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Plasmacytoid myoepithelioma of palate: three rare cases and literature review

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Abstract

Salivary gland myoepitheliomas are rare tumours, accounting for less than 1 per cent of neoplasms of the salivary glands. Myoepithelioma of the palate is very rare, and only a few cases have been reported in the world literature. Histological and immunohistochemical analysis supports the myoepithelial origin of this tumour. Here, we report three cases of myoepithelioma of the palate in adult males and review previously reported cases.

Key words: Salivary Gland Neoplasms; Palate; Myoepithelioma; Pleomorphic Adenoma

Introduction

Myoepithelial cells are present in-between the basal lamina and the acinar and ductal cells in salivary glands and other exocrine glands. These myoepithelial cells have structural features of both epithelial and smooth muscle cells.[1] Myoepithelioma is an uncommon tumour composed entirely of myoepithelial cells. It is estimated to represent 1 per cent of all salivary gland tumours and mainly involves the parotid gland. Sheldon[2] was the first to use the term 'myoepithelioma' to describe these rare, benign tumours. Because myoepithelial cells are difficult to identify on routine microscopic preparations, analysis of the cytoplasmic filament expression and ultrastructural features of these cells is essential to fulfill the diagnostic criteria for myoepithelioma.[3,4]

Case reports

Case one

A 38-year-old man presented with a two-month history of a burning sensation in the throat radiating to the right ear, nasal intonation of voice, painless swelling on the right side of the oral cavity and occasional blood-staining of saliva.

On examination, there was a well circumscribed, firm mass, 3 cm in diameter, arising from the right side of the soft palate and pushing the uvula to the opposite side. The mass was ulcerated over its anterior aspect, probably due to previous biopsy. There was no regional lymphadenopathy in the neck, and the remaining physical examination was unremarkable.

A contrast-enhanced computed tomography (CT) scan showed an isodense, soft tissue mass arising from the right side of the soft palate and hanging into the oropharynx (Figure 1). There was no bony destruction or infiltration of surrounding tissue.

The tumour was excised *in toto* under general anaesthesia through a transoral approach.

The patient was asymptomatic four years after surgery.

Case two

A 35-year-old man presented with a progressive swelling over the right side of the palate, of one year's duration.

Examination revealed a well defined, smooth, firm, nontender, mucosa-covered mass, 5 cm in diameter, arising from the left side of the hard palate. There were no palpable neck nodes, and the general physical examination was normal (Figure 2).

Pre-operative fine needle aspiration cytology diagnosed the mass as a pleomorphic adenoma of the soft palate.

A contrast-enhanced CT scan showed an isodense, soft tissue mass arising from the right side of the soft palate and hanging into the oropharynx (Figure 3). There was no bony destruction or infiltration of surrounding tissue.

Total excision of the tumour was performed under general anaesthesia, preserving the overlying mucosa, through a transoral approach.

Histopathological examination of the excised specimen confirmed the diagnosis of plasmacytoid myoepithelioma.

A six-month follow up period showed no evidence of recurrence.

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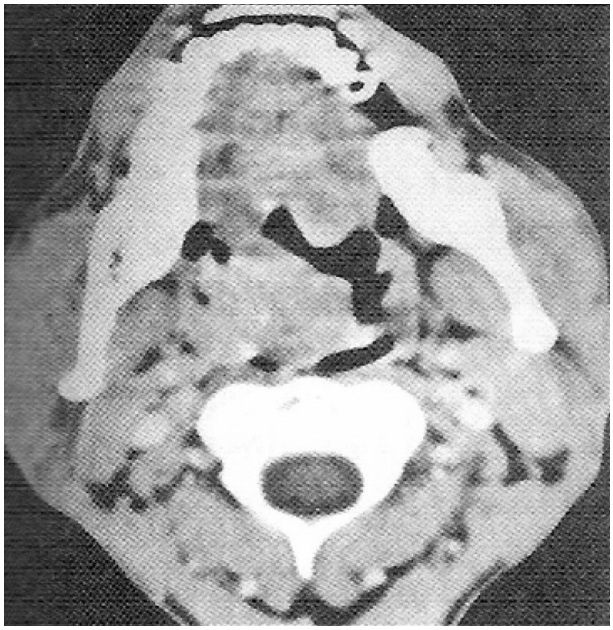

FIG. 1
Axial, contrast-enhanced computed tomography scan, showing an isodense lesion arising from the soft palate. No destruction of surrounding tissues is noted (case 1).

On histological examination in cases one and two, the tumour was observed to be surrounded by a thick, fibrous capsule. The tumour lobes were composed entirely of polygonal cells with eosinophilic cytoplasm and an eccentrically situated, round to oval nucleus. The cells were loosely arranged in a trabecular fashion, with an intervening, acellular myxoid stroma. A very small area of ductal or chondroid differentiation was seen (less than 5 per cent), so a final diagnosis of myoepithelioma, plasmacytoid type, was favoured over that of myoepithelial-rich pleomorphic adenoma (Figures 4 and 5). On histochemical staining, the plasmacytoid cells were diffusely immunoreactive for S-100 protein in both cases.

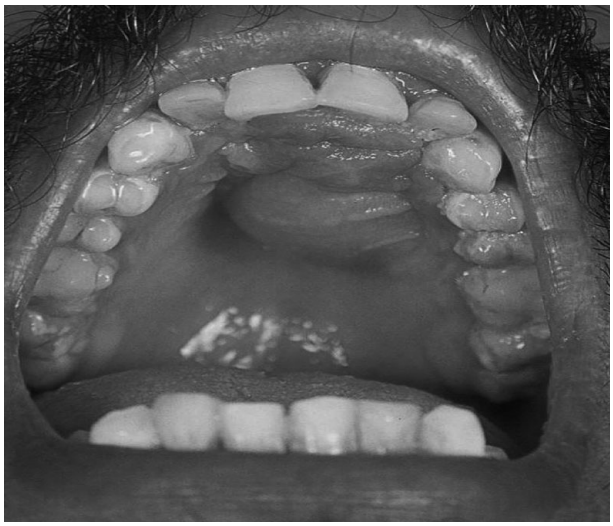

FIG. 2
Intra-oral view, showing a well circumscribed palatal mass (case 2).

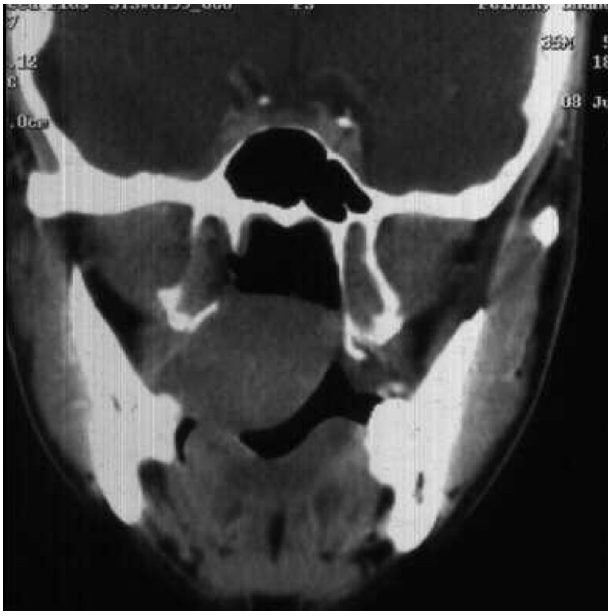

FIG. 3
Coronal, contrast-enhanced computed tomography scan, showing an isodense lesion arising from the hard palate and hanging into the oral cavity (case 2).

Case three

A 65-year-old man presented with a progressive swelling over the right side of the palate, of four months' duration.

Examination revealed a well defined, smooth, firm, nontender, mucosa-covered mass, 2 cm in diameter, originating from the posterior part of the right side of the hard palate. The general physical examination was normal.

Pre-operative fine needle aspiration cytology diagnosed the mass as a pleomorphic adenoma of the soft palate.

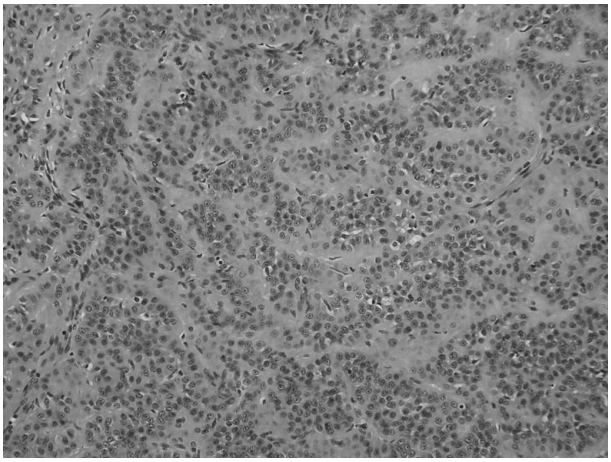

FIG. 4
Photomicrograph showing predominately myoepithelial cells (H&E; original magnification ×20).

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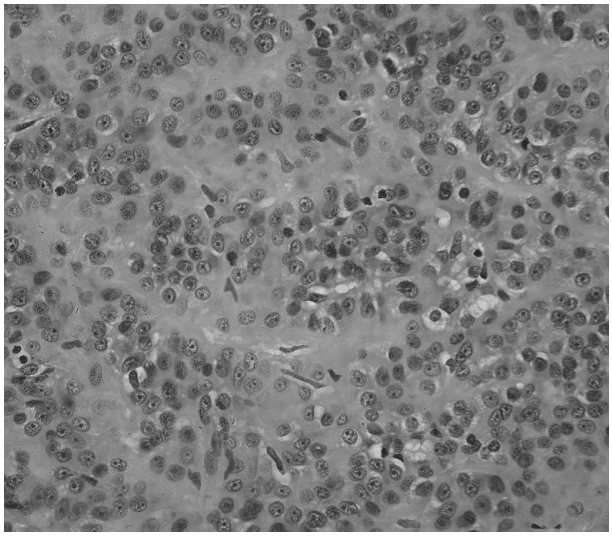

FIG. 5

Photomicrograph showing predominately plasmacytoid myoepithelial cells. (H&E; original magnification ×40).

An excisional biopsy was performed under local anaesthesia through a transoral approach.

Immunohistopathology of the mass was similar to that of the previous two cases, with diffuse immunoreactivity for S-100 protein, favouring a diagnosis of plasmacytoid myoepithelioma.

No evidence of recurrence was observed over a three-month follow up period.

Discussion

The myoepithelial cell, the putative cell of origin of myoepithelioma, is a normal component of salivary glands. Myoepithelial cells are situated along the outer aspect of the acini, the intercalated ducts, and the interlobar and interlobular striated ducts.[5] The myoepithelioma is a tumour mainly composed of myoepithelial cells. Myoepitheliomas are rare tumours, representing 1 per cent of all salivary gland neoplasms.[6] Although previously classified as part of the spectrum of pleomorphic adenomas, myoepithelioma has recently been tentatively proposed to represent a distinct variant of salivary gland adenoma.[7] Soft palatal myoepithelioma arises from the minor salivary glands. On rare occasions, myoepitheliomas arise in the intraoral minor salivary glands, and a number of single cases with sufficient documentation have been published.

In a review of the literature, 12 cases of ultrastructurally and immunohistochemically confirmed myoepithelioma of minor salivary gland origin were found.[8–19] These are summarised, together with the present three cases, in Table I.

The average age at presentation of intraoral myoepithelioma is 36 years. The most frequent site of origin is the palate (93 per cent). The tumour occurs equally in both men and women. Both the soft and hard palate are equally involved. The tumour generally shows a benign clinical course. The common presentation is a painless swelling or mass progressively increasing in size over several months to years. The most frequent type of myoepithelioma is the plasmacytoid type.

Our first case had nasal intonation of voice, probably due to mechanical interference with palatal movement, causing improper closure of the nasopharynx and escape of air through the nostrils during phonation.

The gross appearance of plasmacytoid myoepithelioma has been described as a well circumscribed and solid, encapsulated mass without ulceration; however, our first patient showed ulceration because of a previous biopsy.

Histologically, a thin capsule (which may be partial) surrounds the tumour. A range of microscopic architectural patterns has been reported, including myxoid (pleomorphic adenoma type), nonmyxoid (solid), reticular (canalicular) and mixed. In the myxoid type, a considerable amount of glycosamine is seen in the stroma. Our cases showed a nonmyxoid pattern. Two predominant types of cellular differentiation have been reported, spindle cell

TABLE I

REPORTED CASES OF MYOEPITHELIOMA OF MINOR SALIVARY GLANDS OF SOFT PALATE ORIGIN WITH ULTRASTRUCTURAL OR HISTOCHEMICAL CONFIRMATION

Author	Age (years)/sex	Site	Cell type
Kahn & Schoub[8]	17/F	Hard palate	Plasmacytoid
Luna *et al.*[9]	30/F	Hard palate	Spindle
Sciuba & Goldstein[10]	22/M	Palate	Plasmacytoid
Nesland & Sorbinho-Simoes[11]	18/F	Soft palate	Plasmacytoid
Enmoto *et al.*[12]	57/F	Soft palate	Plasmacytoid
Barnes *et al.*[13]	24/F	Hard palate	Plasmacytoid
Thompson *et al.*[14]	23/M	Oral floor	Plasmacytoid
Ellyn & Gnepp[15]	8/F	Soft palate	Plasmacytoid
Kawabe *et al.*[16]	53/M	Soft palate	Plasmacytoid
Lopez *et al.*[17]	46/M	Soft palate	Plasmacytoid
Kanazawa *et al.*[18]	42/F	Hard palate	Plasmacytoid
Katsuyama *et al.*[19]	67/F	Soft palate	Plasmacytoid
Present case 1	38/M	Soft palate	Plasmacytoid
Present case 2	35/M	Hard palate	Plasmacytoid
Present case 3	65/M	Hard palate	Plasmacytoid

F = female; M = male

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type and hyaline (plasmacytoid) type.[6,7] The spindle cells are arranged in sheets, fascicles or a swirling pattern and contain fibrillar, eosinophilic cytoplasm with elongated nuclei. The hyaline cells are arranged in solid clusters or sheets, as seen in our cases. These cells are round to polygonal and contain fibrillar, eosinophilic cytoplasm and eccentrically placed, round nuclei.

Immunohistochemical studies of myoepitheliomas have demonstrated cytoplasm immunoreactivity for a number of antigens, including cytokeratin, vimentin, myosin, neuron-specific enolase and glial fibrillary protein antigen.[3,20] Normal myoepithelial cells have similarly been shown to co-express immunopositivity for cytokeratin and muscle-specific actin.[3] This expression of markers of epithelial, neural and muscle antigens is of histogenetic interest and potential diagnostic importance. There is considerable variation in the tumour cell expression of muscle-specific actin – spindle cells react strongly with this protein, whereas plasmacytoid cells are generally non-reactive.[3,4,7] Tumours cells were diffusely immunoreactive for S-100, vimentin and cytokeratin in all cases of our series.

Plasmacytoid types of myoepithelioma tend to occur more frequently in the oral cavity, especially the palate, than do other cell types of myoepithelioma, whereas spindle cell types have been reported to occur in the parotid gland.[6,21] Myoepitheliomas have been frequently mistaken for cellular plemorphic adenomas, because both tumours contain abundant myoepithelial cells.[4,5] Several investigators have indicated that myoepithelioma is a variant of pleomorphic adenoma.[6,15,22] However, the latest World Health Organization classification of salivary gland tumours, by Seifert *et al.*,[7,23] clearly separated myoepithelioma from pleomorphic adenoma; it also indicated that myoepitheliomas show myoepithelial but not ductal differentiation and lack chondroid or myxochondroid foci. On histopathological examination, myoepitheliomas are differentiated from pleomorphic adenomas by a lack of ductal or glandular pattern of differentiation and an absence of chondroid stroma, with large numbers of round to oval tumour cells proliferating predominately in a trabecular or sheet-like pattern and containing abundant, eosinophilic cytoplasm with peripherally located, oval nucleus (thus mimicking plasma cells). The diagnosis can be confirmed by immunohistochemical examination.

The majority of reported myoepitheliomas have a benign prognosis similar to that of pleomorphic adenoma. A local recurrence rate of 18 per cent was found in two reported series.[6,15] Myoepithelial carcinoma was recognised in the recent World Health Organization classification. Although cytological atypia and mitotic activity with or without pathological evidence of invasion has occasionally been reported, malignancy (as determined by lethal, locally aggressive disease or distant metastasis) has rarely been reported.[24–26]

The histological parameters of myoepithelioma prognosis are not well recognised.

The treatment of myoepithelioma is primarily complete surgical excision with clear margins and long term follow up.

Conclusion

We present three cases of myoepithelioma arising from a minor salivary gland of the soft palate. Myoepitheliomas are rare, benign tumours with a good prognosis. Their treatment consists of complete surgical excision and regular follow up.

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