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Histopathological study of a mixed intraluminal and mural type of unicystic ameloblastoma

KEYWORDS
Unicystic ameloblastoma;
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Mural type

Unicystic amloblastoma (UA) is a specific type of ameloblastoma presented as a unilocular radiolucent lesion with less aggressive clinical behavior than the conventional solid intraosseous ameloblastoma.[1] This case report presented the histopathological features of a mixed intraluminal and mural type of UA in the left posterior mandible of a 27-year-old male patient.

This 27-year-old male patient came for extraction of the horizontally impacted left mandibular third molar without any clinical symptom and sign. The panoramic radiography showed a 3 × 1.5 cm well-circumscribed cystic lesion surrounding the root of the horizontally impacted left mandibular third molar with partial external root resorption. Because the radiolucent lesion was relatively large, the clinical diagnosis was an odontogenic keratocyst. After discussing with the patient and obtaining the signed informed consent, the extraction of the horizontally impacted left mandibular third molar and total enucleation of the cystic lesion were performed. Microscopically, the lesion was a cyst lined by the ameloblastomatous epithelium with the cuboidal basal ells and suprabasal stellate reticulum-like cells. The most characteristic feature was the projection of several plexiform ameloblastoma-like nodules from the cystic lining into the lumen of the cyst (Fig. 1A–F). In addition, foci of follicular-typed ameloblastoma infiltrating into the fibrous connective tissue wall of the cyst were discovered (Fig. 1G and H). The infiltrated nest of follicular ameloblastoma showed cuboidal or columnar peripheral cells and central loosely arranged epithelial cells that resembled the stellate reticulum of an enamel organ (Fig. 1H). These characteristic histological features confirmed the diagnosis of a mixed intraluminal and mural type of UA.

The cystic lesion of our patient was predominantly an intraluminal type of UA. However, mural invasion of islands of follicular ameloblastoma was noted in focal areas of the fibrous cystic wall. Thus, the final histopathological diagnosis was a mixed intraluminal and mural type of UA. UA frequently presents the clinical and radiographic features similar to either a dentigerous cyst or a radicular cyst.[2–4] Therefore, it is important to search for a method to distinguish UA from dentigerous cyst or radicular cyst. A previous study used immunohistochemistry as well as TaqMan mutation detection qPCR assay and Sanger sequencing to evaluate the expression of BRAFV600E protein and the BRAFV600E mutation, respectively, in 8 UAs, 9 dentigerous cysts, and 9 radicular cysts. The expression of BRAFV600E protein is discovered in 8 (100%) of 8 UAs, 2 (22.2%) of 9 dentigerous cysts, and 2 (22.2%) of 9 radicular cysts. However, the BRAFV600E mutation is detected in 5 of 8 UAs and none of 9 dentigerous cyst and 9 radicular

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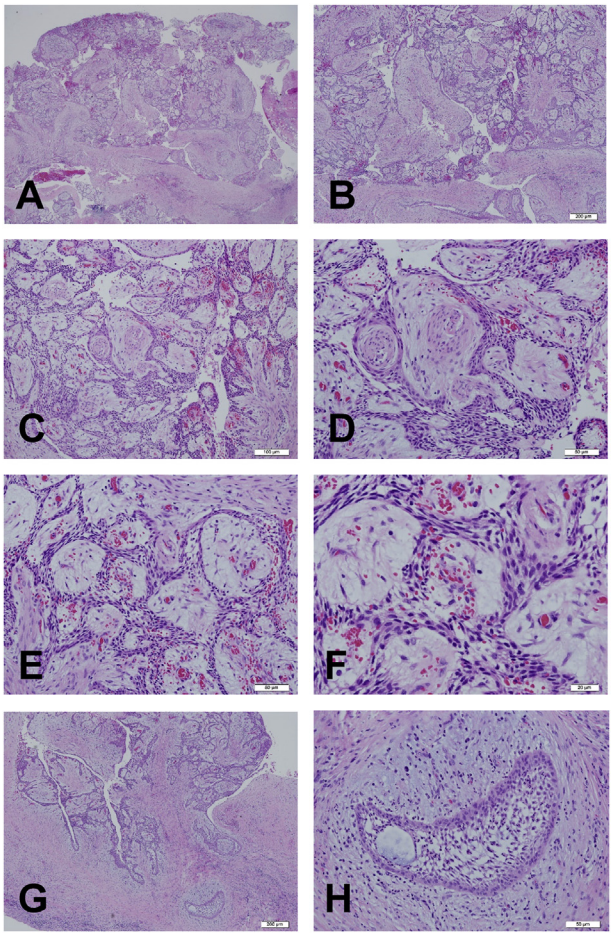

Figure 1 Hematoxylin and eosin-stained histological sections of our case of unicystic ameloblastoma of a mixed intraluminal and mural type. (A and B) Low-power microphotographs exhibiting a cyst lined by the ameloblastomatous epithelium with the cuboidal basal cells and suprabasal stellate reticulum-like cells as well as projection of several plexiform ameloblastoma-like nodules from the cystic lining into the lumen of the cyst. (C, D, E and F) Medium and high-power microphotographs showing the plexiform ameloblastoma composed of a network of long and anastomosing cords of odontogenic epithelium bounded by cuboidal ameloblast-like cells surrounding the loosely arranged central epithelial cells. (G and H) Low- and high-power microphotographs showing projection of several plexiform ameloblastoma-like nodules from the cystic lining into the lumen of the cyst as well as mural invasion of a nest of follicular ameloblastoma with cuboidal or columnar peripheral cells and central loosely arranged epithelial cells that resemble the stellate reticulum of an enamel organ (H). (Hematoxylin and eosin stain; original magnification; A, 2 × ; B and G, 4 × ; C, 10 × ; D, E, and H, 20 × ; and F, 40 ×).

cysts, indicating that the expression of BRAFV600E protein and the detection of the BRAFV600E mutation can be used to differentiate UA from dentigerous cyst and radicular cyst.[5] The treatment for a UA is still controversial. As a general rule, if the UA is a luminal type (i.e. the ameloblastoma is confined in the lining epithelium) or an intraluminal type (i.e. the ameloblastoma is mainly located in the cystic lumen), then the enucleation of the cystic lesion with curettage is an adequate treatment. However, if the UA is confirmed as a mural type (i.e. the ameloblastoma islands have invaded some distance into the fibrous cystic wall) after enucleation, then you may do a subsequent local resection of the lesion area or just keep the patient under a long-term and regular follow-up.[1] In our case, we did not do further surgery for our patient and he was under close radiographic observation once per 6 months.

Declaration of competing interest

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

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