Myoepithelioma of the Soft Palate: a Case Report Giving Special Attention to the Differential Diagnosis

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ABSTRACT

Background: Myoepitheliomas are rare tumours that may generally arise from the minor or major salivary glands. The differential diagnosis of this tumour should be performed along with several benign and malignant soft tissue neoplasms. The present case report describes an asymptomatic mass that arose in the soft palate of 42 year old black woman with duration of the six months.
Methods: An incisional biopsy of soft palate lesion was carried out and submitted for histological evaluation under the clinical hypothesis of salivary gland tumour. To confirm the myoepithelial nature of neoplastic cells the immunohistochemical reactions for smooth-muscle actin, cytokeratins and S100 were performed.
Results: The histological examination revealed the presence of tumour originating from a minor salivary gland and covered by a stratified squamous oral epithelium. The tumour cells were arranged in order to form a myxoid pattern and, individually, small and/or medium spindle-shaped cells with predominantly round or ovoid nuclei, as well as epithelioid and plasmocytoid cells were noted. The stroma was myxomatous and no ductal or syringomatous epithelial structures were observed. Following the histological and immunohistochemical diagnosis of myoepithelioma, the lesion was surgically removed. After the surgery, a follow-up of one year showed no signs and symptoms of reccurrence.
Conclusions: The myoepithelioma should be carefully distinguished from the other soft tissue tumours, especially those arising from salivary glands, such as pleomorphic adenoma and adenoid-cystic carcinoma.

Keywords: myoepithelioma; soft tissue neoplasms; diagnosis, differential; palate; salivary glands, minor; salivary gland diseases.

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INTRODUCTION

Myoepithelioma, also known as myoepithelial adenoma and benign myoepithelial tumour is a neoplasm mainly constituted by ectodermally derived contractile cells that act as smooth muscle cells and are named myoepithelial cells [1,2]. These tumours can present several architectural patterns, which are non-myxoid (solid), myxoid (pleomorphic adenoma-like), reticular (canalicular-like) and mixed [3].

In that way, myoepitheliomas that arise in the oral cavity are very rare, accounting for 1.5% of all tumours in the major and minor salivary glands and representing 2.2% and 5.7%, respectively of all benign major and minor salivary gland tumours [4]. The lesions are generally asymptomatic and may increase slowly in size over a period of several months or years [5].

The myoepithelial cells may be encountered in several human organs, especially in the salivary glands. Thus, the greatest part of myoepitheliomas develop in the major salivary glands, while they are uncommon in the other parts of body, such as soft tissues and the carpal tunnel [3,6-8]. Finally, one of the most frequent salivary gland - myoepitheliomas, was found in the parotid gland (50%), in the sublingual gland (33%), and in the submandibular gland (13%) [9].

Myoepitheliomas and pleomorfic adenomas have very similar clinical outcomes, which makes this distinction not important for the patients. Nevertheless, pathologists must analyze the lesions carefully in order to differentiate them from other malignant neoplasms that have analogous histological features [3].

This case report sought to demonstrate a myoepithelioma arising from the soft palate, as well as its respective histological analysis and clinical management giving special attention to the possible differential diagnosis of this tumour.

CASE DESCRIPTION AND RESULTS

A 42 year old black woman, smoker and nonalcoholic, presented with a 6 months history of asymptomatic mass in the oral cavity. On the clinical examination, a firm and rubbery nodular ovoid in shape with a well delimited border lesion measuring 1.5 cm in diameter was palpated at the left side of soft palate. The tumour was not painful to palpation. An elevated maxillary torus could also be noticed in the anterior part of the hard palate (Figure 1A).

An incisional biopsy of soft palate lesion was carried out with a surgical punch (8 mm in diameter and 7 mm in depth) and was submitted for histological evaluation under the clinical hypotheses of salivary gland tumour. The publication of this case report was approved by the Ethical Committee of the School of Dentistry of the University of São Paulo, Brazil.

The tissue was fixed in 10% formaldehyde for 24 hours. Routine laboratory procedures were followed by paraffin embedding of the material. After this, 5 µm thick sections were obtained and stained with hematoxylin and eosin for histological analysis. The histological examination revealed the presence of tumour originating from a minor salivary gland and covered by a stratified squamous oral epithelium (Figure 1B). The tumour cells were arranged in order to form a myxoid pattern (Figure 1C) and, individually, small and/or medium spindle-shaped cells with predominantly round or ovoid nuclei, as well as epithelioid and palsmocytoid cells were noted (Figure 1D). The stroma was myxomatous and no ductal or syringomatous epithelial structures were observed (Figures 1B and 1C).

To confirm the myoepithelial nature of neoplastic cells the immunohistochemical reactions for smooth-muscle actin, cytokeratins and S100 were performed. Immunohistochemical reactions revealed the positivity against all used antibodies. Following the histological diagnosis of myoepithelioma, the lesion was surgically removed along with accessory minor salivary glands of the palate and the histological diagnosis was reconfirmed. The follow-up of one year showed no signs and symptoms of reccurrence.

DISCUSSION

Distinct cell morphologies have been recognized in the myoepitheliomas. The tumour may be composed by spindle cells, which are arranged in interlacing fascicles that show stroma-like appearance [4,8,10], by plasmacytoid cells that are polygonal cells with eccentric nuclei, or even by epithelioid cells arranged in the nests or cords. Some myoepitheliomas can also show clear polygonal cells with abundant and optically clear cytoplasm [4].

Plasmacytoid cells are distinguishable by their dense, nongranular or hyaline, abundant eosinophilic cytoplasm, whereas epithelioid cells are round to polygonal cells, with centrally located nuclei and a variable amount of eosinophilic cytoplasm. Finally, clear cells contain large amounts of the glycogen but are devoid of mucin or fat [4]. These different types of cells may be arranged in several architectural patterns, which are non-myxoid (solid), myxoid (pleomorphic adenoma-like), reticular (canalicular-like) and mixed [3].

Several lesions participate in the differential diagnosis of

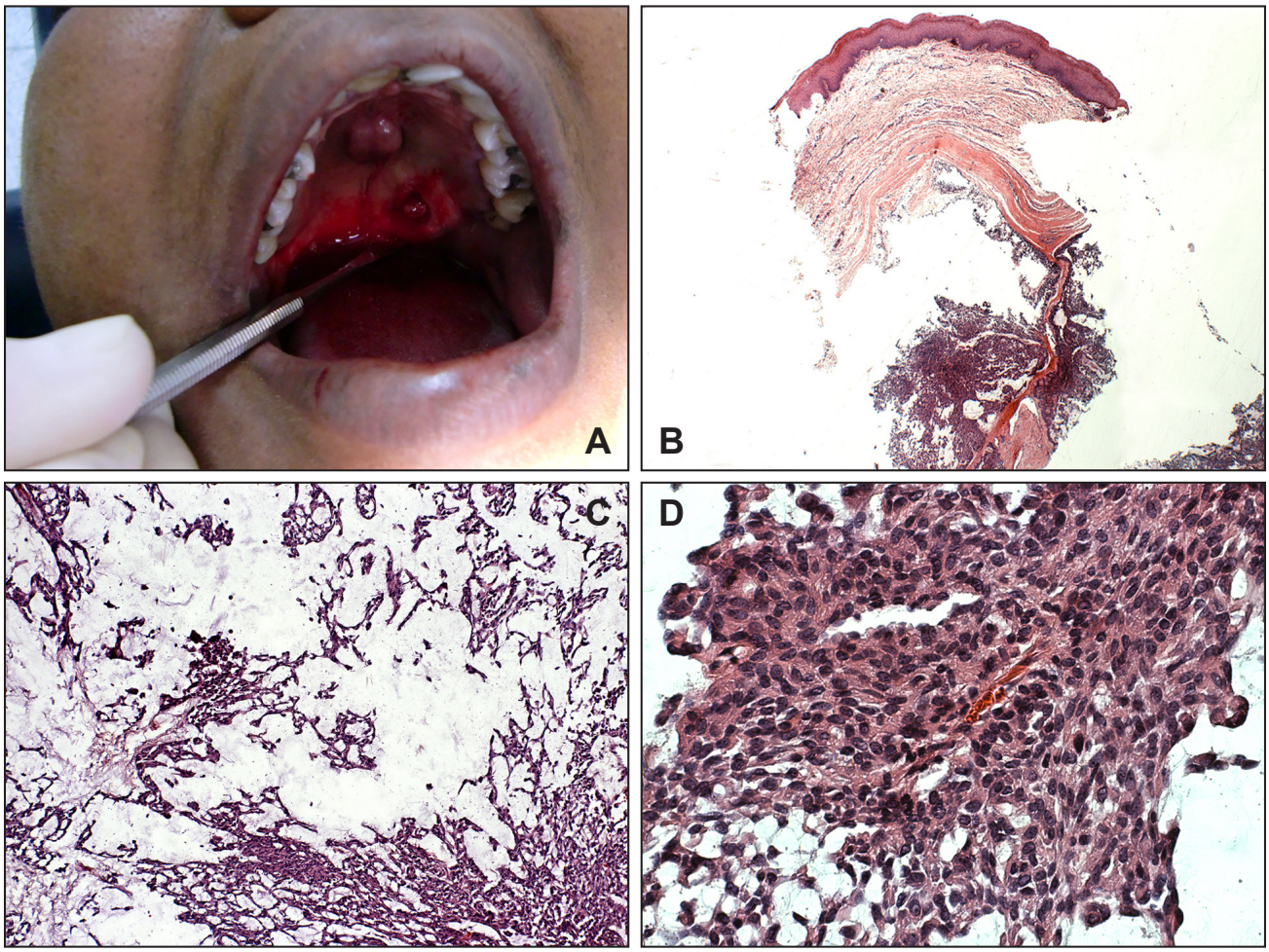

Figure 1. A = Photograph showing clinical appearance of the myoepithelioma located on the left side of the soft palate. An elevated maxillary torus can also be seen in the anterior part of the hard palate.
B = Low power photomicrograph showing histological picture of the myoepithelioma. The lesion can be seen in the submucosa and recovered by a stratified oral squamous epithelium (hematoxylin and eosin stain, original magnification x25).
C = Photomicrograph showing the myxoid pattern of the tumour (hematoxylin and eosin stain, original magnification x100).
D = High power photomicrograph showing the types of tumour cells encountered: spindle, plasmacytoid and epithelioid (hematoxylin and eosin stain, original magnification x400).

myoepitheliomas. Among them, benign and malignant tumours such as pleomorphic adenoma, adenocarcinoma, nerve sheath tumour, fibrous hystiocytoma, nodular fasciitis, synovial sarcoma, leiomyoma, leiomyosarcoma, hemangiopericytoma, solitary fibrous tumour and paraganglioma may be cited. In that way, it is very important to differentiate myoepitheliomas from malignant neoplasms [11,12].

Clinical appearance of the lesions mentioned above can share similarity with myoepitheliomas and the only safe way to differentiate them is by the means of histological evaluation. Myoepitheliomas present rare mitoses and absence of nuclear and cellular pleomorphism as well as a non-infiltrative growth typical of a benign tumour [11]. The tumour cells are also generally positive for smooth muscle actin (SMA) [13].

In the case presented herein, the myoepithelial cells were diffusively stained by the SMA antibody. In addition, tumour showed cytokeratins and S100 protein positivity, which may help the distinction between this lesion and nodular fasciitis, leiomyoma and leiomyosarcoma, once these tumours normally do not express these proteins [11]. Alternatively, plasmacytoma shows positivity for cytoplasmic immunoglobulins, whereas myoepitheliomas do not [5,14].

Besides, the competent observation of the tumour morphology is very important to perform the diagnosis. As mentioned previously, the predominant type of myoepithelial cell in the myoepithelioma may be spindled, epithelioid, clear or plasmacytoid, and one single tumour may also present two or more histological patterns [8,12]. Accordingly, peripheral nerve sheath tumours should participate in the differential diagnosis of spindle cell variant of myoepithelioma [15], whilst the clear cell myoepithelioma should be correctly discerned from the clear cell adenocarcinoma and mucoepidermoid carcinomas [11].

Myoepitheliomas may be considered as a variant of

pleomorphic adenoma in which glanduloductal differentiation is entirely or virtually absent. Thus, the architectural patterns and the cellular differentiation in myoepitheliomas are equally apparent to that of the nonluminal portions of pleomorphic adenomas [3]. In addition to these features, Sciubba and Brannon reported the absence of chondromyxoid or chondroid foci as characteristics of myoepitheliomas [12].

After the tissue samples were analyzed, no ducts were found. In fact, no more than 5 to 10 percent of myoepitheliomas may be composed of ducts [8]. Other authors suggest that no more than one duct every medium to high power field (x200 - 400) or no more than one small cluster of ducts is acceptable in these lesions [16]. Moreover, the myoepithelial cells that compose myoepitheliomas are also found in many organs (mainly the salivary glands), and thus are the major components of salivary gland tumours [17,18].

The tumours that arise from the salivary glands comprehend the already mentioned pleomorphic adenoma, adenoid-cystic carcinoma and epithelial-myoepithelial carcinoma of intercalated duct origin [17,18]. Although these lesions hold the myoepithelial cells as their main neoplastic cells, their clinical management may vary consistently. As a general rule, adenoid-cystic carcinoma, epithelial-myoepithelial carcinoma and mucoepidermoid carcinoma demand surgical excision with postoperative intensity-modulated radiotherapy with or without chemotherapy [19].

On the other hand, the accepted treatment of myoepitheliomas as well as for other benign tumours is the simple surgical excision. These tumours do not present high levels of the recurrence and this clinical case did not show any sign of recurrence after a one year follow-up.

CONCLUSIONS

The myoepithelioma should be carefully distinguished from the other soft tissue tumours, especially those arising from salivary glands, such as pleomorphic adenoma and adenoid-cystic carcinoma.

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