

Extensive Multinodular Recurrence of Pleomorphic Adenoma of Parotid Gland - A Case Report and Literature Review

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Abstract

We present a case of a massive, unilateral, multinodular recurrence of parotid gland Pleomorphic Adenoma (PA). It extended through facial to cervical region which is reported to be a rare area for extension of recurrence. A 37-year-old male complained of dysphagia to solids and breathing difficulties in 2013. The examination of the oral cavity and oropharynx found a mass on the right side wall of the oropharynx. The computed tomography (CT) scan revealed a large expansive oval formation measuring 60 x 45mm with regular edges, localized in the right parapharyngeal space. The patient underwent surgery and the histological exam confirmed the diagnosis of PA with no signs of malignancy. In March 2018, the patient presented with multiple palpable and non-tender masses and subcutaneous nodules of the right parotid and homolateral cervical region. Revision surgery was indicated and excision of the lesions was performed. Histopathology revealed 43 nodular lesions of PA.

The standard treatment of PA is surgery. However, adverse outcomes of the surgical procedures are common and recurrence is a complex problem. A careful dissection in cases of tumours with an intact capsule may decrease recurrence rate which is more evident in cases of incomplete and no meticulous dissections.

Keywords: Pleomorphic Adenoma; Salivary Gland; Head and Neck Surgery

Introduction

Pleomorphic adenoma (PA), also known as benign mixed tumor, is the most common salivary tumor, accounting for almost two-thirds of all salivary gland neoplasms [1]. Mostly, PA are located in the parotid glands (85%), minor salivary glands (10%) and sub-mandibular glands (5%) [2]. It is a slow-growing benign neoplasm, most commonly seen as a solitary lesion and it is most frequently found in middle-aged females. [3-5]. Optimal initial management is essential to reduce the risk of recurrence and malignant transformation [6].

We present a case of a massive, unilateral, multinodular recurrence of parotid gland PA constituted by 43 nodules. It extended through facial to cervical region which is reported to be a rare area for extension of recurrence [7].

Clinical case

A 37-year-old male was referred to our department in January 2013. He complained of dysphagia to solids and breathing difficulties. The beginning of clinical symptoms dated back to 4 months before, with progression of discomfort when swallowing. No significant medical, surgical, social or family history was

reported. The examination of oral cavity and oropharynx found a mass on the right side of the oropharynx repressing the uvula and ipsilateral palatine tonsil inside. This mass was hard, non-painful to palpation, with a normal overlying mucosa and had no cervical expression. Fiberoptic flexible laryngoscopy showed narrowing of the nasopharyngeal and oropharyngeal lumens with no relevant alterations on the hypopharynx. All cranial nerves, most notably the facial nerve, were intact. Computed tomography (CT) scan revealed a large expansive oval formation measuring 60 x 45mm with regular edges, localized in the right parapharyngeal space (Figure 1). Fine-needle aspiration was consistent with PA. The patient underwent surgery and excision of the lesion was performed via S-shaped preauricular and submandibular incision. Histological examination confirmed the diagnosis of PA with no signs of malignancy. The patient was discharged, after 6 days, without signs of facial palsy, and was regularly seen for routine monitoring. In March 2018, the patient presented with multiple painless, well-defined, palpable and non-tender subcutaneous nodules in the right parotid and homolateral cervical region (level II and III). Clinically, he showed no facial paralysis. Magnetic resonance imaging (MRI) showed multiple nodules in the parapharyngeal space, in the lower part of the right parotid space and along the cervical scar (Figure 2). Fine-needle aspiration cytology demonstrate a recurrence of PA. Revision surgery was indicated and excision of the lesion was performed via right latero-cervical approach. Definitive histopathology revealed 43 nodular lesions of PA and no malignancy was identified in the specimen. The patient recovered from the surgery without complications and is currently being seen regularly for routine monitoring.

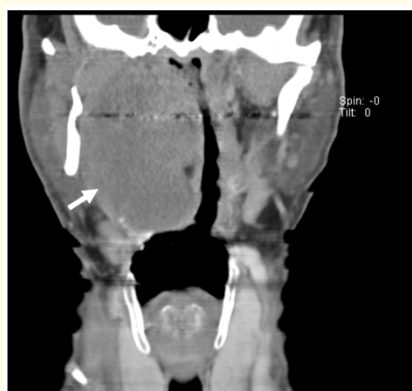


Figure 1: Coronal CT scan revealed a large expansive oval formation localized in the right parapharyngeal space.

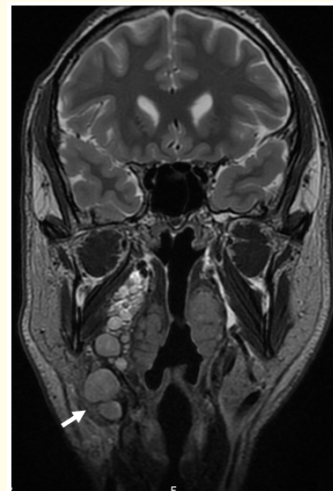


Figure 2: T2 MRI (coronal view) showed multiple nodules in the parapharyngeal space, in the lower part of the right parotid space.

Discussion

Recurrence of PA is a complex problem, occurring 2-15 years after the initial surgery [8]. The standard treatment of PA is a surgical procedure [4,6]. However, adverse outcomes of surgical procedures are common and the recurrence rate has been reported to be 1-45% [4,9]. The high rate of recurrence is considered to be associated with an incomplete initial surgical management, capsular rupture during enucleation, multinodular histological type, proximity to facial nerves and young age [6,8]. In this case, although tumor was completely removed along with the surrounding tissue in the primary surgery, the tumor size was significant (> 4 cm), and therefore capsule rupture during surgical manipulation or an incomplete capsule might have led to the referred recurrence on the surgical field of primary surgery [10]. Probably tumour spillage in our case, might be one of the reasons why recurrence exhibited a multifocal pattern [10]. Although the high local recurrence pattern, and the recurrence extending to the cervical region is rare [4].

Surgical management of recurrent PA is often difficult and it is imperative to inform the patient about the surgical procedures and risks [4]. Recurrent PA should be treated with re-resection but the risk of Frey syndrome, salivary fistula and facial nerve dysfunction increase [4].

MRI remains the gold standard in the diagnosis of neoplasms of salivary glands [6]. Recurrent PA is often multifocal, involving both the tumour bed and the adjacent spaces, as seen in our case (Figure 2). These lesions, often small nodules with diameter less than 1 centimeter, appear strongly hyperintense in T2 sequences, which are easy to identify [3]. In this case, MRI also helped in locating multinodular lesions in fatty areas below the parotid gland and forward and backward of sternocleidomastoid muscle. This has contributed for a better surgical plan in order to make an optimal cervical exploration and excise the multiple nodules. Resection of recurrent multifocal PA must be as complete as possible and adjuvant radiotherapy for a better locoregional control remains controversial [6]. In our patient, taking into account no histological evidence of malignant transformation, long-term morbidity associated to radiotherapy and patient's preference, no adjuvant treatment was performed and he continues under regular surveillance in our department.

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