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Orthokeratinized odontogenic cyst with foreign body giant cell reaction to luminal keratin materials at the exposed fibrous connective tissue wall

KEYWORDS

Orthokeratinized
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Orthokeratinized odontogenic cyst (OOC) is a specific type of odontogenic cyst with a lower recurrence rate.¹ In this case report, we presented an OOC with foreign body giant cell reaction to luminal keratin materials at the exposed fibrous connective tissue wall in the right maxillary third molar region.

This 24-year-old male patient was referred to the dental department of our hospital for treatment of a radiolucent lesion around the crown of the impacted right maxillary third molar. Oral examination showed the missing of the right maxillary third molar in the oral cavity and a slight expansion of the buccal and palatal cortical plates of the right maxillary third molar region. Panoramic radiography revealed a unilocular radiolucent lesion surrounding the crown of the impacted right maxillary third molar. Therefore, a clinical diagnosis of a central type of dentigerous cyst was made. After discussing with the patient, the treatment plan was extraction of the impacted right maxillary third molar followed by total enucleation of the cystic lesion. After obtaining the signed informed consent, extraction of the impacted right maxillary third molar was performed under local anesthesia and the cystic lesion was totally removed. The excised soft tissue specimen was sent for histopathological examination. Microscopically, the specimen showed a cystic lesion lined by the orthokeratinized stratified squamous epithelium of 5–7 layers of

epithelial cells. The superficial granular cell layer was prominent and composed of one to two layers of granular epithelial cells with keratohyaline granules in the cytoplasm. Many keratin shreds were found in the cystic lumen (Fig. 1A, B, C and D). A daughter OOC was discovered in the fibrous cystic wall (Fig. 1B and D). In the subepithelial area with a moderate to severe lymphoplasmic cell infiltrate, loss of superficial granular cell layer and surface keratin layer from the lining stratified squamous epithelium was found (Fig. 1B). In a specific area with complete loss of the lining epithelium and exposure of the underlying fibrous connective tissue to the keratin shreds in the cystic lumen, foreign body reaction with formation of multinucleated foreign body giant cells at the surface of the fibrous connective tissue was noted (Fig. 1E, F, G and H). The multinucleated foreign body giant cells had foamy cytoplasm, indicating the phagocytosis of the cystic keratin materials into the cytoplasm of the surface multinucleated foreign body giant cells (Fig. 1G and H). The characteristic microscopic findings confirmed the histopathological diagnosis of an OOC with foreign body giant cell reaction to luminal keratin materials at the exposed fibrous connective tissue wall.

The keratin layer on the surface of oral mucosa or skin has its function to protect the underlying epithelial and connective tissue cells. However, when the keratin materials are exposed to the stromal connective tissue cells,

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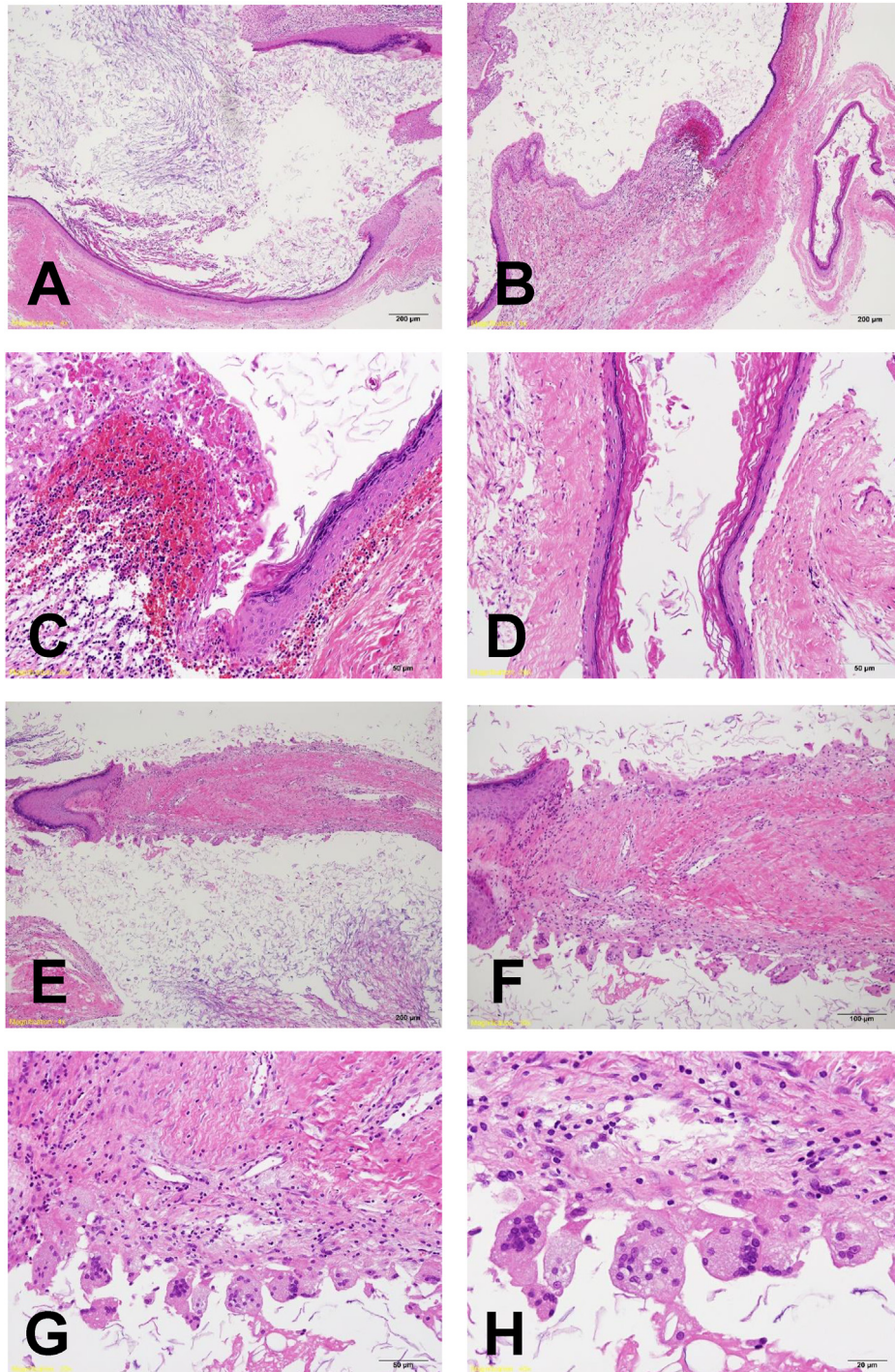


Figure 1 Histopathological photomicrographs of our orthokeratinized odontogenic cyst (OOC). (A, B, C and D) Low- and high-power photomicrographs showing a cystic lesion lined by the orthokeratinized stratified squamous epithelium of 5–7 layers of epithelial cells. The superficial granular cell layer was prominent and composed of one to two layers of granular epithelial cells with keratohyaline granules in the cytoplasm. Many keratin shreds were found in the cystic lumen. A daughter OOC was discovered in the fibrous cystic wall (B and D). In the subepithelial area with a moderate to severe lymphoplasmacytic cell infiltrate, loss of superficial granular cell layer and surface keratin layer from the lining stratified squamous epithelium was found (B). (E, F, G and H) In a specific area with complete loss of the lining epithelium and exposure of the underlying fibrous connective tissue to the keratin shreds in the cystic lumen, foreign body reaction with formation of multinucleated foreign body giant cells at the surface of the fibrous connective tissue wall was noted. The multinucleated foreign body giant cells had foamy cytoplasm, indicating the phagocytosis of the luminal keratin materials into the cytoplasm of the surface multinucleated foreign body giant cells (G and H) (Hematoxylin and eosin stain; original magnification; A, B and E, 4 × ; F, 10 × ; C, D and G, 20 × ; H, 40 ×).

such as in the cases of well-differentiated squamous cell carcinoma with formation of tumor keratin, squamous cell carcinoma with chemotherapeutic drug-induced keratin formation after adjuvant chemotherapy, and the ruptured epidermal cyst or ruptured OOC with exposure of the fibrous connective tissue wall to luminal keratin materials, foreign body reaction with phagocytosis of the keratin materials by the multinucleated foreign body giant cells may occur.^{2–5}

In this case, there was no inflammation in the most parts of the subepithelial fibrous connective tissue wall, suggesting that this OOC is a developmental odontogenic cyst. However, in the minor parts of the fibrous cystic wall, when there was moderate or severe inflammation in the subepithelial connective tissue areas, loss of characteristic superficial granular cell layer and surface keratin layer from the lining stratified squamous epithelium could be discovered.

Declaration of competing interest

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

Acknowledgments

None.

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