Giant parotid pleomorphic adenoma in a Nigerian male

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Abstract
Pleomorphic adenoma (PA) is the most common benign salivary gland tumor, accounting for up to 60% of all tumors of the salivary gland and 75–80% of all parotid tumors. It has a slow but progressive growth pattern and if untreated can become greatly enlarged in size. However, giant PA is uncommon. We report a case of giant parotid PA of more than 10 years duration in a 60-year-old male Nigerian. A 60-year-old male patient with 10-year history of painless, massive, and pedunculated swelling in the left side of lower face measuring 25 cm × 23 cm × 17 cm in dimension. Computerized tomographic scan and percutaneous fine needle aspiration cytology returned a preliminary diagnosis of PA. Superficial parotidectomy with facial nerve preservation via the transcervical approach was performed. The excised mass weighed 5.5 kg while the postoperative healing was satisfactory. Benign PAs may attain a giant size if left untreated. Socioeconomic problems are some of the reasons for late presentation. Adequate excision of the tumor sparing the facial nerve is possible but intraoperative and reactionary hemorrhage are likely complications.

Key words: Challenges, developing country, giant, parotid, pleomorphic adenoma, salivary

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Introduction
Pleomorphic adenoma (PA) is the most common benign salivary gland tumor, accounting for as many as 60.0%1 of all tumors of the salivary gland and 75–80% of all parotid tumors.1,2 Submandibular, sublingual, and minor salivary glands are also affected to lesser extent.1,2

They present as rubbery, often lobulated swellings which run an insidious course. The absence of pain or discomfort in the early stages is characteristic. The main early presenting complaint is usually related to unacceptable and problematic esthetic issues. Without treatment, these tumors can assume huge sizes, or less commonly undergo malignant transformation.1

Microscopically, PA consists of fibrous, myxoid, or cartilaginous mesenchymal elements due to the pluripotent properties of myoepithelial cells.1,3 The different cellular components originate from duct epithelium or myoepithelial cells and are completely varied and disordered in arrangement and in proportions. This histomorphologic pleomorphism in appearance gave it its name.1,3

The diagnosis of PA is hinged on both clinical and histological findings from fine or core needle cytology as well as radiological evidence of the absence of malignant features.1 The mainstay of treatment is wide surgical excision; however, tumor recurrence is common. This is

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attributable to multifocality, intraoperative cellular spillage and incomplete clearance owing to difficulty in completely removing the pseudocapsule.[1]

Giant PA is uncommon, few cases have been reported in the parotid gland (superficial and deep lobes) weighing a few kilograms and of considerable sizes and dimensions.[4‑12] The present report adds to the scientific literature on giant PA of the parotid gland. We highlight the socioeconomic factors associated with the late presentation and challenges associated with surgical resection of a giant parotid mass.

Case Report

A 60-year-old male patient presented to our maxillofacial surgery clinic with progressively increasing, painless, massive swelling in the left side of the jaw of 10 years duration. The patient did not seek medical treatment up until then due to financial reasons. His medical history was however unremarkable. On examination, we found a lobulated, pedunculated, nontender swelling of 25 cm × 23 cm × 17 cm in dimension on the left side of his lower face. The swelling was firm in consistency with normal overlying skin, it was bigger than the size of his head (giving a “double head” appearance), and he had to support the mass with his hands [Figures 1 and 2]. There was neither intraoral swelling nor palpable lymph nodes of the neck. All cranial nerves were intact.

Computerized tomographic (CT) scan was obtained which showed huge soft tissue mass in the parotid gland projecting to the cervical region with multiple amorphous calcifications but no underlying bony involvement [Figures 3 and 4].

Percutaneous fine needle aspiration for cytology (FNAC) showed admixture of epithelial, myoepithelial, and chondromyxoid elements in a hemorrhagic stroma. There were no features suggestive of malignancy. Based on the clinical, radiological, and FNAC findings, a preliminary diagnosis of PA was made, and the patient was counseled for parotidectomy.

Superficial parotidectomy with facial nerve preservation via the transcervical approach was performed after obtaining informed consent. In anticipation of the presence of aberrant, displaced vessels, and neovascularization, the incision line was copiously infiltrated with lidocaine 2%/epinephrine (1:100,000). Although peripheral branches of the facial nerve were displayed, we avoided injury to the cervical and mandibular branches of the facial nerve by identifying these structures during the retrograde dissection. No effort was made to identify the nerve trunk as this can increase the risk of serious nerve damage.[13,14]

At surgery, large vascular channels were seen around the lesion which resulted in brisk bleeding that was successfully arrested with prompt and meticulous ligation. Nonetheless, the patient received 3 units of blood intraoperatively due to bleeding from large tumor bed and many new vascular channels. The excised mass weighed 5.5 kg, its cut sections show gray-yellowish appearance.
with areas of microcalcification, myxoid change, and microcystic areas.

There was an episode of reactionary bleeding in the immediate postoperative period, for which the patient required wound exploration in the operating room. Histological examination of the mass confirmed the diagnosis of PA. A benign mixed tumor composed of epithelial and mesenchymal components was seen under the microscope with mesenchymal components consisting of chondroid and bone on a fibromyxoid background.

The patient had a satisfactory postoperative course with good wound healing. The facial profile was also satisfactory. There was temporary paresis of the buccal branch of the facial nerve which resolved spontaneously [Figure 5].

The patient has been followed up for up to 6 months. He is free of tumor recurrence and has intact facial nerve functions. He is currently being reviewed 3 monthly.

**Discussion**

Although parotid PAs are common, giant sizes are rarely encountered. Late presentations of medical conditions are common occurrences in low-socioeconomic communities. Our patient demonstrated a usual problem associated with living in remote villages in a developing country. Access to health care is poor, and patients do not see the need to travel to big cities for health care unless there is associated pain, discomfort or loss of function which is characteristic of very late stages of the disease. The absence of pain and constitutional symptoms allowed our patient to carry on with his life in his environment in spite of the enormous size of his swelling.

Esthetic complaints are the least of the reasons for presentations in rural dwellers. However, the sheer size of the lesion which is bigger than his head was expected to have produced symptoms linked with weight and posture and compelled him to present earlier. Furthermore, the “double head” appearance could presumably be associated with psychological problems. Our patient however had no pointers to psychological effects from his facial deformity. It is not uncommon for people dwelling in the rural places of Nigeria and Africa to carry such disfiguring benign masses around; a common example is a very large goiter in an endemic area. Socioeconomic factors therefore are still major reasons why appropriate health care is not sought after in rural places of developing countries. The main reason for late presentation in our patient is a financial constraint. Fear of surgery, lack of information, negligence, and other sociocultural factors have been identified as other reasons for late presentation.

Since the 1998 review of the ten largest PAs ever published in the English-language literature, more recent reports have documented lesions of various sizes often affecting the deep lobe of the parotid gland. The present case affecting only the superficial lobe of the left parotid gland weighs 5.5 kg and adds to scientific literature.

FNAC remains a cost-effective, rapid, and minimally invasive aid in diagnosis that is applicable even in resource-poor situations such as the case we presented here. FNAC can breach tumor capsule, but the incidence of tumor seeding is low when compared with open biopsies; however, concerns about reliability, accuracy, and damage to vital structures have been expressed. Ultrasound guided FNAC or core needle cytology may improve accuracy. CT scan and magnetic resonance imaging are essential in tumor delineation and surgical planning, but cost and availability in low socioeconomic surgical practices remain a concern.

Extensive vascularization is common with giant tumors, giant PA did not behave differently. As a result, surgery tends to be associated with excessive hemorrhage as seen in this patient who received 3 units of blood intraoperatively. Embolization...
of the lesional vascular channel will reduce intraoperative bleeding considerably. Anticipation of this during surgical resection and meticulous ligation or cauterization of feeder vessels is essential to minimize blood loss.

The possibility of bleeding into tumor mass giving rise to sudden hemodynamic instability intraoperatively must be borne in mind. Careful ligation of the feeder vessels done well ahead of tumor extirpation was done here to reduce this risk.

There was no evidence of malignant features in the present case despite the long-term history. PA is known to undergo malignant transformation in long-standing cases,[1] especially in cases with repeated trauma or surgery. Although our patient had no history of repeated trauma or surgery, the actual reason for the absence of malignant transformation is not precisely known. The possibility of transformation occurring in future is also not completely ruled out. Therefore, consistent follow-up is still required to diagnose this if it occurs.

In summary, benign PAs may attain a giant size if left untreated. Socioeconomic problems are some of the reasons for late presentation. Adequate excision of the tumor sparing the facial nerve is possible but intraoperative and reactionary hemorrhage are likely complications. Satisfactory functional and esthetic outcome are achievable if standard preoperative and intraoperative surgical protocol are adopted.

Declaration of patient consent
The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest
There are no conflicts of interest.

References