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Calcifying odontogenic cyst coexists with adenomatoid odontogenic tumor and peripheral cemento-osseous reactive proliferation



KEYWORDS

Adenomatoid odontogenic tumor;
Calcifying odontogenic cyst;
Peripheral cemento-osseous reactive proliferation

Hybrid odontogenic lesion (HOL), characterized by two or more defined odontogenic cysts and/or tumors occurring at the same site, is an uncommon disease with only 203 cases being reported until January 2021.¹ The most common HOL is calcifying odontogenic cysts (COC) associated with odontoma, followed by central odontogenic fibroma with central giant cell granuloma, adenomatoid odontogenic tumors (AOT) with calcifying epithelial odontogenic tumor, AOT with dentigerous cyst (DC), and DC with odontoma.¹ Both AOT and COC are uncommon odontogenic tumor and cyst, which account for less than 5% of odontogenic lesions.² The HOL comprised of COC and AOT is extremely rare. Hereby, a case of HOL consisting of COC and AOT with peripheral cemento-osseous reactive proliferation was presented.

A 52-year-old female patient visited Department of Oral and Maxillofacial Surgery of our institution for the swelling at the left mandibular area. Intraoral findings showed a firm swelling mass covered by normal mucosa over the left anterior mandibular gingiva with prominent buccal bony expansion, measuring about 3.0×2.5 cm in dimension. An ulcer measuring 0.5×0.4 cm in dimension at the buccal gingiva of edentulous ridge of tooth 33 was also identified (Fig. 1A). Panoramic radiograph revealed a well-defined

mixed radiolucent and radiopaque lesion over the left mandibular symphysis (Fig. 1B). Cone-beam computed tomography showed a buccal bony expansion and perforation of the buccal plate (Fig. 1C). The clinical impression was a cemento-ossifying fibroma, and enucleation was performed under general anesthesia. Histopathological examination showed an inflamed, cystic lesion with a thick fibrotic wall (Fig. 1D). The cyst was lined by non-keratinized stratified squamous epithelium with focal proliferation. Numerous ghost cells and calcified materials were present in the lumen and fibrotic wall (Fig. 1E and F). A lobule of basophilic cells arranged in rosette-like and duct-like structures in the cystic wall was seen, which revealed the histopathological characteristics of AOT (Fig. 1G). An area with fibro-osseous lesion-like appearance was also discerned at the periphery of the lesion (Fig. 1H). Collectively, the final diagnosis was an AOT in COC with peripheral cemento-osseous reactive proliferation.

The development of HOL might be due to the multipotentiality of the odontogenic epithelium; however, the mechanism has not yet been clarified.³ Cemento-osseous proliferation at periphery of an odontogenic cyst or tumor is rare, and the condition is likely to be a secondary reactive

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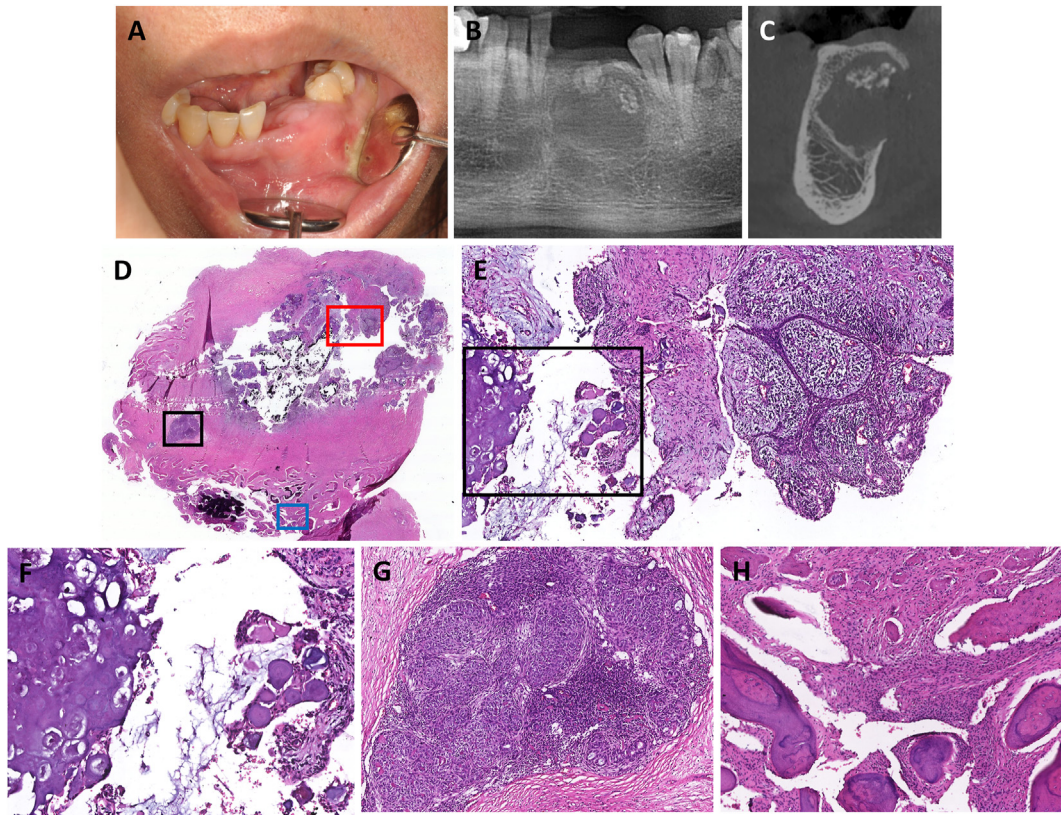


Figure 1 Clinical, radiographic and microscopic photographs of the current case of hybrid odontogenic lesion in the left mandibular symphysis. (A) A firm swelling mass covered by normal mucosa over the left anterior mandibular gingiva with prominent buccal bony expansion. (B) A well-defined mixed radiolucent and radiopaque lesion of the left mandibular symphysis. (C) Buccal bony expansion and perforation of the buccal plate. (D) An inflamed, cystic lesion with a thick, fibrotic wall. (E) The cyst was lined by proliferative, non-keratinized stratified squamous epithelium. Numerous ghost cells and calcified materials were present in the lumen and fibrotic wall (magnified image taken from the red rectangular area in Fig. 1D). (F) Magnification of ghost cells and calcified materials taken from black rectangular area in Fig. 1E. (G) A lobule of basophilic cells arranged in a pattern of the histopathological characteristics of adenomatoid odontogenic tumor (magnified image taken from the black rectangular area in Fig. 1D). (H) An area with fibro-osseous lesion-like appearance at the periphery of the lesion (magnified image taken from the blue rectangular area in Fig. 1D). (Hematoxylin and eosin stain; original magnification; D, 10 \times ; E, 40 \times ; F, 100 \times ; G, 100 \times H, 200 \times).

proliferation rather than a hybrid lesion.⁴ Generally, the prognosis of HOL is good, and recurrence after nonaggressive surgical treatment is uncommon.¹ In the current case, the histopathological findings revealed a collision of three distinct lesions, including COC, AOT, and fibro-osseous proliferation. Recurrence of COC, AOT, and cemento-osseous proliferative lesions (such as cemento-ossifying fibroma and cemento-osseous dysplasia) after conservative surgical treatment is seldom seen.⁵ Simple enucleation was therefore an acceptable approach for the present case.

Declaration of competing interest

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

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