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CASE REPORT

Sclerosing mucoepidermoid carcinoma of the oral cavity

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Sclerosing mucoepidermoid carcinoma (SMEC) with eosinophilia is a rare but distinctive tumor usually affecting the thyroid. SMEC involvement of salivary gland is exceptional, with only six cases in the literature. We present here the first case of an intermediate-grade SMEC, arising from the intraoral minor salivary glands. A particularly interesting finding is the cytoplasmic accumulation of eosinophilic hyaline granules in carcinoma cells, similar to aberrant zymogen-like granules previously described in salivary sclerosing polycystic adenosis. *J Oral Pathol Med* (2005) **34**: 187–9

Keywords: minor salivary gland; sclerosing mucoepidermoid carcinoma; sclerosing polycystic adenosis; tumor-associated lymphoid proliferation

A 28-year-old man presented with a gingival mass, which had been present for 1 year. Examination revealed a non-ulcerated, firm, polypoid, 20 × 20 mm, tumor on the right retromolar pad. The lesion tethered to gingiva, but not fixed to the underlying bone. There was no lymphadenopathy. The mass was easily shelled out and surgeons ensured tumor-free margins by intraoperative frozen section analysis. Postoperative course was uneventful.

A well-demarcated but non-encapsulated tumor was centrally composed of hypocellular sclerotic tissue with a peripheral rim of tumor-associated lymphoid proliferation [TALP (1); Fig. 1a]. A dense, hyalinized stroma segregated nests of carcinoma cells, giving a scirrhous appearance (Fig. 1b). A varying admixture of mucous, epidermoid and intermediate cells was evident in tumor islands distributed along the margins of the lesion. Irregularly shaped and sized neoplastic ducts were filled with mucinous material (Fig. 1c). Most tumor cells reacted with cytokeratin (AE1/AE3, 1:50; Dako, Glostrup, Denmark), while staining for vimentin (V9, 1:200; Dako), α-smooth muscle actin (1A4, 1:800; Dako) and S-100 protein (1:1000; Dako) was negative. In addition to non-neoplastic ductal remnants, there were also small separate clusters of mucoepidermoid carcinoma (MEC) within the TALP, reminiscent of metastatic foci (Fig. 2a). Often, carcinoma cells were filled with intensely eosinophilic, intracytoplasmic hyaline granules suggestive of the cellular degenerative process (Fig. 2b). The rupture of carcinomatous ducts and spillage of mucin into the stroma were readily observed (Fig. 2c). Although increased mitoses, anaplasia and necrosis were absent, perineural invasion was evident (Fig. 2d). In many areas, an eosinophilic infiltrate was lacking or subtle.

Comments

In 1963, Gray et al. (2) in their series of MEC of the salivary glands, briefly described a unique variation in which mucin has spilled into the stroma and incited dense scarring with mononuclear cell infiltration. Presumably, this case is the first description of sclerosing mucoepidermoid carcinoma (SMEC). Clinical summaries of salivary SMEC are as follows (3–6): (i) ages of the patients ranged from 17 to 65 years, with a mean of 43 years; (ii) there were four male and three female subjects; (iii) lesions were slow-growing masses in the parotid gland ($n = 4$), submandibular gland, parapharyngeal space and retromolar gingiva ($n = 1$ each); (iv) tumors varied from 2 to 6 cm in greatest dimension; and (v) two patients developed lymph node metastasis and one of them died of lung metastasis as long as 7 years later (6).

The constellation of histologic features are (i) a well-defined sclerotic lesion with limited but notable foci of low-to high-grade MEC, (ii) broad front, often circumscribed invasion with or without perineural spread, (iii) predominantly solid but a few small cystic structures, (iv) striking fibrous scarring around the mucous pool; (v) constant association and close intermingling with TALP, and (vi) repletion of eosinophils in some but not all lesions (4, 6). The hitherto undescribed finding in our SMEC is that the carcinoma cells were frequently packed with brightly eosinophilic granules. It is of interesting that such a degenerative cytoplasmic granularity is an important diagnostic clue to sclerosing polycystic adenosis (SPA) of the salivary gland (7). Given the concomitance of dense lymphocytic infiltration with germinal center formation in SPA (7), both lesions may share a similar sclerosing basis.

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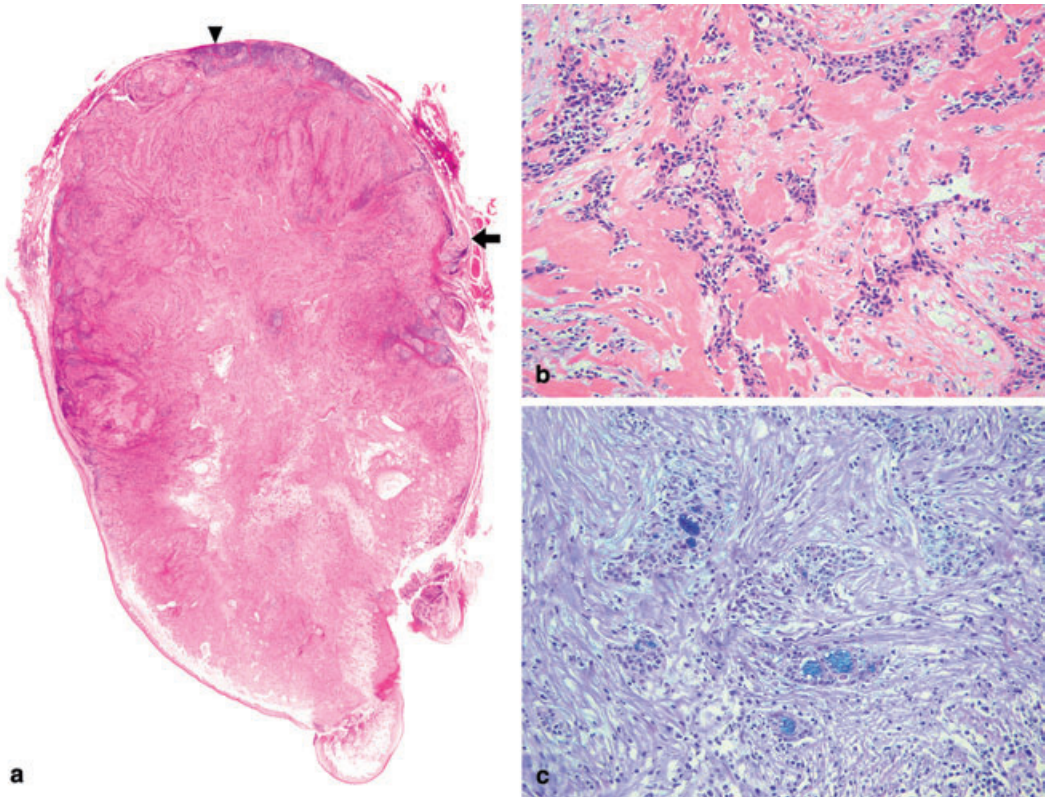

Figure 1 (a) Unencapsulated, circumscribed tumor showing central sclerosis and lymphocytic rim. Area indicated by arrowhead or arrow is enlarged in (2a) or (2d). (b) Scar-like zone containing only foci of carcinoma (hematoxylin and eosin stain; a, ×3.5; b, ×200). (c) Neoplastic ducts widely spaced by desmoplastic stroma (alcian blue/periodic acid-Schiff stain; ×200).

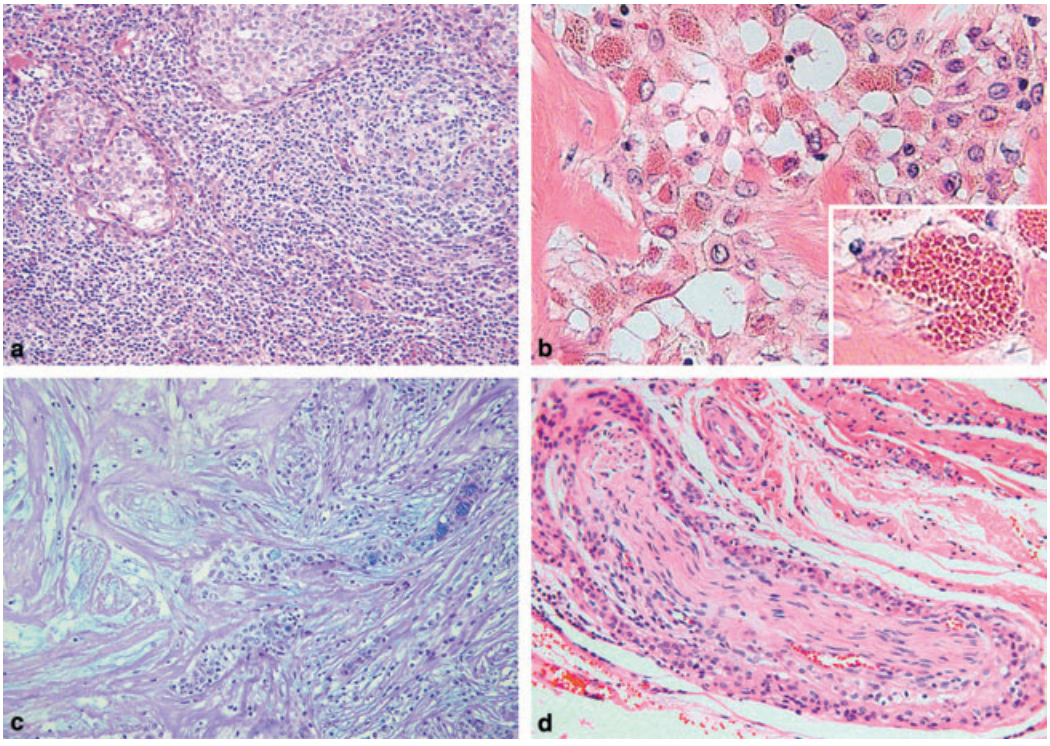

Figure 2 (a) Isolated nests of mucoepidermoid carcinoma embedded in lymphoid tissue. (b) Eosinophilic granules in carcinoma cells. (c) Fibrous scarring around the spilled mucin. (d) Perineural invasion (hematoxylin and eosin stain; a and d, ×200; b, ×400; alcian blue/periodic acid-Schiff stain; c, ×200).

The general consensus is that salivary SMEC can behave in an indolent manner with prolonged survival (3–6). There is interesting debate regarding the histogenesis. The pronounced desmoplasia may be interpreted as an exaggerated mucous escape reaction (3). Unlike thyroid SMEC with eosinophilia, which occurs in the setting of chronic lymphocytic thyroiditis, the significance of TALP for SMEC in the salivary glands remains unexplained. A larger study is required to determine whether this appearance is simply a secondary defensive response to tumor growth (1) or represents an important background lymphoepithelial lesion.

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