucencies on both right and left sides of the mandible

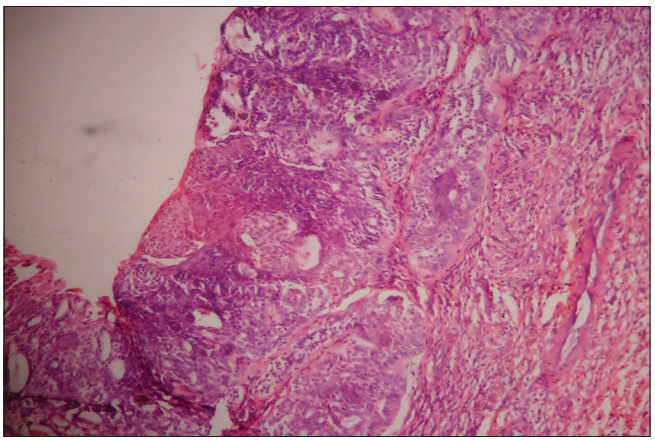

Figure 2: Histopathological image showing cystic lumen surrounded by odontogenic epithelium with ductal or glandular pattern (H&E stain, ×100)

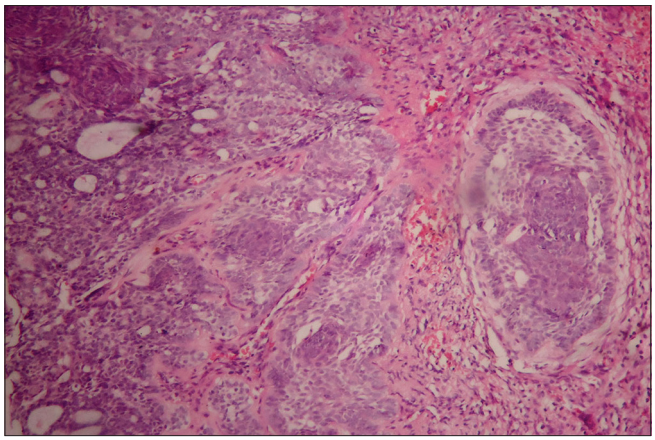

Figure 3: Histopathological image showing follicles lined by cuboidal and columnar cells with centrally placed stellate reticulum-like cells (H&E stain, ×200)

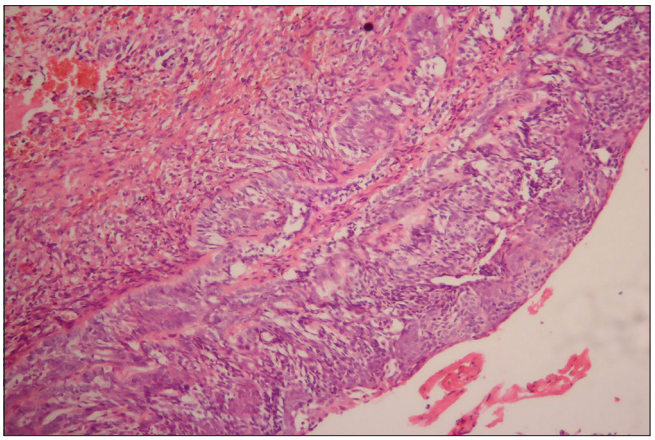

Figure 4: Histopathological image showing strands of epithelium lined by columnar cells and areas of spindle-shaped cells (H&E stain, ×100)

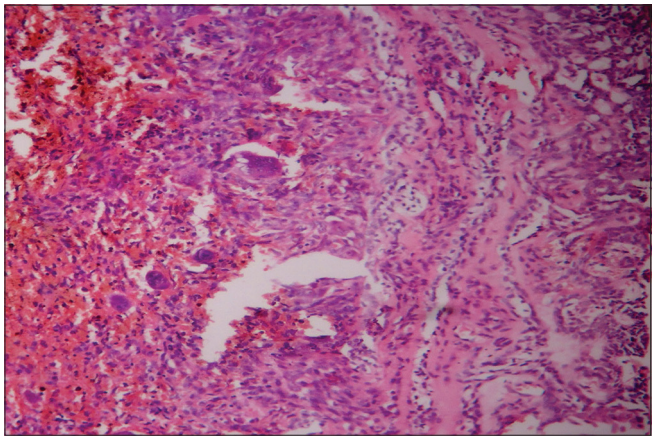

Figure 5: Histopathological image showing eosinophilic dentinoid matrix and multinucleated giant cells at the periphery (H&E stain, ×200)

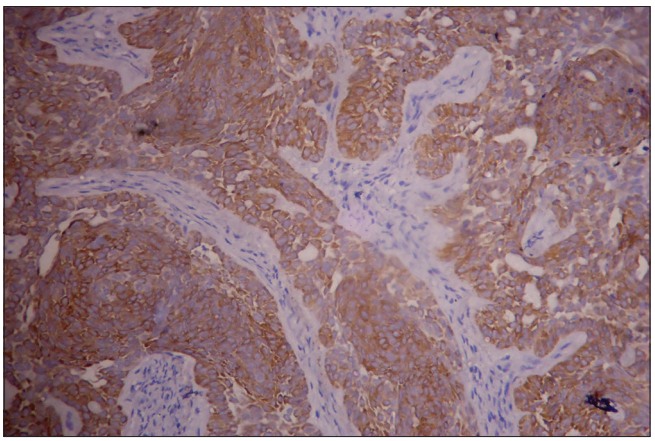

Figure 6: Photomicrograph showing positive immunostaining with cytokeratin-19 (×200)

AOT is not typically associated with dentin formation, although dystrophic calcifications within the tumor are not unusual. As many as twenty different histological patterns of AOT have been described in the literature. Review of literature demonstrates a diversified histological architecture as explained by Philipsen, Sier, Garg, Nomura as nests-like pattern, rosette-like arrangement, trabecular, tubular arrangement, duct-like structures, cribriform pattern and also cystic variant of AOT as described by Mutalik *et al*.[5] Rather than the columnar epithelial cells that typically line the lumen of AOT, cuboidal or flattened cells were predominant and characterized the lesion in our case.

Adenoid ameloblastoma with dentinoid, first reported by Slabbert *et al*.,[6] is a rare odontogenic tumor showing histopathological features similar to ameloblastoma and AOT along with hard tissue formation. Tumor histology demonstrates duct-like structures, epithelial whorls and dentinoid with foci of tubular dentin.[7] It is considered as a neoplasm with decided potential for extension and recurrence. At times, the AOT-like areas predominate, which may overshadow the ameloblastomatous areas leading to benign diagnosis and conservative treatment, which will ultimately result in recurrence.[8] Thus, decision of proper patient care is determined by accurate diagnosis of the tumors.

The present case showed a cystic lesion with areas of ameloblastoma (plexiform pattern) in anastomosing strands and cords and few areas showed AOT-like features with duct-like pattern, spindle and stellate reticulum-like cells and areas of homogenous eosinophilic material which seems to be dentinoid-like material without dentinal tubules. This was similar to cases reported by Evans *et al*.[7] and Saxena *et al*.[8] and differed from that of conventional ameloblastoma. Thus, a definitive diagnosis of adenomatoid ameloblastoma with dentinoid was made.

Slabbert *et al*. used the name "dentino ameloblastoma" as their case demonstrated ameloblastoma showing evidence of dentinoid induction.[6] The diagnostic name "adenoid

ameloblastoma with dentinoid" was used by the Armed Forces Institute of Pathology in 1994.[8] Orlowski *et al*.[9] presented a cystic odontogenic tumor with characteristic features of plexiform type ameloblastoma, containing tubular dentin, with odontoblastic differentiation. Matsumoto *et al*. reported a cystic plexiform-type ameloblastoma containing duct-like structures, tubular dentin and dentinoid in the tumor stroma in a 19-year-old patient. They suggested using the term "adenoid ameloblastoma with dentinoid" for this type of lesions.[10] Evans *et al*. described a recurrent lesion that initially had been diagnosed as AOT. The tumor demonstrated duct-like structures, epithelial whorls and dentinoid with foci of tubular dentin. After the third recurrence and reviewing all histologic findings, they named it adenoid ameloblastoma with dentinoid.[7] In another report of a 19-year-old female with a maxillary tumor, microscopically, it was found to be consistent with previously reported cases of adenoid ameloblastoma with dentinoid.[11]

Formation of dysplastic dentin in epithelial tumors is now considered as a result of a metaplastic process rather than epithelial–ectomesenchymal interaction.[12] Papagerakis *et al*.[13] clearly demonstrated that ameloblastic epithelial cells in mixed odontogenic tumors expressed gene products normally present in ectomesenchymal cells and resulted in conversion and coexpression of mesenchymal phenotype. Thus, it is probable that neoplastic epithelial cells committed to ameloblastic differentiation could produce the dentinoid which exists in some tumors. This was substantiated by immunohistochemical study using CK19 which showed homogenous immunoreactivity of epithelial cells.

Thus, this case report describes adenoid ameloblastoma with dentinoid involving the mandibular region which was accurately diagnosed with the help of extensive histopathological examination and supported by immunohistochemical study. Recognition of the subtypes of ameloblastoma is important because of the aggressive biological behavior than the so-called "conventional" ameloblastoma.

CONCLUSION

Adenoid ameloblastoma with dentinoid is a rare lesion. Due to the diverse histopathological presentation and also the paucity of reported cases, pathologists may face diagnostic confusion on many occasions. Hence, we report this unusual case to avoid any under diagnosis and also to add to the list of the published case reports of rare hybrid variants of ameloblastoma.

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Conflicts of interest

There are no conflicts of interest.

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