Pleomorphic Multicentric Adenoma in the Submandibular Gland

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Abstract Neoplasms of salivary glands represent a small group among the diseases involving the head and neck complex. In this group, the pleomorphic adenoma is the most frequent neoplasm, yet involves the submandibular gland in only 12.3% of cases. A patient presenting a swelling in the region of the submandibular gland was submitted to an incisional biopsy, where a fragment of the gland and one juxtaposed node were removed. Histologically they were defined as pleomorphic adenoma. Later, the patient was submitted to submandibulectomy and two other nodes were found close to the gland and removed. All specimens were histologically defined as pleomorphic adenoma. This multicentric finding is of great interest, perhaps explaining the recurrence rate of this neoplasm. The patient is in continuous follow-up and has not presented signs of recurrence.

Keywords Pleomorphic adenoma · Submandibular gland · Salivary gland tumor · Benign neoplasm · Multicentric tumor

Introduction

Neoplasms of salivary glands represent a significant group of neoplasms that involves the head and neck complex. In this group, the pleomorphic adenoma is the most frequent neoplasm, yet the submandibular gland is involved in only 12.3% of cases. A rare occurrence is its multiple manifestation within the gland, around 2.6%, presenting as multicentric or multifocal. Besides the rarity, this variant represents a determinant characteristic for the recurrence potential, either because of the greater number of lesions or due to its invasive nature [1–5]. This paper reports a case of a pleomorphic adenoma originating from the submandibular gland, in which surgical interventions demonstrated a multicentric behavior of the lesion.

Case Report

A white female patient, 36-years-old, presented with a painless tumefaction in the region of the submandibular gland, reporting 18 months of evolution. Clinical examination disclosed an endophytic nodule about 3.5 cm, localized in the region of the left submandibular gland, with lobulated surface and firm consistency. During manual stimulation of the gland, a slight decrease of salivary flow could be seen, but with no signs of infection. Computed tomography scan demonstrated an increase in the size of the submandibular gland without change of its density, suggesting a neoplasm (Fig. 1). At that time, there was no experienced professional to perform a fine needle aspiration biopsy at our institution, so the patient was submitted to incisional biopsy, in which a node partially superficial to the platysma and a small piece of the gland were removed. The result of the histopathologic examination, in both samples, was pleomorphic adenoma.

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Fig. 1 Computed tomography in axial cut demonstrating the increase of the left submandibular gland's volume

Fig. 2 Superficial nodes adjacent to the submandibular gland (see arrows)

About a month later the patient was treated by a left submandibulectomy through a submandibular approach. Interestingly, during the surgical procedure two additional nodules were observed just beside the gland, and even though they appeared as lymph nodes, both of them were removed (Fig. 2). After the identification of the gland capsule, a subcapsular enucleation was accomplished, avoiding injury to the lingual and hypoglossal nerves at the medial aspect of the gland (Fig. 3). Total removal was done with identification and clamping of the salivary duct at its most anterior portion. All specimens were analyzed and histologically defined as pleomorphic adenoma (Fig. 4).

The associations of these findings characterized the multicentric behavior of the disease. The patient is being followed and after 1 year, presents with good mobility and sensibility of the tongue. No signs of recurrence were found.

Fig. 3 Removal of the submandibular gland

Fig. 4 Hematoxylin–eosin stain histological aspect of the pleomorphic adenoma: gland and tumoral capsule in 10× magnification (a) and adjacent nodes with the ductal and myoepithelial neoplastic components in 40× magnification (b and c, respectively)

Discussion

The term pleomorphic adenoma was initially suggested due to the diversity of histological patterns observed in these tumors, basically characterized by ductal and myoepithelial neoplastic components, immersed in a hyalinized collagenous stroma with incomplete capsular involvement in most cases. This tumor presents itself as a painless and slow growing nodule that can reach great dimensions without

compromising the adjacent anatomical structures [1–5]. Proper treatment is only achieved when the correct diagnosis is obtained. Fine needle aspiration is the preferred initial diagnostic procedure since incisional biopsy for salivary gland lesions may result in seeding tumor beyond the capsule. However, due to the lack of a typical histological pattern and to the morphological variety of myoepithelial neoplasms, fine needle aspiration biopsy, incisional biopsy or frozen biopsy are useful in the differentiation from malignant lesions, but do not always provide for a definitive diagnosis. This confirmation is only obtained from the histopathologic examination of the entire specimen, associated with clinical and image information [4–7]. Treatment success is directly related with total removal of the gland. Once the pleomorphic adenoma presents an incomplete capsule, its enucleation with the preservation of the gland is controversial, probably increasing the possibilities of recurrence [8, 2]. Recurrences generally present as multiple nodes in the topography of the affected gland. This characteristic, however, may not be exclusive to recurrences, being seen in primary lesions as well [2, 9]. Multiple nodes affecting the same region characterize the multicentric or multifocal behavior of a disease. This characteristic is rare and mostly seen in malignant neoplasms, being interpreted, somehow, as local metastasis [10]. Multicentricity will increase the possibility of recurrences, since an invasive characteristic is not supported in the literature [10–12]. In the report of 38 cases of submandibular pleomorphic adenoma presented by Laskawi et al., the only multicentric case reported developed a recurrence [8]. Another variation of the behavior of a lesion is the possibility of its presentation in different locations at the same time, being then called as a synchronous lesion, mostly seen in Whartin tumor [12]. Seeding of the tumor during surgical procedures has been the primary feature that explains multicentric behavior; however, in this case report, multiple presentations were already seen in the incisional biopsy. Another detail is that the submandibular gland maintained capsular integrity; therefore the possibility of local metastasis would be highly questionable. Epithelial remnants of the submandibular gland trapped in local lymph nodes could also explain multicentric and synchronous behavior of the presented case [12]. Due to the reports of this variant behavior of a pleomorphic adenoma, the authors support the importance of removal of nodes adjacent to the affected gland, even

though they seem to be lymph nodes, and they should all be histologically analyzed. This would probably lead to an additional few minutes of surgery, but this approach can avoid secondary surgery. Maybe, some of the recurrences of pleomorphic adenomas are associated with misdiagnosed multicentric variants, increasing the need for careful evaluation of this neoplasm in the perspective for better results [2, 8, 10, 11].

Knowledge of the clinical behavior of a neoplasm is important to promote adequate treatment. Pleomorphic adenomas have a tendency to reoccur but secondary surgery for recurrent lesions carries a worse prognosis.

References

1. Ito FA, Ito K, Vargas PA, de Almeida OP, Lopes MA. Salivary gland tumors in a Brazilian population: a retrospective study of 496 cases. *Int J Oral Maxillofac Surg* 2005;34:533.
2. Erol B, Özer N, Unlü G, Gülsün B. A massive pleomorphic adenoma of the submandibular salivary gland. *J Oral Maxillofac Surg* 2004;55:1011.
3. Alves FA, Perez DEC, Almeida OP, Lopes MA, Kowalski LP. Pleomorphic adenoma of the submandibular gland: clinicopathological and immunohistochemical features of 60 cases in Brazil. *Arch Otolaryngol Head Neck Surg* 2002;128:1400.
4. Simpson RHW. Myoepithelial tumours of the salivary glands. *Curr Diagn Pathol* 2002;8:328.
5. Ravidis AD, Stavrianos S, Lagogiannis G, Faratzis G. Tumors of the submandibular gland: clinicopathologic analysis of 23 patients. *J Oral Maxillofac Surg* 2004;62:1203.
6. Ethunandan M, Witton R, Hoffman G, Spedding A, Brennan PA. Atypical features in pleomorphic adenoma—a clinicopathologic study and implications for management. *Int J Oral Maxillofac Surg* 2006;35:608.
7. Boerner SL. Patterns and pitfalls in fine needle aspiration of salivary gland. *Curr Diagn Pathol* 2003;9:339.
8. Laskawi R, Ellies M, Arglebe C, Schot A. Surgical management of benign tumors of the submandibular gland: a follow-up study. *J Oral Maxillofac Surg* 1995;53:506.
9. da Cruz Perez DE, Pires FR, Alves FA, Almeida OP, Kowalski LP. Salivary gland tumors in children and adolescents: a clinicopathologic and immunohistochemical study of fifty-three cases. *Int J Pediatr Otorhinolaryngol* 2004;68:895.
10. Seifert G, Donath K. Multiple tumours of the salivary glands terminology, nomenclature. *Eur J Cancer B Oral Oncol* 1996;32B:3.
11. Shinohara M, Ikebe T, Nakamura S, Takenoshita Y, Oka M, Mor M. Multiple pleomorphic adenomas arising in the parotid and submandibular lymph nodes. *Brit J Oral Maxillofac Surg* 1996;34:515.
12. Gnepp DR, Schroeder W, Heffner D. Synchronous tumors arising in a single major salivary gland. *Cancer* 1989;63:1219.