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Pleomorphic Adenoma of Submandibular Gland: A Case Report and Literature Review

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ABSTRACT

Pleomorphic adenoma, a benign salivary gland tumor, is commonly encountered in clinical practice in parotid gland affection. His location submandibular region is more uncommon and has the same rate with potential malignant neoplasm. This case report highlights a unique instance of pleomorphic adenoma affecting the submandibular gland, emphasizing the diagnostic challenges, treatment modalities, patient outcomes associated with this specific presentation and review the literature.

Keywords: Pleomorphic adenoma, Submandibular gland, Salivary glands tumors

INTRODUCTION

Pleomorphic adenoma is a benign salivary gland tumor involving mainly major salivary glands; among this, it mainly involves parotid gland in about 84% cases, submandibular gland involvement can occur in about 8% cases, while rarely, it can occur in minor salivary glands for 6,5%.[1,2] Salivary gland tumors though relatively rare account for approximately 5% of all head and neck tumors, while 0.5% of such tumors can be malignant.[2] This paper describes a case of Pleomorphic adenoma involving submandibular gland.

OBSERVATION

A 64-year-old female patient with no notable history who has presented for 10 years with a left cervical swelling mass that has progressively increased in size without any other associated sign, all evolving in a context of general preservation of her condition.

The clinical examination revealed a left latero-cervical mass with no inflammatory signs, multilocular in appearance, with healthy skin of hard, stony consistency, measuring approximately 7 to 8 cm in his long diameter and painful to palpation [Image 1].

The patient underwent cervical ultrasound revealed a left latero-cervical tissue mass associated with a hypertrophic multinodular thyroid gland. Blood tests did not reveal any thyroid disorder.

Given the cost and the availability of the MRI, a CT-scan was made for assessing the relationship with adjacent structure. This exam showed a voluminous circumscribed mass measuring 56x48 mm, developed from submandibular gland, with no vascular compression, no lymph nodes or mandibular bone involvement. [Image 2]

An excision of the tumor under general anesthesia was made, by submandibular approach and the operative specimen was sent for anatomopathological examination, which came back in favour of a pleomorphic adenoma. [Image3]

A five month follow up did mot show any sign of recurrence and the patient is still under surveillance for her thyroid.[Image 4]

Image 1: Facial and profile Vue of the patient before surgery showing the submandibular poly lobular mass.

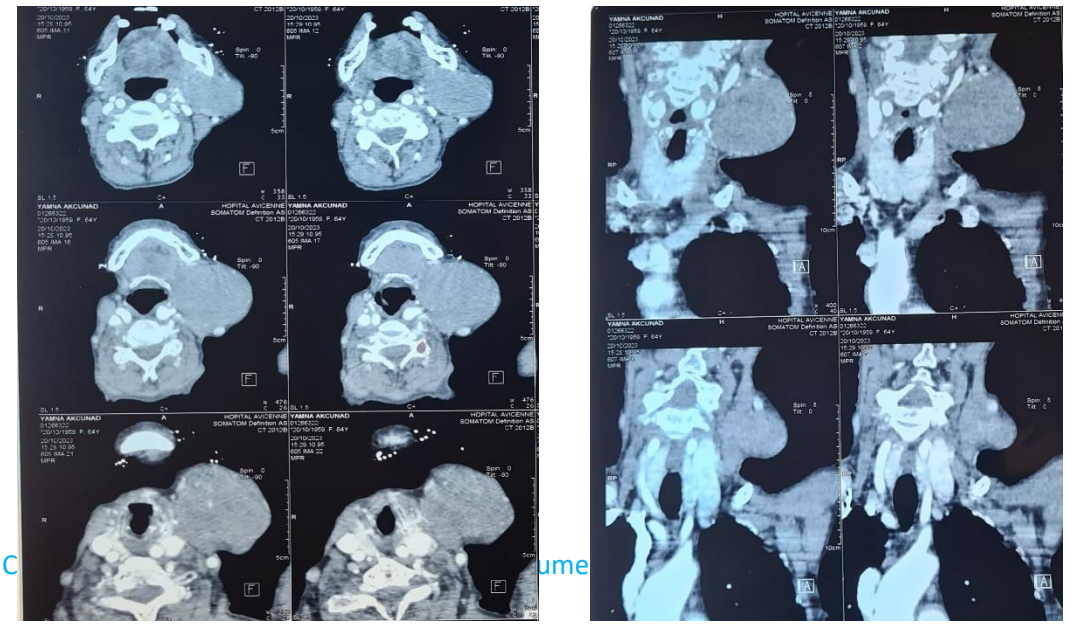

Image 2: Axial and coronal view of an injected CT-scan showing a homogenous oval mass occupying the submandibular fossa.

Image 3: macroscopical aspect of the tumor after surgical excision showing an encapsulated poly lobulated mass.

Image 4: Facial and profile Vue of the patient five months after surgery

DISCUSSION

Salivary gland tumours are relatively rare. Pleomorphic adenoma is by far the most common salivary gland tumor involving both major and minor salivary glands, where it constitutes 60 to 75% of parotid tumors.[1]

Submandibular gland tumours are much less common than parotid tumours and constitute about 10 to 15 % of salivary tumours with an equal distribution of malignant and benign tumours.[1]

Pleomorphic adenoma can be seen at any age, mostly between 30 to 60 years old with a slight female predilection (2F/1M). This statement has been assessed by several retrospective studies.[1,2,3,4]

Clinically, their development is generally slow and painless, which generally delays treatment. Nevertheless, some of them can present a rapid clinical evolution that can cause confusion with potential malignant tumor by the presence of facial nerve weakness.[3]

Macroscopically, it presents as a single polylobate, slow-growing, painless, firm and in some cases with stony appearance, mobile mass localized in the submandibular region without any fixation to the floor of the mouth or the mandible. There is no nerve injury or compression and no lymphadenopathies associated.[5]

Microscopically, the PA in submandibular location is an encapsulated with an intact capsule and generally present as a unique mass without pseudopodia and satellite nodules, occupying the periphery of the gland. It's composed with epithelial and myoepithelial elements arranged in variable mucoid, myxoid, or chondroid mesenchymal components.[5,6] This architectural diversity is the base of several subtypes which can be encountered in histopathological exams. It is from this diversity that the name mixed tumor commonly given to pleomorphic adenomas derives.

In our case, the anatomopathological exam found an encapsulated mass with a capsule of variable thickness. The cellularity was made mix of epithelia and myoepithelial cells associated with a fibro-chondro-myxoid contingent.

The exploration of submandibular abnormalities can be made by a scope of imaging. Ultrasound can easily differentiate solid from cystic lesions in the salivary glands. It is the examination of choice for ruling out non-tumoral lesions, such as normal lymph nodes, inflammatory adenitis or chronic sialadenitis nodules.[7]

Pleomorphic adenomas are typically hypoechoic and may show posterior acoustic enhancement. It is also useful in guiding a biopsy (both FNAC and core biopsies). Most salivary gland tumors are investigated by Fine-needle aspiration cytology (FNAC); it is frequently practiced as part of the preoperative work-up for salivary gland lesions in replacement of biopsy. With a sensitivity and specificity more than 90%.[7,8]

Computerized Tomography (CT) scan and Magnetic Resonance Imaging (MRI) are the gold standard radiological tools for lesion arising from the salivary glands they have similar sensitivity and specificity for determining tumour location, tumour margin and tumour infiltration. Even if CT has lower resolution than MRI for soft tissue, CT offers an advantage in detecting early cortical bone invasion of the mandible.[1,2,7]

The final pathologic diagnosis is always established based on the histopathological findings after surgical excision. The per operative specimen examination can be required for distinction between a malignant and benign tumor but expose to the possibility of recurrence and to unclear diagnosis by the lack of enough material.[7]

Resection of the tumor and the involved gland is the treatment of choice for pleomorphic adenoma of the submandibular gland. The key to surgery is to not disrupt the capsule otherwise the risk of recurrence increase.[9,10,11]

Enucleation alone carries a high risk of recurrence due to pseudopod-like extensions of tumour. Injury to the marginal mandibular nerve is the most common complication leading to temporary or permanent paralysis due to the stretching or compression of the nerve.[6,7,9,11]

Malignant transformation of pleomorphic adenoma is rare and occurs most frequently in patients with long-standing tumour. The risk of malignant transformation in pleomorphic adenoma is 1.5% within the first 5 years of diagnosis but increases to 10% if observed for more than 15 years. Therefore, early definitive treatment is strongly recommended.[6,7,12,13]

CONCLUSION

Pleomorphic adenoma is the commonest benign neoplasm of salivary glands. His low-silent growing can lead to voluminous tumor and potentially malignant tumor. That's why his surgery must be sufficient and carefully done to avoid with recurrence or malignant transformation.

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