Case Report

Giant Pleomorphic Adenoma of the Parotid Gland

SHAHZAD ALAM SHAH, MOHAMMAD SARFRAZ
Department of Surgery, Lahore General Hospital, Lahore
Correspondence to Dr. Shahzad Alam Shah

Pleomorphic adenomas account for the majority of parotid masses arising mostly in the superficial part of the gland and enlarging slowly over time. The vast majority are 2 to 6 cm in size when resected. We report resection of a huge mixed parotid tumor. A 40 year-old man had a 15-year history of an enlarging right periauricular mass that had developed areas of discoloration of the overlying skin due to underlying necrosis The patient ultimately underwent resection of the mass, which measured 24 cm in diameter, weighed 4.65 kg, and proved on pathologic examination to be a benign mixed tumor without malignant degeneration. The implications of this unusual case for the management of mixed tumors are discussed, and a review of the world literature on giant pleomorphic adenomas is presented.
Key Words: Pleomorphic adenoma

Approximately 80% of parotid masses are benign; of these, 80% are pleomorphic adenomas[1]. These tumors are almost uniformly characterized by a slow-growing, painless mass in the preauricular or retromandibular area with no associated facial weakness. Treatment consists of excision with wide margins, typically via superficial parotidectomy. The excision of a benign tumor in the parotid, with the associated risk of facial nerve injury, Frey's syndrome and other complications, is generally justified by the risk of malignant degeneration, which is quoted to be in the range of 1.4-6.3%[2]. An unusual case of a patient who came to the Department of Surgery Lahore General Hospital has prompted us to reevaluate the behavior of those benign mixed tumors that remain benign. We propose that this case and others like it demonstrate sufficient morbidity to justify the early excision of all pleomorphic adenomas, despite the relatively low risk of malignant transformation.

History. An 40-year-old man presented to us with a huge lump on his neck. A few days back he started having pain in it and in addition, he had become febrile within the 24 hours preceding presentation. He stated that the mass had begun to develop 15 years ago in the region of his left ear and had slowly enlarged since.

Clinical findings. Physical examination on admission revealed a middle age man with a massive right-sided neck tumor (Figure 1). The mass was nodular, tensely cystic in places, and had prominent veins near the base. It lay on the patient's shoulder making it nearly impossible for his neck movement. Some of the surface had become focally necrotic. The patient had no cranial nerve deficits and the remainder of his physical examination was unremarkable. Admission laboratory values revealed an Hb% of 8mg/dl and the patient was given two units of blood. He was admitted in the ward and after preliminary investigation resection of the tumour was planned. Computed tomography (CT) was performed to assess the extent of the mass (depth), which was noted to abut the carotid sheath in the mid-neck.

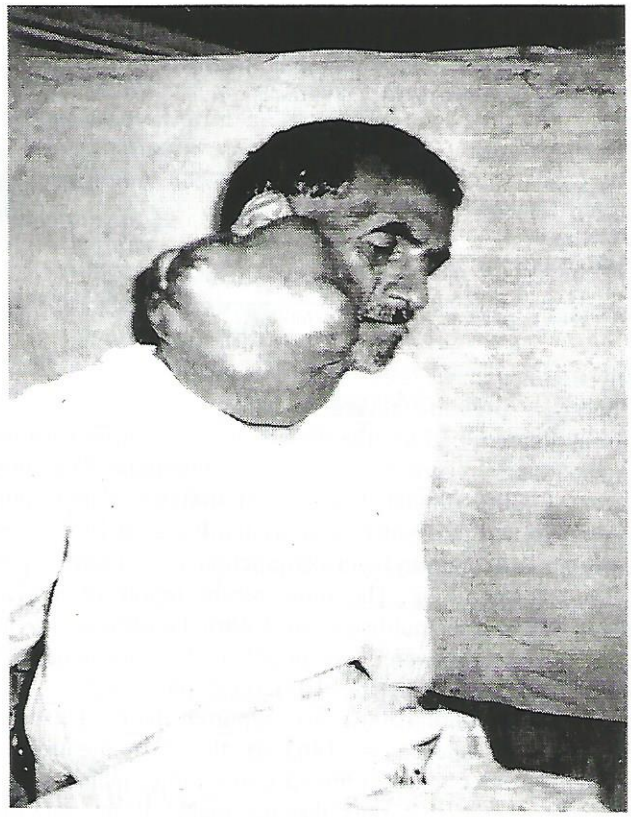

Fig. 1

Operative Findings. Skin flaps were raised off the sides of the mass to provide sufficient tissue for closure. Despite the age and size of the mass a plane of dissection was clearly apparent around the tumor base. The sternocleidomastoid muscle was greatly hypertrophied on the left and the mass displaced it posteriorly and inferiorly, coming into contact with the vagus nerve and the carotid sheath. Numerous vessels were ligated as dissection proceeded superiorly. Due to years of traction the mass had descended well below the angle of the mandible and the facial nerve was avoided without difficulty. The mass was ultimately attached only by a fibrous band extending

deep to the posterior belly of the digastric muscle, just below the angle of the mandible; this was ligated and the specimen delivered. The mass was sectioned and demonstrated large necrotic cystic spaces. The wound was closed without difficulty. Histologic examination revealed extensive necrosis and cartilaginous metaplasia, but no evidence of malignancy. The patient had no cranial nerve deficits, required no additional transfusions, and his postoperative recovery was uneventful. At one-year follow-up, he was doing well, without evidence of recurrence.

Table. Giant Pleomorphic Adenomas (World Literature).

Author	Year	Patient Age/ Gender	Tumor Wt (kg)
Frylinck	1956	63/F	26.50
Cotterill	1907	46/M	11.40
Skurczynski et al	1978	71/F	8.50
Zhang	1983	46/F	7.00
Buenting	1994	85/F	6.85
This Case	2001	40/M	4.65
Schultz-Coulton	1989	78/F	4.50
Banerjee et al	1978	70/F	4.50
DeRonville et al	1904	44/F	2.95
Pullian	1963	25/F	2.83

(Ref: John E. Buenting, Timothy L. Smith.) .[3]

Discussion

This case presents several unusual features worthy of discussion. At 4.65 kg, the mass is the sixth largest benign mixed tumor recorded in the world literature. The most recent English language review of massive pleomorphic adenomas was published by Short and Pullar in 1956 (cited by John and Timothy)[3], in conjunction with a case report of a 2.3 kg tumor. The most recent report of a giant pleomorphic was published in 1996[4]. Unfortunately, the size and weight were not specified. The accompanying table shows data on the 10 largest pleomorphics ever resected[3], including the case reported here. The case reported by Spence in 1863 is the first mention of successful resection of a mixed tumor larger than 1 kg[3]. A common theme that runs through many of the reported cases is the patients' inappropriate fear of the risks of resection. Although in our case the patient had not sought the advice of a physician, fear of surgery was certainly the underlying reason he had allowed his neck mass to progress to such an extent. The gross specimen clearly showed cystic degeneration in the center of the tumor. Indeed, the patient's presenting complaint had been of pain in the mass. Spontaneous infarction of benign mixed tumors is considered a rare occurrence, and raises concern about malignant degeneration[6,7]. In massive tumors, however, even slow growth eventually outstrips the blood supply, resulting in hemorrhagic degeneration of the central portion of the mass[8]. The cyst is then 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 hemorrhagic 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 tumors is rare only because most are resected while still small.

Conclusions

Although there was no evidence of malignancy in our specimen, and the patient suffered no major morbidity, he is fortunate in this regard. Neglecting even a benign parotid mass carries an ever increasing risk of facial nerve injury when surgery is finally undertaken. The bony and muscular deformity associated with such tumors is uniformly disfiguring and ultimately incapacitating. Although it is generally accepted that more than 95% of all pleomorphics remain non-malignant, this case and others like it serve to remind the surgeons that the clinical course of such growths can be far from benign. [2] Despite the reliability of clinical examination in determining the non-malignant character of parotid masses,[1] we therefore continue to advocate the early excision of parotid masses in all patients who will tolerate surgery.

References

1. Phillips DE, Jones AS. Reliability of clinical examination in the diagnosis of parotid tumors. J R Coll Surg Edinb 1994; 39(2):100-2.
2. Duck SW, McConnel FM. Malignant degeneration of pleomorphic adenoma -- Clinical Implications. Am J Otolaryngol 1993;14(3):175-8.
3. John E. Buenting, Timothy L. Smith. Giant parotid tumors: With report of a case. Ear, Nose & Throat Journal 77(8): 634, 637-638, 640, 1998.
4. Lomeo PD. Giant pleomorphic adenoma of the parotid. Ear Nose Throat J 1996;75(7):402.
5. Schultz-Coulton HJ. Pleomorphe Riesenadenome der Glandula Parotis. Laryngo Rhino Otol 1989;68:445-9.
6. Layfield LJ, Reznicek M, Lowe M, et al. Spontaneous infarction of a parotid gland pleomorphic adenoma: Report of a case with cytologic and radiographic overlap with a primary salivary gland malignancy. Acta Cytol 1992;36(3):381-6.
7. Abiko Y, Kaku T, Shimono M, et al. Large cyst formation in pleomorphic adenoma. Bull Tokyo Dent Coll 1993;34(1):9-14.
8. Banerjee AK, Ahmad M. A grand parotid tumor. J Indian Med Assoc 1978;70:258-9.