

Case Report

Plasmacytoid Myoepithelioma of the Palate with Rapid Growth: A Case Report

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ABSTRACT

Myoepitheliomas are benign tumors which account for less than 1% of all salivary gland tumors. In this article, we report the clinical, pathologic, and immunohistochemical features of plasmacytoid myoepithelioma of the hard palate in a 73-year-old man having a painless hard palate mass with progressive growth for one month who was admitted in Alzahra Hospital of Isfahan University of Medical Sciences. The patient underwent a complete surgical resection of the tumor. Light microscopy showed a nodule with sharp margins of large cells with plasmacytoid aspect presenting round eccentric nuclei and an abundant and homogenous eosinophilic cytoplasm arranged in sheets and groups separated by an abundant, loose, myxoid and hyalinized matrix. Immunohistochemistry was performed on formalin-fixed and paraffin-embedded tissue with a panel of immunohistochemical markers comprising cytokeratin (CK), vimentin, S100, actin and EMA. Tumor cells showed strong and diffuse positivity for S100 and also CK, vimentin, and actin, but confirming the myoepithelial origin of the tumor, EMA immunostaining was negative.

Key words: Hard palate, Mass, Plasmacytoid myoepithelioma, Rapid growth

Introduction

Myoepitheliomas are benign tumors which account for less than 1% of all salivary gland tumors (1). The affected patients ranged in age from 6 to 85 years with a mean age in the early to mid-40s and without gender predilection. The parotid gland is the most commonly affected site. Two thirds of the minor gland tumors occur in the palate. Similar to the most benign salivary gland tumors, myoepitheliomas most often present as slowly enlarging, asymptomatic masses (2). Few cases of plasmacytoid variant of this tumor of the hard palate have been reported.

In this article, we reported the clinical, pathologic,

and immunohistochemical features of plasmacytoid myoepithelioma of the hard palate in an adult patient that was presented as a rapidly enlarging mass.

Case report

A 73-year-old man having a painless hard palate mass with progressive growth for one month was admitted in Alzahra Hospital of Isfahan University of Medical Sciences. A good oral hygienic condition was found in oral examination and also it was found out that the patient used removable denture. In addition, a well-circumscribed, firm and sub-mucosal nodule

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measuring 3 cm 3 cm was observed in the right side of midline in hard palate which was not tender. The tumor presented a smooth and erythematous surface without ulceration. Computed tomography revealed an increase in soft tissue in the right side of oral roof, but no space occupying lesion or bone involvement was seen. The primary clinical hypothesis upon diagnosis was that this mass was a benign tumor of the salivary gland. Afterwards, an incisional biopsy was performed and the diagnosis of pleomorphic adenoma was suggested. The patient underwent a complete surgical resection of the tumor. Grossly, the mass was well circumscribed measuring 4 cm 2 cm 2 cm. Light microscopy showed a nodule with sharp margins of large cells (Fig.1) with plasmacytoid aspect presenting round, eccentric nuclei and an abundant and homogenous eosinophilic cytoplasm arranged in sheets and groups separated by an abundant, loose, myxoid and hyalinized matrix (Fig.2). Small foci of cells with mild atypia but without mitosis were seen. Ductal differentiation was noted in less than 2% of epithelial component.

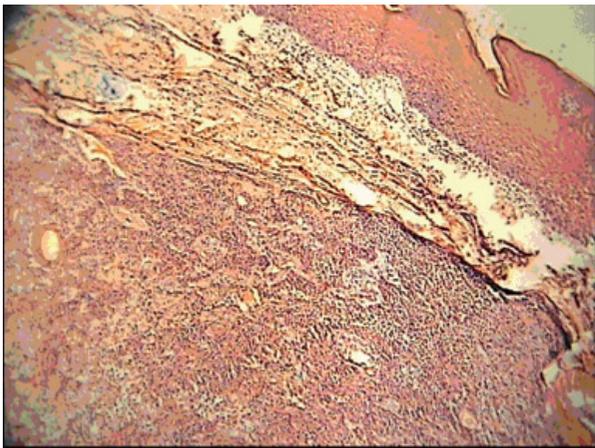


Figure 1. Sharp margin of the tumor in light microscopy (H&E x40)

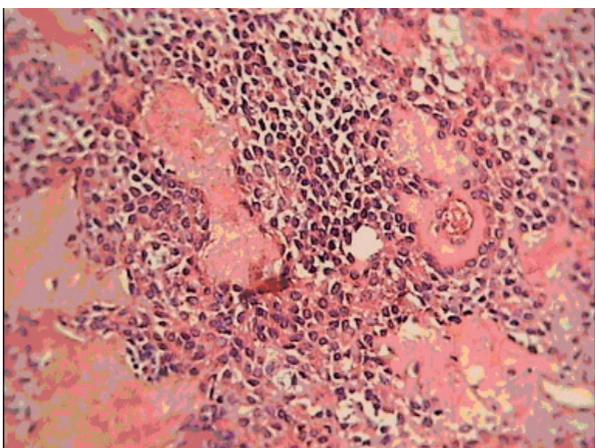


Figure 2. Myoepithelioma composed of large plasmacytoid cells (H&E x400)

Immunohistochemistry was performed on formalin-fixed and paraffin-embedded tissue with a panel of immunohistochemical markers comprising cytokeratin (CK), vimentin, S100, actin and EMA. Tumor cells showed strong and diffuse positivity for S100 and also CK, vimentin and actin, but confirming the myoepithelial origin of the tumor, EMA immunostaining was negative (Fig. 3-5).

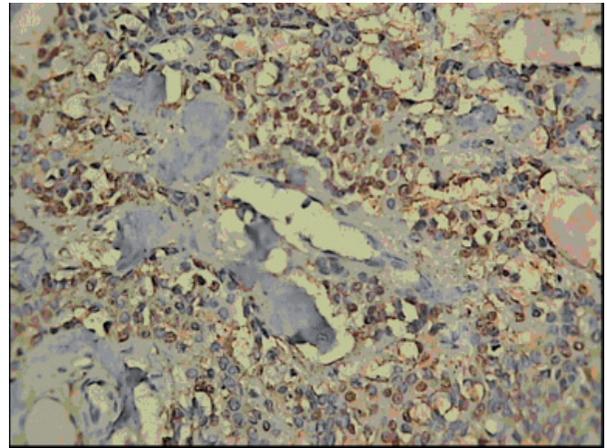


Figure 3. Positive immunostaining for S-100 (x400)

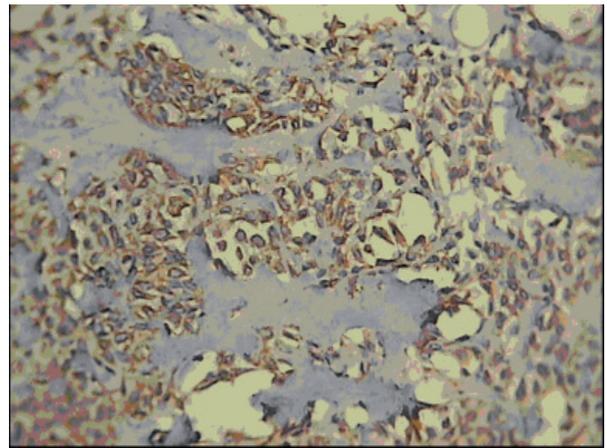


Figure 4. Positive immunostaining for vimentin (x400)

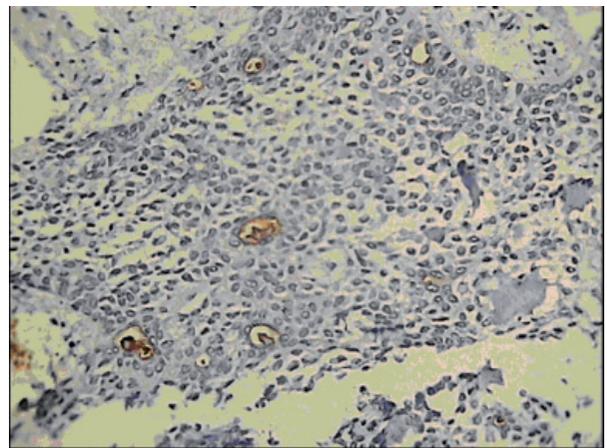


Figure 5. Negative immunostaining for EMA (x400)

Discussion

Myoepithelioma is a tumor composed entirely or predominantly of myoepithelial cells. Most authors affirm that these represent one extreme of the histological spectrum of pleomorphic adenoma with either no ductal differentiation (1) or up to 5% of the tumor composed of ductal epithelium (2). Myoepitheliomas represent about 1% of salivary gland neoplasms (3). Approximately 42% of myoepitheliomas occurred in the minor salivary glands or in seromucinous gland sites including nasal cavity and larynx (2). Most of the minor gland tumors occurred in the palate (2, 4). Epithelioma most often present as slowly enlarging, asymptomatic masses (2). However, in our case it had rapid and progressive growth during one month. Myoepithelioma is a well circumscribed or encapsulated tumor (4, 5). Microscopically, these tumors fall into five patterns: Spindle cell type, plasmacytoid cell type, reticular type, clear cell type and a combination of these types (2, 7). Spindle cell subtype is the most common pattern and has a predilection for the parotid glands of older individuals (1-2). The plasmacytoid type has a predilection for the palate of younger individuals and may be arranged in sheets, however, it is often less cellular and the cells may be arranged in groups separated by an abundant, loose myxoid matrix that is predominantly composed of hyaluronic acid (2). The plasmacytoid cells are round, oval or polyhedral with eosinophilic cytoplasm and eccentric nuclei (1-2). Positive staining for cytokeratin, muscle specific actin, smooth muscle actin, calponin or S100 protein confirms its myoepithelial nature. Vimentin is also frequently expressed, however, EMA immunostaining is negative in this type of tumor (2). A few cases of plasmacytoid variant have been reported. Danyle Elias et al have reported a plasmacytoid myoepithelioma of the palate in a child (8). Also, Kern et al have described a spindle cell myoepithelioma of the minor salivary gland (9). Sarafs et al have presented a plasmacytoid myoepithelioma of oral cavity as well (10). Furthermore, few cases of plasmacytoid myoepitheliomas of the palate are reported (11-12, 13).

Various benign mesenchymal neoplasms such as fibroma, fibrous histiocytoma, leiomyoma and schwannoma may be considered in the histologic differential diagnosis of the spindle cell type of myoepithelioma. Of these tumors, only myoepithelioma would be expected to express cytokeratin, muscle specific or smooth muscle actin, calponin and S100

protein. Also, plasmacytoma may be confused with plasmacytoid type of myoepithelioma, although the nuclear morphology of these two is different (2). In our patient, light microscopy showed a well-circumscribed nodule that was composed of large round or oval cells with eosinophilic cytoplasm and eccentric nuclei with ductal differentiation in less than 5% of epithelial component. In normal myoepithelial cells, muscle specific actin and cytokeratin (14) are the two most important immunopositive antigens (7). But EMA immunostaining is negative in this tumor. Nevertheless, the tumor cells of myoepithelioma, particularly of the plasmacytoid variant showed little or no normal myoepithelial features (15). Despite the fact that vimentin is not expressed in normal myoepithelial cells, it is an important marker for neoplastic myoepithelial cells (7, 14). S100 has also been identified as a marker of neoplastic myoepithelial cells (3, 7), but it is not expressed in normal myoepithelial cells. In this case, the tumor cells showed strong and diffuse positivity for S100, vimentin, cytokeratin and actin. Therefore, plasmacytoid myoepithelioma was confirmed for our patient. The treatment of these tumors consists of surgical excision with tumor-free margins (3) that was performed for the reported patient.

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