Adenoid Ameloblastoma with Dentinoid: A Case Report

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Adenoid ameloblastoma with dentinoid is a rare odontogenic tumor showing histopathologic features similar to ameloblastoma and adenomatoid odontogenic tumor along with hard tissue formation. In this report, a 19-year-old female with a maxillary tumor, microscopically consistent with previously reported cases of adenoid ameloblastoma with dentinoid is presented.

Keywords: Adenoid ameloblastoma • adenomatoid odontogenic tumor • ameloblastoma with dentinoid

Introduction

Ameloblastoma is one of the most common odontogenic tumors, with benign but aggressive clinical features. This tumor microscopically simulates the primary enamel organ and can occasionally demonstrate unusual histopathologic features.1 With the exception of odontoameloblastoma, there is no dental hard tissue formation in most of ameloblastomas.2 However, dentinoid formation without concomitant enamel formation has been reported in a few cases.2

The adenomatoid odontogenic tumor (AOT) represents 3 – 7% of all odontogenic tumors and was once considered as a variant of ameloblastoma. Microscopically, AOT exhibits tubular characteristic and duct-like structures that led to the term “adenoameloblastoma”, which previously used to designate this lesion.3 In contrast to ameloblastoma, AOT is a circumscribed lesion with slow growth.

A few extraosseous variants of AOT have been reported. In one of them the basal cells of oral epithelium were a potential source of origin.4 In the latest edition of WHO classification of odontogenic tumors in 2005, AOT was classified into the first group of tumors (odontogenic epithelium without ectomesenchyme) instead of the second group (odontogenic epithelium with ectomesenchyme).5 Because of the absence of ectomesenchyme in immunohistochemical staining, dysplastic dentin in AOT, is now considered as the result of a metaplastic process rather than epithelial-ectomesenchyme interaction.5

In this article, a rare odontogenic tumor with histopathologic features resembling both ameloblastoma and AOT along with hard tissue formation is reported. The Armed Forces Institute of Pathology (AFIP) in the U.S.A. described a similar neoplasm with recurrence potential and suggested the term “adenoid ameloblastoma with dentinoid” for these lesions.2 A few other examples with different names have been reported in the literature.

Case Report

The patient was a 19-year-old female, visited in a local surgical clinic in Guilan, complaining of a painless swelling on the right side of the maxilla, which had been present for more than two months. Her medical history was unremarkable. Intraoral examination disclosed a nontender expansion of the right maxilla, covered by normal mucosa. Panoramic radiograph showed a unilocular radiolucency with relatively well-defined borders extending from the right canine to the right maxillary first molar. There was root divergence.
but no root resorption (Figure 1).

Using local anesthesia, an excisional biopsy was performed. The surgical sample was fixed in formalin, embedded in paraffin, and stained with hematoxylin-eosin using the standard method. According to the clinical and surgical findings, the lesion was diagnosed as an odontogenic cyst. Unfortunately, follow-up of the patient was not feasible.

**Histopathologic findings**

The tumor displayed a cystic pattern with characteristic features of a plexiform-type ameloblastoma, containing microcysts formation. Sheets and cords of epithelial cells were observed, which demonstrated a loose arrangement similar to stellate reticulum, intermixed with focal areas showing a whorled appearance. Reverse polarity of peripheral cells was prominent and tubular or duct-like structures lined by cuboidal cells were observed in some areas (Figure 2).

The tumor exhibited large amounts of eosinophilic matrix material in the stroma, consistent with dentinoid (Figure 3). A round calcified mass of dentin with clear dentinal tubules was seen in one of the sections (Figure 4). Formation of enamel matrix and primitive-appearing connective tissue was not present. There were no ghost cells in the specimen.

**Discussion**

Ameloblastomas are epithelial odontogenic tumors with no induction of dental hard tissue formation. However, there are few reports of ameloblastomas containing hard tissue. In reviewing the literature, we found only 13 reports of cases that were histologically similar to ameloblastoma and AOT, which exhibited hard tissue formation.

![Figure 1](image1.png)  
**Figure 1.** A panoramic radiograph demonstrates a unilocular radio-lucency of the right maxilla.

In the cases reported by Evans et al,2 Orlowski et al,6 Tajima et al,7 Slabbert et al,8 Matsumoto et al,9 and in the present report, the tumor was diagnosed as ameloblastoma, while in six other reports the tumor was considered to be AOT.9

Bone formation was observed in one neoplasm but in other reports the hard tissue had been interpreted as dentin or dentinoid.9 Interstitial ossification has been reported in two cases of polycystic ameloblastoma,10 and enamel matrix formation was described in two reports.9

Slabbert et al claimed the first case of such unusual tumors. They described a unicystic neoplasm containing typical follicles of ameloblastoma with large amounts of dentinoid and psammomatous-type dystrophic calcifications in an Asian male patient.8 A similar case was reported by another author and the term “adenoid ameloblastoma with dentinoid” was suggested for these lesions by the AFIP.2

Orlowski et al presented a cystic odontogenic

![Figure 2](image2.png)  
**Figure 2.** Duct-like structures lined by cuboidal cells (hematoxylin-eosin, original magnification ×20).

![Figure 3](image3.png)  
**Figure 3.** Matrix material consistent with dentinoid (hematoxylin-eosin, original magnification ×10).
tumor with characteristic features of plexiform-type ameloblastoma, containing tubular dentin, with odontoblastic differentiation.\textsuperscript{6} 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 lesion.\textsuperscript{9} In addition, Evans et al described a recurrent lesion that initially had been diagnosed as AOT. After the third recurrence and reviewing all histologic findings, they named it adenoid ameloblastoma with dentinoid. The tumor demonstrated duct-like structures, epithelial whorls, and dentinoid with foci of tubular dentin.\textsuperscript{2}

Having considered the previous reports, the histologic findings of the present lesion resembled strongly the cases reported by Orlowski et al,\textsuperscript{6} Matsumoto et al,\textsuperscript{9} and Evans et al.\textsuperscript{2} Therefore, the term “adenoid ameloblastoma with dentinoid” for the current tumor is the most acceptable and logical description. It was proposed by one author that these tumors could be considered as a variant of ameloblastoma with the potential for extension and recurrence.\textsuperscript{2} However, in the third edition of WHO histologic typing of odontogenic tumors in 2005, this term is not included, probably because of the rarity of the reports.\textsuperscript{5}

If further investigation with immunohistochemical staining clarifies the absence of ectomesenchyme, they may be classified in the first group of the new WHO classification of odontogenic tumors (odontogenic epithelium without ectomesenchyme).\textsuperscript{5} Whether hard tissues in these lesions should be regarded as a metaplastic process, like AOT,\textsuperscript{7} or represent a true inductive effect is still to be clarified.

Because of the paucity of reported cases of adenoid ameloblastoma with dentinoid, the behavior and proper management of these tumors are uncertain. However, an example of this lesion with three times recurrences have been reported by Evans et al.\textsuperscript{2}

References