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CASE REPORT

Ameloblastoma ex calcifying odontogenic cyst (dentinogenic ghost cell tumor)

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Calcifying odontogenic cyst (COC) has shown to be of extensive diversity in its clinical and histopathological features, as well as in its biological behavior. In this report, a rare case is described of ameloblastoma ex COC (dentinogenic ghost cell tumor) and the relevant literature is briefly reviewed.
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Keywords: ameloblastoma; calcifying odontogenic cyst; dentinogenic ghost cell tumor

Case report

The patient, a 79-year-old male, reported painless swelling of edentulous posterior region of the right mandible. Radiographs showed a well-demarcated, multilocular mixed radiolucent-radiodense lesion. After enucleation, no recurrence has been recorded in the ensuing 5 years. Histologically, cystic structures were lined with an epithelial lining exhibiting stellate reticulum-like arrangement and polarization of basal layer. Ghost cells, dentinoid and calcified bodies were abundant (Fig. 1a). Within the thick fibrous wall were many large follicles of ameloblastoma (Fig. 1b). All mural follicular tumor islands consisted of peripheral palisading of tall columnar cells fulfilled the ameloblastic histologic criteria (1) and in no instance was the formation of ghost cells and dentinoid observed (Fig. 1c).

Comments

Several reports in the literature have documented the combined microscopic features of calcifying odontogenic cyst (COC) and ameloblastoma, merging from one to the other (1). However, the photomicrographs said to show ameloblastoma would be better classified nowadays as so-called ameloblastomatous transformation (2, 3). The existence of an ameloblastoma indeed arose in association with COC has been accepted since 1991, when Hong et al. (4) described two cases of plexiform ameloblastoma occurring in neoplastic variant of COC and strengthened a distinct difference between ameloblastoma ex COC and ameloblastomatous COC. There is on record at least one other example of what appears to be the same as ameloblastoma ex COC in the literature (5). In the present lesion, the diagnosis of ameloblastoma is straightforward, being the first well-described case of follicular type ex COC.

As mentioned above, ameloblastoma ex COC and ameloblatomatous COC are morphologically entirely different and easily distinguishable (1, 4, 5). In our opinion, there seems to be no justification for subtyping simple cystic (non-neoplastic) COC. The term *ameloblastomatous* adds further to the confusion about its true nature as a cyst and could thus be abandoned. Our notion is supported by the observations that the proliferating epithelium of COC invariably shares features with ameloblastoma (5) and ameloblastomatous elements never alter its biological behavior (1, 4, 5). Unfortunately, the mural development of bona fide ameloblastoma in COC is of unknown clinical significance at this time because of the limited number of cases and limited follow-up information (4, 5). The questions as to whether ameloblastoma ex COC behaves differently from COC alone can be resolved after publication of a large series.

Because of its complexity, the nomenclature of COC is largely confusing (1, 5). The term *COC* is not covered in the forthcoming World Health Organization classification of odontogenic tumors (1, 6). In accordance with the above mentioned most recent terminology (1, 6), the present benign tumor variant belongs to dentinogenic ghost cell tumor.

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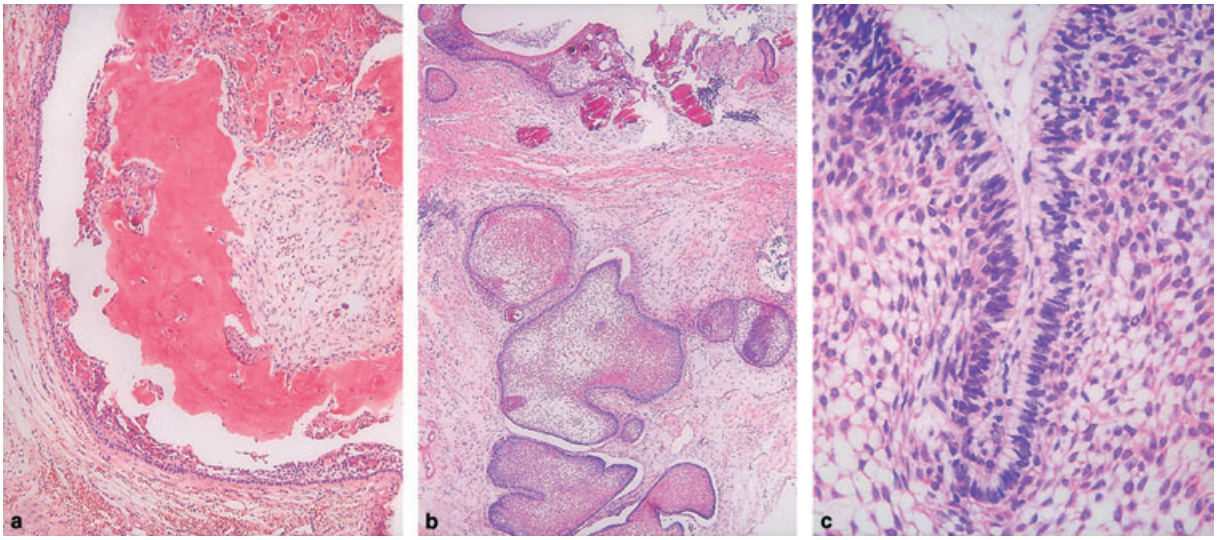

Figure 1 (a) Cystic architecture with ghost cells and dentinoid. (b) Mural follicular ameloblastoma lacking ghost cells and dentinoid. (c) Tumor follicle fulfilling the ameloblastic histopathologic criteria [hematoxylin and eosin stain; (a), ×100; (b), ×40; (c), ×400].

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