

Case Report

Giant Pleomorphic Adenoma of the Parotid Gland: Report of a Case in the Reference Health Center of Commune VI of BAMAKO

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Abstract

Salivary glands tumors are rare and account for less than 3% of all tumors occurring in the head and neck region. They are generally benign, with pleomorphic adenoma (PA) of the parotid gland being the most common. PA typically occurs in adult females, and giant forms are still considered rare. Thus, the present report describes a case of giant Pleomorphic Adenoma of the parotid gland in a healthy 60-year-old male who came for consultation at the ENT department of the Reference Health Center (CSR  ) of Commune VI in the District of Bamako, presenting with a giant, slow-growing, asymptomatic parotid mass that had evolved over 20 years. Physical examination revealed a firm, multisegmental, painless preauricular swelling. The scan showed a right parotid tissue mass that was heterogeneous, multi-septated, measuring 119/104 mm, without any osteolytic lesions. Management consisted of extracapsular dissection of the tumour. Histological examination of the surgical specimen revealed a giant pleomorphic adenoma of the parotid gland. The patient had an uneventful postoperative recovery and was followed for seven years without recurrence. PA can disrupt a patient's daily life or develop into malignancy if not managed early. Practitioners therefore need to be aware of these different characteristics in order to make a diagnosis that justifies appropriate therapeutic decisions, as well as educating patients.

Keywords

Giant Pleomorphic Adenoma, Parotid Gland, ENT Department, Bamako/Mali

1. Introduction

Salivary glands tumors are rare, accounting for less than 3% of all reported tumours in the head and neck region [1, 2]. They are benign in around 80% of cases, and frequently occur in the main parotid gland in 70-80% of cases [3, 4]. These

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tumours are slow-growing, painless, rarely aggressive or transform into malignancy, in contrast to tumours of the minor salivary glands, although also rare, have a preponderance of malignancy in 80-90% of cases [1].

Pleomorphic adenoma (PA) is the most common Salivary Gland Tumor (SGT) [5], accounting for 60-80% of all benign salivary gland tumors and 60-70% of all parotid gland tumors [6, 7]. This tumour may be solitary or synchronous with other cervicofacial spaces [8]. Pleomorphic adenomas can occur at any age, but are frequent between the thirties and sixties, and occur more frequently in women, with a 2:1 ratio. The etiology of pleomorphic adenoma is poorly understood, but irradiation and the oncogenic simian virus (SV40) may play a role in its appearance or progression [5].

The diagnosis of PA is mainly based on clinical and histological findings, and the mainstay of management is surgical excision [1, 9].

In its clinical presentation, it is an irregular, rubbery, lobed, slow-growing mass, with no associated pain or discomfort [9]. Typically, the patient's complaint is of an unpleasant or unsightly facial appearance, or discomfort due to the weight of the mass [9, 10]. Specifically in the parotid gland, signs of weakness of the facial nerve appear in giant cases or in cases progressing to malignancy [5]. The risk of malignant transformation is low, and is inherent in factors such as advanced age, radiotherapy, large size and recurrent tumours. This risk can be suspected by signs such as pain, peripheral facial paralysis and, in advanced stages, lymph node metastases, skin infiltrations, ulcerations, massive tumor hemorrhage, dysphagia and mandibular invasion [5, 11].

Histologically, by name, it is a benign mixed tumor (pleomorphic) of ductal and myoepithelial cell origin. Due to the multipotential nature of the myoepithelial cells, the tumor is composed of epithelial and fibrous, myxoid and cartilaginous mesenchymal elements surrounded by a pseudocapsule, with occasional squamous metaplasia [9].

Surgical management is either by superficial or total parotidectomy, or by Extra-Capsular Dissection (ECD). All the above procedures carry a risk of postoperative facial paralysis and Frey's syndrome [6, 11]. Recurrence is generally associated with inadequate removal and incomplete removal of the pseudocapsule [9, 11]. The resected dimensions of the AP are frequently in the range of 2 cm to 6 cm and rarely reach 25-35 cm or 26.5 kg [1, 12].

Giant pleomorphic adenomas of the parotid gland are rare. However, cases or case series have been reported in the literature since 1995, with sizes of up to 28-33 cm and weights of

up to 26.50 kg [1, 10, 13].

The aim of this article is to add to the existing scientific literature a case of giant parotid PA treated surgically by extra capsular dissection, followed by uneventful postoperative recovery.

2. Observation

A 60-year-old man with no known medical or surgical history presented with swelling of the right parotid region. The onset of symptoms was about 20 years ago, marked by the progressive appearance of a parotid swelling, with no other associated signs. Physical examination revealed a giant swelling, approximately 30 cm in extension, painless, multi-nodular, irregular and mobile, with apparently normal overlying skin. The swelling extended superiorly-inferiorly from the level of the external ear to the lower edge of the mandible, and anteroposteriorly from the angle of the mouth to the posterior edge of the mandible. On endo-buccal examination, the right tonsillar compartment was pushed back. There was no lymph node involvement or facial nerve deficit.

A computed tomography (CT) scan was ordered, revealing a large, heterogeneous, multi-partitioned right parotid tissue mass measuring 119/104 mm, with no osteolytic lesions.

The patient's general examination was unremarkable.



Figure 1. Macroscopic image showing the mass in the right parotid region.

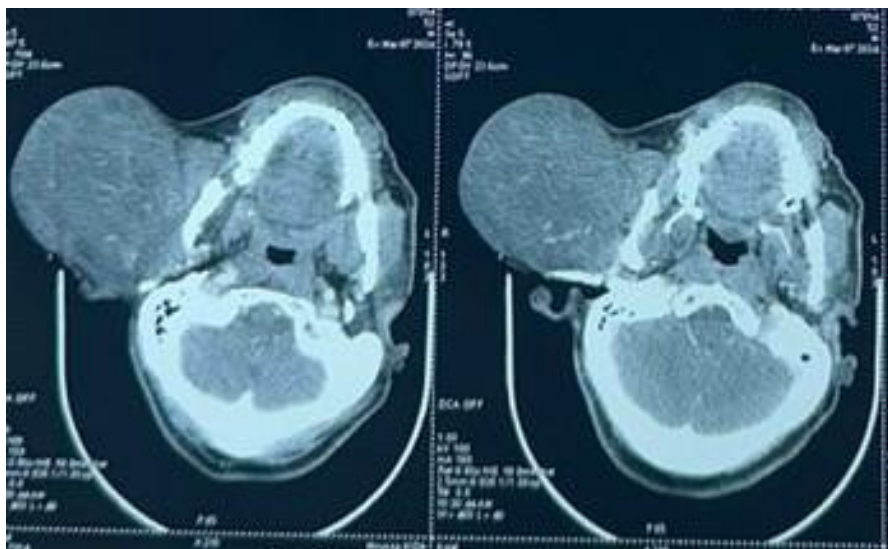


Figure 2. CT image showing a heterogeneous, multi-partitioned right parotid tissue mass measuring 119/104 mm, with no osteolytic lesion.

Surgical removal of the right parotid gland under general anaesthesia was planned and explained to the patient. The lesion was excised completely by DCE, with preservation of all facial nerve branches. A clearly discernible dissection plane around the tumor was used to dissect the tumor mass, without any iatrogenic damage to the facial nerve branches. Due to the ancient nature of the tumor, several small feeder vessels had to be ligated circumferentially around the tumor to achieve hemostasis. The intraoperative period was uneventful, and the patient did not require a blood transfusion. The resected mass was bilobed and ovoid in shape, with a final dimension of 8 × 14 × 8 cm and weighing 1.7 kg, and the patient's blood was not transfused.



Figure 3. The excised tumor specimen.

Pathological examination concluded a giant pleomorphic

adenoma of the parotid gland.

At 6 months post-op, the patient showed normal healing with no neurological deficit of the facial nerve.

3. Discussion

Pleomorphic adenoma is the most frequent mixed form of TGS occurring in the parotid gland, and several cases have been reported in the literature, the first dating back to 1863 [14]. Giant PAs of the parotid gland are considered rare, but cases or case series have been reported in the literature since 1995 [1, 10, 13]. Usually solitary, but can be synchronous with the parapharyngeal space [8]. The present report details a case of giant PA of the right parotid gland, together with its surgical management, histological presentation and long-term recurrence-free follow-up.

Giant PA usually occurs in adult females. Of 43 cases reported in the literature, the majority were female, aged 45 [1]. Also in the case series by Pareek et al., the female sex dominated at 66.67% of cases and patient age ranged from 30 to 81 years, with a mean age of 50.33 ± 15.30 years [13]. In the present report, the patient's age was 60, but he was male. This condition is slow-growing and asymptomatic, although the patient's complaints could be an unpleasant, unsightly facial appearance, facial nerve paresis or discomfort due to the weight of the mass [5, 9, 10].

The swellings, which vary from a few millimetres to several centimetres, averaging 2-6 cm, can progress over a long period without treatment, reaching 33-35 cm or 26.5 kg [1, 12, 15]. In this study, the delay was 20 years and the mass measured 30 cm. At resection, it was bilobed and ovoid in shape, with a final dimension of 8 × 14 × 8 cm and weighing 1.7 kg. Authors have reported that the evolution of giant parotid APs can range from 5 to 20 years before appropriate management [13]. In the literature review by AlKindi et al.,

tumors ranged from 3 to 5 cm in diameter, up to 35 x 28 cm 27 cm, in two perpendicular planes, and resected tumor weight varied from 0.12 kg to 7.3 kg [1]. Others report up to 9.7 kg after extraction over an evolutionary period of 11 years [10].

According to the author [10], the delay in consultation is linked to the low socio-economic level in developing countries, with difficult geographical accessibility to specialized care services, inadequate social security coverage and, above all, as long as the swelling does not hinder or disrupt the daily activities of patients living in rural areas, they do not care about aesthetics alone.

This delay is also explained by the common practice in African countries of treating tumours of the face and neck as witchcraft, thus encouraging recourse to traditional therapists before consulting a doctor [10]. There is also a fear of surgery [9].

The diagnosis of PA is mainly based on clinical and histological findings, and the mainstay of management is surgical excision [6, 9].

Apart from malignant transformation or concern for aesthetics or perceived weight, PA is asymptomatic. The risk of malignant transformation is low, and is inherent in factors such as advanced age, radiotherapy, large size and recurrent tumours [5]. Signs that should attract attention are pain, peripheral facial paralysis and, in advanced stages, lymph node metastases, skin infiltrations, ulcerations, massive tumour haemorrhage, dysphagia and mandibular invasion [11].

This is why advanced imaging is so useful. CT scans with IV contrast can highlight mandibular or temporozygomatic bone lesions, and the gold standard is cervical magnetic resonance imaging, which is the most sensitive in detecting malignant tumours and allows clearer delineation of tissue planes [11].

Tissue sampling procedures, including fine needle aspiration (FNA) and needle biopsy, are associated with very low rates of tumor dissemination. PAF pinpoints malignancy with an approximate sensitivity of 90%, while needle biopsy is more invasive, but gives a histological and diagnostic accuracy of around 97%. The need for immunohistochemistry to delineate the various cellular components [5]. In our diagnostic approach we used CT, which showed a heterogeneous, multi-partitioned right parotid tissue mass measuring 119/104 mm, with no osteolytic lesion, and histology after extraction confirmed the diagnosis of a giant pleomorphic adenoma of the parotid gland.

The management of APs of all types is exclusively surgical, with giant APs involving the superficial lobe of the parotid gland treated either by superficial parotidectomy or by ECD [1]. In this study, the patient was managed surgically by ECD, thus avoiding iatrogenic damage to the facial nerve.

Patrick et al. had found a postoperative complication of Housse and Brackmann grade II temporary facial paresis in the context of total parotidectomy with preservation of the facial nerve, and total facial paralysis in the context of total parotidectomy without preservation of the facial nerve in a

case of invasive carcinoma within a pleomorphic adenoma [10]. Extracapsular dissection combined with postoperative follow-up is a shorter, less costly and safer alternative to traditional superficial parotidectomy in the management of benign trimmed lesions [1]. The longest recurrence-free follow-up for giant parotid AP was 5 years [14]. This was even lower than the 7-year recurrence-free follow-up observed in our case.

4. Conclusion

Giant parotid AP is rare and even less common in men than in women. Thus the present case is reported for its long-standing clinical presentation, successful surgical management by ECD without postoperative complications or facial nerve deficit, atypical histological presentation and long-term follow-up without recurrence. Although generally benign, these tumors can disrupt the patient's daily life or develop into malignancy if not managed in time. Practitioners therefore need to be aware of these different characteristics in order to make a diagnosis that justifies appropriate therapeutic decisions. It also requires the implementation of public health procedures to raise patient awareness.

Abbreviations

PA	Pleomorphic Adenoma
ECD	Extra-Capsular Dissection
PAF	Fine Needle Puncture
TGS	Tumors of the Salivary Glands
TDM	Tomodensitometry

Author Contributions

Mohamed Saydi Ag Mohamed Elmeahdi Elansari: Conceptualization, Supervision

Mariam Sangare: Conceptualization, Project administration, Supervision

Lassana Keita: Conceptualization, Methodology, Writing – review & editing

Abdoul Wahab Haidara: Supervision, Visualization

Moussa Keita: Visualization, Writing – original draft

Mamadou Ouattara: Resources

Diaffe Drame: Resources

Fatogoma Issa Kone: Supervision, Visualization

Kadidiatou Singare: Supervision, Visualization

Conflicts of Interest

The authors declare no conflicts of interest.

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