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CASE REPORT

Myxolipomatous pleomorphic adenoma: an unusual oral presentation

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The first case of a myxoid variant of lipomatous pleomorphic adenoma arising in the intraoral minor salivary gland is presented. A well-encapsulated tumor was composed almost entirely of myxolipomatous tissue with honeycomb-like spindled cellular areas, which contained only a scant glandular element. Immunohistochemistry confirmed the myoepithelial nature of spindle cells intimately admixed with mucoadipose component. We propose the term *myxolipomatous pleomorphic adenoma* for this peculiar lesion.
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Keywords: lipomatous pleomorphic adenoma; minor salivary gland; myoepithelioma; myxolipomatous pleomorphic adenoma

A 35-year-old woman presented with a 10-year history of a painless lump in the left buccal mucosa. Intraoral examination revealed a soft, freely movable mass separated from the Stensen's duct and the parotid gland. No recurrence has been experienced 8 years after surgery. Grossly, the smooth surfaced lipomatous mass measured 4 cm × 3 cm × 3 cm and had a uniform yellow slimy cut surface.

Microscopic examination revealed a thick hyalinized capsule completely surrounding the lesion (Fig. 1A). The tumor consisted predominantly of mature adipose tissue (more than 95% of the entire tumor) with extensive myxoid change containing dispersed irregularly shaped spindled cellular foci (Fig. 1B). Adipocytes were univacuolar in shape, and no lipoblasts were identified. Spindly to stellate cells surrounding adipocytes or embedded in mucoid stroma had bland nuclei, lacking substantial atypia (Fig. 1C,D). Transition between the adipocytes and vacuolated spindle cells was apparent. At the periphery, there was a scant epithelial component arranged in distorted ducts, squamoid islands or septum-like cords (Fig. 1E).

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Most spindle cells were immunoreactive for vimentin (V9, 1 : 200, Dako, Glostrup, Denmark), cytokeratin (AE1/AE3, 1 : 50; Dako) (Fig. 2A), S-100 protein (polyclonal, 1 : 1000; Dako) (Fig. 2B) and calponin (CALP, 1 : 400, Dako) and some of them for α-smooth muscle actin (1A4, 1 : 800; Dako) (Fig. 2C) and glial fibrillary acidic protein (polyclonal, 1 : 1000; Dako). They were negative for CD34 (MY10, 1 : 50; Becton Dickinson, San Jose, CA, USA).

Comments

The term 'lipomatous pleomorphic adenoma (LPA)' was coined by Seifert et al. (1) in 1999, who defined this tumor as an otherwise typical pleomorphic adenoma (PA) with an adipose component of more than 90% of the tumor area. This newly described variant is extremely rare and only one submandibular (2) and two parotid cases (1, 3) exist in the literature. Recently, four cases of lipomatous myoepithelioma of the parotid gland have been added (4).

We are unaware of any publication describing an example of LPA of the minor salivary glands. Submucosal PAs of the cheek are common but none with lipomatous features (5). Another interesting finding is the presence of widespread mucoid stroma. It is well known that occasional PAs are predominantly mucoid with a scant epithelial component (5); however, the extent of myxoid change in the previously reported LPA was only a minor (1–3).

The present tumor broadens the morphological spectrum of LPA. This may represent a diagnostic challenge and potential pitfall. The differential diagnosis includes all lipomatous tumors with significant myxoid change, such as myxolipoma, vascular myxolipoma, spindle cell lipoma, and dendritic fibromyxolipoma. Myxoid liposarcoma is an additional possibility. In addition to the immunohistochemical analysis, a thorough investigation of sections from multiple blocks helps to confirm the diagnosis.

In summary, the available data to date suggest that spindle myoepithelial tumor cells are prone to produce both the mucoid and adipose tissues (2, 4, 6). Neoplastic myoepithelium other than the plasmacytoid (hyaline) cells in PA is a dynamic cell capable of altering its phenotype along with lipogenic differentitation.

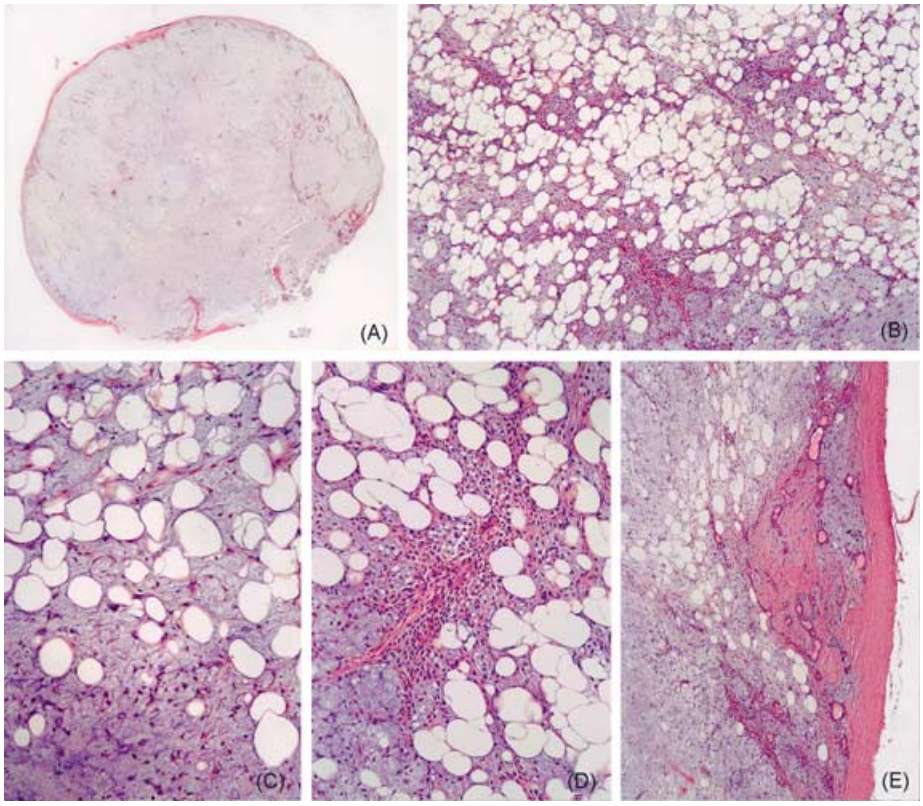

Figure 1 Hematoxylin and eosin appearance. (A) Encapsulated lipomatous tumor containing only scant epithelial components. (B) Variable mixture of mature adipose tissue, spindled cellular areas and myxoid stroma. (C) Myxolipomatous area. (D) Spindle cell lipoma-like area. (E) Typical pleomorphic adenoma area (A, ×3; B and E, ×40; C and D, ×100).

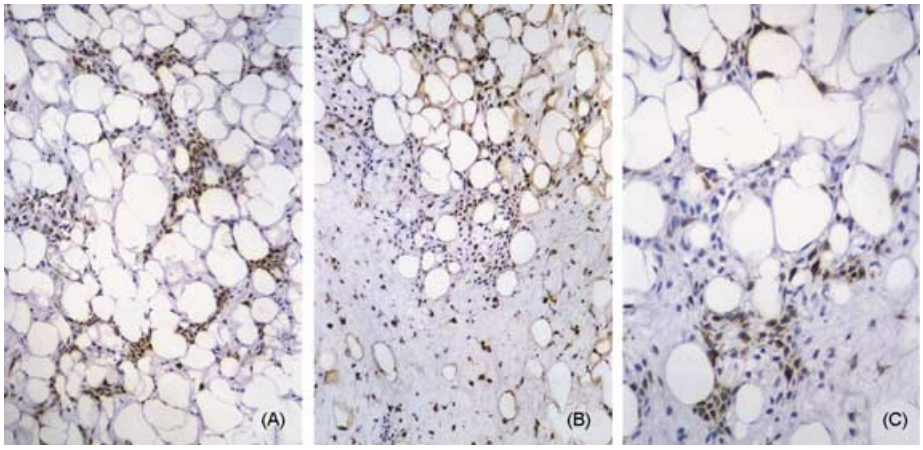

Figure 2 Spindle tumor cells are immunoreactive for cytokeratin (A), S-100 protein (B) and α-smooth muscle actin (C) (avidin–biotin–peroxidase complex method; A and B, ×100; C, ×200).

Addendum

After the original version of this manuscript was submitted, two additional cases of parotid LPA were reported (*Histopathology* 2002; **40**: 487–8; *Ann Pathol* 2002; **22**: 219–21).

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