

Benign Myoepithelioma of the Hard palate: A Diagnostic Challenge. Case and Review of the literature

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ABSTRACT

Myoepithelioma is a rare benign tumor of the salivary glands, considered a variety of pleomorphic adenoma, the palatal location of which is most common in the oral cavity. Our work reports a case of a myoepithelioma of the accessory palatine salivary glands in a 63-year-old man, clinically he presented a lobulated, firm palatal swelling covered with a healthy mucosa. The diagnosis was based on clinical and pathological arguments. The interest of imaging lies in the appreciation of the extension of the tumor process to the mucosa and to deep tissues. Tumor excision must be large to avoid the risk of recurrence and malignant transformation. Through this observation and in the light of the literature we will focus on the anatomo-clinical aspects.

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Introduction

Myoepithelioma is a rare benign tumor that accounts for less than 1% of all salivary gland tumors. It was first described by Sheldon in 1943, is considered a variety of pleomorphic adenoma as well as a separate clinical entity according to the WHO since 1991. Currently, most authors have thought that it is of a relatively aggressive tumor. This tumor exhibits a different biological behavior from that of mixed tumors. (1,2)

Palatal localization is most common in the oral cavity. They are more frequently found in young adults between the ages of 30 and 50. The clinical appearance of the tumor suggests the diagnosis of myoepithelioma; in fact, most cases present in the form of a firm, painless mass of small size with slow growth. Confirmation is done by a biopsy with microscopic and immunohistochemical studies. Pleomorphic adenoma and adenoid cystic carcinoma are the main differential diagnoses of this tumor. (2,3)

The aim of our work is to focus on the anatomo-clinical and histological aspects and the diagnostic difficulties of benign myoepithelioma through an observation and in the light of the literature.

Observation

It was a 63-year-old patient, diabetic, smoking at a rate of 15 packs / year unweaned, amputated of the right lower limb for PAD, presenting for 2 years a palatal swelling gradually increasing in volume without associated signs. The clinical examination objectified, a palatal mass lateralized on the left, bumpy with telangiectasias opposite without mucous ulceration, of firm consistency measuring about 3cm from long axis, painless with a broad base of implantation extending forward and to the left to the upper left hemi arch, extending beyond the midline and not reaching the soft palate posteriorly. (Fig.1) The remainder of the endo-oral examination finds an edentulous patient without any other lesion. The rest of the clinical exam is normal, to note no palpable cervical lymphadenopathy. Faced with this clinical

picture, we suspected a very probable benign tumor of the accessory palatal salivary glands.



Figure 1. Pre-operative view: palatal mass lateralized on the left, well-circumscribed, with a sessile base. The overlying mucosa with telangiectasias, without mucous ulceration.

A cervico-facial computed tomography with injection of contrast product revealed a tumor of the hard palate, of tissue density slightly enhanced after injection of the contrast product, well limited, of regular contours, with lysis of the palatal and alveolar process of the bone maxilla on the left associated with a cortical invasion. (fig.2)

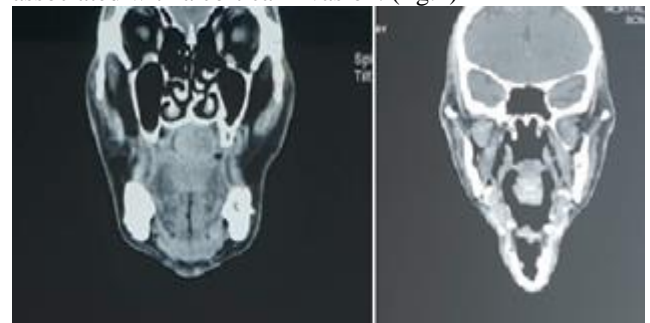


Figure 2. Computed tomography scan on coronal plane shows a mass of the hard palate, well limited, with regular contours and heterogeneous contrast enhancement.

A biopsy was carried out, the anatomopathological study of which objectified a tumor proliferation made up of sheets lined with myoepithelial cells of plasmacytoid phenotype. These structures are bathed in a fibro hyaline background. Myoepithelioma was diagnosed.

The patient was operated on, under general anesthesia by endo-oral route, consisting of a complete resection of the mass. Histological and immunohistochemical analysis of the operative specimen confirmed the diagnosis of benign myoepithelioma. The patient did not present any postoperative complications. At a follow-up of 12 months, no sign of recurrence was noted.

Discussion

Myoepitheliomas are rare, benign tumors which develop at the expense of the minor or major salivary glands, they represent less than 1% to 1.5% of all these tumors. (1,3) About 40% of myoepitheliomas develop in the parotid gland, while accessory salivary glands are affected in 21%. This tumor affects men and women equally. The average age is variable and varies between 39 and 53 years. (4,5)

Its clinical presentation is similar to that of any benign palatal tumor. It should be noted the interest of trying to differentiate benign myoepithelioma from its malignant counterpart, which is more aggressive and invasive with a tendency to very important recurrence. Clinically, it grows rapidly, and manifests as an ulcerated mass that often invades adjacent tissues causing bone erosion with distant metastases; however, benign myoepithelioma presents as a slow growing, asymptomatic mass, well circumscribed, lobulated, rarely with ulceration except in cases of repeated trauma or prosthetic interference without signs of nerve damage or lymphadenopathy. (6,7)

Computed tomography can accurately determine the extent of the tumor process to the mucosa and deep tissues, knowing that the bone involvement is exceptional (6)

Faced with the clinical and tomodensitometric aspect of this tumor, the question that arises and which is far from easy: is it a benign tumor, distinct from myoepithelial carcinoma or any other variety of salivary cancer with cells myoepithelial? the diagnosis is made on the exeresis piece, it is defined morphologically by a proliferation formed by groups of myoepithelial cells closely arranged in cords with an eccentric nucleus and an eosinophilic cytoplasm. Myoepithelial cells are surrounded by an edematous or fibrohyaline stroma. (5,8)

A precise diagnosis on a biopsy material is often difficult in view of this aspect which can also be observed in other salivary tumors with a prognosis different from that of myoepithelioma. In our opinion, the extemporaneous examination has a crucial interest in salivary pathology. (6)

Immunohistochemical techniques confirm the diagnosis, myoepithelial cells express the S100 protein and the glial fibrillar acidic protein (GFAP) (2,6). Neoplastic myoepithelial cells consistently demonstrate immunoreactivity to cytokeratin AE1 and AE3 and to smooth muscle actin. (2,5)

The closest differential diagnosis of this tumor is pleomorphic adenoma, but it also involves many salivary

lesions containing myoepithelial cells. The malignant form can occur de novo or appear secondarily in a benign myoepithelioma or a pleomorphic adenoma. (6) The treatment is surgical consists of a large excision of the tumor avoiding the risk of recurrence and malignant transformation. Although this tumor does not present a high risk of recurrence, so regular monitoring is particularly indicated. (5,9)

Conclusion

Myoepithelioma is a rare benign tumor of the salivary glands today considered to be a specific entity. This tumor should be carefully distinguished from other salivary gland tumors including pleomorphic adenoma and adenoid cystic carcinoma. The diagnosis is based on a set of clinical arguments; radiological and especially histological and immunohistochemical.

Competing interests

The authors declare no competing interests.

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