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Case Report

Management of Recurrent Parotid Pleomorphic Adenoma: A Case Report

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Abstract

Pleomorphic adenoma is the most common neoplasm of the salivary glands with an incidence of 45% to 60% [1]. The recommended treatment for the pleomorphic adenoma is surgery. The recurrence of pleomorphic adenoma is uncommon and recurrence rates depend on the initial surgery. Magnetic Resonance Imaging is the imaging study of choice for recurrence. Management of recurrence is challenging and there is no standardized treatment. It must be selected for every patient and there is a high probability of needing multiple treatment procedures. We present the case of a 54 year old patient with a history of a parotid tumor resection in 2016 that concurres to the Head and Neck Surgery office with a recurrent left parotid mass. A left parotidectomy and left jugular-carotid lymphadenectomy is performed in which the pathology reports recurrence of pleomorphic adenoma.

Keywords: Adenoma; Parotid; Recurrence; Pleomorphic; Parotidectomy

Abbreviations

PA: Pleomorphic Adenoma; MRI: Magnetic Resonance Imaging

Introduction

Pleomorphic adenoma (PA) is the most common neoplasm of the salivary glands, constituting up to two-thirds of all salivary tumors [2]. It is a solitary benign lesion that usually presents as a painless, slow growing mass. This tumor can be present during many years but has a risk of malignant transformation. The risk increases proportionally to the time of lesion. Therefore, surgical resection of the tumor is recommended. Other factors that increase the risk of malignization include: radiation, advanced age, size and recurrent tumors [1].

Recommended treatment for the PA is surgery. At present, it is treated with total or superficial parotidectomy. PA is no longer enucleated in surgical practice because of the high recurrence rate. The recurrence of PA is usually during the 7 to 10 years after the first surgery. The risk of recurrence is higher in younger patients, after enucleation or when margins are invaded [1].

Case Report

We report the case of a 54-year-old female with a history of enucleation of a parotid tumor in 2016 that consulted for a recurrent left parotid mass. The pathology of the tumor resection reported pleomorphic adenoma. On physical examination the patient presented a left parotidectomy scar and a polylobulated lesion in the anterior portion of the left parotid gland. The lesion was 6cm x 3cm, mobile and without skin involvement. She presented hypoesthesia of the auricular nerve with no lymphadenopathy or involvement of the facial nerve.

Ultrasound revealed a 56 cm x 38 mm hypoechoic, multilobulated mass with internal vascularization. Fine needle aspiration of the lesion guided by ultrasound confirmed recurrence of pleomorphic adenoma. A magnetic resonance imaging (MRI) with contrast of the head and neck was also performed. It revealed confluent nodular formations with homogeneous enhancement with gadolinium involving subcutaneous tissue and upper pole of the left parotid gland that could correspond to recurrence of pleomorphic adenoma (Figure 1).

Figure 1: a. MRI T1 with contrast, coronal section. 1.b. MRI T1 with contrast, sagittal section. Confluent nodular formation with homogeneous enhancement with gadolinium involving subcutaneous tissue and upper pole of the left parotid gland.

With a presumptive diagnosis of recurrence of pleomorphic adenoma a surgical exploration was performed. The patient underwent a left parotidectomy and left jugular carotid lymphadenectomy (Figure 2). During the procedure, a tumor involving the auricular nerve and lymphadenopathies on level IIa were found. Resection of the lesion and a lymphadenectomy of jugular-carotid dissection was performed. Frozen pathology of the lymphadenopathies evidenced absence of neoplasia. Dissection and resection of the tumor was completed with use of neurostimulation. Frozen pathology was compatible with pleomorphic adenoma. At the end of the surgery, a positive neurostimulation response was evidenced in the branches of the facial nerve.

Figure 2: a. Dissection of tumor with identification of facial nerve. 2. b. left parotidectomy and left jugular carotid lymphadenectomy.

The patient evolved with a favorable outcome. Six months postoperatively, the patient presented an adequate healing wound, no facial paresia and adequate pain management. The permanent pathology reported recurrence of pleomorphic adenoma.

Conclusion

The recurrence of PA is uncommon, and its occurrence is associated with the selection of the first surgery [2]. The time interval from the initial treatment can be as long as 15 years [3]. It occurs usually during the 7-10 years after the first surgery. It is reported that the recurrence rates depend on the initial surgery. After a superficial parotidectomy the recurrence rate is below 3% [3]. The recurrence time rate is shorter in patients in which an enucleation is performed.

The recurrence of parotid adenoma is multifocal. There are multiple hypotheses about the recurrence of these tumors. It has been attributed to: the surgical procedure, the rupture of the capsule during surgery, the size of the tumor, the multicentric origin, the misdiagnosis and the undetected tumor extension [4].

The recurrence of PA is associated with a risk of metastasis and malignant transformation of 5-15% [4]. This risk might increase with the time and the number of recurrences and may be preceded by pleomorphic adenoma with severe dysplastic change [1]. This increase in malignant transformation situates this carcinoma ex pleomorphic adenoma as the fourth most common malignant salivary tumor.

Patients usually present an asymptomatic mass. The image of choice for evaluation of pleomorphic adenoma recurrence is the MRI. This type of image can identify the multinodular nature, the depth, extension of the tumor and the residual parotid gland. MRI may not identify all nodules. When a patient presents with recurrence of PA, the number of nodules is probably much higher than is clinically evident or even detectable by MRI [1].

Management of recurrence of PA is challenging and there is no standardized treatment. The surgical treatment is usually difficult because the tumor is multinodular and can be associated with compromise of the facial nerve [3]. Surgery for recurrent PA is associated with significant risk of functional and cosmetic sequelae, further episodes of recurrence, and increased risk of malignant transformation [5].

The treatment of choice will depend on the type of patient, the surgery performed previously, the extension of the recurrence and the facial nerve affection.

The first thing that should be considered is the type of patient. In elderly patients or no surgical candidates observation is recommended, expecting for the tumor not to grow. Second, if the previously performed surgery was an enucleation or a partial parotidectomy, the treatment of choice is a superficial parotidectomy or a total parotidectomy. The surgery should be selected depending on the involvement of the facial nerve and location of the tumor. When the previously performed surgery was a total parotidectomy the surgical treatment should be a localized resection of the tumor with the objective of preserving the facial nerve. Last thing to be considered is the affection of the facial nerve. In patients who present infiltration of the branches or main trunk a radical or extended parotidectomy should be performed. In less than 30% of the patients there is a need to sacrifice a branch of the facial nerve [1]. Extensive or radical parotidectomies with sacrifice of the facial nerve do not prevent further recurrences.

Recurrence of PA is unusual and it is fundamental a first adequate approach of de PA. Management of recurrent PA is challenging and there is no standardized treatment. It is important to take in account the type of patient, facial nerve involvement and past surgeries in the time of deciding a surgical approach.

Conflicts of Interest

None declared.

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