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actaorlgallega@gmail.com





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Caso Clínico

Pleomorphic Adenoma of the mobile tongue

Adenoma Pleomórfico da língua móvel

Alexandra Correia, Alcinda Reis, Maria Bastos, Hugo Amorim, Carlos Carvalho

Centro Hospitalar entre o Douro e Vouga

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Abstract

Pleomorphic adenoma of the tongue is a rare condition that should be considered in the differential diagnosis of a tongue neoplasm. Excision is the main therapeutic option and a conservative surgical approach can be performed in the elderly and in cases where the tumor is well circumscribed. The authors present a case report and review of the literature.

Keywords: Pleomorphic adenoma, tongue neoplasm, minor salivary gland tumor

Resumo

O adenoma pleomórfico da língua é uma entidade rara que deve ser considerada no diagnóstico diferencial das neoplasias linguais. A cirurgia é a principal opção terapêutica e uma abordagem conservadora pode ser realizada em pacientes mais idosos e quando a neoplasia se encontra bem circunscrita. Os autores apresentam um caso clínico e revisão da literatura.

<u>Palavras-chave</u>: Adenoma pleomórfico, neoplasia da língua, neoplasia das glândulas salivares minor.

Correspondencia: Alexandra Correia Centro Hospitalar entre o Douro e Vouga Correo electrónico: costacorreia.alex@gmail.com

Introduction

Pleomorphic adenoma is the most common benign tumor of the salivary glands. However, it is more common in the major salivary glands, with only 8% involving the minor salivary glands. In the minor salivary glands, the palate, followed by lips and maxillary sinus are the most affected structures. Occurrence in the tongue is very rare and very few cases have been described in the literature.

Case Report

An eighty-year-old female arrived at the clinic with a painless right tongue mass that had been growing for the past 2 years. She also complained about sialorrhea, dysphagia and difficulty speaking. She had no alcohol or smoking habits.

On physical examination, a firm and smooth mass of about 3 cm was observed on the right side of the mobile tongue, covered by a normal mucosa. No cervical lymph nodes were palpable.

A contrast-enhanced computed tomography (CT) was performed, revealing a well-defined, moderately enhancing soft tissue mass on the right side of the mobile tongue, between the middle and anterior third, with no calcifications or cystic areas, and measuring 32x29x25 mm (figure 1).

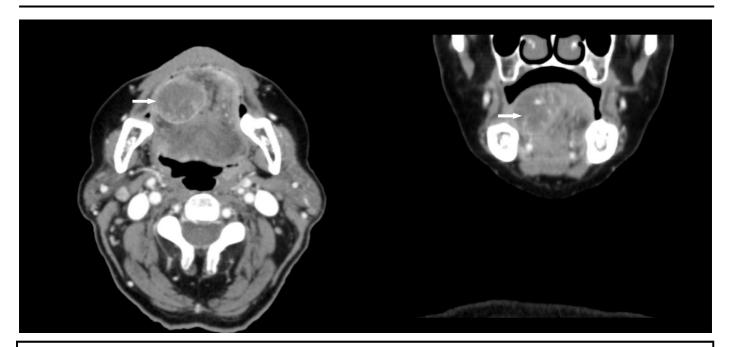


Figure 1: Axial and reformatted coronal CT sections of the tumor, which is blended in the intrinsic tongue muscles, with no involvement of the genioglossus and geniohyoid muscles, or the vasculonervous bundle.

The transoral fine needle aspiration for cytology revealed squamous cells and basal epithelial cells with a pseudoglandular pattern and a mucoid stroma. Definitive conclusions were not obtained due to fragmentation of the material.

We proposed surgical excision under general anesthesia. The lesion was resected with a clear dissection plane, through a transoral approach (Figure 2). The patient started oral feeding on the first postoperative day.

Pathology reported a nodular lesion, not intersected by the resection plane, with 15.6 g and 3.3x2x2.5 cm and with histology of pleomorphic adenoma (Figure 2).

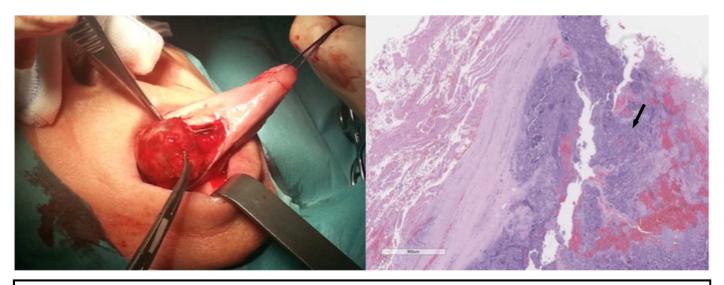


Figure 2: Tumor dissection (left). Histologic section (right) showing epithelial and myoepithelial cells surrounded by a condromyxoid stroma (hematoxylin-eosin stain).

A follow-up magnetic resonance_ was taken 2 months after resection, which confirmed complete removal of the tumor.

The patient, two and half years later, is still free of disease and with no sequelae.

Discussion

Pleomorphic adenoma of the tongue is a rare condition. Lingual salivary gland tumors are more likely to be malignant, with adenoid cystic carcinoma being the most common.⁶ In a series of 737 cases of minor salivary gland tumors presented by Wang et al.⁶, only 5 were found in the tongue and none of them was a pleomorphic adenoma. In another series of 543 cases by Pires et al.⁷, only 3 pleomorphic adenomas were attributed to the tonsillar area, tongue, mandible or other, not specifying the lesion site.

It is often a painless, slow-growing lesion and a worsening dysphagia can be the first clinical sign, or it can be incidentally detected upon routine examination by general practitioners.^{3,4}

The differential diagnosis includes all the minor salivary gland tumors and also benign and malig-

nant mesenchymal lesions, such as lipoma, neurofibroma and rhabdomyosarcoma.⁸ The histopathologic appearance of a pleomorphic adenoma is mainly composed of epithelial and myoepithelial elements, which are arranged in a variety of patterns and embedded in mucopolysaccharide stroma. The capsule is usually false, because its formation is caused by the fibrosis of the surrounding salivary parenchyma.⁴

Surgery is the main therapeutic option.^{3,4,8} Resection of the tumor with an adequate margin is key to avoid recurrence, and the chosen surgical approach must consider the age of the patient, the dimensions and the site of the lesion.⁴ However, these tumors are often well encapsulated and as such, if a clear dissection plane with the surrounding structures is seen, a more extensive surgery can be avoided.⁴ In this case, a resection without partial glossectomy was performed, strongly reducing the morbidity to an elderly patient.

Recurrence is uncommon and, when it does occur, may be attributed to partial excision or a multi-focal origin of the tumor.^{3,4,8} Spiro reported a recurrence rate of 6% in patients with benign, minor salivary gland tumors.¹ However, patients should continue to be seen for follow-up visits for a long period after, due to the possibility of late recurrence.

Conflict of interest: The authors declare that there is no conflict of interest.

References

- 1- Spiro RH. Salivary neoplasms: Overview of a 35-year experience with 2,807 patients. Head Neck Surg 1986;8:177-84
- 2- Grewal DS, Pusalkar AG, Phatak AM. Pedunculated pleomorphic adenoma of the tongue base manifesting with dyspnoea: A case report. J Laryngol Otol 1984;98:425-7
- 3- Nascimento LA, Vilela TGP. Pleomorphic adenoma of the tongue base: Case report and review. Int Arch Otorhinolaryngol. 2014;18(3):328-331
- 4- Berry S, Tay H, Puentes CP. Pleomorphic adenoma of the base of the tongue. Ear Nose Throat J. 2004;83(9):646-648
- 5- Tanigaki Y, Mikami Y, Ono M, Tsukuda M. Pleomorphic adenoma of the lateral side of the tongue. Acta Oto-laryngol.2004;124(5);649-51
- 6- Wang D, Li Y, He H, Liu L, Wu L, He Z. Intraoral minor salivary gland tumors in a Chinese population: a retrospective study on 737 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2007;104(1):94-100
- 7- Pires FR, Pringle GA, de Almeida OP, Chen SY. Intra-oral minor salivary gland tumors: a clinicopathological study of 546 cases. Oral Oncol.2007;43:463-70
- 8- Pires FR, Alves FA, Perez DE, Kowalski LP, Lopes MA, Almeida OP. Juvenile intraoral pleomorphic adenoma: report of five cases and review of the literature. Int J Oral Maxillofac Surg. 2002;31(3):273–275.