



Giant recurrent pleomorphic adenoma of the parotid gland with facial nerve palsy: A case report

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Abstract

Context: Pleomorphic adenoma is the most common benign salivary gland tumour, typically involving the parotid gland. Giant pleomorphic adenomas with prolonged progression and associated facial nerve palsy are rare and present management challenges.

Case presentation: We report the case of a 46-year-old male with a long-standing recurrent giant pleomorphic adenoma of the right parotid gland following previous excision, presenting with facial nerve palsy. The patient had undergone previous surgery approximately 12 years earlier, with recurrence reportedly occurring approximately four months later. Delayed presentation was attributed to financial constraints. The patient had total parotidectomy. Histopathologic examination showed pleomorphic adenoma without identified malignant transformation. Early postoperative recovery was satisfactory; however, longer-term assessment was limited because the patient was lost to follow-up after three months.

Conclusion: This case highlights the potential for neurological complications and extensive tumour growth in long-standing untreated pleomorphic adenoma. Early intervention and long-term follow-up improve overall outcome and quality of life.

Introduction

Pleomorphic adenoma is the most common benign tumor of the salivary glands. Approximately 75% of all salivary gland tumors occur in the parotid gland, about 85% of which are benign; of these, 80% are pleomorphic adenomas. Pleomorphic adenomas are slow-growing, painless mass, arise in the superficial lobe of the parotid gland but can arise from other major and minor salivary glands and rarely from the oral

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cavity, palate, neck, and nasal cavity². Histologically, it exhibits a combination of epithelial and myoepithelial elements embedded within a mesenchymal-like stroma, hence the term "pleomorphic".

A giant pleomorphic adenoma is defined as an unusually large tumour that exceeds 10cm in greatest dimension; exceptionally large tumours

may also weigh more than 1kg³. They are rare and often due to prolonged growth from delayed presentation, lack of access to health care, or patient neglect due to the painless nature of the mass. Giant recurrent pleomorphic adenoma may clinically mimic carcinoma ex pleomorphic adenoma, particularly when accompanied by fixation, rapid change, pain, ulceration or facial nerve dysfunction. Giant recurrent tumours with preoperative facial nerve palsy represent an unusual and particularly challenging presentation requiring careful assessment of tumour extent, facial nerve involvement, and the possibility of malignant transformation.

In this report, we present a case of long-standing giant recurrent pleomorphic adenoma of the parotid gland complicated by right-sided facial nerve palsy. The patient experienced a delayed presentation due to financial limitations.

Case presentation

A 46-year-old male trader presented with a painless recurrent right cheek mass of about 15 years' duration. The mass started like a nodule just inferior to the earlobe and gradually increased in size. It was painless, non-itching, no hearing impairment, toothache, jaw pain or weight loss. No cough or prolonged contact with a chronically coughing individual. He had an excision of the mass about 12 years ago in a private hospital but the patient had no histopathology report on presentation and does not know if the specimen was sent for histology also the operative details could not be accounted for, however, the mass recurred about 4 months post-surgery and has markedly increased in size over the years, with a feeling of heaviness, dragging sensation, and difficulty chewing on the same side. Also, mild facial deviation on the same side. He no longer finds it comfortable attending public functions due to stigmatization and was also divorced by his wife, due to patient's condition. The patient had previously sought care at multiple healthcare facilities; however, surgical intervention was deferred due to concerns regarding his perioperative risk and the potential for poor postoperative outcomes.

Examination revealed a marked facial asymmetry. A well-defined, roughly ovoid, multilobular 14cm x 12cm x 13cm right parotid mass in diameter, with

overlying 5cm hypertrophic scar on the summit. The swelling extends superoinferiorly from the right zygomatic arch to about 2cm below the angle of the mandible and anteroposteriorly from the posterior border of the sternocleidomastoid muscle to about 5cm from the oral commissure, with large distended veins all over the mass. The right ear lobule was slightly everted with a flattened nasolabial fold, with a deviated angle of the mouth and loss of nasolabial fold on the same side (House-Brackmann grade II).

There was no differential warmth, no tenderness; it was firm, nodular, attached to the skin and the underlying structures. There was no intraoral extension, with no lymphadenopathy. The left cheek is essentially intact.

Diagnosis was recurrent right parotid tumour with mild right facial nerve palsy.

Preoperative investigations requested for were ultrasound-guided FNAC, CT scan of the neck, FBC, serum electrolyte, urea and creatinine, blood glucose, LFT, coagulation profile, blood group and screen/crossmatch. The laboratory test results were all within normal ranges and packed cell volume was 40%. The patient could not do the ultrasound-guided FNAC after the CT scan due to financial challenges.



Preoperative picture



The Excised tumour

Neck CT-scan: Soft tissue mass with well-defined, regular outer margins but ill-defined inner margins. Soft tissue mass shows minimally heterogeneous enhancement measuring 13.5cm x 11.8cm x 12.9cm. There is no calcification or cystic component within it. The mass could not be well delineated from the neck muscles and the right parotid gland. There is an increase in vascularity around the mass compared to the left. There is no obvious cervical lymphadenopathy. The left neck and the surrounding structures are normal.

The diagnosis and surgery were discussed with the patient, including the benefits and risks. Preoperative evaluation and optimization were done, and informed consent was obtained from the patient. Right-sided total parotidectomy was done under general anaesthesia, with careful dissection, the facial nerve trunk was identified but not the branches due to fibrosis from the previous surgery. The excised tumour was 14cm x 12cm x 13cm in dimension and weighed 2.8kg. The wound was irrigated, haemostasis achieved, and the wound closed in layers with an active drain.

Histologic sections showed a pleomorphic adenoma with a well-circumscribed epithelial and myoepithelial differentiation within a variably myxoid, chondromyxoid, chondroid and/or fibrous

stroma. The former consists of glandular structures lined by two or more cell layers, trabecular structures, and occasional solid islands which demonstrate squamous metaplasia. The stromal element consists of myxoid/ chondromyxoid matrix, dense fibrous stroma and foci showing cartilaginous change. Although, multinodularity, satellite nodules and variable capsular abnormalities were noted; extensive histopathological examination revealed no overt malignant epithelial component, marked cytological atypia, increased or atypical mitotic activity, tumour necrosis, destructive or infiltrative growth, capsular or extracapsular invasion, perineural invasion or lymphovascular invasion, thereby excluding carcinoma ex pleomorphic adenoma.



Postoperative picture.

Mild facial nerve palsy (House-Brackmann grade II) was observed postoperatively with affection of the marginal mandibular and buccal branch causing

deviation of the mouth, mild loss of nasolabial fold, and slurring of speech. The post operative weakness observed was the same as preoperative state (House-Brackmann grade II). The patient was discharged home on postoperative day 8 and was lost to follow-up after 3 months with improved speech. He was subsequently lost to follow up.

Discussion

Pleomorphic adenoma, as defined by the World Health Organization (5th WHO edition 2022) as a benign salivary gland epithelial tumour composed of variable proportions of epithelial and myoepithelial cells embedded in a characteristic myxoid, chondroid, or mucoid stromal matrix⁴.

It is the most common benign salivary gland tumour, accounting for about 60-80% of cases⁵. Typically located below the ear lobe and overlying the angle of the mandible. Approximately 80% occur in the superficial lobe with peak incidence of 30-50 years of age and a slight female preponderance, although the index patient is a male^{6,7}. Less than 20% of pleomorphic adenomas arise in the deep lobe⁸.

The size may vary from a few millimeters to even a giant size. The first published case of a giant pleomorphic adenoma (>1kg) was reported in 1863 by Spence; afterwards, several studies have revealed giant pleomorphic adenoma being reported in different parts of the world, with the largest weighing 8.03kg⁹. The excised tumour in our patient weighed 2.8kg.

Pleomorphic adenoma arises from pluripotent intercalated duct reserve cells capable of both epithelial and mesenchymal differentiation. It presents as a mobile slow-growing, painless, asymptomatic firm swelling without ulceration of the underlying mucosa, irregularly shaped, with a pseudo capsule and microscopic extensions through the capsule, increasing risk of recurrence if incompletely excised^{5,9}. Facial nerve weakness is a rare finding except in large, neglected, and recurrent cases, which may present with facial nerve weakness as seen in our case⁵. The mild facial nerve palsy (House-Brackmann grade II) observed could be due to facial nerve injury from previous surgical intervention, or the mass effect from the long-standing huge tumour.

Malignant transformation to carcinoma ex

pleomorphic adenoma is relatively rare, but rapid increase in size of a long-standing previously slow-growing swelling, pain or tenderness, ulceration of overlying skin, fixation to skin or underlying structures or presence of regional lymphadenopathy can suggest malignant change. Given the recurrent nature, massive size, apparent fixation and facial nerve dysfunction in the index patient, the histopathological examination revealed no overt malignant epithelial component, marked cytological atypia, increased or atypical mitotic activity, tumour necrosis, destructive or infiltrative growth, capsular or extracapsular invasion, perineural invasion or lymphovascular invasion, thereby excluding carcinoma ex pleomorphic adenoma.

Magnetic resonance imaging (MRI) is preferred to other radiological studies based on better soft tissue definition, detailed tumour margin description, and tumour relationship with the surrounding tissues⁵. However, it was not done in our case as it was not available. The histopathological report is consistent with pleomorphic adenoma, showing epithelial and myoepithelial cells with myxoid and cartilaginous differentiation as appreciated in the stromal components.

Early intervention is essential in giant tumours of long standing duration like this patient due to high risk of malignant transformation. Adequate surgical care for involvement of both superficial and deep lobes entails total parotidectomy with or without facial nerve preservation^{9,10}. Injury to the facial nerve during dissection can result in a post-operative facial deformity and its impact on the quality of life. We made an effort to locate and preserve the facial nerve trunk, but the tumour was firmly adherent to the surrounding structures, thus making it nearly impossible to distinguish the nerve branches.

Although pleomorphic adenoma is generally recognized as a benign tumor, recurrence can occur, as seen in this index patient before presenting to us. The recurrent rate depends on the excision margin, rupture of the capsule with subsequent spillage during excision; and follow up⁵. The patient's recurrence cannot be attributed to any of the above mechanisms as the details of the previous surgery was not known. The patient was lost to follow-up after 3 months post-surgery; the possibility of

recurrence cannot be ruled out.

The limitations include short follow-up and loss to follow-up, non-availability of the photomicrograph of the histology, and the histology report of the previous surgery.

Conclusion

This case underscores the clinical and socioeconomic challenges associated with the management of long-standing giant pleomorphic adenomas. The unusual presentation with facial nerve palsy, secondary to delayed intervention, highlights the potential for neurological complications in otherwise benign tumours. Also, large size, recurrence, fixation and facial nerve dysfunction in a pleomorphic adenoma warrant careful evaluation for malignant transformation, but these clinical features do not themselves establish malignancy; adequate histopathologic sampling remains essential.

Furthermore, it demonstrates how financial constraints and perceived surgical risks can significantly impact timely treatment in resource-limited settings.

References

1. Tarsitano A, Pizzigallo A, Giorgini F, Marchetti C. Giant pleomorphic adenoma of the parotid gland: an unusual case presentation and literature review. *Acta Otorhinolaryngol Ital.* 2015;35(4):293-6.
2. Kalwaniya DS, Meena R, Kumar D, Tolat A, Arya SV. A Review of the Current Literature on P l e o m o r p h i c A d e n o m a . *Cureus.*2023;15(7):e42311.
3. Pareek YK, Gupta D, Aseri Y, Rawat DS, Singh BK, Verma PC. Giant Pleomorphic Adenomas of Parotid Gland: a case series. *Indian J Otolaryngol Head Neck Surg.* 2022;74(2):2008-2013
4. Skalova A, Hyrcza MD, Leivo I. Update from the 5th Edition of the Whorld Health Organization Classification of Head and Neck Tumours: Salivary Glands. *Head and Neck Pathol.* 2022;16(1):40-53.
5. Jain S, Hasan S, Vyas N, Shah N, Dalal S. Pleomorphic Adenoma of the Parotid Gland: Report of a case with Review of Literature. *Ethiop J Health Sci.* 2015;25(2):189-194
6. Mahesh Melkundi, Prashant Babaji, Rashmi Saikhedkar, Manjunath B Chaluvaiah. Pleomorphic adenoma of parotid gland: A case Report. *Oral and Maxillofacial Pathology Journal (OMPJ).* 2012;3(1):228-231
7. Omitola O.G, Soyele O.O, Butali, A, Akinshipo, A.O, Okoh, D, Sigbeku, O, et al. Descriptive Epidemiology of Salivary Gland Neoplasms in Nigeria: An AOPRC multicenter Tertiary Hospital study. *Oral Disease.* 2019;25:142-149
8. Farrh FM, Alharbi A. Pleomorphic Adenoma of the Deep lobe of Parotid Gland Presenting With Dysphagia: A case Report. *Cureus.* 2023;15(3):e36314.
9. Gobishangar S, Raviraj S, Vinojan S, Dias SJT, Theepan JMM, Dinoshiga K. Surgical management of a giant parotid gland pleomorphic adenoma in Northern Sri Lanka: A case report. *Int J Surg Case Rep.* 2023;111:108865.
10. Bhattacharya A, Singh M, Shah A, Varghese LL. Partial superficial parotidectomy for pleomorphic adenoma of the parotid gland. *BMJ Case Rep.* 2021;14(6):e238759