



Mark Laurence B. Barrios, MD
Neil Aldrine I. Peñaflo, MD

Department of Otolaryngology-Head and Neck Surgery
Bicol Medical Center

Giant Pleomorphic Adenoma in a 64-Year-Old Woman: A Case Report

ABSTRACT

Objective: To report a case of a giant pleomorphic adenoma in a 64-year-old Filipino woman, its management and surgical outcome.

Methods:

Design: Case Report
Setting: Tertiary Government Training Hospital
Patient: One

Results: A 64-year-old woman presented with a 50-year history of a slow growing, painless, left infra-auricular mass, not associated with facial weakness, xerostomia, or hearing loss. Computed tomography revealed a 14 x 15 x 19 cm large lobulated complex enhancing mass with calcifications and septations, with no enlarged lymph nodes identified in the neck. Fine needle aspiration cytomorphology was consistent with pleomorphic adenoma. The patient underwent superficial parotidectomy with facial nerve preservation. The facial nerve was identified using standard landmarks. Final histopathological findings were consistent with pleomorphic adenoma measuring 23.5 cm x 11.5 cm x 15 cm and weighing 2177 grams.

Conclusion: Pleomorphic adenoma can grow to a gigantic size if left untreated. It often presents as a chronic, slow growing and painless swelling. The approach to its diagnosis is mainly clinical and can be confirmed by fine needle aspiration biopsy and computed tomography scan. In our case, the standard landmarks for facial nerve identification were still reliable despite the size of the mass, producing good post-surgical outcomes.

Keywords: *giant pleomorphic adenoma; parotid gland; superficial parotidectomy*

Although uncommon, giant pleomorphic adenomas of the parotid gland have been reported.¹ To the best of our knowledge, a search of HERDIN Plus, the Western Pacific Region Index Medicus (WPRIM), the Directory of Open Access Journals (DOAJ), MEDLINE (PubMed and PubMed Central) using the search terms “pleomorphic adenoma,” “giant pleomorphic adenoma,” “parotid gland,” and “case report” showed no locally reported case of giant pleomorphic adenoma to date. We report one such case.

Correspondence: Dr. Neil Aldrine I. Peñaflo
Department of ENT - Head and Neck Surgery
Bicol Medical Center
Concepcion Pequeña, Naga City 4400
Philippines
Phone: +63 917 513 8823
Email: bmc2017ent@gmail.com

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CASE REPORT

A 64-year-old Filipina from Masbate noted onset of a 1 cm x 1 cm hard, nontender, movable mass on her left infra-auricular area with no associated signs and symptoms such as pain, xerostomia, facial weakness, dysphagia, trismus, or hearing loss, 50 years prior. There was gradual growth of the mass with doubling of size every eight years, still without associated symptoms. Thirty-four years prior to admission, the mass grew to approximately 6 cm x 6 cm in diameter. At this time, she consulted a local rural health unit in Burias where she was advised further work up and surgery. However, due to financial constraints, she was lost to follow up. Four months prior to admission, due to further increase in the size of the mass, now with associated neck discomfort, she consulted in our department. On physical examination, there was a prominent 23 cm x 12 cm x 10 cm hard, multilobulated, nontender, movable mass occupying the left preauricular area extending to the left lateral neck causing downward displacement of the lateral canthus. (Figure 1) There was limitation of neck mobility towards the ipsilateral side due to the mass effect of the tumor. There were no palpable neck lymph nodes, trismus or signs of facial nerve palsy. The patient was a previous smoker with a 3-pack year history, denied habitual alcoholic beverage intake, illicit drug use or betel nut use. The past medical history and family history were unremarkable.

Fine needle aspiration biopsy (FNAB) of the parotid gland revealed hypocellular smears showing cords and clusters of epithelial cells with intimately associated spindly myoepithelial cells enmeshed in chondromyxoid stroma, consistent with pleomorphic adenoma. A contrast enhanced facial and neck CT scan showed a large lobulated complex enhancing mass with calcifications and septations at the left side of the neck measuring 14 cm x 15 cm x 19 cm. (Figure 2) No focal masses were seen in the visualized right parotid and submandibular glands, but there were incidental findings of bilateral maxillary sinusitis, chronic mastoiditis left and multiple thyroid nodules.

The patient underwent left superficial parotidectomy with facial nerve preservation. (Figure 3) The excised tumor was 23.5 x 11.5 x 15.0 cm and weighed 2.18 kg. (Figure 4) On gross examination, the mass was grayish to tan, firm, well-circumscribed, multilobulated, with notable areas of infarction. (Figure 5) Final histopathologic findings were consistent with pleomorphic adenoma. (Figure 6) Throughout the postoperative period, the patient showed no signs of facial nerve injury or other acute post-surgical complications, and was discharged on the fourth hospital day. (Figure 7)



Figure 1. Preoperative photos showing a 21 cm x 11 cm x 9 cm hard nontender movable mass arising from the left infra-auricular area: **A.** anterior view, and **B.** lateral oblique view. Photos published with permission.

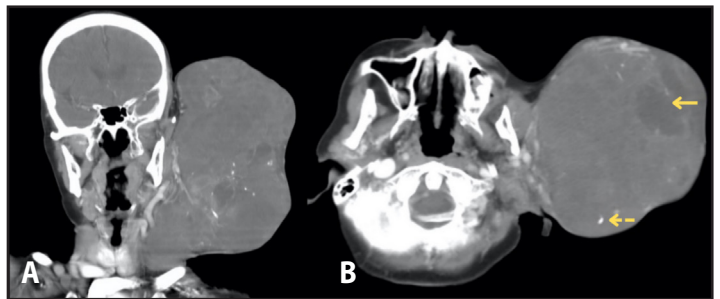


Figure 2. Facial computed tomography scans, **A.** coronal view, and **B.** axial view, reveal a 14 x 15 x 19 cm large lobulated complex enhancing mass with calcifications (dashed arrow) and septations (solid arrow), with no enlarged nodes identified on both sides of the neck.

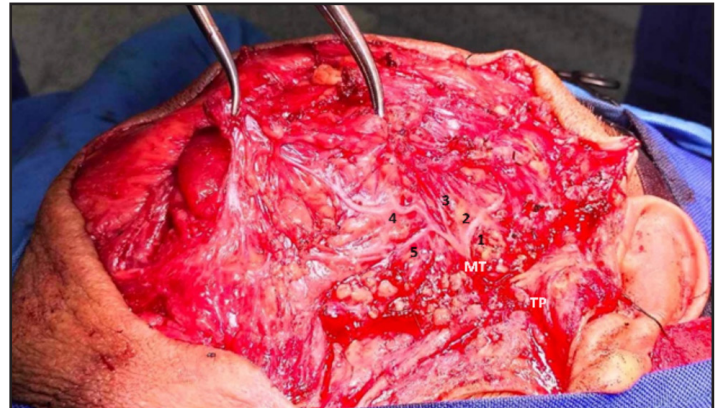


Figure 3. The facial nerve with all its terminal branches identified and preserved: TP, tragal pointer; MT, main trunk; 1, temporal branch; 2, zygomatic branch; 3, buccal branch; 4, marginal mandibular branch; and 5, cervical branch

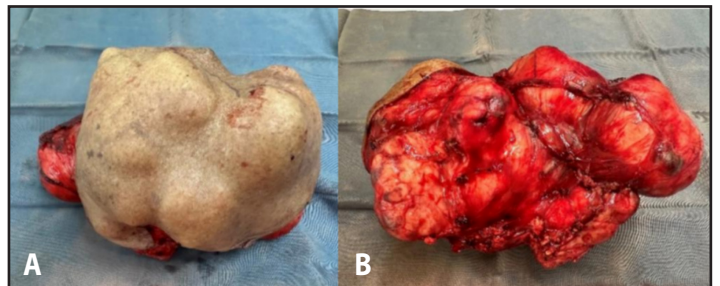


Figure 4. Gross surgical specimen measuring 23.5 x 11.5 x 15 cm and weighing: 2.177 kg; **A.** outer surface; and **B.** inner surface

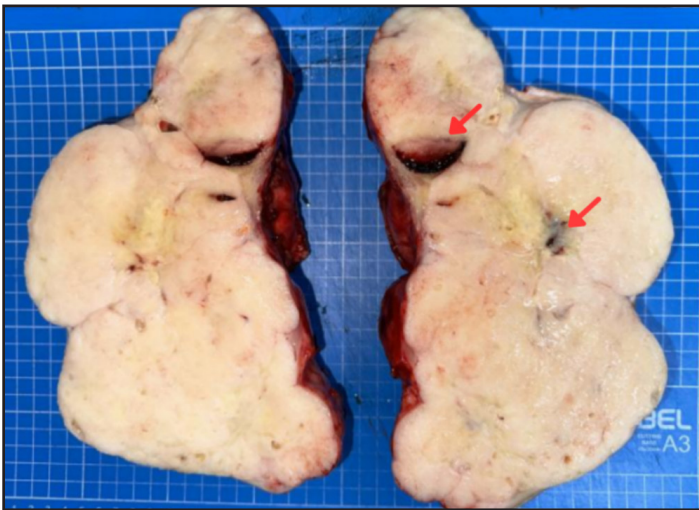


Figure 5. Cut surface showing firm, multilobulated, well-circumscribed grayish to whitish mass with areas of infarction (arrows)

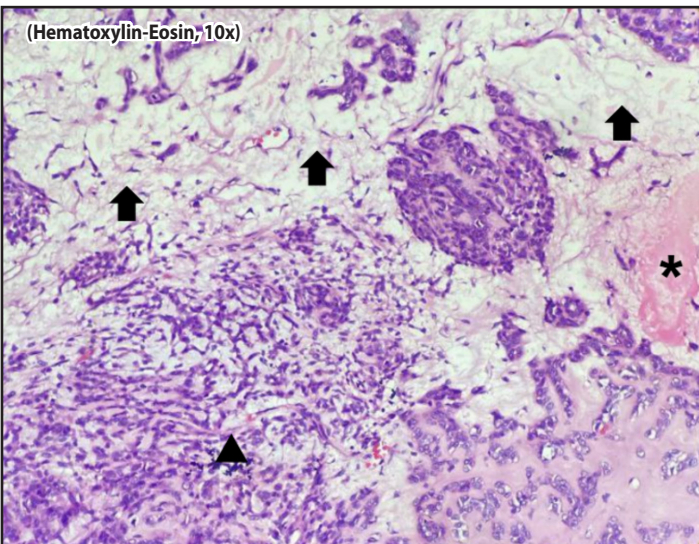


Figure 6. Histopathologic slide, hematoxylin-eosin stain, low-power view (10x magnification) showing focal cystic areas filled with mucin (arrows), focal calcified area (arrowhead), and a focal hemorrhagic/necrotic area (asterisk), consistent with pleomorphic adenoma



Figure 7. Postoperative facial nerve assessment at one month showing symmetric: **A.** eyebrow elevation; **B.** eye closure; and **C.** smiling. Photos published in full with permission.

DISCUSSION

Pleomorphic adenomas (PA) involve approximately 2 - 3.5 cases per 100,000 population, occur in all ages, are more common among females, and predominantly affect the superficial lobe of the parotid gland (84%), followed by the submandibular gland (8%) and minor salivary glands (6.5%).¹ They have a slow progressive growth pattern and if left untreated, may progress to a massive size.² There are several reasons for reaching enormous size.³ In our case, financial constraints and geographic distance were the main reasons for the delay. While there was limitation in neck movement, this functional limitation only became apparent in the latter years.

Although there are no specific physical criteria outlined in the literature to classify giant parotid PA, the earliest recorded case report dates back to 1863.³ Pleomorphic adenomas of the parotid gland are considered "giant" if they exceed 10 cm in widest diameter.² The largest tumor described was 35 cm and the heaviest weighed 7.3 kg.³ Our case exhibited typical characteristics of a giant pleomorphic adenoma with female sex predilection, slow rate of growth, late presentation and clinical dimension.

The etiology of PA is unknown, but the incidence may be increasing in the last 15-20 years in relation to supposed radiation exposure and an association with simian virus 40.¹ Whether these factors are truly associated with PA, our patient resides in a remote island in Masbate, with no known exposure to them.

Most guidelines recommend ultrasound as the initial imaging modality of choice for the assessment of palpable abnormalities of the salivary gland; however, CT was more useful in our case for assessment of extensive tumor extent.⁴ A contrast-enhanced facial CT scan was chosen as the diagnostic modality for preoperative diagnosis and surgical planning, due to the volume of the tumor, its availability in our institution and affordability for our patient. In addition to volumetric information, CT with contrast provides knowledge about vascularity of the tumor.⁴ Our CT findings leaned towards a benign entity hence, superficial parotidectomy was considered.

Intraoperative findings showed a clear surgical plane and nonadherence to adjacent structures. Identification of the facial nerve trunk was a concern pre-operatively with the possibility of distortion from its usual path due to the mass effect. The standard landmarks in identifying the facial nerve trunk were proven to be reliable, and the superficial parotidectomy with facial nerve preservation was performed with favorable postoperative outcomes.

The malignant potential of PA tumors is proportional to the time the lesion is in situ (1.5% in the first five years, 9.5% after 15 years).¹



Our patient carried the tumor for 50 years. Fortunately, the final histopathologic findings were still consistent with pleomorphic adenoma.

It has been proposed by Joson, *et al.* that parotid cases be concentrated in centers where surgeons have enough experience in handling the complex problems in the management of such tumors.⁵ Besides a clear understanding of parotid surgical anatomy, pre-operative preparedness, patient preparedness and surgical experience contribute to achieving a good surgical outcome.

In conclusion, pleomorphic adenoma can grow to a gigantic size if left untreated. It often presents as a chronic, slow growing, and painless swelling. The approach to its diagnosis is mainly clinical and can be confirmed by fine needle aspiration biopsy and computed tomography scan. In our case, the standard landmarks for facial nerve identification were still reliable despite the size of the mass, producing good post-surgical outcomes.

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