



Correspondence

Glandular odontogenic cyst in the anterior mandible



KEYWORDS

Glandular odontogenic cyst;
Mandible;
CK19;
Mastermind-like 2 (MAML2) gene rearrangement;
Fluorescent in situ hybridization (FISH)

Glandular odontogenic cyst (GOC) is a rare and aggressive developmental odontogenic cyst with a strong predilection for the anterior region of the jaws. This report presented a case of GOC in the anterior region of the mandible.

This 62-year-old female patient visited the oral and maxillofacial surgery department of our hospital for treatment of a mucoepidermoid carcinoma in the anterior mandible, which was confirmed histopathologically in another hospital. Oral examination showed no definite swelling of the anterior mandible (Fig. 1A), but mobility of teeth 31, 41, 42, and 43 and percussion pain of tooth 42 were noted. Panoramic and periapical radiographs revealed a multilocular radiolucent lesion at the anterior mandibular region from tooth 31 to tooth 44 (Fig. 1B and C). Because the initial biopsy of the lesion showed a mucoepidermoid carcinoma, marginal mandibulectomy from tooth 31 to tooth 45 was performed to remove the tumor. Histopathological examination of the specimen showed a cystic lesion lined by non-keratinized stratified squamous epithelium of varying thickness and papillary surface. Mucin-secreting cells and microcysts were discovered in the lining epithelium (Fig. 1D). Moreover, small satellite cysts with mucous cells and microcysts in the lining epithelium and a row of eosinophilic cuboidal cells on the lining epithelial surface were found in the fibrous cystic wall (Fig. 1E and F). In

addition, spherical nodules were occasionally observed in the lining epithelium (arrows, Fig. 1F). The lining epithelium was diffusely and strongly positive for CK 19 (Fig. 1G). However, the lining epithelial cells were negative for Mastermind-like 2 (MAML2) gene rearrangement detected by fluorescent in situ hybridization (FISH) (Fig. 1H). All these findings supported the diagnosis of a GOC. The clinical course of the patient was uneventful and no recurrence of the lesion was found 3 months after the operation.

The characteristic histological features of GOC include the lining epithelium of variable thickness, mucous cells and microcysts in the lining epithelium, papillary projections, epithelial spheres, and eosinophilic cuboidal cells on the surface of the lining epithelium; all of these can be detected in our GOC case. The immunohistochemical stains can be used for identification of tumor cell type and origin.^{1–4} The lining epithelial cells of our GOC case were all positive for CK 19 which was a biomarker of odontogenic epithelial cells. In addition, approximately 80% of mucoepidermoid carcinomas are positive for MAML2 gene rearrangement.⁵ However, our GOC case was negative for MAML2 gene rearrangement. Therefore, the histological features as well as immunostaining and FISH findings all favored the diagnosis of a GOC for this patient.

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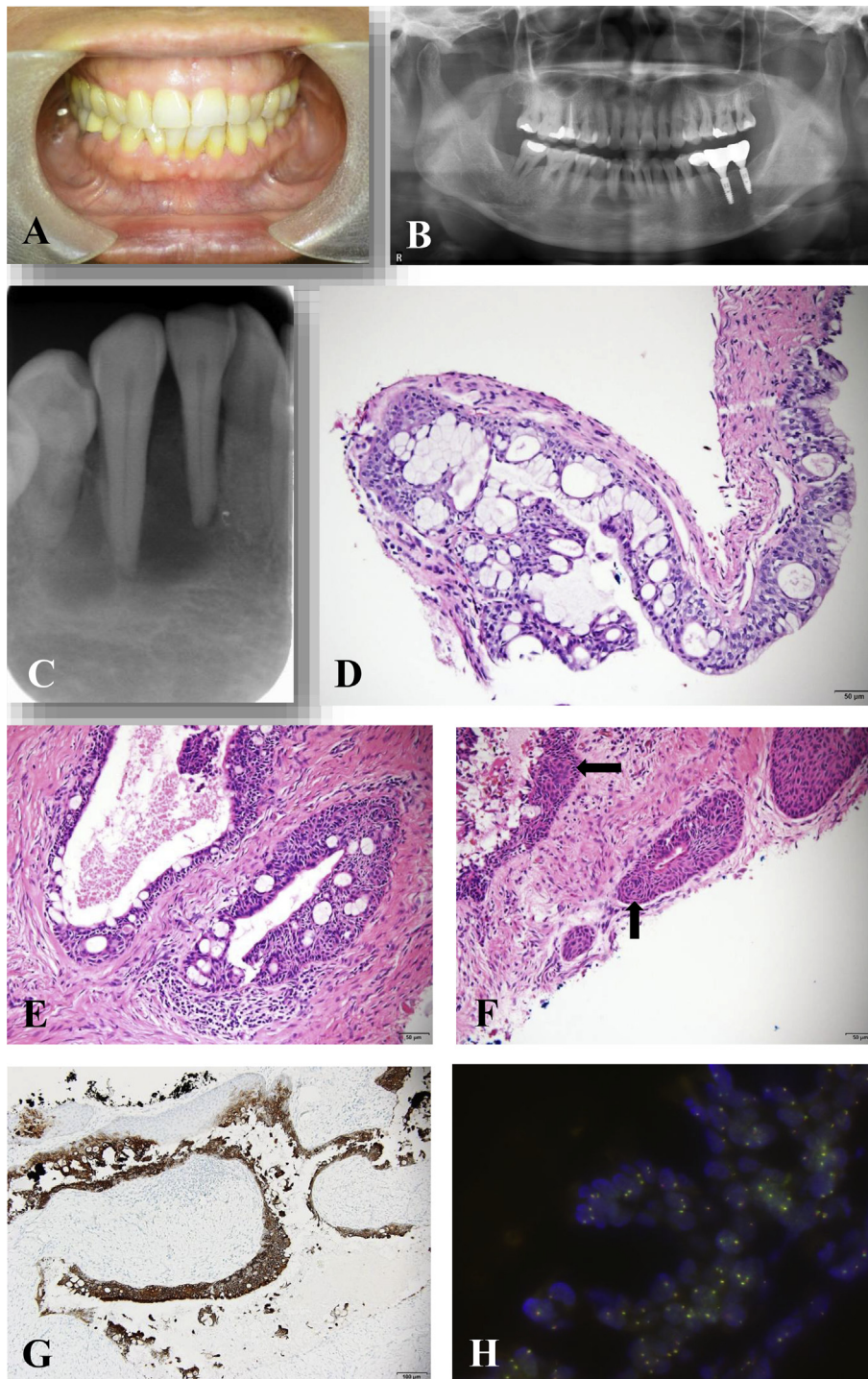


Figure 1 Clinical, radiographic, histological, immunostaining, and fluorescent in situ hybridization (FISH) photographs of our glandular odontogenic cyst case. (A) Clinical photograph showed no definite swelling of the anterior mandible. (B and C) Panoramic and periapical radiographs revealed a multilocular radiolucent lesion at the anterior mandibular region from tooth 31 to tooth 44. (D) High-power view demonstrated a cystic lesion lined by non-keratinized stratified squamous epithelium of varying thickness and papillary surface. Mucin-secreting cells and microcysts were discovered in the lining epithelium (H&E stain, original magnification, 20 \times). (E) High-power view showed small satellite cysts with mucous cells and microcysts in the lining epithelium and a row of eosinophilic cuboidal cells on the lining epithelial surface (H&E stain, original magnification, 20 \times). (F) Spherical nodules were occasionally observed in the lining epithelium (arrows, H&E stain, original magnification, 20 \times). (G) The lining epithelium was diffusely and strongly positive for CK 19 (immunostain, original magnification, 20 \times). (H) The lining epithelial cells were negative for Mastermind-like 2 (MAML2) gene rearrangement detected by FISH (original magnification, 100 \times).

Our GOC case was initially misdiagnosed as a mucoepidermoid carcinoma. This led us to use the marginal mandibulectomy for treating our patient. The multilocular GOC is supposed to be an aggressive lesion that is usually treated by a more radical surgery. Therefore, although our patient was misdiagnosed in the beginning, the marginal mandibulectomy was still a proper treatment for our patient.

Conflicts of interest

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

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