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CORRESPONDENCE

Keratin debris-filled ruptured salivary duct cyst



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Salivary duct cyst (SDC) is a cystic lesion with the lining epithelium arising from the excretory duct of salivary glands. SDC is thought to develop from salivary duct obstruction. The major content of the SDC is mucous proteins. However, keratin debris may be identified in the SDC or mucocele occasionally.¹ Here, we report a case of ruptured SDC with abundant keratin debris filled in the lumen of the cyst.

This 22-year-old female patient bit her left labial mucosa 4 months ago. Subsequently, a swelling developed in the left lower labial mucosa. She tried to regress the swelling by massage, but the swelling persisted. Therefore, she came to our hospital in search of treatment. Clinical examination revealed a swelling in the left lower lip, measuring 1.0 cm × 0.7 cm, with a smooth surface and normal mucosal color (Figure 1A). The consistency was elastic and no fluctuation or palpation pain was noted. The differential diagnosis included mucocele, salivary duct cyst, salivary gland tumor, fibroma, and other rare soft-tissue tumors. A mucocele was provisionally diagnosed and an excisional biopsy was performed under local anesthesia.

Microscopically, multiple cystic spaces were seen in the submucosa and the cystic spaces were majorly lined with a

thick layer of connective tissue containing epithelioid histiocytes and multinucleated giant cells (which were immunoreactive to CD68, data not shown) in the inner layer, and an infiltrate of chronic inflammatory cells in the outer layer, minorly lined by focally keratinized stratified squamous epithelium (Figure 1B). In some areas, the cystic content was composed of numerous spherical, trabecular, or filamentous acellular eosinophilic materials (Figure 1C). Some of these irregularly-shaped materials were found to be positive by alcian blue (Figure 1D) and mucicarmine stains (Figure 1E). In addition, some of the materials were cytokeratin-positive (Figure 1F), indicating they were exfoliated keratin debris from the cystic lining epithelium. Thus, a keratin debris-filled ruptured SDC with foreign body reaction is considered.

The majority of SDCs are caused by ductal obstruction, and thus are filled with mucinous materials that cannot be excreted into the oral cavity. Because the SDC is usually lined by nonkeratinized stratified squamous epithelium, the cystic cavity is rarely filled with keratin debris. However, our SDC was lined by focally keratinized stratified squamous epithelium and the cystic content was confirmed to be keratin proteins by the immunostain. When the keratin debris-filled SDC ruptured, the keratin proteins were released into the adjacent soft tissue and finally resulted in the formation of a cystic lesion surrounded by a thick layer of foreign body granuloma-like tissue composed predominantly of epithelioid histiocytes and multinucleated foreign body giant cells similar to those seen in this case. In actual fact, the immunostains may help to confirm the tissue origin of the cells; for example, they are useful for identification of Langerhans cells in central granular cell odontogenic

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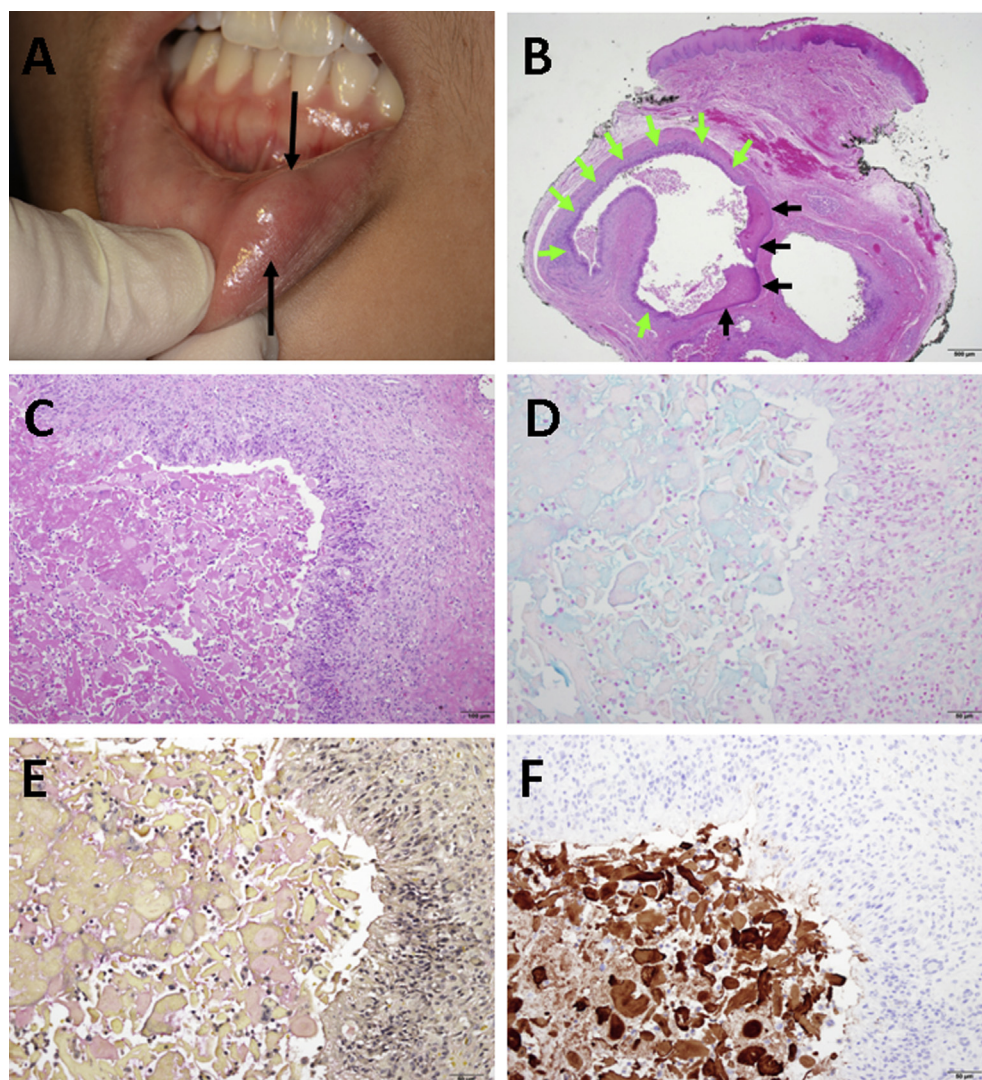


Figure 1 Clinical, histopathological, and immunostained photographs of a keratin debris-filled ruptured salivary duct cyst (SDC) with foreign body reaction. (A) Clinical photograph showing a nodule in the left lower labial mucosa. (B) Histopathological microphotograph of the SDC revealing multiple cystic spaces majorly lined by a thick layer of connective tissue containing epithelioid histiocytes and multinucleated giant cells in the inner layer, and an infiltrate of chronic inflammatory cells in the outer layer (green arrows) minorly lined by focally keratinized stratified squamous epithelium (black arrows; hematoxylin and eosin (H&E) stain; original magnification, 4 \times). (C) High-power view of the ruptured SDC exhibiting the cystic content composed of numerous spherical, trabecular, or filamentous acellular eosinophilic materials surrounded predominantly by aggregates of epithelioid histiocytes and multinucleated giant cells (H&E stain; original magnification, 20 \times). Some of these irregularly shaped materials were positive by (D) alcian blue (original magnification, 20 \times) and (E) mucicarmine stains (original magnification, 20 \times). (F) Immunohistochemical stain showing cytokeratin-positive keratin debris in the cyst lumen (original magnification, 20 \times).

tumors, in lining epithelia of odontogenic cysts, and in odontogenic epithelia of odontogenic fibromas.^{2–5} In our case, the cystic contents were positive by the mucicarmine stain, and stratified squamous epithelial lining was found in focal areas. These findings further proved that our cystic lesion was of salivary gland origin and actually an SDC.

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