

Volume 4 Issue 2 February 2020

# Metachronous Bilateral Pleomorphic Adenoma of the Parotid Gland: Case of Rarity

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DOI: 10.31080/ASDS.2020.04.0769

Received: January 17, 2020 Published: January 28, 2020 © All rights are reserved by Nitin Ghuge., *et al* 

## Abstract

Bilateral pleomorphic adenomas of parotid gland are extremely rare and are reported in 37 cases till date. Its rarity contributes to its poorly understood etiology, pathogenesis and behaviour. Diagnosis based on imaging and microscopic examination helps in early diagnosis. Surgical excision is the gold standard treatment of pleomorphic adenoma (PA) preserving the facial nerve. We present a rare case of bilateral metachronous PA of parotid gland in a 65-year-old woman.

Keywords: Bilateral Tumour; Parotid Gland Tumors; Pleomorphic Adenoma; Parotidectomy

## Abbreviations

PA: Pleomorphic Adenoma; FNAC: Fine Needle Aspiration Cytology; MRI: Agnetic Resonance Image.

## Introduction

Salivary gland tumors account for about 3% of all of the head and neck tumors of which 70% - 80% occurs in major salivary glands [1] Parotid gland involvement is the most common with pleomorphic adenoma (PA) being the most frequently occurring tumor. Pleomorphic adenomas usually presents as solitary, unilateral, slow growing asymptomatic nodules. Bilateral tumors may be seen in 0.6 - 3.5% cases. Depending on the chronology of the tumor they may occur synchronously or metachronously. Due to the rarity of bilateral pleomorphic adenomas very little is known about its etiology and development [2]. Here we report a rare case of metachronous bilateral PA of the parotid gland.

#### **Case Report**

A 65-year-old female reported to the Department of oral and maxillofacial Surgery, Sawangi (M) with a complain of swelling on right angle of mandible. Patient gave history of asymptomatic enlarging swelling on right side since 6-year for which she underwent FNAC in 2011 r/s/o "pleomorphic adenoma" of right parotid gland. Patient did not undergo any intervention and reported back in 2017 for the same complain. Patient also gave history of small asymptomatic swelling on left side since 1 year. On examination, a well-circumscribed lobulated swelling of about 6 x 4 cm, firm, mobile and non-tender in nature was noted on right side with ear lobe elevation. There was no history of bleeding or pus discharge or nerve paraesthesia. A similar swelling of size 2 x 1 cm seen in left angle region. There was no evidence of other neck masses or lymphadenopathy. (Figure 1) FNAC was performed using a 23-gauge needle over swelling on both sides that showed similar

cytomorphology [3]. Smear showed sheets cuboidal polygonal epithelial cell, at places entrapped and surrounded by fibro myxoid material. The cells carry uniform nuclei, at places imparting honeycomb looks. These cell sheets also contain myoepithelial cells. Background shows isolated fibro myxoid flaks, bare nuclei and few hyalinised flaks with satellite nuclei. All these findings were suggestive of bilateral pleomorphic adenoma (Figure 2). Radiographic examination was done on magnetic resonance image (MRI) examination that showed e/o ill-defined large lobulated mass lesion in the right parotid gland predominantly in right superficial lobe showing the extension into the deep lobe. The mass is showing intermediate signals on T1W1 and hyper intense on T2W1 and vivid contrast enhancement. The mass is measuring approx. 6.9 x 5.78 x 5.6 cms. Neurovascular bundles to be displaced posterolateral. Similar lesion was approx. size 1.8 1.7 x 1.6cms is seen in left parotid gland predominantly in the superficial gland with extension into the deep lobe with similar enhancement pattern (Figure 3 and 4). After the final diagnosis, patient was prepared for surgical removal of bilateral tumors. After obtaining the fitness of the patient, GA was induced and bilateral tumors were opened by preauricular incision. Facial nerve identification was performed by retrograde method on right and anterograde method on left side. Preserving the facial nerve on both sides, complete functional parotidectomy of right side and partial superficial parotidectomy of left was performed. Closure was then done in 2 layers. Post-operative facial nerve function was observed and was found normal on both sides. Patient is on follow up for 2 years and no complications or recurrence is seen.



**Figure 2:** MRI aspects of the tumors in the parotid gland. Both the lesions are homogeneous and presented well-defined limits, contrasting with the neighboring tissues.



Figure 3: Intra operative Pic showing Right and Left Preserving all branches of facial Nerve.



Figure 1: Clinical aspects of the bilateral metachronous pleomorfic adenoma of the parotid glands.



Figure 4: Right and left resected tumour mass.

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## Discussion

Pleomorphic adenomas are most common parotid gland tumors accounting for about 70 - 80% of all parotid tumors. PAs are usually unilateral, slow growing painless tumors. Seifert and Donath [4] in 1996 classified these tumors as unilateral and bilateral, and according to its chronology - Synchronous or metachronous. Metachronous salivary gland tumors can be of 3 types - the bilateral tumors, the unilateral multifocal ones and combination of bilateral with unilateral multifocal ones. Bilateral tumors are rare and only 37 cases have been reported in literature till date of which 21.6% were metachronous tumors. We report one such rare case of bilateral parotid gland in this article which had an initial presentation of unilateral tumors of right side and later showed similar swelling on left side after 6 years. Similar case was reported by Silva., et al. in 2006. The etiology and development of bilateral tumors is not well known, however head and neck radiation, ill-defined environmental factors and genetic susceptibility have been suggested. chromosomal translocation of t (3;12) (p21; q15) have been demonstrated by Ahn., et al [5].

Histologically, no difference between unilateral and bilateral tumor were reported. Treatment of choice for pleomorphic adenoma is total or superficial parotidectomy preserving the facial nerve with a recurrence rate of 2 - 5%. Malignant transformation is rare and only 5% of cases have been reported [6]. In the present case total parotidectomy of right side and superficial parotidectomy of left side was performed with preservation of bilateral facial nerve. No reoccurrence have been reported on follow up of 2 years.

#### Conclusions

Pleomorphic adenomas though are benign tumors, should be diagnosed at early stage and must undergo surgical removal. Metachronous bilateral tumor may be missed at first presentation and therefore careful evaluation of imaging is mandatory to detect early and unsuspected lesions.

#### **Conflict of Interest**

None.

#### Acknowledgements

None.

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