

Hybrid ameloblastoma and adenomatoid odontogenic tumor: report of a case and review of hybrid variations in the literature

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Hybrid odontogenic tumors including 2 or more different histologic types have been documented, but their occurrences are not very common. We present a case of hybrid odontogenic tumor composed of ameloblastoma and adenomatoid odontogenic tumor (AOT) arising in the mandibular molar region of a 31-year-old Japanese woman who had a history of familial adenomatous polyposis. The tumor, measuring 10 mm in diameter, was surgically removed from the alveolar bone. Histopathologically, the tumor consisted of both follicular and plexiform types of ameloblastoma in which multiple and smaller foci of AOT were intermingled. There have been 3 reported cases of hybrid ameloblastoma and AOT, all of which presented unicystic types as ameloblastoma components. This, however, is the first report of a hybrid tumor containing an authentic solid-type ameloblastoma compartment and an AOT compartment in a patient with a background of familial adenomatous polyposis. (Oral Surg Oral Med Oral Pathol Oral Radiol 2013;■:e1-e7)

Odontogenic tumors are characterized by their diverse histopathologic spectra, because the tooth germs themselves develop through complicated interactions between epithelial and mesenchymal elements,¹ which are critically regulated in time-dependent manners.²⁻⁴ Based on their combination of predominant histologic

elements, benign odontogenic tumors have roughly been classified into 3 categories: (1) odontogenic epithelial tumors without odontogenic ectomesenchyme; (2) odontogenic epithelial tumors with odontogenic ectomesenchyme; and (3) odontogenic ectomesenchymal tumors with or without included odontogenic epithelium.⁵ The histopathologic diagnosis of odontogenic tumors can be challenging, owing to complicated clinicopathologic features and nomenclatural confusion. Furthermore, unusual combinations of neoplastic components, namely hybrid odontogenic tumors, are occasionally observed. In fact, the presence of hybrid odontogenic tumors which contain 2 or more different histologic types is well recognized, and various cases of hybrid odontogenic tumors have been documented in the literature. However, their frequencies and component combinations are not well understood due to ambiguous histologic classifications.⁶

In this article, we report a rare case of hybrid ameloblastoma and adenomatoid odontogenic tumor (AOT) arising in the mandible of a 31-year-old woman with familial adenomatous polyposis (FAP). We show the characteristic histopathologic features with immunohistochemical profiles, and we discuss the possible association with FAP. In addition, we review the varieties of hybrid odontogenic tumors documented in the literature to analyze their frequencies.

CASE REPORT

A 31-year-old Japanese woman, who had undergone total colectomy and ileoanal anastomosis for FAP and colon adenocarcinoma 3 months earlier, was referred to the Division of Oral and Maxillofacial Surgery, Niigata University Hospital, for detailed examination of FAP-related jaw lesions, although she had no obvious symptoms. She had a history of extraction of supernumerary teeth in her childhood. On intraoral

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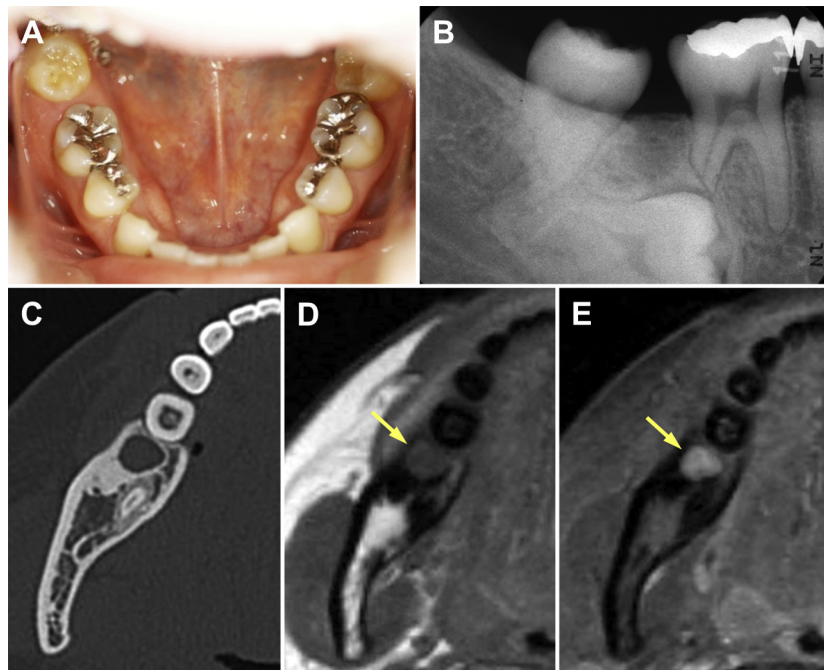


Fig. 1. Clinical findings of hybrid ameloblastoma and adenomatoid odontogenic tumor arising in the molar region of the mandible of a 31-year-old Japanese woman. **A**, Intraoral view. **B**, Dental radiograph. **C**, Computed tomography (CT) image, bone window, axial section. **D**, T1-weighted magnetic resonance image (MRI), axial scan. **E**, Fat-suppressed contrast-enhanced T1-weighted MRI, axial scan. The patient had a 2-mm space between the right first and third molars of the mandible with the second molar unerupted (**A**). Radiographically, her second molar was shown to be impacted horizontally in the alveolar bone, and above the impacted tooth there was a round radiolucent lesion. The distal root of the first molar was sharply resorbed (**B**). CT revealed a spherical unilocular mass between the 2 molars in the mandible (**C**). T1-weighted MRI showed muscle signal intensity in the lesion (**D**, *arrow*), but it was significantly enhanced after contrast medium administration (**E**, *arrow*).

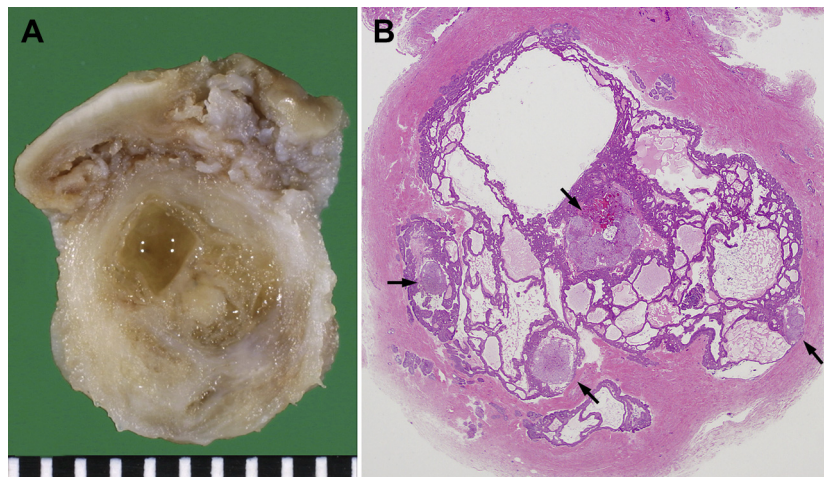


Fig. 2. Macroscopic findings of hybrid ameloblastoma and adenomatoid odontogenic tumor of the mandible. **A**, Gross view of the cut surface of the surgical specimen. **B**, Loupe view of the histologic section at the same cut surface (hematoxylin-eosin). On the cut surface, there was a tumorous mass with cystic spaces circumscribed with a thick fibrous capsule beneath the gingival mucosa (**A**). Histologically, the tumor was mainly composed of ameloblastoma with cystic changes of stromal space, and there were multiple small adenomatoid odontogenic tumor foci within the ameloblastoma (**B**, *arrows*).

inspection, her mandibular right second molar was unerupted, resulting in a 2-mm-long interdental space between the first and third molars (Figure 1, A). Plain radiographic examinations

revealed an oval radiolucent lesion circumscribed with an irregular-shaped radiopaque rim in the alveolar bone between the first and third molars (see Figure 1, B), below which

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