Journal of Dental Sciences 17 (2022) 595–597

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journal homepage: www.e-jds.com

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# A hybrid odontogenic tumor of calcifying odontogenic cyst, ameloblastic fibroma, and complex odontoma

**KEYWORDS**
Hybrid odontogenic tumor;
Calcifying odontogenic cyst;
Ameloblastic fibroma;
Complex odontoma

Calcifying odontogenic cyst (COC) is an odontogenic cyst with locally aggressive clinical behavior.[1,2] Sometimes, it may occur in combination with other odontogenic tumors.[3,4] In this study, we presented a large hybrid odontogenic tumor of COC, ameloblastic fibroma, and complex odontoma in the right maxilla with invasion into the right maxillary sinus of a young male patient.

This 12-year-old young male patient was brought to our hospital for management of a progressive and painful right facial swelling for about 4 months. The computed tomography (CT) of face was arranged and showed a marked periosteal mucosal thickening of the right maxillary sinus with polypoid lesions and fluid collection (Fig. 1A). Further magnetic resonance imaging (MRI) study revealed polypoid cystic lesions over the right maxillary sinus with fluid collection and peripheral thin-layer enhancement of the sinus (Fig. 1B). The clinical impression was an extensive odontogenic cyst or tumor. After discussing with the patient and his parent and obtaining the signed informed consent, surgical excision of the lesion with Caldwell Luc's operation was performed. The specimen was sent for histopathological examination. Microscopically, the large lesion contained multiple cysts lined by the ameloblastomatous epithelium with the well-defined cuboidal basal cells and suprabasal stellate reticulum-like cells. The most characteristic feature was the presence of many "ghost cells" with eosinophilic cytoplasm and pale centrally-located nuclear shadow in the suprabasal and superficial areas of the lining epithelium. Some of the ghost cells underwent calcification (Fig. 1C and D). In addition, multiple nodular areas with many long, narrow cords of odontogenic epithelium or multiple islands of odontogenic epithelium with peripheral columnar cells and central loosely arranged angular cells resembling the stellate reticulum of an enamel organ in a primitive cell-rich mesenchymal tissue stroma were found (Fig. 1E and F). Multiple areas of hyalinized acellular tissue were also discovered (Fig. 1E, right half). Furthermore, a complex odontoma composed of dentin, enamel matrix, and keratinized or calcified ghost cells in the proliferating reduced enamel epithelium was also noted (Fig. 1G and H). The overall characteristic histological features confirmed the histopathological diagnosis of a hybrid odontogenic tumor of COC, ameloblastic fibroma, and a complex odontoma. After operation, the patient was discharged under stable condition and was arranged for regular follow-ups at the outpatient clinic.

The COC is a locally aggressive odontogenic cyst. In our patient, the multiple COCs grew progressively with invasion into the right maxillary sinus. A concomitant presence of multiple nodules of ameloblastic fibroma and a complex odontoma was also noted. COC has been reported in association with different odontogenic tumors including odontoma, ameloblastoma, adenomatoid odontogenic tumor, odontoameloblastoma, ameloblastic fibroma, and odontogenic myxoma.[3,4] By immunohistochemical staining, the epithelial components of the COC and ameloblastic fibroma are positive for cytokeratin (CK)14, CK19, CK5-6, CD138 (syndecan-1), p63, and epidermal growth factor receptor

https://doi.org/10.1016/j.jds.2021.07.007

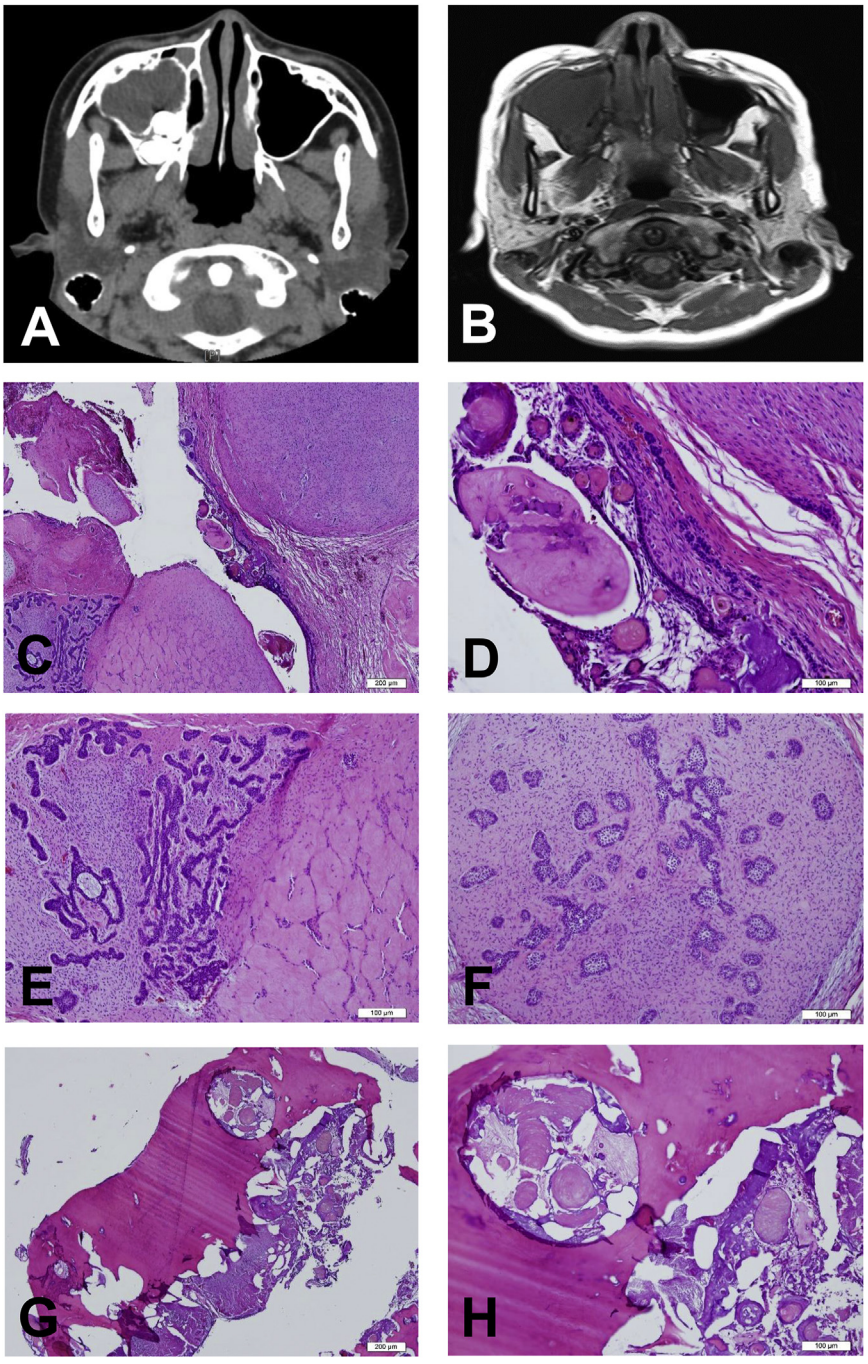


**Figure 1** Hematoxylin and eosin-stained histological sections of our case of a hybrid odontogenic tumor of calcifying odontogenic cyst (COC), ameloblastic fibroma, and complex odontoma. (A) Computed tomography (CT) showed a marked periosteal mucosal thickening of the right maxillary sinus with polypoid lesions and fluid collection. (B) Magnetic resonance imaging (MRI) revealed polypoid cystic lesions over the right maxillary sinus with fluid collection and peripheral thin-layer enhancement of the sinus. (C) Low-power microphotograph exhibiting a COC in the center and two nodules of ameloblastic fibroma at the right upper part and the left lower part of the microphotograph. (D) Medium-power microphotograph showing a COC lined by the ameloblastomatous epithelium with the well-defined cuboidal basal ells and suprabasal stellate reticulum-like cells. Many eosinophilic keratinized ghost cells with some of them undergoing calcification were found in the suprabasal and superficial areas of the lining epithelium. (E and F) Medium-power microphotographs showing many long, narrow cords of odontogenic epithelium or multiple islands of odontogenic epithelium with peripheral columnar cells and central loosely arranged angular cells resembling the stellate reticulum of an enamel organ in a primitive cell-rich mesenchymal tissue stroma. Multiple areas of hyalinized acellular tissue were also discovered (E, right half). (G and H) Low- and medium-power microphotographs showing a complex odontoma composed of dentin, enamel matrix, and keratinized or calcified ghost cells in the proliferating reduced enamel cells. (Hematoxylin and eosin stain; original magnification; C and G, 4 $\times$ ; D, E, F and H, 10 $\times$ ).

(EGFR), and are negative for glial fibrillary acidic protein (GFAP) and p53.[4] In addition, the ghost cells in the lining epithelium of the COC are positive for amelogenin and CK6, but are negative for CK19.[5] Although the hybrid odontogenic tumor in our patients was quite extensive, basically it was a benign odontogenic tumor and can be treated by conservative excision with relatively good prognosis. However, long-term and regular follow-ups are needed for our patient.

## Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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Received 18 July 2021
Available online 26 August 2021