

Ghost Cell Differentiation and Calcification in Ameloblastic Fibroma

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Ghost cell differentiation within an ameloblastic fibroma is extremely rare. The ghost cells found in an ameloblastic fibroma in previously reported cases were all associated with a typical calcifying odontogenic cyst. Here, we report a case of an ameloblastic fibroma with focal ghost cells and calcifications in some neoplastic epithelial islands, but without other histologic manifestations consistent with a calcifying odontogenic cyst. The patient was a 13-year-old Chinese boy who presented with a bony-hard swelling in the posterior mandibular region over a 6-month period. Radiographs showed a well-defined multilocular radiolucency associated with an unerupted tooth. The lesion was mostly cystic-solid and comprised of odontogenic epithelial strands, islands and myxoid ectomesenchymal component microscopically. Small groups of ghost cells and calcification were noted in the epithelial islands.

Key words: ghost cell, ameloblastic fibroma

According to the 2005 WHO classification for odontogenic tumors, an ameloblastic fibroma is a rare odontogenic tumour, in which both epithelial and ectomesenchymal components are neoplastic¹. Mixed odontogenic tumours with ameloblastic and fibromatous components comprise a heterogeneous group of neoplasms. If there is dysplastic dentin or dentin plus enamel deposition in an ameloblastic fibroma, due to the inductive interactions of neoplastic epithelium and mesenchyme, the lesions are referred to as ameloblastic fibrodentinoma and ameloblastic fibro-odontoma, respectively. Even more rarely, ameloblastic fibromas may be combined with other odontogenic tumours, such as ameloblastomas and calcifying odontogenic cysts²⁻⁴. Here we report a case of an ameloblastic fibroma with ghost cell differentiation and calcification in the epithelial component, but with no cyst wall that resembled a calcifying odontogenic cyst as previously reported.

Case report

A 13-year-old boy came to the Department of Maxillofacial Surgery, Peking University Stomatologic Hospital for an evaluation of a mass located in the right posterior mandible. The patient noticed a slow, enlarging mass over the past 6 months and a painless losing tooth recently. He also reported having no numbness of lip and tongue, preexisting medical conditions, fever, or other signs of infections.

A panoramic radiograph revealed a relative large, expansile, multilocular radiolucent lesion extending from the mandible body corresponding to the lower right second premolar area to the sigmoid notch of the ascending ramus. The lesion had absorbed the lower right first molar, lower right second molar and contained an unerupted third molar. A supernumerary tooth was located in the region of upper right incisors (Fig 1). Physical examination revealed facial asymmetry, with an increase in volume of posterior mandibular body. Buccal and lingual cortical expansion of posterior mandibular body and ramus was evident and the overlying mucosa seemed normal. The lesion was generally well circumscribed, but the buccal cortical palate was not intact and fluctuant on palpation, whereas the lingual cortex was intact. There was obvious mobility of the lower right second molar. The patient demonstrated no

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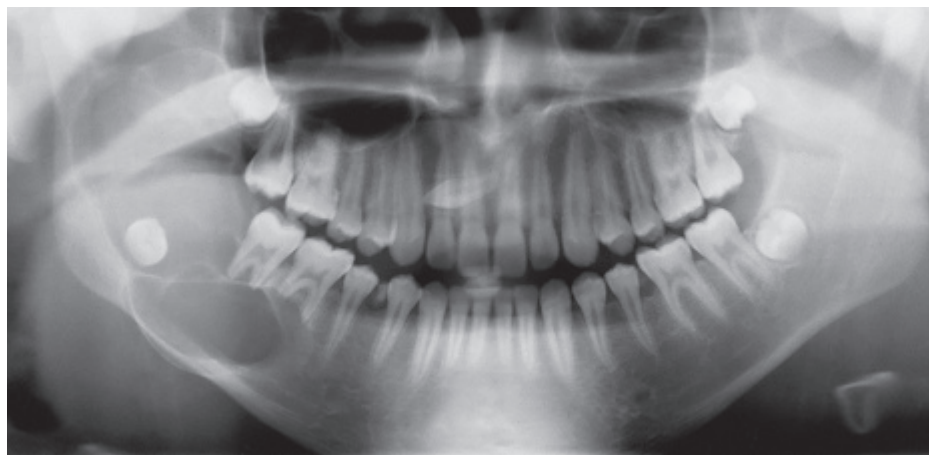


Fig 1 Panoramic radiograph revealing multilocular radiolucency of right mandible associated with an unerupted tooth and roots resorption of teeth 46 and 47.

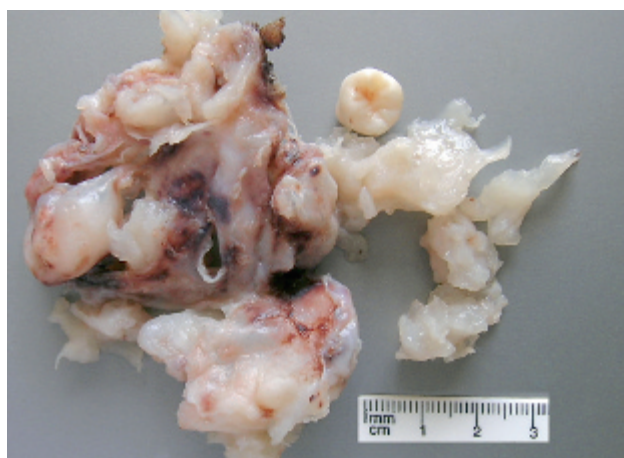


Fig 2 The enucleated lesion consisting of a pale grey cystic-solid tissue.

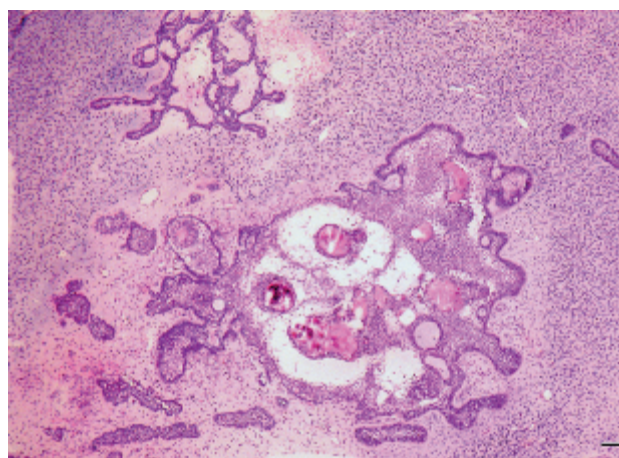


Fig 3 Low-power photomicrograph showing islands and strands of odontogenic epithelium and cellular mesenchymal tissue resembling dental papilla, bar = 250 μ m.

pain on palpation and had no sensory deficit in the region of the mandibular division of the trigeminal nerve.

Preoperative laboratory exams were ordered and they were all within normal parameters. Based on these findings, the clinical diagnosis was a cystic lesion involving the posterior mandible including the ramus.

During the operation under general anaesthesia, the surgeons found the lesion had perforated the buccal cortex and the inferior alveolar nerve had displaced inferiorly by the lesion. The lesion was thoroughly separated along the perforated bone wall by blunt dissection and curettage.

The lower right first and second molars involved by the lesion were vital and were not extracted. The specimen was fixed in formalin and submitted for microscopic

examination. The lesion consisted mostly of a pale grey cystic-solid tissue measuring 6.0 \times 5.0 \times 2.0 cm. A bisection showed multilocular cavities in varied size. Most of the portion of the lesion was white, semitransparent and solid in density. The crown of the lower right third molar was impacted in the lesion (Fig 2).

Microscopic examination showed a typical ameloblastic fibroma in the solid part. The epithelial component consisted of interconnecting strands, buds and islands (Fig 3) with a peripheral layer of cells exhibiting reverse nuclear polarity and a distinct basement membrane (Figs 4 and 5). In some sections, stellate cells similar to the stellate reticulum of enamel organ were encompassed in the centres of epithelial nests. These epithelial elements were embedded in a cellular

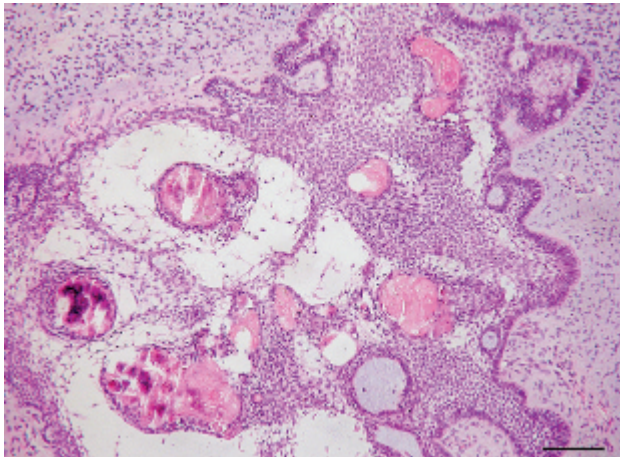


Fig 4 Small groups of ghost cells and calcification within the odontogenic epithelial nests characterized by a palisaded peripheral columnar cells enclosing loose stellate reticulum-like tissue, bar = 150 μm .

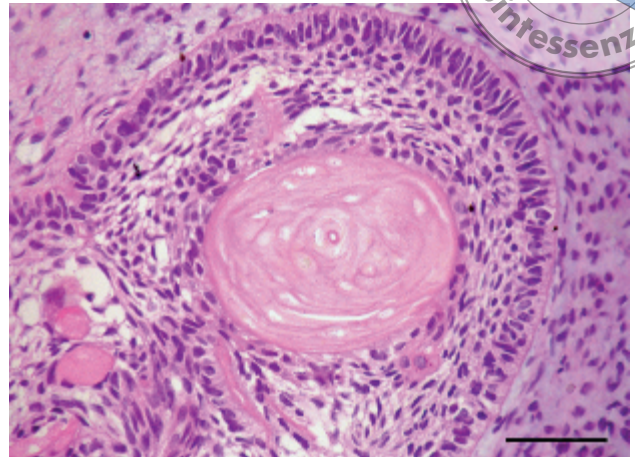


Fig 5 High-power photomicrograph showing typical ghost cells in a ameloblastic epithelial island, bar = 50 μm .

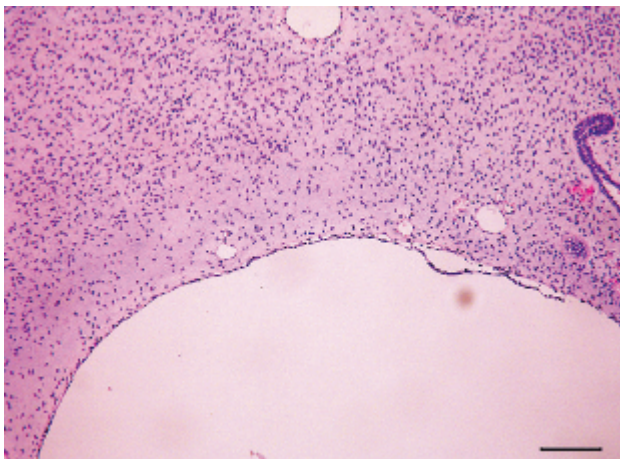


Fig 6 The cystic region is lined by a single layer of epithelium, bar = 150 μm .

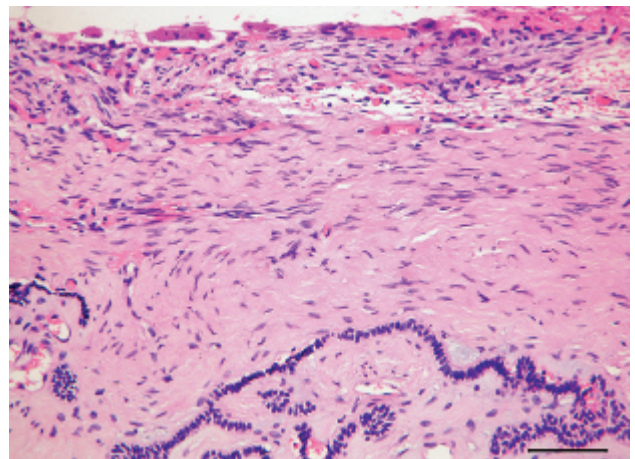


Fig 7 Collagenous fibrous tissues capsule with osteoclasts in the bony side, bar = 100 μm .

mesenchymal component resembling dental papilla, consisting of fibroblasts and colloid ground substance with little collagen fibres. Occasionally there were some mitoses among the fat fibroblasts.

The most prominent characteristics were the ghost cells and the calcifying deposition that appeared in the neoplastic epithelial nests. The ghost cells, just like those in the calcifying odontogenic cystic tumour, tended to gather in small groups and the calcification seemed to occur in the ghost cell background (Fig 5). In the cystic region, the cyst wall varied from lined with a single layer of flatting epithelial cells to without any epithelial lining (Fig 6). The tumour was capsulated with collagenous fibrous tissues and osteoclasts could be seen in the bony side of the capsule (Fig 7).

Discussion

An ameloblastic fibroma is a rare odontogenic tumour with both neoplastic epithelial and ectomesenchymal components. Ameloblastic fibromas containing ghost cells such as those in calcifying odontogenic cysts are an even rarer event³⁻⁶. So far there have only been 7 such cases reported in English literature. They all had a structure of a calcifying odontogenic cyst with a lining epithelium showing ghost cell differentiation and calcification, whereas the components of the ameloblastic fibroma existed in the cyst wall or some solid area. In Yoon's reported case, ghost cell masses were also found within the ameloblastic epithelium in the area of the ameloblastic fibroma⁴. Although our case also was



cystic with a solid region, the cystic wall did not show any histological features of a calcifying odontogenic cyst. The ghost cells and calcification were only found within the ameloblastic epithelial components, locating themselves in close association with stellate cells in the nests centers.

Generally, ghost cells appear especially in a group of neoplasms, including calcifying cystic odontogenic tumours, dentinogenic ghost cell tumours and odontogenic ghost cell carcinomas. But they had been occasionally reported to associate with other odontogenic tumours – for example in a calcifying cystic odontogenic tumour – such as odontomas^{7,8}, ameloblastomas⁹, adenomatoid odontogenic tumours¹⁰ and ameloblastic fibromas. As to calcifying cystic odontogenic tumours with ameloblastic fibromas, there are only 7 previously documented cases in the English literature. The mechanism of such a combination is unknown. They might occur either as a collision of 2 separated tumours or as a new development of one tumour from another⁴. Although some investigators believe that some odontogenic tumours could occur secondarily from calcifying cystic odontogenic tumours, as reported by Praetorius et al⁵ and Farman et al⁶, the ghost cells and calcification might differentiate secondarily from ameloblastic epitheliums of preexisting ameloblastic fibromas under certain stimulation. Because in our case the ghost cells and calcification were only found in a few ameloblastic epithelial islands and the lesion did not show other features of a calcifying cystic odontogenic tumour, such as a ghost cell containing epithelial lining, the histogenesis and pathogenesis are worth further investigation.

The behaviour of ameloblastic fibromas with ghost cell differentiation is not clear. The treatment is conservative and similar to that of an ameloblastic fibroma, such as enucleation or curettage. Recurrence has not been reported. Information accumulation is needed from further reporting cases to clarify the long-term outcome of the tumour.

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