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Giant Pleomorphic Adenoma of the Parotid Gland: A Rare Case Report And Literature Review

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Abstract: *Pleomorphic adenoma is the commonest type of all benign and malignant salivary gland neoplasm majority involving the parotid gland. It is a slow growing and benign tumour which can reach an enourmous size and may turn malignant if left untreated. Although uncommon, most cases of giant pleomorphic adenoma were reported before 1980's, with few publications recently. This paper presents a rare case of giant pleomorphic adenoma in the parotid gland measuring 3.1 kg by weight in a 78 year old female with a history of 10 years. Pre operative assessment was done by thorough clinical examination, histological and radiological findings. Treatment comprises Total conservative parotidectomy by the Transcervical Transparotid approach with post operative excellent aesthetic and functional results.*

Keywords: *giant pleomorphic adenoma, parotid gland, parotidectomy*

I. Introduction

Salivary gland neoplasms are very rare comprising 1- 4 % [1] of all neoplasms and 3 – 5 % [2,3,4] of overall head and neck neoplasms . About 70% of all salivary gland tumours arise in the parotid gland out of which 80% are benign. Pleomorphic adenomas, also known as benign mixed tumour represents 45-74% of all salivary gland tumours, majority of which about 65% occurring in the parotid gland[(5,6,7)]. There is a female sex preponderance and a peak age range of 5 th to 6 th decade for all pleomorphic adenomas.[(5,8,9)] Most cases of giant pleomorphic adenoma were seen before the 1980's in English literature. where mean tumour weight 7.81 Kg and mean age 56.2 years was reported.[(10,11)]. Malignant change has been reported to occur in 2-7% of cases.[(12)]. Diagnosis is usually by a combination of clinical history, Fine needle aspiration biopsy for cytology and histological confirmation of excised tumour. Computerized Tomography (CT) scan, Magnetic Resonance Imaging (MRI) and Ultrasonography Scan may be necessary to assess extent of gland involvement. Treatment is usually by superficial parotidectomy with facial nerve preservation although some authors have treated by total parotidectomy with facial nerve preservation.[(13).] This paper presents a rare case of giant pleomorphic adenoma occurring in the parotid gland, alongwith prompt and meticulous surgical management to avoid recurrence and malignant transformation. Finally to prevent the patient from social morbidity and provide with better aesthetic.

II. Case Report

Our patient, a 78 year old female came to the emergency department of gauhati medical college and hospital,with complaints of a huge left sided painless parotid swelling with bleeding from the surface . Inspite of the massive enlargement the patient and the family members were not much bothered about the size of the swelling, but more concerned for the bleeding. The swelling was insidious in onset,and has gradually progressed over the span of 10 years, with history of rapid growth in last 5 years. She developed bleeding from surface of mass since last 10 days . There was no history of fever, abnormal swelling in any other part of the body, or loss of weight. No h/o any chronic illness like hypertension, diabetes or asthma. Her general condition was fair and vitals were stable.

On clinical examination the swelling was giant, irregularly shaped, mobile, nontender , firm and cystic in consistency, multinodular with bosselated surface . Edges were clearly defined. Skin over the swelling was stretched and shiny with multiple venous engorgement. There was one bleeding point on anteroinferior surface which was ligated. Skin over the swelling was pinchable and there were no signs of regional lymphadenopathy or signs of facial nerve palsy.

It was extending vertically from left tragus upto left 2nd rib and attaching below in middle 1/3 rd of neck and horizontally extending from 6 cm away from left angle of mouth to the post auricular region till the hairline (for about 4cm). The swelling measures about 14.2 cm antero-posteriorly , 10.6 cm transversely and

16 cm vertically. Diagnosis was confirmed by a combination of fine needle aspiration biopsy for cytology , computerized Tomography (CT) scan and ultrasonography.

After obtaining the informed consent of the patient, total conservative parotidectomy was performed under general anesthesia by the transcervical transparotid approach. The peripheral nerve branches were identified and preserved following a retrograde approach. The postoperative course was uneventful.

Macroscopically, the excised mass measured 15 x18 x 10 cm and weighed 3.1 kg (Fig. 4).

Microscopically the tumor was composed of islands and strands of epithelial cells immersed in a hyaline stroma. Areas presenting spindle and plasmocytoid myoepithelial cells in a myxoid stroma were also abundant,with no evidence of malignant changes. The patient presented excellent aesthetic and functional results, without signs of facial nerve palsy or recurrence (Fig. 6).

III. Fig 1. Pre operative photos (intact facial nerve function):

Fig 2. CT Scan images

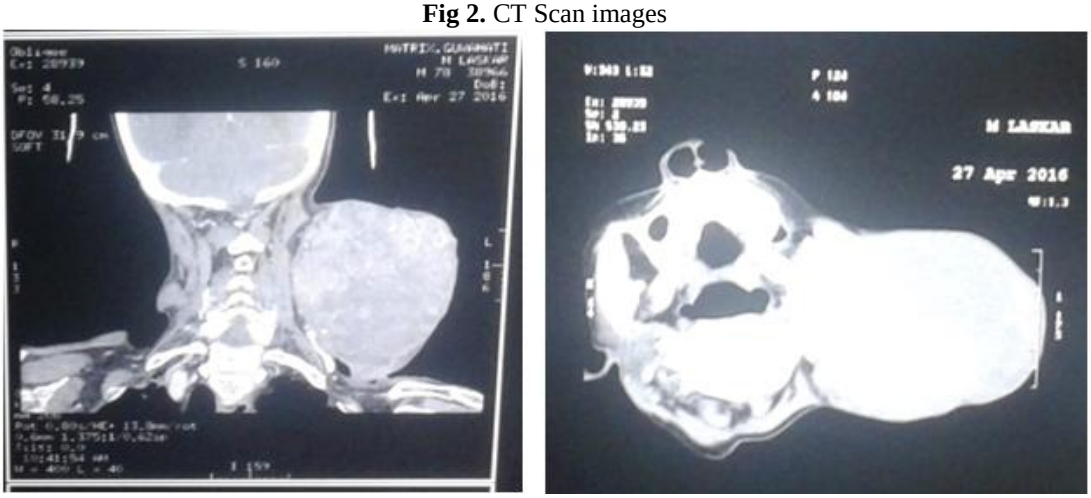
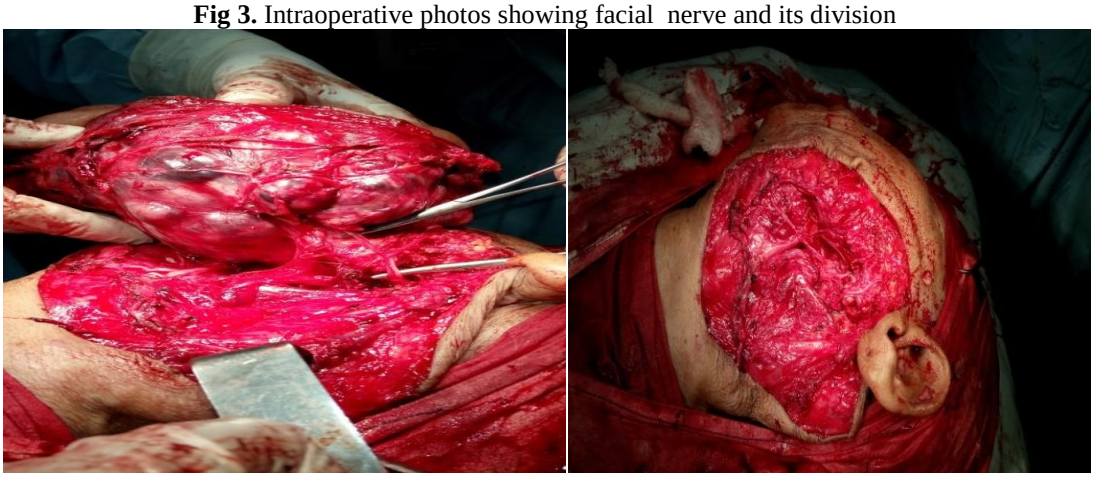
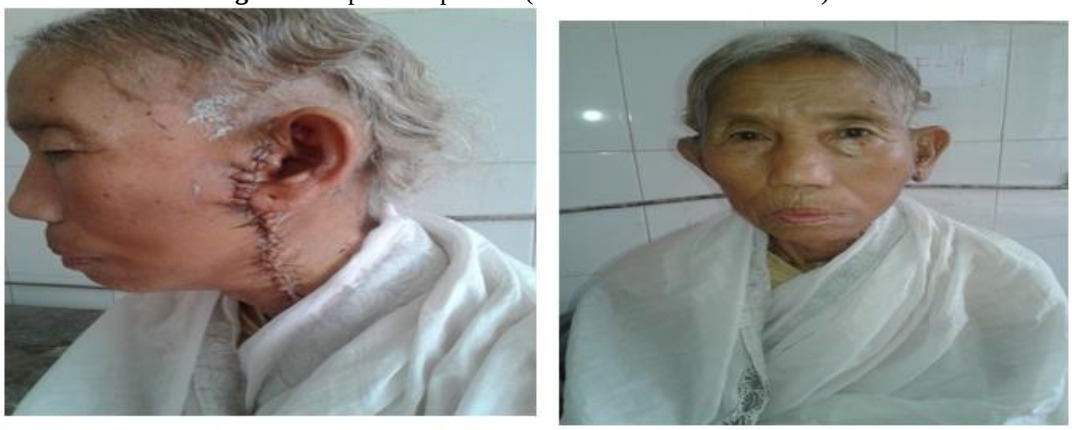


Fig 3. Intraoperative photos showing facial nerve and its division



4. Images showing excised specimens.

Fig 6. Post operative photos (Intact facial nerve function):



IV. DISCUSSION

Pleomorphic adenoma can be defined according to World Health Organization(1972) as a well defined tumour characterized by its pleomorphic or mixed appearance. There is intermixing of the epithelial component with myxoid, mucoid, and chondroid component [14] Inspite of several histological features due to different tissue elements and cellular components, it is generally considered to be a benign neoplasm.[15,16] Pleomorphic adenoma is known to be the most common salivary gland neoplasm. It occurs most commonly in the parotid gland. [17,18,19] .Classical parotid tumour present just below the lobule of the ear, above the angle of mandible [17,18] . In present case the site of occurrence is concurrent with the literature.(Fig1)

In 1863[19], Spence reported the first case of giant pleomorphic adenoma published in the English language literature , who described the treatment of a mixed tumour > 1 kg.

In 1956, Short and Pullar [20] published an English language review of huge pleomorphic adenomas and a case-report of an adenoma of 2.3kg.

In 1989, Schultz-Coulon[21] reviewed 31 cases of massive pleomorphic adenomas of the parotid gland and found a female preponderance (64.5%), with an age range from 20 to 40 years old, and a tumour weight between 1 to 27 kg.

Buenting described the 10 largest pleomorphic adenomas published in the medical literature. The author found a mean tumour weight of 7.8 kg; 9 out of 10 occurred in females, with a mean age of 56 years. The author reported 5th largest pleomorphic adenoma (6.85 kg).

In our case the patient was a female of 78 years with a history of 10 years and tumour weight3.1 kg which agrees with the literature reviews.

The incidence of malignant transformation in adenomas ranges from 1.9% to 23.3% [22]. The risk of malignancy increases with longer duration, post operative recurrence, elderly age group and location in a major salivary gland[23]. Some authors postulated that the risk of malignant transformation increases from 1.6% in tumours with less than 5 years of evolution, to 9.5% for those presenting for more than 15 years [24].

The classical history of carcinoma ex-pleomorphic adenoma is a slowly-growing mass for many years, with a recent rapid growth phase. This typical history of a case of a giant adenoma with malignant transformation was reported in 2005 by Honda[4] in a 72-year-old female with a long history for 20 years,and a rapid increase in the last 3 months.

In the review literature of Schultz-Coulon [21] , 3 of 31 cases of giant adenomas showed malignant transformation. In our case, although the patient presented all the characteristics for an increased risk of malignancy, clinically and histologically there was no such evidence.

However the period of evolution of growth into a giant tumor in our

patient was long similar to most literature reports.[25]

Two main metastatic variant, carcinoma ex pleomorphic adenoma and metastasizing benign mixed tumor are observed. [5]

Macroscopically the tumour show mainly irregular, multinodular lesion with bosselated surface with the complete or incomplete capsule like structure. The surface remain firm in consistency and the softness in nodule suggesting cystic degeneration of the lesion.[26] In our case report all the macroscopic features are in consistent with the literature reviewed [Figure 4].

The radiological findings are essential in diagnosis of pleomorphic adenoma. Mostly the benign tumors shows a hyperdense image on the C.T. scan of the lesion.[27] In our presented case also a hyperdense image is found within the structure of the parotid gland [Figure 2]

The treatment of choice for the pleomorphic adenoma of the superficial lobe of parotid gland is superficial parotidectomy with adequate excision of the capsule and preservation of the facial nerve.[28,29,30,31] In two recent cases of giant PA reported in the english literature, the resection of the tumors were performed with preservation of the facial nerve.[11] Our case was treated by total conservative parotidectomy because of the tumor size, with excellent aesthetic and functional results. Because of the presence of incomplete and thin capsule, the tumour buds like pseudopods may extend beyond the capsule and is responsible for the high recurrence.[30] A recurrence rate of up to 40% was noticed in treated cases of pleomorphic adenoma.[32] The recurrence is debatable and subjected to the treatment. The main casues for recurrence is believed to be the intra-operative tumour spillage and incomplete resection.[33]. However in our case no recurrence is noticed upto 2 years of follow up. Prognosis for pleomorphic adenoma is excellent with a cure rate of 95% which is concurrent with our case.[8]

V. Conclusion

Our above case report shows us that pleomorphic adenoma can occur as massive as the size of 15 x 18 x 10 cm and still be benign tumour even over the huge span of 10 years. However, an extensive histolopathological assessment of excised tumor is mandatory to rule out malignant transformation.

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