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CASE REPORT

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Myoepithelioma in accessory parotid gland: report of a rare case

Mioepitelioma em glândula parótida acessória: relato de um caso raro

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ABSTRACT

Myoepithelioma is a benign tumor of the salivary gland that mainly affects the parotid gland. The presence of an accessory parotid gland is an anatomical variation and neoplasms in this structure are extremely rare. This paper describes a case of a myoepithelioma arising in the accessory parotid gland of a 29-year-old woman. To the author´s knowledge this is the seventh case reported in English. The patient´s complaint was about a five year growing mass in the left cheek with a sudden growth outbreak. The patient underwent surgery via an intra-oral approach and the histopathology and the immunohistochemistry disclosed a myoepithelioma.

KEYWORDS

Parotid gland; Myoepithelioma; Salivary gland, neoplasms.

RESUMO

Mioepitelioma é um tumor benigno de glândula salivar que acomete, principalmente, a glândula parótida. A presença de uma glândula parótida acessória é uma variação anatômica e neoplasias nessa estrutura são extremamente raras. Este artigo descreve um caso de mioepitelioma ocorrendo em uma glândula parótida acessória de uma mulher de 29 anos de idade. No entendimento dos autores este é o sétimo caso relatado em Inglês. A queixa da paciente era uma massa na bochecha, com cinco anos de evolução, apresentando súbito surto de crescimento. A paciente foi submetida à cirurgia por meio de uma abordagem intra-bucal e a histopatologia e a imunohistoquímica revelaram um mioepitelioma.

PALAVRAS-CHAVE

Glândula parótida; Mioepitelioma; Glândula salivar, neoplasia.

INTRODUCTION

Salivary gland neoplasms are a rare group of neoplasms accounting for nearly 3% of tumors of the head and neck [1]. Myoepitheliomas are uncommon benign neoplasms that occur, mainly, in the parotid gland. [2,3]They comprise, approximately, 1% of all salivary gland tumors [2,3].

The accessory parotid gland (APG) is an anatomical variation consisting of a separate salivary tissue of the main body of the parotid gland [4-7]. The APG is located to the anterior of the parotid gland [4-7]. It is estimated that APG occurs in 21 to 69% of individuals [4-7]. Tumors in APG are extremely rare [3-7]. According to Iguchi et al. [3], until 2014, only 6 cases of myoepitheliomas in APG were reported in English, including their report. Given the rarity of the neoplasm site of presentation, many other diagnoses are hypothesized instead of a myoepithelioma [3-7].

This paper aims to show the seventh case of myoepithelioma in APG.

CASE REPORT

A 29-year-old caucasian woman was referred for a consultation with an oral and maxillolofacial surgeon, in January 2013. She reported a five year growing mass in the left cheek.

Upon physical examination a palpable, hardened and mobile lesion, anterior to the left parotid, was noted in the cheek. The approximate measures were 2.0 x 2.0 cm, according to the report.

A provisional diagnosis of a pleomorphic adenoma or lymph node hypertrophy was established. Ultrasonography revealed a lesion measuring, approximately, 1.5 x 1.2 x 1.0 cm (Figure 1). A Color Doppler showed signs of blood flow inside the lesion and a second ultrasound revealed a mass that was diagnosed as a lymph node.

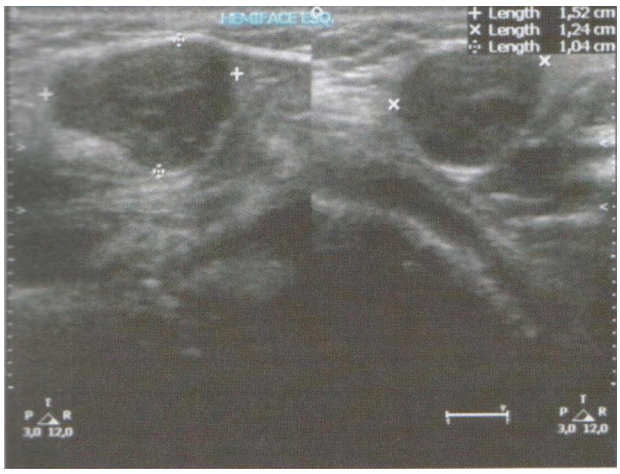

Figure 1 - Ultrasonography revealed a lesion measuring, approximately, 1.5 x 1.2 x 1.0 cm.

A computed tomography scan, in the axial aspect, revealed a nodular image, measuring 1.4 x 1.1 cm, located superficially to the masseter muscle (Figure 2).

The sum of anamnesis, the physical examination, and the image exams indicated the presence of a benign tumor in an APG. In February 2013, the patient was informed about her condition. In May 2013, the patient returned and surgery was proposed.

In December 2013, the patient was operated on. The lesion was excised via an intra-oral approach. The tumor showed a hard consistency and was attached to the muscle planes nearby the Stensen's duct. A transient left facial paralysis was present for 24 hours in the postoperative period.

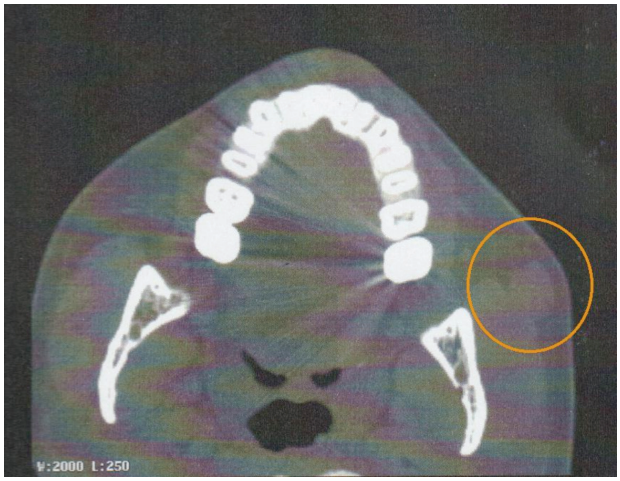

Figure 2 - 2D Computed tomography, in the axial aspect. Nodule measuring 1.4 x 1.1 cm, located superficially to the masseter.

The surgical specimen was sent for histopathological examination. The microscopy showed the presence of spindle cells in bundles (Figure 3). In order to further diagnose the tumor, an immunohistochemical panel was proposed. Antibodies and their presentation were as follows: α-smooth muscle actin (+) (Figure 4), P63 (+), cytokeratin 5 (+), pan-cytokeratin AE1 / AE3 (+) (Figure 5), S-100 protein (+) and Ki-67 (under 2%). The histopathology associated with immunohistochemistry favored the diagnosis of a myoepithelioma arising in an APG.

Ten days following the surgery, the sutures were removed. The recovery of the operated area was uneventful. There was no purulent discharge and the left parotid gland produced a copious flow of saliva. One month after surgery the patient returned for clinical evaluation. The patient did not come to the other follow-ups.

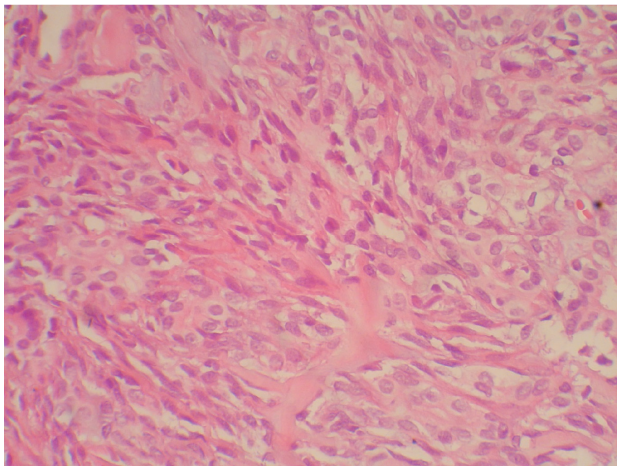

Figure 3 - Spindle cells arranged in fascicles, with stroma-like presentation (HE staining, 400x).

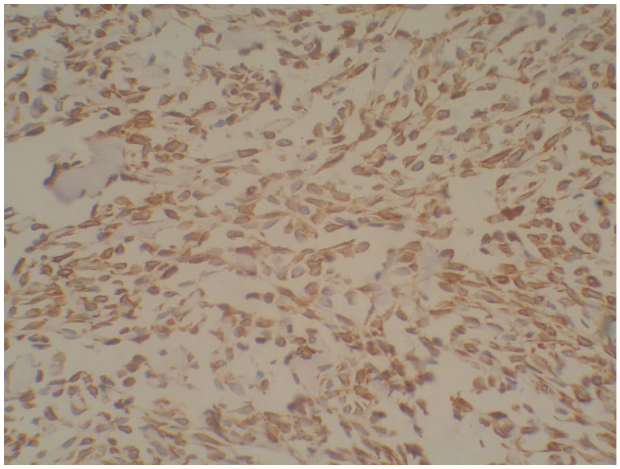

Figure 4 - Spindle cells stained positive for - smooth muscle actin (Immunohistochemistry, 400x).

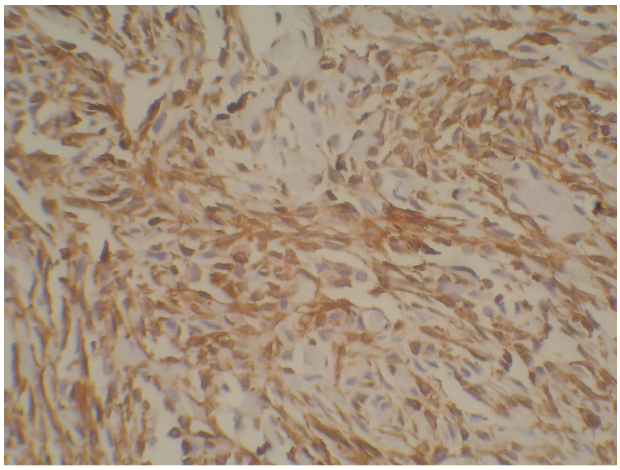

Figure 5 - Spindle cells stained positive for AE1/AE3 (Immunohistochemistry, 400x).

DISCUSSION

Myoepithelioma is a tumor that can affect any salivary gland, major or minor. However, the parotid gland originates 40% of cases. [2] The neoplasm is rare, accounting, approximately, for 1% of all salivary gland tumors, and it is composed, exclusively, by myoepithelial cells. [2,3] The mean age at diagnosis is 44 years (range 9 to 85), with its peak occurrence in the third decade of life. [2] There is no gender predilection. [2]The differential diagnosis of myoepithelioma is the pleomorphic adenoma [2,3], the most common tumor of the parotid gland and of the APG [1-7].

The APG is considered an anatomical variation and tumors in this structure are extremely rare. [3-7]The occurrence of APG tumors ranges between 1% to 7.7% among all the parotid gland tumors [4-7].

According to Lukšić et al. [4], after 24 years of clinical experience, 488 patients with primary tumors of the parotid glands underwent surgical treatment. Tumors in APG were found in six patients (1.23%). Only one disclosed a myoepithelioma after microscopic examination[4]. Hamano et al. [5], reported that 53 patients with parotid gland tumors were operated on during a period of three years (2000-2003) in the Department of Otorhinolaryngology (Tokai University School of Medicine). A single case of APG (1.8%) occurred, revealing a pleomorphic adenoma. In the studies of Klotz and Coniglio [6] and De Riu et al. [7], the pleomorphic adenoma was the most common benign tumor reported in APG, in accordance with the findings of Hamano et al. [5]. However, no myoepithelioma was found by the authors [5-7].

The diagnostic features of the APG neoplasms do not differ from the manifestation in the parotid gland and, usually, there is no pain at the time of the presentation[3-7]. A detailed medical history and a diligent physical examination are not sufficient to suggest a

myoepithelioma in the APG since the site of presentation could indicate other common lesions like the pleomorphic adenoma or a hypertrophic lymph node [3-7].Therefore, the use of imaging exams is imperative [3-7]. Magnetic resonance imaging and computed tomography scans can provide some provisional diagnosis [2-7]. However, the histopathology and the immunohistochemistry are of utmost importance in diagnosing myoepitheliomas and other lesions [2-7].

A variety of cellular morphological types have been recognized in myoepitheliomas, such as spindle cells, plasmacytoid cells, epithelioid cells and clear cells [2,3]. Most myoepitheliomas consist of a single cell type, but some combinations might occur as well. [2] The tumor cells are typically positive for some markers, including cytokeratins, S-100 protein, vimentin, calponin, muscle specific actin, and glial fibrillary acidic protein;in contrast, they are negative for carcinoembryonic antigen [2,3]. The reactivity of spindle cells is variable for α-smooth muscle actin, muscle specific actin,calponin, S-100 and smooth muscle myosin heavy chain. [2]. In the present study, the tumor was composed of spindle cells in bundles. The cells showed positivity for α-smooth muscle actin, pan-cytokeratin AE1 / AE3, cytokeratin 5, S-100 and p63. Ki-67 was below 2% indicating a low mitotic profile. The sum of the microscopic observations favored the diagnosis of a myoepithelioma, in accordance with literature.

In order to avoid damage to the facial nerve, some researchers perform the surgical removal of the tumor and the APG tissue inside the cheek by an external pre-auricular approach. [4,7] However, the intra-oral access or the direct skin approach right over the mass can also be used for the excision of the mass. [4,7] The latter technique is briefly mentioned in literature [4,7]. Klotz and Coniglio [6],and De Riu et al. [7] indicate the pre-auricular approach because they consider that intra-oral surgery could jeopardize the integrity of the facial nerve [6,7]. According to Hamano et al. [5] and De Riu et al. [7], the pre-auricular access provides sufficient exposure of the surgical site aiding the dissection of the facial nerve, although causing greater injury when compared to the intra-oral approach [5,7]. De Riu et al. [7] recommend the intra-oral access only when the surgeon has mastered the technique [7]. The same authors affirm that the advantages of this procedure were related to less surgical time required by the intra-oral direct approach, the reduction of surgical trauma and less aesthetic morbidity [7]. The actual case report was operated via an intra-oral access achieving the aforementioned benefits. The lesion was completely removed and the patient presented 24 h transient facial paralysis. Despite the transient nerve injury, the recovery was satisfactory.

Pertinent literature states that relapses of myoepitheliomas are less common than pleomorphic adenoma and are related to a compromised margin in the first surgical excision [2,3]. Iguchi et al. [3] reported no sign of relapse in the 3.5 years after the tumor in the APG was surgically removed. The possibility of malignant myoepitheliomas exists, especially in long-term neoplasms or in tumors with multiple recurrences [2]. Because this case report was followed for only one month, the authors understand that in the actual case, it is not known whether or not a long-term relapse occurred. However, the tumor was entirely removed, suggesting a positive prognosis.

CONCLUSION

Although the APG presents itself as a common anatomical variation, neoplasms in this structure are extremely rare. Myoepithelioma is a benign neoplasm of the salivary glands and, until the year 2014, only 6 cases of myoepithelioma in APG were reported in English.To the best of our knowledge, the case presented is the seventh report of myoepithelioma in APG.

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