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Case Report

Giant pleomorphic adenoma of the parotid gland with a history more than forty years without malignant transformation

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Abstract: Pleomorphic adenoma (PA) is the most common type of all salivary gland tumors. Although PAs are benign in pathology, they are malignant in biological behavior sometimes. The tumor could transform to malignancy as malignant pleomorphic adenoma (MPA), if the preexisting remains for long time or after multiple recurrent. Cases of giant pleomorphic adenomas (GPA) are rare, according to previous medical literatures, the majority of which were involving in the parotid gland. We presented a 73-year old woman with a giant pleomorphic adenoma in left parotid gland who had kept the mass for more than forty years and was successfully treated with surgery without any facial nerve injury.

Keywords: Giant pleomorphic adenoma, parotid gland, malignant transformation, facial function, parotidectomy

Introduction

Pleomorphic adenoma (PA) is the most common type of salivary gland tumors, and more than half arise in the parotid gland. This type of tumor is marked by slow-growing, painless mass, and tumor can achieve different sizes and weights if untreated. Diagnosis of PA is based on clinical and cytological features. It is a benign disease characterized with some malignant biological behaviors, like proliferation, recurrence, infiltration, diffusion and metastasis. Entire surgical excision and facial nerve preservation of the tumor are the chief principles of operation. Fear of surgery or sociocultural factors may lead to the delayed diagnosis and intervention, resulting in enormous size of the tumor. Giant pleomorphic adenomas (GPA) are not seen frequently and usually present in patients with a long history of the tumor, even several decades.

In this work, we described a case of a GPA arising in left parotid gland along with a more than four decade successfully treated and facial nerve function reservation with surgery.

Case report

Patient presentation

A 73-year-old woman living in a rural area admitted to our department in July 2016 with a giant and multilobular mass for more than forty years in her left parotid gland. During the last month, she felt interrupted needle-like pain during nights, while the first few years she didn't have any complaints. Before this present, she has undergone several times fine-needle aspiration in other departments, but the diagnosis was unknown. She did not receive any surgery because of economic hardship and fear of surgery. Physical examination revealed a multilobular, non-tender, painless mass involving in her left parotid gland area vertically extending from front cheek to retroauricular and horizontally from earlobe to the cervical region (**Figure 1**). It measured approximately 20 cm×13 cm×12 cm and covered by vascularized skin. However,

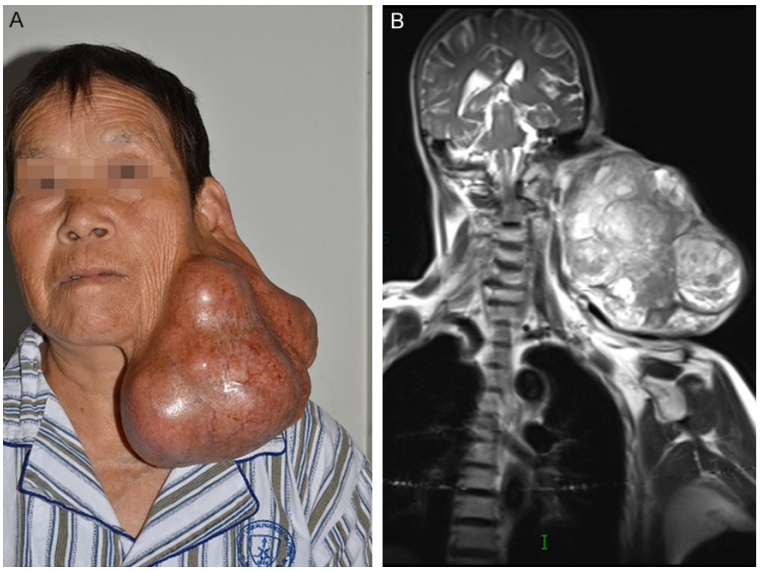

Figure 1. Preoperative clinical feature and MRI image of the patient. A. Clinical view of the giant mass from front aspect, extending down to the neck. B. MRI demonstrated a heterogeneous solid encapsulated tumor. The tumor originated from superficial lobe of the left parotid gland.

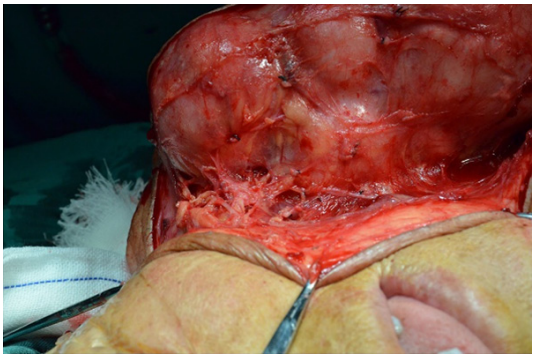

Figure 2. Main trunks and the branches of facial Nerve. A clear plane of dissection was found, and the facial nerve was detached carefully.

there were no abnormal facial nerve functions or pathological-enlarged lymph nodes.

Examination

Magnetic resonance imaging (MRI) revealed a giant, heterogeneous mass originated from the left parotid gland, comprising part of the tumor tightly adhered to left sternocleidomastoid muscle. The lesion had highly vascular components, which were mainly supplied by the left external carotid artery and venous drainage in the subclavian vein (**Figure 1**). Fine-needle aspiration was performed and reported as diagnosis of pleomorphic adenoma or mucinous adenocarcinoma showing suspicious salivary gland tumor showing many suspicious cancer cells.

Treatment

Based on clinical manifestations and examination, a superficial parotidectomy and facial nerve dissection was performed. Although the tumor with a large volume, a clear border between the tumor and health tissue was found, and the main trunks and branches of facial nerve were detached carefully (**Figure 2**). Tumor was resected completely and skin adjacent to the mass was reserved to provide sufficient tissue for incision closure and ear reconstruction.

Pathological examination

Macroscopically, the mass was measured 20 cm×12 cm×13 cm with an intact capsule. It weighted 2.5 kg. The cut section was heterogeneous, grayish white, nodular and gelatinous, composed of necrosis and cystic degeneration areas (**Figure 3**). Microscopically, the histopathological specimens consisted with a diagnosis of pleomorphic adenoma, comprising cystic-necrotic foci. No tumor cells residual surrounded the surgical margin. No identifiable evidence to support malignant transformation (**Figure 4**).

Postoperative appearance

The post-operative recovery was uneventful. She got obvious cosmetic outcomes and facial nerve functions were normal. Postoperative MRI showed no recognizable tumor residual (**Figure 5**).

Discussion

Parotid gland is the foremost site occurrence of PA, accounting for as many as 85% of all cases, most frequently affecting patients in the fifth to sixth decades of life [1, 2]. The tumor originates from not only superficial lobe of the

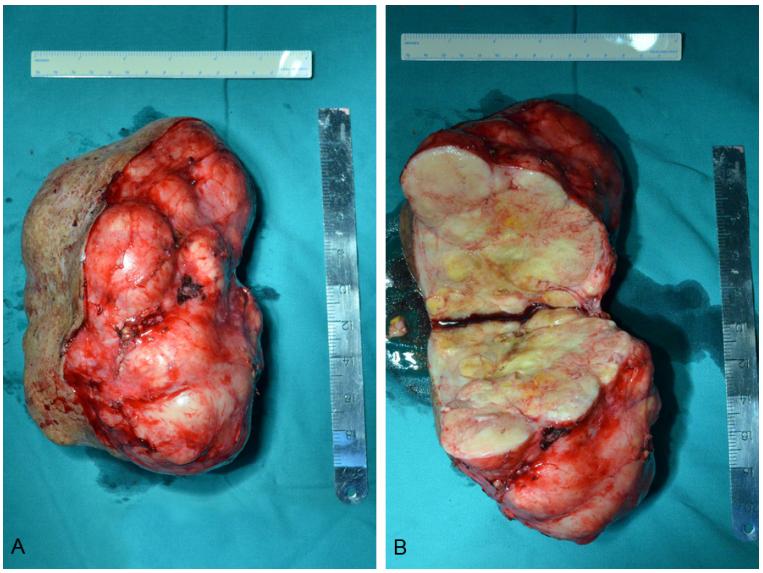

Figure 3. The macroscopical images of excisional mass. A. The mass measure 20×12×13 cm with intact capsule and weighted 2.5 kg. B. The cut surface was grayish white, nodular and gelatinous, with local necrosis and cystic degeneration.

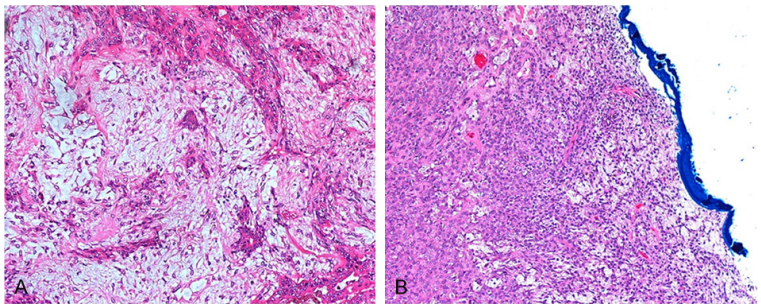

Figure 4. Post-operative histopathological image of the giant tumor. The histopathological result consisted with pleomorphic adenoma, comprising cystic-necrotic foci. No tumor cells residual surrounded the surgical margin. (H&E, original magnification ×100).

The tumor also has small protrusions that extend beyond the central masses. Based on these characters, it is a challenge for doctors to remove the tumor completely for these fragile "false capsule" and protrusions during surgery. In order to avoid rupture of the pseudocapsule and plantation of tumor cells, the mass has to be resected expansively with surrounding tissue [2].

Neglecting a benign parotid tumor causes aesthetic and social impacts, also gives rise to an increasing risk of facial nerve injury both tumor compression and operation. The bony and muscular deformity associated with such tumor is uniformly disfiguring and incapacitating.

In our case, the patient was a woman presented for a giant mass with a more than four-decade history of evolution and weighted 2.0 kg. Mostly, lack of medical knowledge and patient's negligence are considered as the most pertinent factors leading to the delayed treatment [6]. In our case, pervious economic hardship is regarded as the major reason.

parotid gland but also the deep lobe, even from the whole gland, while the former one is much common [3-5]. PA is generally considered a benign neoplasm that could assume gigantic dimensions if without treatment. Although uncommon, reports of giant PA have been described and the majority involved the parotid gland [1, 6-8].

PA shares some same biological features with malignant tumors for its special histological structures. PA is typically well demarcated from the surrounding tissue by a fibrous capsule, which is referred to as "false capsule" and varies both in thickness and completeness. The most important reason we focus on PA is that there is malignant transformation possibility in tumors with long-term evolution, recurrence, advanced age of patients or location in a minor salivary gland [6, 9], which ranges from 1.9%-23.3% [2]. Malignant changes include three different types: carcinoma ex-pleomorphic adenoma (CXPA, also called malignant mixed tumor), carcinosarcoma, and metastasizing PA, and the last twos are relatively rare [10]. The classic clinical history of CXPA is a slow-growing mass for many years, which experienced a sudden fast growth phase. Like other malignant tumors, CXPA could spread through direct extension, distance metastasis through

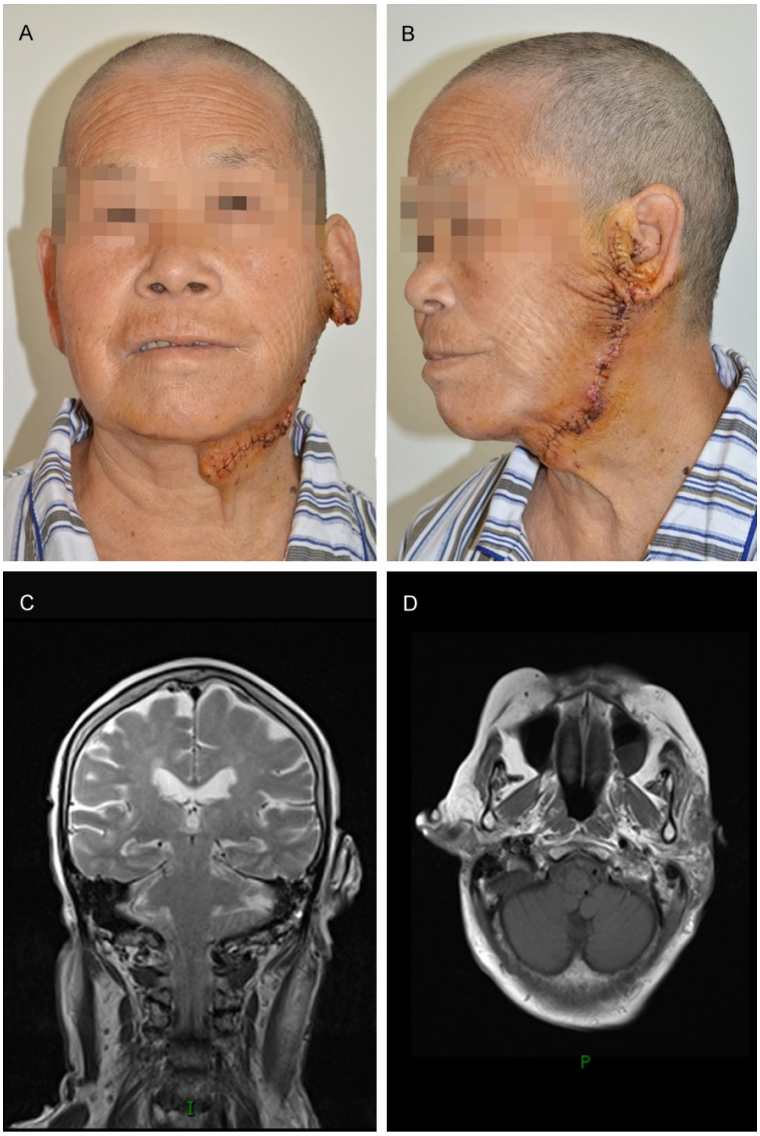

Figure 5. Post-operative clinical images and MRI images. The patient got obvious cosmetic outcomes. A. Front view. B. Left side photograph. MRI showed no obvious tumor residual. C. Axial section. D. Coronal section.

lymphatic vessels and hematogenously [11]. In our case, the patient presented characteristics for high risk of malignant transformation, but clinically and histologically here didn't provide such evidence.

Conclusion

Although it's generally considered that the majority of all GPA remain non-malignant, it should serve to remind us that the clinical course of these masses would be far from benign [1, 7, 9, 10, 12]. In conclusion, early diagnosis and management of the PA are desirable if possible. As for treatment, completely excision of tumor with surrounding tissue is necessary prevention against recurrent or distant metastasis [2, 13-15]. Otherwise, function impairment, aesthetic problems and social handicaps will accompany with delays in treatment.

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Disclosure of conflict of interest

None.

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