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Case Report

Myoepithelioma of parotid gland: a case report

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ABSTRACT

Myoepithelioma (ME) is a benign tumor, arising predominantly from major and minor salivary glands (SG). Clinically, it presents as a painless mass with a slow growth. In our case report, this lesion occurred in a 51-year-old man, and presented as a well-defined oval mass on the right parotid region. The magnetic resonance imaging study confirmed the presence of a lesion with benign features that was excised surgically by partial superficial parotidectomy. The histopathological and immunohistochemical analysis confirmed the diagnosis of parotid ME. Other SG tumors such as pleomorphic adenoma are differential diagnoses of ME. Complete surgical excision reduces the rate of recurrence.

Keywords: Myoepithelioma, Benign tumor, Salivary glands

INTRODUCTION

Myoepitheliomas (ME) are rare benign tumors, that consist almost entirely of myoepithelial cells. They are present in several organs such as the SG, breast, larynx and cutaneous sweat glands, accounting for 1-1.5% of all glandular tumors.[1,2]

ME arises predominantly from major and minor SG, affecting parotid gland in 50% of cases, sublingual gland in 33% of the cases and submandibular gland in 13%.[3-5]

The term ME emerged in 1943 to describe a group of tumors histologically composed of solitary myoepithelial cells, initially described as a variant of pleomorphic adenoma. In 1991, the world health organization classified the ME as a distinct entity. ME were defined as neoplasms with poor ductal structure and without chondroid or myxochondroid stroma and, according to the pattern of cellular organization, they can be solid, myxoid, reticular or mixed. In cases of malignant ME, myoepithelial carcinoma, the cells present with cellular atypia, cellular pleomorphism, cell necrosis, increased mitotic figures, invasive growth pattern or a combination of these and are clinically characterized by a more aggressive behavior and a higher recurrence rate.[1,4,6]

Although ME can appear at any age, with cases described between 8 and 35 years old, the usual age of presentation is between the 3rd and 5th decades of life.[1,4] It typically presents as painless mass, with slowly progressive growth and which, depending on its location, can cause different types of symptoms usually related to obstruction caused, mostly at advanced stage. Prognosis is usually favorable and recurrence is rare after complete excision.[4,6]

Although the rate of malignancy is very low, metastases can arise from apparently benign lesions and recurrence should be considered as potentially unrecognized malignancy.[1,4]

A rare case of ME of the parotid gland is presented below, with a discussion of its imaging and histological characteristics and treatment implemented.

CASE REPORT

A 51-year-old male patient, with no history of smoking or alcohol and no relevant personal or surgical history, was

referred to the otorhinolaryngology (ENT) consultation of our hospital with a history of an unilateral parotid swelling over a period of 6 months. The patient denied complaints of nasal obstruction, anterior or posterior rhinorrhea, odynophagia, dysphagia, dyspnea, dysphonia, change in body weight, anorexia or fever. On clinical examination, he had a painless, 2.2×2×1.7 cm well-defined oval mass, located in the right parotid gland. The mass was firm, non-tender and adherent to the deep planes, with no signs of overlying ulceration. The patient had no facial paralysis or palpable adenopathies.

A flexible nasofibroscopy did not show the presence of any other alteration. The patient underwent a magnetic resonance imaging (MRI), which showed a large exophytic and lobulated mass (Figure 1 and 2). Figure 3 shows the study with gadolinium uptake, with greater peripheral enhancement and a well-circumscribed mass (2.2×2.0×1.7 cm) in the right parotid gland without enlarged cervical lymph nodes.

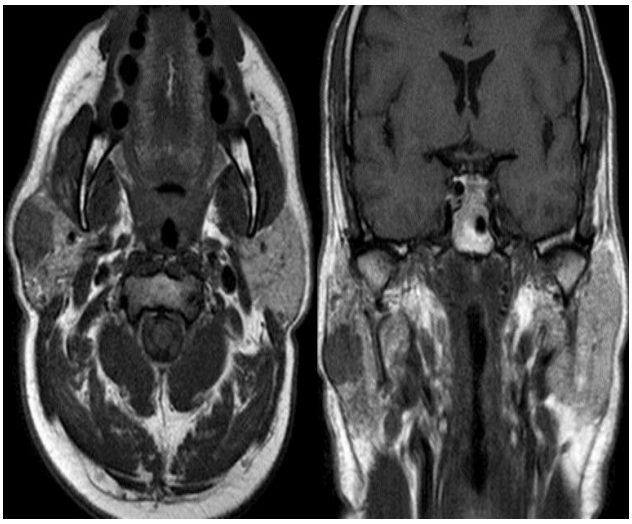

Figure 1: Parotid MRI, T1, axial and coronal sections.

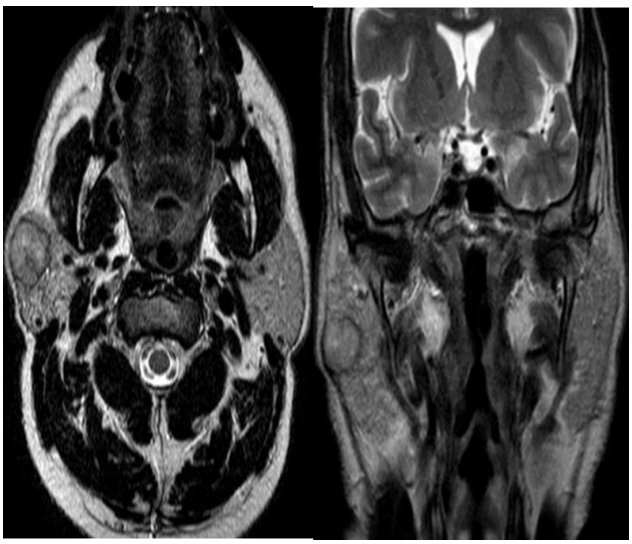

Figure 2: Parotid MRI, T2, axial and coronal sections.

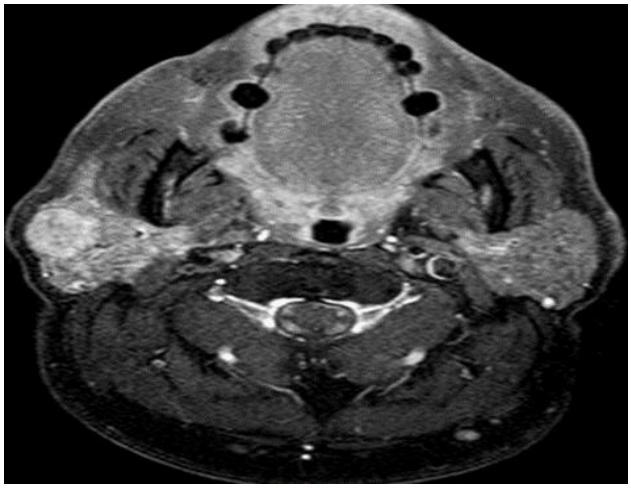

Figure 3: Parotid MRI, axial section after gadolinium administration.

Based on the clinical and imaging features, the main diagnostic hypothesis was a benign parotid tumor.

A partial superficial parotidectomy for complete excision of the lesion and parotid tissue was performed. The resected mass was a brownish nodular formation, with 5 gr and 2.3×2×1.6 cm, with a rough external surface and a compact, yellowish cut surface, with focally hemorrhagic areas.

The histopathological analysis showed the presence of a neoplasm of the SG, consisting of cells of plasmacytoid morphology with ample and eosinophilic cytoplasm, arranged in a trabecular pattern. There were no ductal structures and no necrosis, capsular or vascular invasion, pleomorphism or increased mitotic activity were observed.

The immunocytochemical study showed immunoreactivity for cytokeratin 8 and 18 (CK8 and CK18), S-100, and focal smooth muscle actin (SMA). This set of findings allowed the diagnosis of plasmacytoid myoepithelioma of the salivary gland.

After the surgery, the patient is being followed in an ENT consultation to date and there is currently no clinical evidence of symptom worsening or local signs of recurrence.

DISCUSSION

ME account for less than 1.5% of all SG tumors and are usually located in the parotid gland. ME frequently presents as asymptomatic, slowly growing masses. Thus, a clinical examination and a high degree of suspicion are essential for the diagnosis, which is histopathological.[7,8] The imaging study helps to determine the intraglandular or extra-glandular origin, the relationship between the lesion and the facial nerve and, in some cases, whether the lesion is benign or malignant, as verified in this case.[8]

From a microscopic analysis, different types of patterns and cellular constitution do not appear to be associated with different rates of recurrence.[9]

ME express cyto-keratins so their absence should raise the suspicion of another diagnosis. The S-100 protein which is normally not present in normal myoepithelial cells, is a very sensitive marker for myoepithelial neoplasms, as seen in this case. In addition to the S-100 protein, vimentin and smooth muscle actin (SMA) can also be used to the diagnose of myoepithelial neoplasms.[2]

ME is a differential diagnosis with pleomorphic adenoma, neurinoma, hemangioma, lymphomas, solitary fibrous tumors, myelin sheath tumors, fibrous histiocytomas, paragangliomas, leiomyomas and leiomyosarcomas, hemangiopericytoma, among others. Many of these differential diagnoses share clinical and radiological features with each other so biopsy is essential for diagnostic confirmation.[4]

The recurrence rate of ME is described as 15-18%, with possible malignant transformation associated to long term cases or recurrences, which has been attributed to the overexpression of c kit receptors and mutation of the gene p-53.[1,2,10] Thus, even after complete excision of the lesion, follow-up of these patients for an extended period of time is recommended.

Our case report is considered a typical case of a benign salivary gland tumor, since it presents with a slowly growing and asymptomatic mass. The appropriate radiological study before the surgical intervention made it possible to anticipate the diagnosis and program the therapeutic strategy that was considered most appropriate.

Although there is a possibility of recurrence or malignancy, in this specific case a favorable prognosis is expected, and follow-up of the patient is recommended.

CONCLUSION

ME are rare, mostly benign tumors that often present as asymptomatic, slowly growing masses. They make a differential diagnosis with other SG tumors such as pleomorphic adenoma or Wharton tumor.

The treatment of choice is complete surgical excision with free margins, with radiotherapy being an option in cases where surgical intervention is not feasible. Recurrence is rare.

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