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

Felipe Fornias Sperandio<sup>1</sup>, Fernanda Salgueiredo Giudice<sup>1</sup>, Décio dos Santos Pinto-Junior<sup>1</sup>, Suzana Cantanhede Orsini Machado de Sousa<sup>1</sup>

<sup>1</sup>Department of Oral Pathology, School of Dentistry, University of São Paulo, São Paulo, Brazil.

#### **Corresponding Author:**

Felipe Fornias Sperandio Department of Oral Pathology, School of Dentistry, University of São Paulo Avenida Professor Lineu Prestes, 2227 - CEP: 05508-900, São Paulo Brazil

Phone: +55 21 11 30917912 Fax: +55 21 11 30917894 E-mail: sperandio@usp.br

#### **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.

Accepted for publication: 27 December 2010

To cite this article:

Sperandio FF, Giudice FS, Pinto-Junior DD, de Sousa SC. Myoepithelioma of the Soft Palate: a Case Report Giving Special Attention to the Differential Diagnosis.

J Oral Maxillofac Res 2011 (Jan-Mar);2(1):e4

URL: http://www.ejomr.org/JOMR/archives/2011/1/e4/v2n1e4ht.pdf

doi: 10.5037/jomr.2011.2104

### **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  $[\underline{1},\underline{2}]$ . These tumours can present several architectural patterns, which are non-myxoid (solid), myxoid (pleomorphic adenoma-like), reticular (canalicular-like) and mixed  $[\underline{3}]$ .

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).

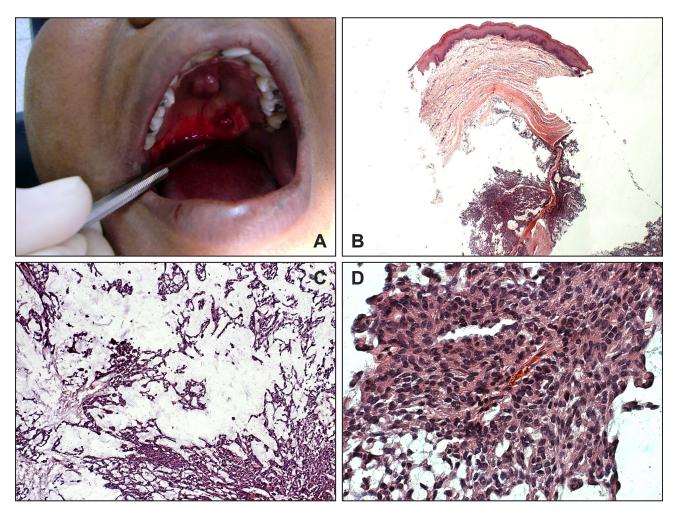
To confirm the myoepithelial nature of neoplastic cells the immunohistochemical reactions for smoothmuscle actin, cytokeratins and S100 were performed. Immunohistochemical reactions revealed the positivity against all used antibodies. Following the histological diagnosis of myoepithelioma, the lesion 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

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 epithelialmyoepithelial 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,

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.

# ACKNOWLEDGMENT AND DISCLOSURE STATEMENTS

The authors wish to thank Dr. Leoneti S. S. Asahi for providing the clinical picture presented in this study. The authors report no conflict of interests.

#### REFERENCES

adenoid-cystic carcinoma,

1. Hamperl H. The myothelia (myoepithelial cells). Normal state; regressive changes; hyperplasia; tumors. Curr Top Pathol. 1970;53:161-220. Review. [Medline: 4323195]

epithelial-myoepithelial

- Hübner G, Klein HJ, Kleinsasser Schiefer HG. Role of myoepithelial in the development of salivary gland tumors. Cancer. 1971 May;27(5):1255-61. 4325746] [Medline: [doi: 10.1002/1097-0142(197105)27:5<1255::AID-CNCR2820270533>3.0.CO;2-1]
- 3. Dardick I. Myoepithelioma: definitions and diagnostic criteria. Ultrastruct Pathol. 1995 Sep-Oct;19(5):335-45. Review. [Medline: 7483010] [doi: 10.3109/01913129509021906]
- 4. Cardesa A, Alos L. Myoepithelioma. In: Barnes L, Eveson JW, Reichart P, Sidransky D, editors. World Health Organizartion Classification of Tumors. Pathology and Genetics of Head and Neck Tumors. Lyon: IARC Press; 2005. p. 259-60.
- 5. Barnes L, Appel BN, Perez H, El-Attar AM. Myoepithelioma of the head and neck: case report and review. J Surg Oncol. 1985 Jan;28(1):21-8. [Medline: 2982059] [doi: 10.1002/jso.2930280107]
- 6. Kilpatrick SE, Hitchcock MG, Kraus MD, Calonje E, Fletcher CD. Mixed tumors and myoepitheliomas of soft tissue: a clinicopathologic study of 19 cases with a unifying concept. Am J Surg Pathol. 1997 Jan;21(1):13-22. [Medline: 8990137] [doi: 10.1097/00000478-199701000-00002]
- 7. Hornick JL, Fletcher CD. Myoepithelial tumors of soft tissue: a clinicopathologic and immunohistochemical study of 101 cases with evaluation of prognostic parameters. Am J Surg Pathol. 2003 Sep;27(9):1183-96. [Medline: 12960802] [doi: 10.1097/00000478-200309000-00001]
- 8. Dardick I, Thomas MJ, van Nostrand AW. Myoepithelioma--new concepts of histology and classification: a light and electron microscopic study. Ultrastruct Pathol. 1989 Mar-Jun;13(2-3):187-224. Review. [Medline: <u>2544051</u>] [doi: <u>10.3109/01913128909057442</u>]
- 9. Lee MW, Nam SY, Choi HJ, Choi JH, Moon KC, Koh JK. Myoepithelioma of parotid gland presenting as infra-auricular subcutaneous mass. J Cutan Pathol. 2005 Mar;32(3):240-4. [Medline: 15701087] [doi: 10.1111/j.0303-6987.2005.00293.x]
- 10. Luna MA, Mackay B, Gamez-Araujo J. Myoepithelioma of the palate. Report of a case with histochemical and electron microscopic observations. Cancer. 1973 Dec;32(6):1429-35. [Medline: 4357077] [doi: 10.1002/1097-0142(197312)32:6<1429::AID-CNCR2820320621>3.0.CO;2-7]

- 11. Ellis GL, Auclair PL. Benign Epithelial Neoplasms. In: Ellis GL, Auclair PL, editors. Tumors of the salivary glands. Maryland: ARP Press; 2008. p. 123-33.
- 12. Sciubba JJ, Brannon RB. Myoepithelioma of salivary glands: report of 23 cases. Cancer. 1982 Feb 1;49(3):562-72. [Medline: 6277451] [doi: 10.1002/1097-0142(19820201)49:3<562::AID-CNCR2820490328>3.0.CO;2-6]
- 13. McLaren BK, Smith J, Schuyler PA, Dupont WD, Page DL. Adenomyoepithelioma: clinical, histologic, and immunohistologic evaluation of a series of related lesions. Am J Surg Pathol. 2005 Oct;29(10):1294-9. [Medline: 16160470] [doi: 10.1097/01.pas.0000164615.38200.86]
- 14. Katsuyama E, Kaneoka A, Higuchi K, Takasu K. Myoepithelioma of the soft palate. Acta Cytol. 1997 Nov-Dec;41(6):1856-8. [Medline: 9390158]
- 15. Gibbons PJ, Powers CN, Reiter ER. Pathology quiz case: plasmacytoid variety of myoepithelioma of minor salivary gland. Arch Otolaryngol Head Neck Surg. 2001 Aug;127(8):1003-4. [Medline: 11493214]
- 16. Lomax-Smith JD, Azzopardi JG. The hyaline cell: a distinctive feature of "mixed" salivary tumours. Histopathology. 1978 Mar;2(2):77-92. [Medline: 78883] [doi: 10.1111/j.1365-2559.1978.tb01697.x]
- 17. Corio RL, Sciubba JJ, Brannon RB, Batsakis JG. Epithelial-myoepithelial carcinoma of intercalated duct origin. A clinicopathologic and ultrastructural assessment of sixteen cases. Oral Surg Oral Med Oral Pathol. 1982 Mar;53(3):280-7. [Medline: 6950345] [doi: 10.1016/0030-4220(82)90304-8]
- 18. Thompson SH, Bender S, Richards A. Plasmacytoid myoepithelioma of a minor salivary gland. J Oral Maxillofac Surg. 1985 Apr;43(4):285-8. [Medline: 2984389] [doi: 10.1016/0278-2391(85)90289-7]
- 19. Schoenfeld JD, Sher DJ, Norris CM Jr, Haddad RI, Posner MR, Balboni TA, Tishler RB. Salivary Gland Tumors Treated With Adjuvant Intensity-Modulated Radiotherapy With or Without Concurrent Chemotherapy. Int J Radiat Oncol Biol Phys. 2010 Nov 12. [Epub ahead of print] [Medline: 21075557]

#### To cite this article:

Sperandio FF, Giudice FS, Pinto-Junior DD, de Sousa SC. Myoepithelioma of the Soft Palate: a Case Report Giving Special Attention to the Differential Diagnosis.

J Oral Maxillofac Res 2011;2(1):e4

URL: http://www.ejomr.org/JOMR/archives/2011/1/e4/v2n1e4ht.pdf

doi: 10.5037/jomr.2011.2104

**Copyright** © Sperandio FF, Giudice FS, Pinto-Junior DD, de Sousa SC. Accepted for publication in the JOURNAL OF ORAL & MAXILLOFACIAL RESEARCH (<a href="http://www.ejomr.org">http://www.ejomr.org</a>), 27 December 2010.

This is an open-access article, first published in the JOURNAL OF ORAL & MAXILLOFACIAL RESEARCH, distributed under the terms of the <u>Creative Commons Attribution-Noncommercial-No Derivative Works 3.0 Unported License</u>, which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work and is properly cited. The copyright, license information and link to the original publication on (<a href="http://www.ejomr.org">http://www.ejomr.org</a>) must be included.