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Intraosseous mucoepidermoid carcinoma arising from odontogenic keratocyst

KEYWORDS

Intraosseous;
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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-thirds 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.

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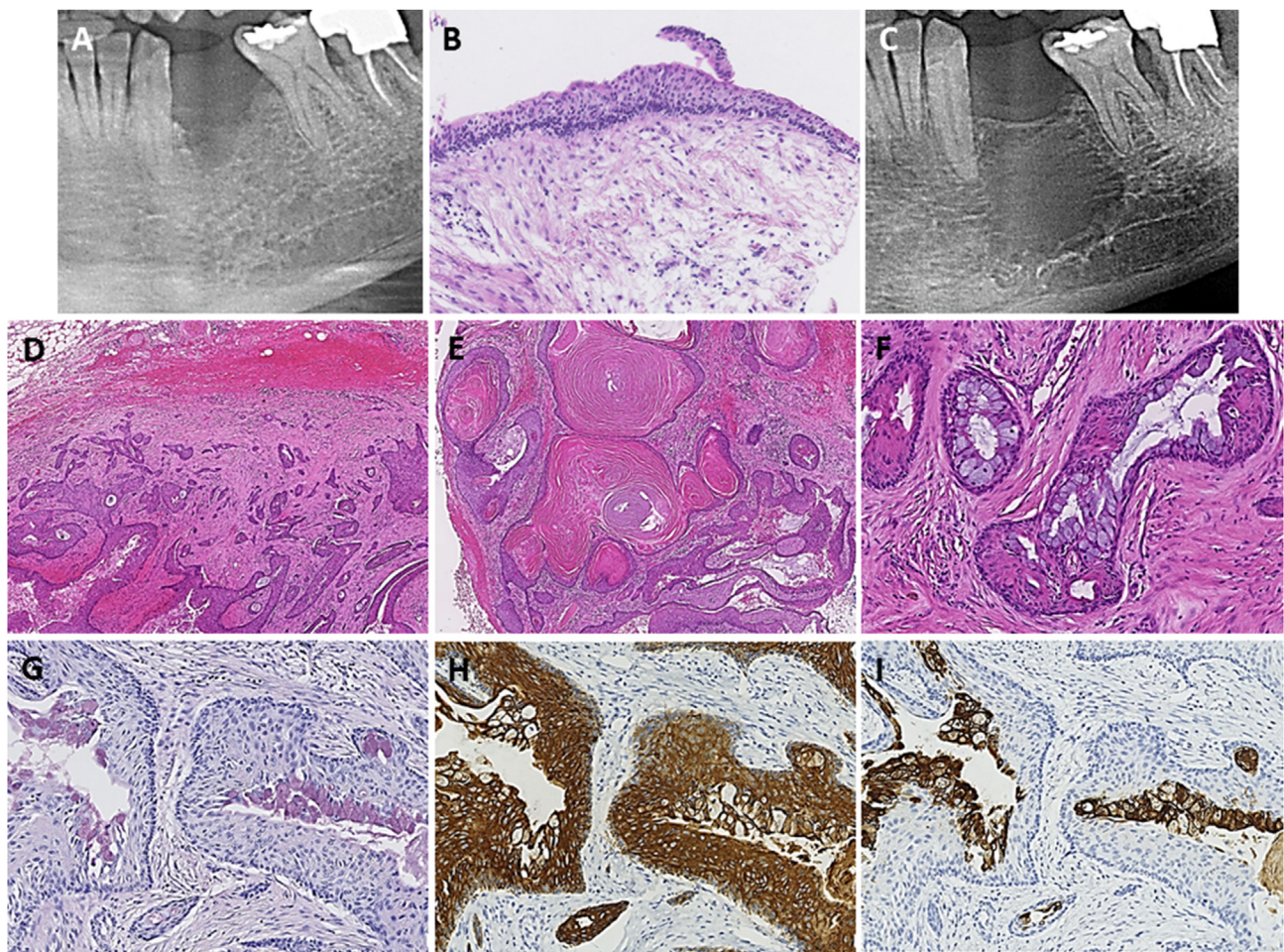


Figure 1 Radiographic and microscopic photographs of the current case of intraosseous mucoepidermoid carcinoma in the mandible. (A) A bony defect of alveolar crest of teeth 34 and 35 area. (B) A cystic lesion lined by parakeratinized stratified squamous epithelium with palisaded and hyperchromatic basal 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 \times ; E, 10 \times ; F, 40 \times). (G) The mucous cells of luminal area were highlighted by the mucicarmine stain (magnification, 40 \times). (H) All of the tumor cells with squamous differentiation were positive for CK19 (magnification, 40 \times). (I) Whereas, the luminal, glandular compartment was positive for CK7 (magnification, 40 \times).

Declaration of competing interest

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

References

1. Woo VI, Chi AC, Neville BW. 10 - odontogenic cysts and tumors. In: Gnepp DR, Bishop JA, eds. *Gnepp's diagnostic surgical pathology of the head and neck*, 3rd ed. Oxford: Elsevier, 2021: 827–80.
2. Bouquot JE, Gnepp DR, Dardick I, Hietanen JH. Intraosseous salivary tissue: jawbone examples of choristomas, hamartomas, embryonic rests, and inflammatory entrapment: another histogenetic source for intraosseous adenocarcinoma. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2000;90:205–17.
3. de Souza LL, Pontes FSC, Pontes HAR, Neto NC, de Carvalho WRS, Guimarães DM. Central mucoepidermoid carcinoma: an up-to-date analysis of 147 cases and review of prognostic factors. *J Cranio-Maxillo-Fac Surg* 2018;46:162–7.
4. Bishop JA, Yonescu R, Batista D, Warnock GR, Westra WH. Glandular odontogenic cysts (GOCs) lack MAML2 rearrangements: a finding to discredit the putative nature of GOC as a precursor to central mucoepidermoid carcinoma. *Head Neck Pathol* 2014;8:287–90.
5. Bell D, Lewis C, El-Naggar AK, Weber RS. Primary intraosseous mucoepidermoid carcinoma of the jaw: reappraisal of the MD Anderson Cancer Center experience. *Head Neck* 2016;38(Suppl 1):E1312–7.

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