Giant pleomorphic adenoma of the parotid gland: a case report

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Abstract

Pleomorphic adenomas account for the majority of parotid masses, typically arising in the tail of the gland and enlarging slowly. Most are 2 to 6 cm in size when resected. We report the resection of a benign mixed tumour of the left parotid gland with a history of bleeding. The resected tumour measured 21 cm in diameter, weighed 1.81 kg, and on pathologic examination was a benign mixed tumour without malignant degeneration. The implications of this unusual case for the management of mixed tumours are discussed with a review of the literature.

Key words: Giant pleomorphic adenoma, parotid tumour, neck mass, Namibia

Introduction

About 80% of parotid masses are benign and of these, 80% are pleomorphic adenomas. They are slow-growing, painless in the preauricular or retromandibular area with no associated facial paralysis. Treatment consists of excision with wide margins, typically via superficial parotidectomy. Parotid tumours are surgically removed because there is always a risk of malignant degeneration. Also, if there is excessive growth this leads to facial deformities, ulceration, bleeding and functional problems.^[1-3]

There is a move at present to consider a more conservative surgical approach for parotid tumours. However, parotid pleomorphic adenomas have a multicentric growth. Hence with very conservative treatment the risk of recurrence is high. Further surgery then more complicated since the facial nerve is difficult to identify and separate. If there is subsequent malignant change it may be necessary to scarify the facial nerve with a radical parotidectomy and postoperative radiotherapy.

This unusual case presented to the maxillofacial surgery service at Oshakati Intermediate Hospital of Namibia prompted us to re-evaluate the behaviour of these benign mixed tumours. We propose that this case, demonstrated sufficient morbidity to justify the early excision of all pleomorphic adenomas, despite surgery being difficult and the risk of bleeding. When these tumours grow massively, they are accompanied by neoformation of blood vessels with increased bleeding during parotidectomy.^[4-,6]

The relatively low risk of malignant transformation is a factor to consider when planning surgery. Tumours of this large size are uncommon and require a working knowledge of the anatomy and approach to surgery.^[7,8]

Case Report

An 83-year-old woman complained of bleeding from a lump on her face. She reported no past illnesses and had not consulted a clinician for ten years apart from traditional healers. She stated that the mass had slowly enlarged over a long period. It had not troubled her until one of the nodular areas had broken down and begun to bleed.



Figure 1. The patient at initial examination, showing a large, multinodular left parotid mass extending from the ear into the neck.



Figure 2. View after surgery showing the posterior flap to close the wound

Clinical examination on admission revealed an elderly woman with a massive left sided facial tumour (Figure 1). It was nodular, soft to palpation, lobulated, painless, well circumscribed; tensely cystic in places, and had prominent veins surrounding the mass. The ear lobe was displaced upwards and the tumour extended onto the neck. The surface was focally necrotic with bleeding (Figure 1). There were no cranial neuropathies and the remainder of the physical examination was normal.

She was admitted for assessment and preparation for surgery. Haematological tests (Table 1), a coagulation screen, renal and liver tests were normal.

Table 1. Routine blood tests

Test	Result	Reference
White blood cell count	6.34	3.39-8.86×10^9/L
Haemoglobin	12.80	11.1 – 14.7 g / 100ml
Platelets	207	171-380×10^9/L



Figure 3. The excised tumour.

A CT scan was taken to check the relationship of the tumour with the great blood vessels and the nerve structures in the neck. Involvement of these structures would markedly increase the risk of surgery.

The CT scans showed no close relationship to the great vessels or the parapharyngeal space (not shown as the images were of too low resolution to publish). The patient was cross-matched in case blood transfusion became necessary.

Surgery was performed under general anaesthesia with oral intubation. Parotidectomy technique was difficult because of bleeding and maintaining haemostasis.

The mass was removed conserving the integrity of facial nerve, which it was identified and properly dissected during the surgery. The wound was closed successfully by creating a posterior rotation flap to achieve a primary closure (Figures 2 and 3).

Histological examination revealed a pleomorphic adenoma with no evidence of malignancy – see Surgical Histology Report.(Table 2.)

The facial nerve function remained intact, no blood transfusions were needed, and postoperative recovery was uneventful.

After seven days, the patient was well but some borders of

Table 2. Surgical Histology Report

Clinical details	Tumour 20 cm in diameter, bleeding, mobile, encapsulated, lobulated, fixed to the skin, surface ulcerated.
Macroscopy	Tissue mass 70/30/60 cm with overlying ulcerated nodular skin. On cut section is haemorrhagic nodular tumour with cystic space.
Microscopy	Section shows overlying epidermis, benign salivary gland and lymphoid tissue with relatively well a circumscribed tumour. The tumour is mainly composed of tubular structures lined by a double layer of epithelium and myoepithelial cells. Papillary structures, cystic spaces, haemorrhage and fibrosis are also noted throughout the tumour. No features of malignancy.
Diagnosis	Left parotid gland tumour: Pleomorphic adenoma.



Figure 4. Seven days postoperative, showing vitality of the skin flaps apart from necrosis at the border due to tension of the wound.

the wound became necrotic and were removed. (Figure 4). The patient was discharged from the maxillofacial service and a follow-up was arranged for three months later, but the patient never returned to the hospital, probably because her village was more than 300 km away.

Discussion

This case presents several unusual features. At 1.83 kg, the parotid mass is a big tumour. One review of massive

pleomorphic adenomas was published by Short and Pullar in 1956 who reported a similar tumour weighing 2.3 kg tumour. This includes the case reported by Spence in 1863 which is an important and successful resection of a mixed tumour larger than 1 kg. A common theme that runs through many of the reported cases is the patients' fear of the surgery and the culture of seeing traditional healers. This was a factor with our patient.^[9,10]

Our patient's presenting complaint had been of bleeding and leakage of turbid fluid from the mass; usually caused by necrosis of the tumour on the surface which did not have enough blood supply.

Spontaneous infarction of benign mixed tumours is unusual and raises concern about malignant change. In massive tumours, however, even slow growth eventually outstrips the blood supply, resulting in haemorrhagic degeneration of the central portion of the mass.^[11,12]

The cyst is lined only with necrotic debris, as was the case in our specimen. This distinguishes such cystic spaces from true cysts within pleomorphic adenomas, which arise from squamous metaplasia or abnormal ductal elements within the mass. While haemorrhagic degeneration in a parotid mass should always raise the index of suspicion for malignancy, our case and others like it suggest that central necrosis in benign tumours is rare only because most are resected while still small.^[13,14]

With surgery of this complexity, it is important to have a thorough knowledge of the anatomy of the local structures. The parapharyngeal space lies medial to the parotid gland and includes:

- Maxillary artery and ascending pharyngeal artery
- Glossopharyngeal nerve (IX)
- Vagus nerve (X)
- Internal carotid artery.
- Internal jugular vein in the carotid sheath.

- Accessory nerve (XI)
- Hypoglossal (XII)
- Sympathetic trunk and superior cervical ganglion of the trunk.
- Ascending pharyngeal artery.

When parapharyngeal space is invaded by parotid tumours the surgery becomes very risky, because it maybe impossible to control due to damage of the jugular vein or carotid artery.

Conclusions

Although there was no evidence of malignancy in our specimen, and the patient suffered no major morbidity, she is fortunate in this regard. Neglecting even a benign parotid mass carries an increasing risk of facial nerve injury when surgery is finally undertaken. The bony and muscular deformity associated with such tumours is disfiguring. Although more than 95% of all pleomorphic adenomas remain benign, it is important to bear in mind that the clinical course of such growths can be far from benign. Although it can be clinically fairly certain that a tumour is benign we advocate the early excision of parotid masses in all patients who will tolerate surgery.

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All images from the authors.

References

- Periasamy S, Manoharan A, Garg H, et al. (August 03, 2019) Pleomorphic Adenoma of the Cheek: A Case Report. Cureus 11(8): e5312. doi:10.7759/cureus.5312. https://www.cureus. com/articles/21911-pleomorphic-adenoma-ofthe-cheek-a-case-report
- Arumugam P, Christopher PJ, Kumar S, Kengasubbiah S, Shenoy V. Pleomorphic Adenoma of the Palate: A Case Report. Cureus. 2019; 11(3):e4308. Published 2019 Mar 25. doi:10.7759/cureus.4308. https://www.cureus. com/articles/18174-pleomorphic-adenoma-ofthe-palate-a-case-report.
- Adiyodi NV, Sequeira J, Mehra A. Twinning of Pleomorphic Adenoma: A Case Report. Cureus. 2020; 12(1):e6608. doi:10.7759/cureus.6608 . https://www.ncbi.nlm.nih.gov/pmc/articles/ PMC7008752/
- 4. Alnofaie, H., Alshammeri, T., Alturkistany, Y. et al. Giant Pleomorphic Adenoma of Parotid Gland in Saudi Arabia: a Rare Case Report. SN Compr. Clin. Med. 2, 1258–1263 (2020). https://doi. org/10.1007/s42399-020-00359-0

- 5. Al Kindi M, Ramalingam S, et al. "Giant Parotid Pleomorphic Adenoma with Atypical Histological Presentation and Long-Term Recurrence-Free Follow-Up after Surgery: A Case Report and Review of the Literature", Case Reports in Dentistry, vol. 2020, Article ID 8828775, 18 pages, 2020. https://doi.org/10.1155/2020/8828775
- 6. Rai A, Sharma S, Shrivastava P, et al. A huge pleomorphic adenoma of the submandibular salivary gland. Case Reports. BMJ Journal 2018. https://casereports.bmj.com/content/2018/bcr-2017-222249.
- Choińska AO, Bruzgielewicz A, et al. Synchronous multiple unilateral parotid gland tumors of benign and malignant histological types: case report and literature review. Braz. j. otorhinolaryngol, 85(3): 388-392. http:// www.scielo.br/scielo.php?script=sci_arttext&pid =\$1808-86942019000300388
- Mittal G, Aggrawal A, Garg R, et al. Pleomorphic adenoma: A case report. International Journal of Applied Dental Sciences 2017; 3(2): 154-155. https://www.oraljournal.com/pdf/2017/ vol3issue2/PartC/3-1-22-625.pdf
- John E. Buenting MD, Timothy L, Smith MD. Giant Pleomorphic Adenoma of the parotid gland: case report and review of the literature. ENT-Ear, Nose & Throat Journal 1998. https://journals.sagepub.com/doi/ pdf/10.1177/014556139807700812
- 10. Short DW, Pullar P. Giant parotid tumours; with report of a case. J R Coll Surg Edinb. 1956 Mar;1(3):240-248. PMID: 13307671.
- Woo C, Son SM. Carcinosarcoma of the parotid gland with abdominal metastasis: a case report and review of literature. World J Surg Onc 16, 103 (2018). https://doi.org/10.1186/s12957-018-1406-6
- Sangwan S, Sufian, Kaur N. Pleomorphic adenoma of tongue: A common entity at unusual location. Int J Oral Health Sci 2020; 10:60-3
- Tandon A, Jaiswal R, Siddiqui S, Bordoloi B. Keratinizing pleomorphic adenoma: An unusual case report. J Oral Maxillofac Pathol [serial online] 2018 [cited 2021 Feb 7]; 22, Suppl S1:69-72. https://www.jomfp.in/text. asp?2018/22/4/69/224617
- 14. Sunil Kumar Sharma.2019. "Extensive nasopalatine pleomorphic adenoma:-a case report", Asian Journal of Science and Technology, 10, (02), 9414-9416.