Journal of Dental Sciences 18 (2023) 486–488

Available online at www.sciencedirect.com

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journal homepage: www.e-jds.com

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Intraosseous mucoepidermoid carcinoma arising from odontogenic keratocyst

KEYWORDS
Intraosseous;
Mucoepidermoid carcinoma;
Odontogenic keratocyst

Intraosseous mucoepidermoid carcinoma (MEC) arising in jaws is extremely rare, which accounts for only 2%–3% of all MECs. The possible origins of central MEC include the epithelial lining of an odontogenic cyst or the epithelial rests of dental lamina, entrapped salivary gland tissue during embryonic development, salivary choristoma, iatrogenically entrapped salivary tissue, and epithelium of maxillary sinus.[1,2] In the current study, we presented an intraosseous MEC arising from an odontogenic keratocyst (OKC).

A 55-year-old male patient was referred from a local dental clinic for a swelling in the left mandibular area for two months. Intraoral examination showed a painless, firm mass with a smooth surface over the left lower buccal vestibule corresponding to edentulous ridge of teeth 34 and 35 area, measuring 2.0 × 1.0 cm in dimension. Trace back the history; the patient had an extraction of teeth 34 and 35 in a local dental clinic five years ago. Due to poor healing of the extraction sockets, he was referred to our oral surgery department for further management. The panoramic radiography showed a bony defect of alveolar crest of teeth 34 and 35 area (Fig. 1A). When surgical debridement was performed, the histopathological findings revealed an OKC (Fig. 1B). After treatment, the patient was lost from follow-up. Five years later, panoramic radiography displayed a partially ill-defined radiolucent lesion over the left mandibular body with an intact alveolar crest (Fig. 1C). Combining the history and clinical findings, a recurrent OKC was suspected. Then, the patient received a marginal resection of the left mandibular body. The histopathological examination shows a polycystic lesion lined by variable thickness of parakeratinized stratified squamous epithelium. Some of the tumor cells are arranged in nests or strands infiltrating in the desmoplastic stroma (Fig. 1D and E). Multifocal glandular differentiation of the cystic epithelium was noted, which included the presence of mucous cells, microcysts, ciliated cells, apocrine snouting, tufting, and vacuolated cells (Fig. 1F). The mucous cells were highlighted by the mucicarmine stain (Fig. 1G). Immunohistochemically, the tumor cells with squamous differentiation were positive for CK19 (Fig. 1H), and the luminal, glandular compartment was positive for CK7 (Fig. 1I). No MAML2 rearrangement was identified by fluorescence in situ hybridization (FISH). The cystic components of the tumor were >20%, and no neural invasion, necrosis, > 4 mitoses/10 high-power-fields (HPF), and anaplasia were noted. Based on these findings, a low-grade intraosseous MEC arising from an OKC was diagnosed.

Central MECs of the jaws mostly occur in patients with 50–70 years of age, and about two-third s of cases affect the mandible. No prominent gender predilection is identified. The clinical manifestations include swelling, pain, and numbness.[3] The differentiation of glandular odontogenic cyst (GOC) from central MEC is difficult. The FISH for MAML2 might help to differentiate between these two entities, the previous study found no MAML2 rearrangement in GOCs.[4] However, only about 50% of intraosseous MECs harbor MAML2 rearrangement.[5] Considering the infiltrative growth pattern and invasion to adjacent stroma, an intraosseous MEC was rendered in this case.

https://doi.org/10.1016/j.jds.2022.10.015

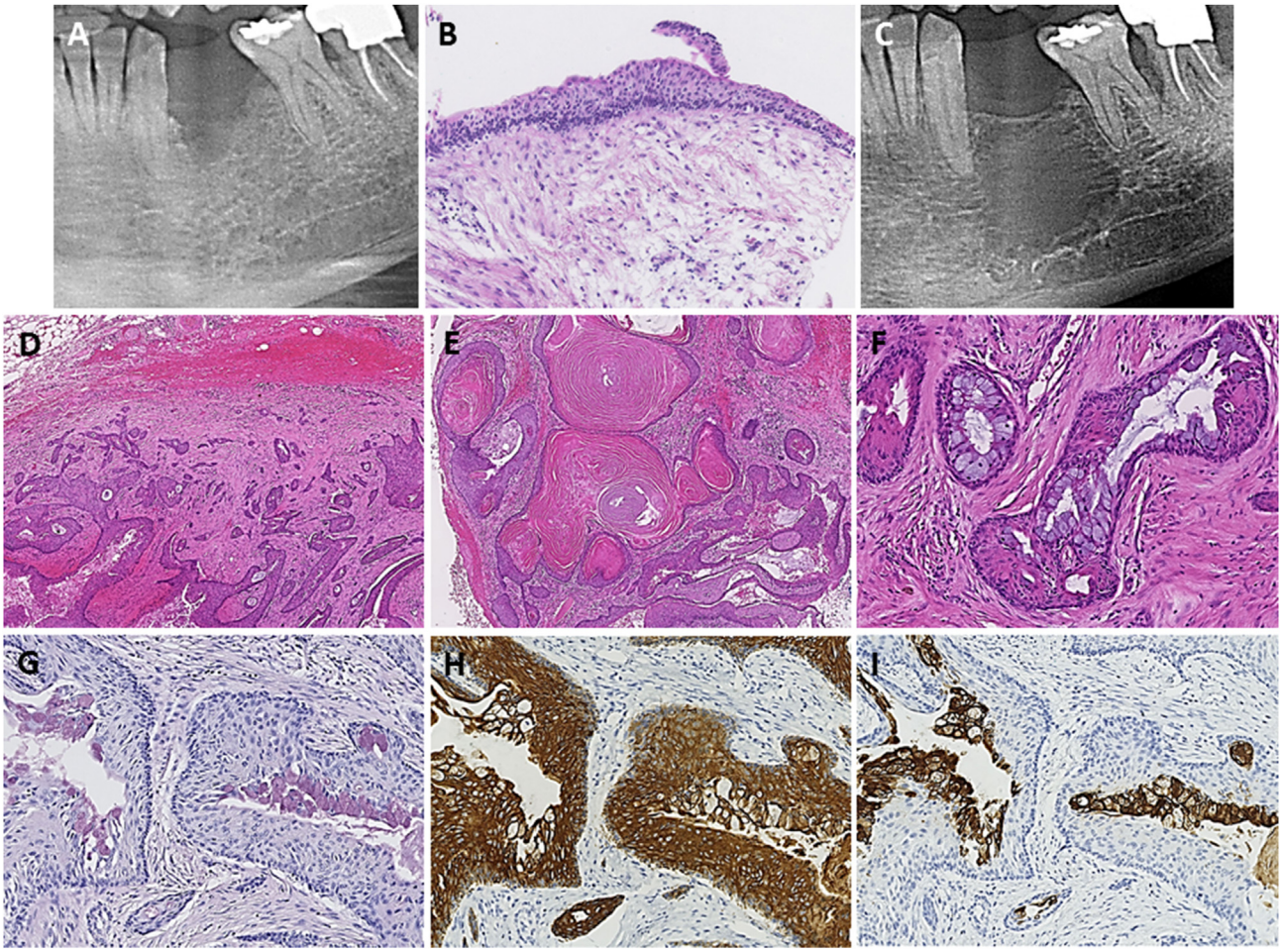

Figure 1 Radiographic and microscopic photographs of the current case of intraosseous mucoepidermoid carcinoma in the mandible. (A) A bony defect of alveolar crest of teeth 34 and 35 area. (B) A cystic lesion lined by parakeratinized stratified squamous epithelium with palisaded and hyperchromatic basal cells. (C) A partially ill-defined radiolucent lesion of the left mandibular body with an intact alveolar crest. (D and E) A polycystic lesion lined by variable thickness of parakeratinized stratified squamous epithelium; tumor cells were arranged in nests or strands infiltrating in the desmoplastic stroma. (F) Glandular differentiation in the luminal part of the cystic epithelium (Hematoxylin and eosin stain; original magnification; D, 10×; E, 10 × ; F, 40 ×). (G) The mucous cells of luminal area were highlighted by the mucicarmine stain (magnification, 40×). (H) All of the tumor cells with squamous differentiation were positive for CK19 (magnification, 40×). (I) Whereas, the luminal, glandular compartment was positive for CK7 (magnification, 40×).

Declaration of competing interest

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

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Received 14 October 2022
Final revision received 16 October 2022
Available online 30 October 2022