

**NEHA VYAS\***, **SACHIN DALAL\*\***, **SAURABH JAIN\*\*\***, **MEGHA VYAS\*\*\*\***

**ABSTRACT**

Pleomorphic adenoma, the most common salivary gland tumour, accounts for a majority of parotid gland neoplasms. Untreated pleomorphic adenoma can gradually enlarge in size and weigh several kilograms. Surgery of pleomorphic adenoma presents an increased risk of facial nerve injury and a considerable recurrence rate. We report a case of pleomorphic adenoma in a 50 years-old female who complained of a large growth on the left side of the face, which enlarged gradually over a period of over 5 years. The excised specimen was 8 cm x10 cm x 12 cm in dimensions and 1.89 kg in weight.

**Keywords:** Pleomorphic Adenoma, Facial Nerve Damage, Parotid Gland, Parotidectomy

**Received:** 01-01-2014; **Review Completed:** 15-02-2014; **Accepted:** 26-02-2014

**INTRODUCTION:**

Parotid gland tumors constitute 3% of all head and neck tumors, with the vast majority being benign. The most common is pleomorphic adenoma, which represents approximately 80% of all benign masses. Although pleomorphic adenoma is benign in nature, it has an unusual propensity for recurrence. Recurrent tumors may be malignant; this is observed in up to 17% of recurrent cases. Surgical resection is the treatment of choice, but the extent of resection varies from enucleation of the tumor to superficial or total parotidectomy. The superficial lobe of the parotid gland, located laterally from the facial nerve trunk and its branches, contains 80% of gland parenchyma. Consequently, the vast majority of parotid tumors are located in the superficial lobe. Therefore, superficial parotidectomy is advised in the surgical treatment of pleomorphic adenoma. Paralysis of the facial nerve is one of the most feared complications of surgery in the parotid region.

**CASE REPORT**

A 50 year old female patient came to the department of oral & maxillofacial surgery with chief complain of swelling in left side of face since 5 years. Swelling was asymptomatic & gradually increased

in size since 5 years. Clinically, there was a well defined 8x10 cm ovoid shape swelling present over left side of face region extending superioinferiorly from left zygomatic arch to about 1 cm below lower border of mandible & anteroposteriorly from midbody region to posterior border of mandible with normal colour of overlying skin, multilobular with loss of wrinkling of skin and visible veins.(Figure 1) Swelling was fixed to underlying structure and overlying skin, warm, nonfluctuant, noncompressible, nonreducible on palpation. Also, external examination revealed that eye movements and visual acuities were normal and no facial damage. On intraoral examination, no swelling, tenderness or pus discharge was found in relation to left stenson's duct region.



Figure 1 a: Frontal View

Figure 1 b: Lateral View

Figure 1: Photograph of Patient's extraoral profile oral cavity showing swelling in relation to left side of face.

\*Professor and Head of the Dept., \*\*Reader, \*\*\*Reader, \*\*\*\*Post Graduate Student

DEPARTMENT OF ORAL AND MAXILLOFACIAL SURGERY AHMEDABAD DENTAL COLLEGE & HOSPITAL GANDHINAGAR, GUJARAT.

ADDRESS FOR AUTHOR CORROSPONDENCE:

Sialography of left parotid gland was done in filling and evacuation phases with lateral oblique radiographs. The filling phase showed the ductal filling with dye and a ball in hand appearance suggesting benign tumour of parotid gland.

Ultrasonography of local parts left cheek showing mixed echogenic area in subcutaneous fat plane with calcification suggesting malignant mass.

MRI report suggested well defined large heterogenous lesion seen arising from superior lobe of left parotid gland. Lesion on T1W1 appeared Figure 3 :Panoramic Radiograph of patient revealed an unusual large lesion invading large maxillary sinus.hyperintense and on T2W1 appeared hyperintense which on post contrast study showed variable enhancement. Lesion was not extending into parapharyngeal space and also neck.



Figure 2: CT image and MRI image.

FNAC was done from the swelling associated with left parotid gland, suggesting pleomorphic adenoma with acute inflammation.

From history, clinical, histological and radiographic examinations, a provisional diagnosis of pleomorphic adenoma of left parotid gland was made. It was decided to excise the superior lobe of left parotid gland with tumor mass under general anesthesia. Modified Blair Incision was given on left preauricular region, platysma muscle and superficial musculoaponeurotic layer was dissected, blunt dissection was done over parotid fascia up to anterior border of tumor (Figure 3 a), retrograde approach was done to identify and

preserve peripheral nerve branches. Stenson's duct on left side was ligated, (Figure 3 b). Nerves were separated from underlying parotid gland tissue, tumor was separated from the facial nerve and the masseter muscle, and tumor mass was removed (Figure 3 c); The flap was closed in three layers, subcutaneous tissue and platysma with 4-0 vicryl and skin with 4-0 ethilon. (Figure 3 d)



Figure 3 a : Exposed tumour mass

Figure 3 b: Highlighting preserved peripheral branches of facial nerve



Figure 3 c: Excised tumour mass

Figure 3 d: Skin closure

Figure 3: Operative Procedure

Enucleating biopsy of the tumor mass was diagnosed histologically (Figure 4) as pleomorphic adenoma with a well capsulated (fibrous) highly cellular tumor mass consisting of epithelial and myoepithelial cells. Epithelial cells were in form of islands and sheets. At some places ducts were also seen. Myoepithelial cells were rounded with eosinophilic cytoplasm. Accumulations of mucoid material between the tumour cells gave a myxomatous background. Chondroid area, fat and osteoid were also seen. The healing was uneventful without swelling or other complications. Alternate

sutures were removed 1 week postoperatively and other sutures were removed on 10th day postoperatively. Follow up was done on every alternative week for a month and every month up to 1 and 1/2 years postoperatively, no complications or recurrence were noticed with normal facial nerve function. (Figure 5 a, b)

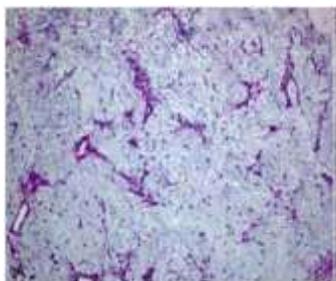


Figure 4 : Histological Section



Figure 5 a: follow up lateral view



Figure 5 b: Examination of post operative facial nerve function

## DISCUSSION

Pleomorphic adenoma are the most common salivary gland tumours, accounting for 45–74% of all salivary gland tumours.<sup>1</sup> They occur most frequently in the parotid gland<sup>1</sup> accounting for a majority of parotid masses<sup>2</sup> and 65% of all neoplasms arising in the parotid glands. They typically present in the lower pole and superficial (lateral) lobe of the parotid gland.<sup>1</sup> Though localization in the deep lobe is rare, approximately 10% of all parotid pleomorphic adenoma are thought to originate in the deep lobe of the gland<sup>4</sup> and can expand intraorally into the para pharyngeal space.<sup>1</sup> The submandibular gland and minor salivary glands, particularly minor salivary glands

of the palate, buccal mucosa and upper lip are frequently affected sites as well.<sup>1</sup> Pleomorphic adenoma usually present as slow growing asymptomatic discrete swellings, which gradually increase in size.<sup>1</sup> The vast majority of these tumours are 2–6 cm in size when resected.<sup>2</sup> Large tumours often form a single but irregularly nodular mass which stretches the overlying skin or mucosa.<sup>1</sup> The weight of the tumour can vary from several grams to more than 8 kg<sup>5</sup> and the weight appears to increase with the duration of the tumour. The weight of the giant tumours reviewed by Schultz-Coulon varied between 1–26.5 kg.<sup>6</sup> In our case the resected tumour was 8cm x 10 cm x 12 cm in dimensions and the weight of the excised specimen was 1.89 kg. In a review of 31 giant pleomorphic adenomas occurring in the parotid gland over a period of 140 years by Schultz-Coulon in 1989, most occurred in females (64.5%) and only 35.5% occurred in males<sup>6</sup> which is consistent with the gender variation of other salivary gland tumours (except Warthins tumour) and particularly with that of pleomorphic adenoma.<sup>1</sup> The age of first tumour manifestation varied between 20 and 40 years. However, paediatric patients could also be affected<sup>7</sup>. Malignant changes can occur in pleomorphic adenoma and include three distinct pathologic entities: Carcinoma arising in pleomorphic adenoma, carcinosarcoma and benign metastasizing pleomorphic adenoma.<sup>1</sup> The clinical characteristics of malignant transformation have been reported in the literature as (1) a long history of pleomorphic adenoma, (2) advanced age, (3) location in a major salivary gland and (4) history of rapid growth associated with pain or ulceration.<sup>12</sup> Development of secondary carcinoma in pleomorphic adenoma in preexisting pleomorphic adenoma is 3–4% according to the literature. Malignant changes were found in only 10% (n ¼ 3) of the giant pleomorphic adenoma cases reviewed

by Schultz-Coulon. 6 The incidence of malignancy frequently shows a correlation between the length of the history of pleomorphic adenoma and the development of a carcinoma. 8 It has been reported that the risk of development of malignancy is only about 1.5% up to 5 years but increases to 9.5% after more than 15 years. 9 However, in other series, 45% of carcinomas arising in pleomorphic adenoma were present for less than 3 years. In the series reported by Spiro et al, some tumours had been present for 2 years or less. 11 The usual presenting symptom of carcinoma arising in pleomorphic adenoma is a slowly growing mass which has been present for a long time, with the average duration of 23 years. 10 Generally, malignant transformation can be suspected with a sudden change in growth and local signs of malignancy including pain, ulceration, spontaneous bleeding, and superficial and deep tissue invasion. 10 Furthermore, multiple recurrences and the size of the tumour may play a role in the malignant transformation of pleomorphic adenoma. Our patient had all the above clinical characteristics of malignant transformation, except a history of rapid growth associated with pain. However, histopathology showed no evidence of malignant changes. In conclusion, paralysis of the facial nerve is one of the most feared complications of surgery in the parotid region. Surgeons with little experience prefer more conservative local surgery. But damage to the facial nerve and risk of recurrent

pleomorphic adenoma will just increase in these situations.

## CONCLUSION

Pleomorphic adenoma of the parotid provides a therapeutic dilemma. While this tumour is benign, recurrence can occur up to 30 years post-treatment. Additionally the risk of malignant degeneration has been established. Preoperative evaluation with imaging, or possibly fine needle aspiration, helps in treatment planning for the adequate resection of this tumour. Recurrences, when they happen, are difficult to treat, with increased risk of facial nerve injury and development of subsequent recurrences throughout the patient's lifetime. Often these patients require multiple procedures, with possible radiation therapy for tumor control. Since ninety percent of pleomorphic adenomas occur in the superficial lobe, lateral to the facial nerve, superficial parotidectomy with facial nerve dissection and preservation is the treatment of choice for pleomorphic adenoma. Untreated pleomorphic adenoma can enlarge gradually up to several kilograms in weight. These giant pleomorphic adenoma are more common in females and may enlarge over a period of several decades. Some of these long standing tumours show malignant changes. Therefore, early diagnosis and treatment of pleomorphic adenoma is essential.

## REFERENCES

1. Ellis GL, Auclair PL. Atlas of tumor pathology. Tumors of the salivary glands. Washington, DC: Armed Forces Institute of Pathology; 1995. pp. 39–41.
2. Buenting JE, Smith TL, Holmes DK. Giant pleomorphic adenoma of the parotid gland: case report and review of the literature. *Ear Nose Throat J* 1998;77(8):643, 637–8, 640.
3. Speight PM, Barrett AW. Salivary gland tumours. *Oral Dis* 2002;8(5):229–40.
4. Morita N, Miyata K, Sakamoto T, Wada T. Pleomorphic adenoma in the parapharyngeal space: report of three cases. *J Oral Maxillofac Surg* 1995;53(5):605–10.
5. Guerriere CN, Goff JJ, Cummings GH, Auber

- AE. An unusually large, solid tumor of the parotid gland. *Ann Plast Surg* 1999;43(5):529–32.
6. Schultz-Coulon HJ. Pleomorphic giant adenomas of the parotid gland. *Laryngorhinootologie* 1989;68(8): 445–9.
7. Uslu SS, Inal E, Ataoglu O, Sezer C. Pleomorphic adenoma of an unusual size in the deep lobe of the parotid gland. *Int J Pediatr Otorhinolaryngol* 1995;33(2):163–9.
8. Mizui T, Ishimaru J-I, Miyamoto K, Toida M. Malignant transformation of a gigantic pleomorphic adenoma of the submandibular gland: a case report. *J Oral Maxillofac Surg* 2000;58:1422–4.
9. Thackray AC, Lucas RB. Atlas of Tumor Pathology. Tumors of the major salivary glands. Washington DC: Armed Forces Institute of Pathology; 1974. p. 37.
10. Boneu F, Gonzalez-Lagunas J, Huguet P, Bassas C. Massive malignant pleomorphic adenoma of the palate. *J Oral Maxillofac Surg* 1998;56:91–6.
11. Spiro RH, Huvos AG, Strong EW. Malignant mixed tumor of salivary origin: a clinicopathologic study of 146 cases. *Cancer* 1977;39(2):388–96.
12. Yamamoto Y. Clinical signs and histology of carcinoma in pleomorphic adenoma. *Otologia* 1994;87:1320–4.