Case Report

A Fibrous Histiocytoma of Intermediate Malignancy Arisen from the Parotid Gland

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The light microscopic and immunohistochemical characteristics of a case of fibrous histiocytoma of intermediate malignancy arising from the parotid gland are presented. This neoplasm is rare in this site and must be distinguished from other spindle cell tumors of the parotid gland, particularly those of epithelial and myoepithelial origins. Histologic characteristics similar to those displayed by dermatofibroma and dermatofibrosarcoma protuberans help to differentiate this tumor from other spindle cell tumors. The absence of cytochemical epithelial markers is useful for establishing the diagnosis. This tumor appears to have arisen from mesenchymal elements within the gland.

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Introduction

Most of salivary gland neoplasms are epithelial tumors. Mesenchymal tumors in this anatomical site are rare.[1] Lipoma, hemangioma, and lymphangioma are the most common types of these tumors. Fibrohistiocytic tumors comprise only a small number of mesenchymal tumors in salivary glands.[1]

Fibrohistiocytic tumors contain spindle cells, and so are categorized as spindle cell tumors. Fibroblasts and smooth muscle cells can be the origin of spindle cells. Therefore, myoepithelioma and fibrohistiocytic tumors are subtypes of spindle cell tumors in the salivary glands and can be a differential diagnosis for each other.[2]

The fibrohistiocytic tumors of intermediate malignancy are uncommon mesenchymal tumors.[2] These lesions can present a variety of histologic features. These tumors occur in several sites, mainly skin and subcutis.[2,3]

Herein, we describe a fibrous histiocytoma of intermediate malignancy, arising from the parotid gland, and its features are similar to those reported by Wiley et al[1] for fibrous histiocytoma of the parotid gland and by Fletcher[4] for benign fibrous histiocytomas of subcutaneous tissue.

Case Report

A 26-year-old woman presented with an intermediate-growing solid nodule in her left parotid gland for 3 months. The nodule had no pain and the facial nerve was intact.

Fine needle aspiration biopsy (FNAB) showed a few spindle cells in myxoid background material. So, we decided to excise the tumor.

After elevating the parotid flap, we excised two lymph nodes adjacent to the parotid gland. The tumor was in the superficial part of the gland but encased the lower branches of the facial nerve. We freed the nerve and excised the tumor with a rim of healthy salivary gland and performed total parotidectomy for the patient.

Grossly, the tumor was a pale tan-gray nodule measuring 2 × 1.5 × 1 cm with neatly marked margin and having a moderately firm to rubbery consistency. Histologic examination showed

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spindle cell proliferation in the form of intersecting bundles in richly vascular and myxoid stroma (Figure 1). Occasional multinucleated giant cells and focal stromal degenerative changes were noted. Mitotic figures were 5 in 10 high power fields without any atypical mitosis. No necrosis was seen. Extravasations of large number of RBCs and scattered small lymphoid cells were also noted in the stroma (Figure 2). The nodule was focally bordered by fibrocollagenous band. Sections of the salivary gland including the deep lobe showed mild fatty infiltration of the stroma.

Two small lymph nodes with benign non-specific reactive changes were also noted. Immunohistochemistry (IHC) revealed patchy positive reaction for CD68 (with 1/50 – 1/100 dilution) (Figure 3) and no reaction for S-100 protein (with 1/25 – 1/50 dilution), vimentin (with 1/100 – 1/200 dilution), smooth muscle actin (SMA) (with 1/25 – 1/50 dilution), muscle specific actin (MSA) (with 1/50 – 1/100 dilution), CD34 (with 1/50 – 1/100 dilution), factor VIII (with 1/50 – 1/100 dilution), CD117 (C-Kit) (with 1/50 – 1/100 dilution), and CD31 (with 1/20 – 1/40 dilution). All above markers were checked with Dako kits.

A diagnosis of "low-grade malignant spindle cell tumor compatible with fibrohistiocytic tumor of intermediate malignancy" was made.

Because of complete surgical excision of the tumor with total parotidectomy, the patient was followed. After 14 months of follow-up, the patient is well and free of disease with full function of her facial nerve.

Discussion

The purpose of this case report is to document the existence of a fibrous histiocytic tumor arising from the parotid gland. Fletcher reported some fibrous histiocytomas in deep subcutaneous soft tissue with benign features. Our tumor had features found in deep subcutaneous fibrous hisiocytoma such as those reported by Fletcher.[4] Although a similar tumor with the same diagnosis but a benign feature was reported by Wiley et al[1] in the parotid gland, our tumor, like most fibrous histiocytomas arising from deep organs or glandular tissues, displayed aggressive behavior with intermediate malignancy.[2]

Fibrohistiocytic tumors are rare in the parotid gland, so we performed many histochemichal studies to exclude myoepithelial and epithelial tumors, such as the spindle cell variant of myoepithelioma.[5] The spindle cell myoepithelioma is keratin and S-100 positive,[6] whereas this tumor was composed of fibroblasts and histiocytes and

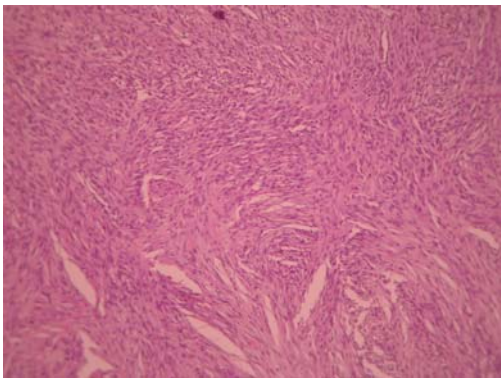

Figure 1. Spindle cell proliferation in the form of intersecting bundles in richly vascular and myxoid stroma.

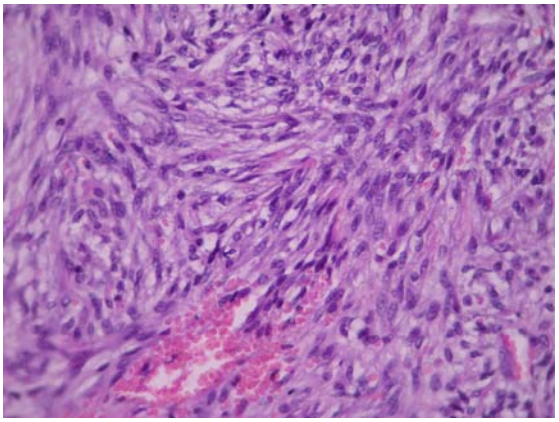

Figure 2. Extravasations of large number of RBCs, scattered small lymphoid cells, mitotic figures, and multinucleated giant cells in the background of focal stromal degenerative changes.

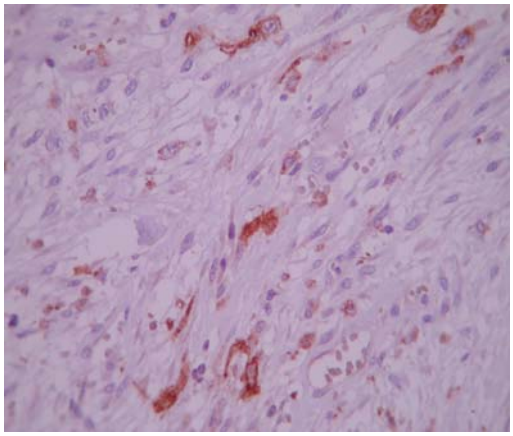

Figure 3. Patchy positive reaction for CD68.

Table 1. IHC markers in salivary gland tumors.

Tumor	Positive markers	Negative markers
Benign mixed tumor	Keratin (ck 19.ck 14) secretory component CEA. Lysozyme alpha 1-antitrypsin, GCDFP-15, alpha 1-antichymotrypsin, lactoferrin, IL-6 steroid C-21 hydroxylase, actin, myosin, fibronectin, S-100, collagen (I, II)	Amylase, PSA, PSAP (50% positive & 50% negative)
Malignant mixed tumor	B72.3 in addition to markers of benign mixed tumor	
Warthin's tumor	S-100, B & T cell, CEA, keratin (ck7, ck8, ck18, ck19), secretory components, mitochondrial associated markers, ribonuclease, lactoferrin, lysozyme, sebaceous cell, somatostatin, musin-secreting cell	
Basal cell adenoma	Keratin, alpha 1-antichymotrypsin, CEA, S-100, vimentin, actin, myoepithelial cell	
Myoepithelioma	Actin, keratin, S-100, vimentin (in some cases), myosin, glycogen	Many of them fail to react with smooth muscle actin
Mucoepidermoid carcinoma	Simple mucin-type carbohydrate Ag(T,Tn syalosyl-th), ck7, ck14, mitochondrial antibodies	
Acinic cell carcinoma	Glycogen, keratin, amylase, alpha1-antichymotrypsin, transferrin, lactoferrin, IgA, secretory component, argyrophilia, densecore granules, vasoactive intestinal peptide	Fat, mucin
Adenoid cystic carcinoma	Keratin, CEA, lysozyme, lactoferrin, alpha 1-antichymotrypsin, S-100, CD-117 (C-kit), actin, collagen IV, laminin, integrin ligands, heparin sulfate proteoglycan, entactin, alpha 1- antitrypsin, hormone receptors	

was keratin and S-100 negative. Also in this tumor factor VIII, CD31, and C-Kit were negative, which excluded extraintestinal stromal tumor, Kaposi sarcoma, and vascular tumors.[7] As a review, we mention IHC results in salivary gland tumors (Table 1). [2]

Eusebi et al[8] and Balogh and his colleagues[9] reported cases of giant cell tumors arising from parotid glands, which had some features in common with the tumor we reported. However, the tumor reported by Eusebi et al[8] had an appearance that was reminiscent of giant cell tumor of bone with osteoid formation, tumor thrombi in blood vessels, and a diffuse growth pattern. Balogh et al's[9] case was a carcinoma of the parotid gland with a component of osteoclast-like giant cells.

Auclair and colleagues[10] reported 67 malignant mesenchymal tumors of salivary glands of which the most common tumors were malignant schwannoma and fibrosarcoma and four were classified as malignant fibrous histiocytomas. Benjamin and co-workers[11] reported two malignant fibrous histiocytomas of salivary glands. One tumor was histologically very similar to the tumor we have reported and had entrapment of the facial nerve within the tumor mass. At follow-up 5 months later, there was no tumor recurrence or lymphadenopathy.

The case we have reported can draw the attention to the occurrence of a primary fibrous histiocytoma of intermediate malignancy of the parotid gland.

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