

Bilateral Pleomorphic Adenomas Of The Parotid Glands: Discussion Over Two Cases

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Abstract

The occurrence of multiple distinct tumors in major salivary glands is quite rare. Although the most common tumor with bilateral synchronous or metachronous development is the Warthin's tumor, pleomorphic adenomas have been very rarely diagnosed simultaneously as well.

The authors report two consecutive cases of patients with bilateral pleomorphic adenomas of the parotid glands discussing clinical features, diagnostic challenges and therapeutic options.

INTRODUCTION

Bilateral tumors of the parotid glands are unusual and quite rare.

Histologically, they are divided into unifocal or multifocal lesions. They can be also distinguished into synchronous or metachronous tumors regarding the time of their detection, even if it might be very difficult to establish.

The most common bilateral tumor of the parotid glands is Warthin's tumor, with an incidence of 5-14%, although pleomorphic adenoma remains the most common unilateral neoplasm (1).

In this paper two cases of bilateral swelling located near the mandibular angle are presented of which the first one resulted as synchronous whereas the latter was referred as metachronous. Afterwards, clinical features, diagnostic challenges and therapeutic options are discussed.

CASE REPORTS

CASE 1

A 35-years old young white woman first presented at our Department in March 2003 complaining about a 4-years history of two slowly growing, painless swellings which appeared quite simultaneously, bilaterally located near the angle of the mandible.

The patient never gave importance to "her lumps" until swellings became too evident thus causing her aesthetic discomfort. She never referred either symptoms of

inflammatory or microbial parotid involvement or signs of autoimmune or hematological disease. Moreover, she had never been under any drug treatment. No history of head and neck malignancy or prior radiation exposure was referred at first presentation.

Bilateral clinical examination showed at the inferior aspect of the parotid region two painless, relatively small, nodular, well-defined, firm, movable masses, without overlying skin changes. The patient did not present facial nerve involvement, ear anesthesia, clinically evident cervical lymphadenopathy or displacement of the soft palate.

An ultrasound assessment of the parotid regions already performed by the patient, Bilaterally swellings were referred as mixed tumors.

The patient underwent serological testing for Sjogren syndrome, tuberculosis, cytomegalovirus, human immunodeficiency virus (HIV), Epstein-Barr virus (EBV), which resulted negative.

US guided fine-needle aspiration cytology (FNAC) of the clinically evident lesions, suggested the presence of two pleomorphic adenomas located at the inferior aspect of the parotid gland, on both sides. The patient even underwent a magnetic resonance imaging (MRI) to better establish extent and limits of the lumps. MRI confirmed the presence of two well defined, fluid-containing masses at the inferior aspect of the parotid superficial lobe.

Hence, surgical removal of the masses was planned.

On May 2003 the patient underwent one-time bilateral superficial conservative parotidectomy. The postoperative course of the patient was uneventful except for a slight transient deficit of the right marginalis nerve. Pathologic analysis of the surgical specimens established the presence of bilateral unifocal pleomorphic adenoma without infiltrative aspects, completely removed. Two months postoperatively the patient showed good facial nerve functional recovery and surgical wound repair was satisfactory on both sides.

CASE 2

A 66 years old woman with arterial hypertension first came to our observation on July 2004 complaining about two, slowly growing, non-tender lumps located on the parotid regions causing her aesthetic discomfort and slight but persistent pain. The patient did not refer either symptoms of inflammatory or microbial parotid involvement or signs of autoimmune or hematological disease and no history of head and neck malignancy or prior radiation exposure resulted.

Notably, she referred the occurrence of the left parotid region mass almost 20 years before the right inferior parotid region lump.

Thus a metachronous bilateral parotid tumor was suspected.

The patient underwent FNAC of the lumps which showed the presence of pleomorphic adenoma on both sides.

Pre-operative MRI assessment confirmed the presence of two, well defined, solid masses, both located on the inferior aspect of the superficial lobe of the parotid glands.

Hence, surgical removal of the masses was planned. On September 2004 the patient underwent the first surgical intervention which consisted on removal of the left side neoplasm via a conservative superficial parotidectomy. Post-operative course was completely uneventful and the definitive histology confirmed FNAC findings, showing unifocal pleomorphic adenoma, completely removed. The right neoplasm was treated in March 2005 using the same surgical technique. Post-operative course was uneventful and the definitive histology confirmed FNAC findings, showing a completely excised unifocal pleomorphic adenoma.

DISCUSSION

Pleomorphic adenoma, called mixed tumor because of its either epithelial and connective component, accounts for 80% of all parotid tumors. It is mostly located at the superficial lobe of the parotid gland. The average age of

onset is between 30 and 50 years. Pleomorphic adenoma clinically presents a benign painless mass slowly growing and locally invasive. Some authors refer mean duration of symptoms prior to diagnosis as 22.9 months, 36.5 in male patients and 22.9 in female patient. A statistically significant association between preoperative duration of symptoms and tumor size was not found.⁽³⁾

Bilateral either neoplastic or non neoplastic parotid swellings are not so unusual in the clinical practice. The rarity of bilateral synchronous pleomorphic adenoma is confirmed by several studies. Turnbull and Frazell⁽⁶⁾ found multiple neoplasms in only 1.3% of cases, but no one was referred as pleomorphic adenoma. Another series published in 2004 only 4 cases of synchronous bilateral pleomorphic adenoma were described⁽⁷⁾.

Nevertheless, the development of a parotid mass is often found in patient with history of hematological or autoimmune diseases.⁽²⁾ Thus differential diagnosis appears to be pivotal to choose the most appropriate treatment.

The most widely used surgical procedure for the excision of a superficial lobe benign parotid tumour is nowadays represented by superficial parotidectomy which nowadays is referred as the most widely used technique by the International Literature. Other inappropriate surgical treatments, such as enucleation, are strongly associated with higher tumour recurrence rate^(3,4).

Simultaneous surgical approach for parotid tumours has not been extensively discussed in International Literature. Nevertheless, some authors stated that simultaneous parotidectomy for bilateral benign parotid glands tumours should be avoided to prevent possible bilateral facial nerve palsy⁽⁵⁾.

Our patients did not notably refer major post-operative surgical complication except for a slight transient right marginalis nerve palsy for case 1 patient which completely recovered 3 weeks post-operatively.

CONCLUSIONS

In the occurrence of a bilateral pre-auricular swelling a complete clinical and instrumental evaluation should be mandatory. The most associated systemic conditions to bilateral pre-auricular swellings appear either haematological or immunological. Thus, a complete evaluation of the patient should ever be performed in such cases.

Over all neoplastic bilateral parotid swellings, Warthin's tumor is indeed the most frequent histological type, whereas pleomorphic adenoma remains the most common unilateral occurrence.

Pleomorphic adenoma is referred as a benign locally invasive pseudo-capsulated slowly-growing tumor. It is supposed to have pseudopodia infiltrating the surrounding glandular tissue that might increase the risk of recurrence in case of inappropriate surgical treatment. Otherwise, some authors reported an overlap of multifocal parotid tumors incidence possibly due to this histological features.

Superficial conservative parotidectomy is definitely the treatment of choice according to the latest studies showing a decreased recurrence rate with subsequent minimized risk of malignant transformation of residual tumor. In these cases the patients underwent simultaneous bilateral conservative superficial parotidectomy and the post-operative course was fairly uncomplicated.

Thus, we would suggest simultaneous bilateral superficial parotidectomy as the most indicated surgical approach, particularly in healthy patients with assured clinical and

cytological diagnosis and without evidence of any other systemic disease.

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