# GIANT PLEOMORPHIC ADENOMA OF SUBMANDIBULAR GLAND : A RARE CASE REPORT

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**ABSTRACT:**

Pleomorphic adenomas of the submandibular glands are exceedingly rare tumors. Patients usually present with a painless and mobile mass without any other associated symptom. Recurrences are rare with complete en bloc excision of the tumor and the submandibular gland. Except for the rare cases of malignant transformation, the prognosis is excellent. In this paper, we highlight a case of giant submandibular pleomorphic adenoma that was managed with surgical excision.

## INTRODUCTION:

Salivary gland tumors are rare and make up to 3% of head and neck tumors.[1]Pleomorphic adenoma (PA) is the most common tumor of the benign salivary gland neoplasms. Approximately 90% of the benign neoplasm of the major salivary gland is associated with the parotid gland. The submandibular gland is the second most common site of PA. Pleomorphic adenoma of the submandibular and sublingual gland is quite uncommon and comprises rest (8–10%) of thegroup.[1]It is also the most frequent benign tumor arising in submandibular gland. PAs are benign tumors with a well-documented transformation to malignancy (carcinoma ex pleomorphic adenoma). It is estimated that up to 25% of untreated PAs undergo malignant transformation.[2] Therefore, early definitive treatment is strongly recommended.This case report is being highlighted for its rarity, unusual presentation and initial diagnostic dilemma as thyroid swelling.

## CASE REPORT:

A 63-year-old female presented with lobulated swelling in front of neck arising from left side of neck and left submandibular region for last 16years. It was insidious in onset, slow growing and painless. It was not associated with swallowing and breathing difficulty, change in voice, fever or weight loss. There was no family history. She attained menopause 10years back.

On Local Examination, lobulated mass of size 30cm×25 cmx25cmwas located in the left side of neck and submandibular region and hanging in front of anterior chest wallupto upper two-third of sternum.It was firm,non-tender and freely mobilearound its base. Prominent veins were seen over the swelling.

No local rise of temperature or pulsation felt.It was not translucent and not adherent to underlying skin. There was no restriction of neck movement.

On General Examination, she was conscious. No palor seen. Her vital signs were as follows- BP: 130/80 mmHg. Pulse: 88 beats/min. Temperature: 98.7◦F. Blood investigations showed Hemoglobin= 11.3 g/dL and other parameters were normal.

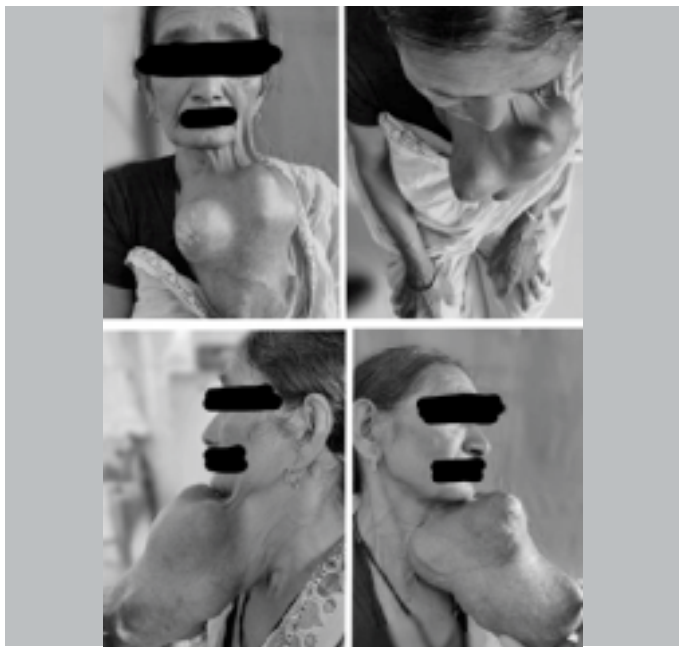

*Fig 1: preoperative pics*

FNAC reported clusters of ductal epithelial cells and myoepithelial cells in background of chondromyxoid stroma suggesting Pleomorphic Adenoma. Chest X-Ray showed tracheal deviation to right side. CT Neck reported exophytic heterogenous lesion of size 15cmx13cm arising from left lobe of thyroid.

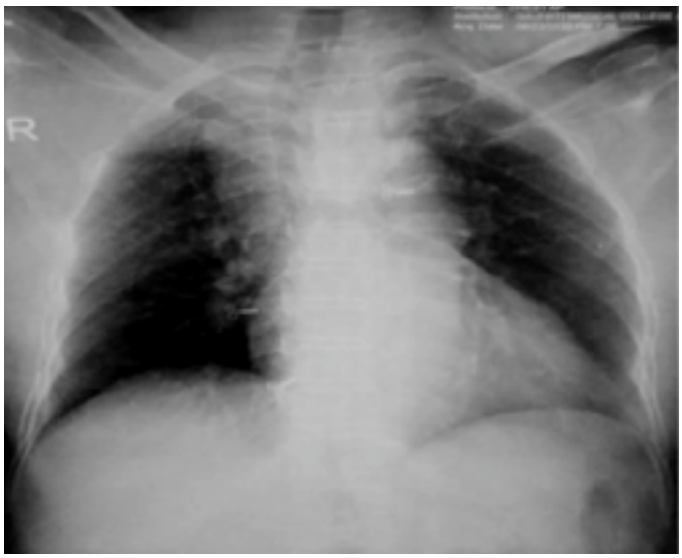

*Fig 2 : CXR showing right sided tracheal deviation*

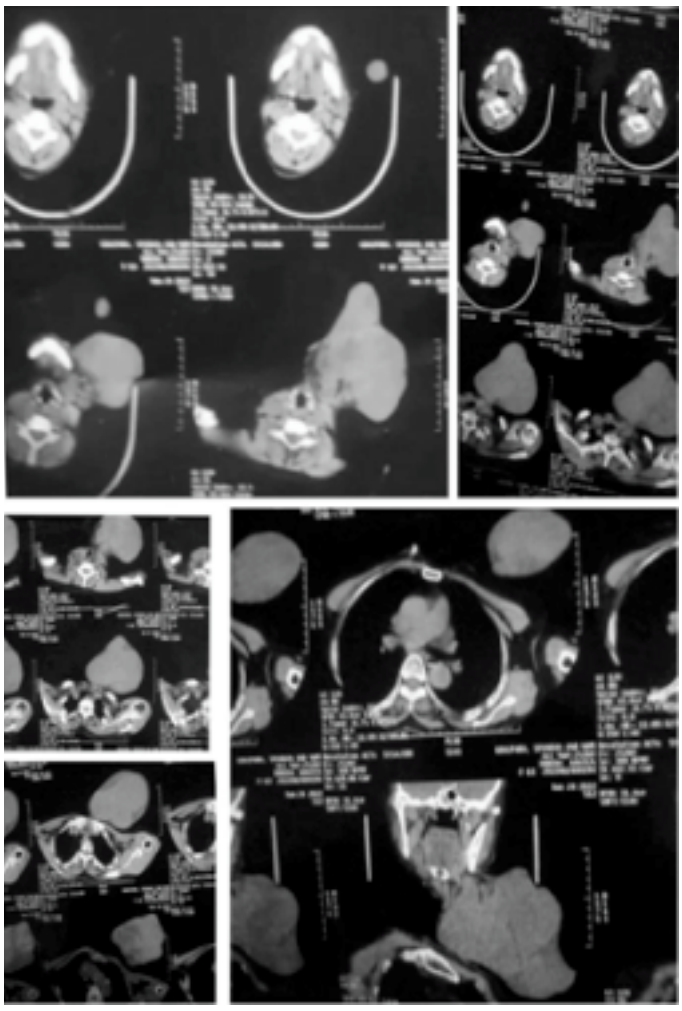

*Fig 3: CT neck*

She was planned for excision of mass under general anesthesia. With circumferential incision around the base of the mass at its pedicle, the mass was excised in toto after ligating large vessels. Anatomy of submandibular region and neck was distorted due to the mass. Immediate post op, there was no deviation of angle of mouth.

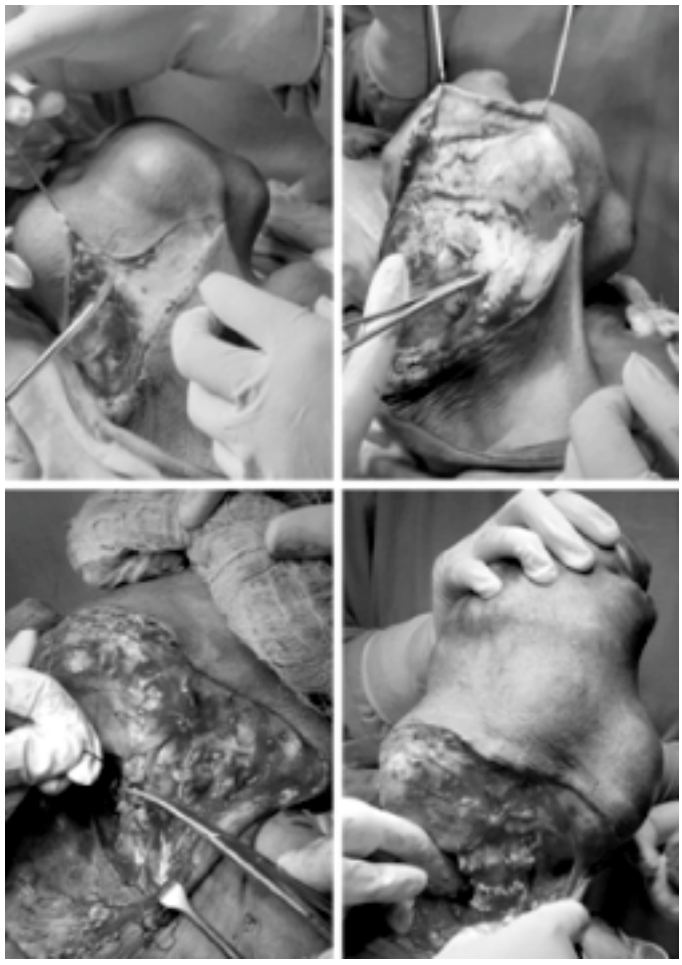

*Fig 4: Intraoperative pics*

*Fig 5: Specimen weighing 2.2kg*

The patient is under follow up for last 6months and is doing well. Post op HPE was confirmatory of Pleomorphic Adenoma.

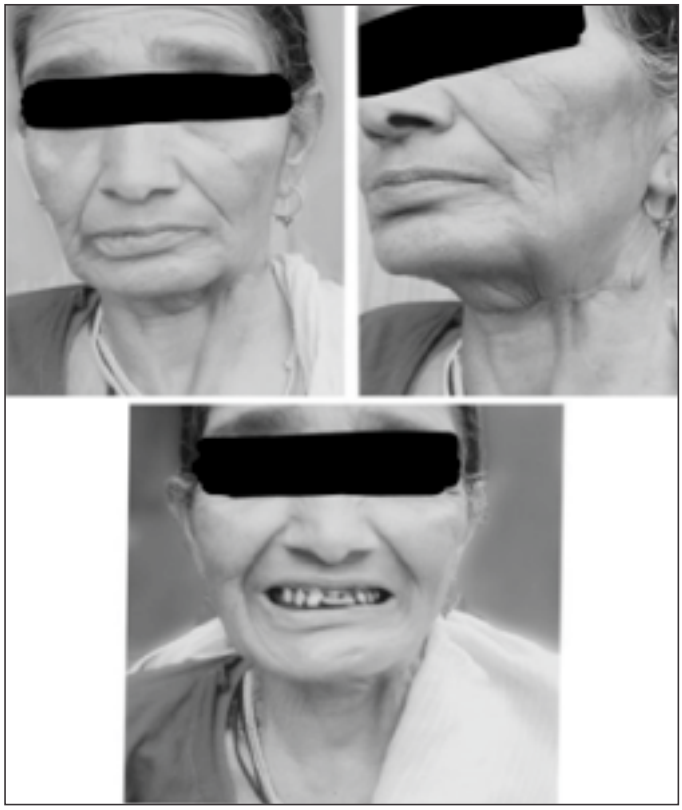

*Fig 6: Postoperative pics*

## DISCUSSION:

Pleomorphic Adenoma is an epithelial tumor of complex morphology, comprising epithelial and myoepithelial elements intermingled with mucoid, myxoid, or chondroid tissue arranged in a variety of patterns and embedded in a mucopolysaccharide stroma.[1]It is found mostly in parotid gland in middle-aged women. The most frequent neoplasms of submandibular glands are: PA (36%), adenoid cystic carcinoma (25%), mucoepidermoid carcinoma (12%) and malignant mixed tumor (10%).[3]

*Becerril-Ramírez et al.* in their 10 year study found out of 22 cases of submandibular gland neoplasms,19 cases (86%) were benign and 3 cases (14%) were malignant. PA accounted for 18 out of 19 cases. The mean age of PA was 39.8 years with female to male ratio of 3.5:1.[3]

*de Oliveira et al.* found that in Brazilian population, salivary gland tumors affected females more often, with a male: female ratio of 1:1.6 in benign tumors and 1:1.5 in malignant tumors. The mean age for benign tumors was 43 years and for malignant tumors was 55 years.[4]

*Adeyemo et al*. reviewed submandibular salivary gland tumors from 1990 to 2006. Out of 36 patients,17 cases were benign and 19 cases were malignant. PA (36.1%) was the most frequent tumor, followed by adenoid cystic carcinoma (11.1%), anaplastic carcinoma (11.1%) and malignant lymphoma (11.1%). Progressive painless swelling (80.6%) was the most common presentation and cases which presented with painful mass (11.1%) and ulceration (8.3%) were malignant.[5]

*Munir and Bradley* reviewed 32 cases of submandibular gland PA from 1988 to 2004. Out of 32 cases,22(69%) cases were female and mean age was 54 years. All patients presented with clinically visible and palpable mass of submandibular fossa among which 84% of cases were asymptomatic and 16% presented with pain.[6]

*Rapidis et al.* analyzed clinicopathologic features of 23 patients with submandibular gland tumors and found 9 were benign and 14 were malignant.PA wasmost frequent benign tumor.[7]

*Alves et al.* reviewed clinicopathological and immunohistochemical features of 60 cases of PA in Brazil and found that PA occurred commonly between 3rd and 5th decades of life and 37/60 (62%) of them were women. Tumor sizes varied from 1 to 10 cm. Only one patient experienced local recurrence, 3 years after treatment.[8]

FNAC provides evidence forpre-operative diagnosis that is 70-80% accurate and also helps to differentiate between tumor and inflammatory conditions or enlarged lymph nodes.[11]An incisional biopsy can be taken initially if the lesion is of large size.The final pathologic diagnosis is always established based on findings from surgical excision.CT scan or magnetic resonance imaging (MRI) are the gold standard for lesion arising from major or minor salivary glands.[11]

The treatment of choice for submandibular gland PA is total submandibular gland excision in toto along with tumor.[9] Recurrence rate of submandibular gland tumors are less than parotid gland since entire gland is excised. Incomplete removal of glandular tissue can lead to definitive recurrence. Injury to marginal mandibular nerve is the most common complication. Temporary paralysis may resolve spontaneously within a period of 3 months.[10]

## CONCLUSION:

Although there are few studies that have been conducted exclusively on submandibular gland, the clinical findings in this present case are in agreement with findings of the existing studies with PA being most common benign tumor affecting submandibular gland, occurring commonly in female between the 3rd and 5th decade of life and presenting as slow growing asymptomatic swelling. However, in the present case, PA was large and was confused with thyroid swelling initially.

## CONSENT:

Written and informed consent was obtained from the patient regarding the use of the patient's picture and the reports of the investigations that were conducted.

## CONFLICT OF INTEREST:

The authors declare no conflicts of interest during the course of the making of this paper.

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