



Giant Pleomorphic Adenoma of Parotid Gland in Saudi Arabia: a Rare Case Report

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Abstract

Pleomorphic adenoma is the most common type of all benign and malignant salivary gland tumors, involving more frequently the parotid gland. It is a benign painless and slowly growing tumor that if not treated, can continue growing to reach a massive size. We report a case of a giant pleomorphic adenoma of right parotid gland in a 25-year-old mentally challenged female, being the largest in size to be excised in Saudi Arabia in recorded literature measuring 18 × 17 × 11.5 cm and weighted 1.5 kg. Clinical examination revealed a giant mass on the right side of the face without any sign of facial nerve injury. The tumor was excised by superficial parotidectomy with preservation of the facial nerve. The patient was followed up to 1 year with good postoperative results and no signs of facial nerve injury or recurrence. Neglect, lack of information, or medical resources and fear of surgery allow such tumors to grow bigger in size which might complicate the treatment and increase the risk of malignant transformation.

Keywords Pleomorphic adenoma · Tumors · Parotid gland · Neck mass · Giant · Saudi Arabia

Abbreviations

PA Pleomorphic adenoma

Introduction

Salivary gland tumors are relatively rare, comprising less than 3% of all of head and neck neoplasms. Around 70% of all salivary gland tumors take place in the parotid gland where approximately 85% are benign [1]. Pleomorphic adenoma (PA), also known as benign mixed tumor, is the most common

salivary gland tumor, constituting up to two-thirds of all salivary gland neoplasms [2].

PA is usually located in the parotid glands (85%), minor salivary glands (10%), and the submandibular glands (5%) [3], and it presents clinically as an asymptomatic, slow-growing mass, without facial nerve involvement [4, 5]. Mostly, tumors of the parotid gland occur in the superficial lobe, but in occasional cases, they may involve the deep lobe and the parapharyngeal space [6]. Minor salivary gland tumors are frequently encountered on the palate, followed by the lip, cheek, tongue, and floor of the mouth [4, 7].

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PAs are best treated by a wide local excision with good safety margins and follow-up for at least 3–4 years [8]. They usually vary in size from 2 to 6 cm when resected [4]. Since PAs are asymptomatic and slow-growing benign tumors, they can reach massive sizes with time if left unoperated.

Most cases of giant PA of parotid gland were reported before 1980's, yet some cases have been recently published [4, 9, 10]. These published cases of giant PA have occurred in the parotid gland, presenting as an irregular multinodular mass that can weight more than 8 kg, and the weight seems to increase with duration of the tumor [1, 11]. Such cases of long standing giant tumors are treated by parotidectomy, but the possibility of positive surgical margins and malignant transformation must be considered [12].

According to the literature, giant PA of parotid gland mostly occurs in elderly patients, and no similar cases have ever been reported in Saudi Arabia or Arab countries. So, the purpose of this article was to report a rare case of a giant PA of parotid gland in a young female patient which is the first case to be reported in Saudi Arabia with a follow-up history of 1-year with signs of recurrence or malignancy.

Case Report

A 25-year-old female, mentally challenged, presented to the Oral and Maxillofacial Surgery Department, King Saud Medical City, Riyadh, Saudi Arabia, complaining of an asymptomatic and slow-growing mass of the right face and neck with a history of 10-year duration. The patient has intellectual disability, and therefore, the history was obtained from the mother. The mother declared that the mass had started to grow about 10 years ago and had slowly enlarged over the years with no history of weight loss, loss of appetite, fever, or painful episodes. She also reported a low socioeconomic status, and her daughter was very uncooperative to be brought to the hospital, and now she forced her to come when the mass has become too large to exercise normal life activities. Family and genetic histories were unremarkable.

Clinical head and neck examination showed a remarkable right facial and neck asymmetry in the form of a giant mass which was firm in consistency, multilobulated and irregular in shape, with sessile base, not compressible and nontender on palpation. There were no signs of facial nerve palsy, and the overlying skin was mildly erythematous with prominent vessels seen all over the swelling. No bleeding or discharge was observed. The mass was extending inferiorly from ala tragus line until the lower border of right mandible and from the right posterior auricular region until 2 cm lateral to the corner of right lip measuring approximately 25 × 25 × 15 cm. This mass was also fixed to the underlying tissues with no palpable regional lymph nodes (Fig. 1). Clinical intraoral examination was unremarkable. Computed tomography (CT) scan with contrast and

magnetic resonance imaging (MRI) of the neck were performed to evaluate the extent of the mass (Figs. 2 and 3).

A provisional diagnosis of benign tumor of the right parotid gland, mostly pleomorphic adenoma, was taken into consideration. Fine needle aspiration (FNA) biopsy was performed, and histopathology results confirmed the suspicion of pleomorphic adenoma.

Ethical approval from Research Centre of King Saud Medical City was obtained under Reference Number H2RI-17-Dec18-01. Informed consent was taken from the patient's parents. On the day of surgery, the patient was prepared for regional flap in case the remaining soft tissue and skin would not be sufficient for direct closure. Excision of the superficial lobe of the right parotid gland along with the tumor mass was done under general anesthesia. A modified Blair's incision was made to the right preauricular region persevering posterior pedicle; platysma muscle and superficial musculoaponeurotic layer were dissected. The main trunk facial nerve was identified and preserved following a retrograde approach. Facial nerve branches were dissected from the overlying parotid mass. The tumor was resected after separation from the facial nerve and the masseter muscle. Despite the size of the mass, a clear plane of dissection was found. The excised mass measured 18 × 17 × 11.5 cm and weighed 1.5 kg. Finally, excision of the redundant skin was performed then followed by wound closure (Fig. 4).

Histopathology results showed triphasic tumor with ductal (epithelial), myoepithelial, and stromal components which is typically myxoid/chondromyxoid. These findings are consistent with pleomorphic adenoma of right parotid gland (Fig. 5).

The patient was followed up to 3-months and 1-year post-operatively and surgical site showed good healing with no signs of recurrence, facial nerve palsy, inflammation, or infection. Patient showed overall good esthetic results and reduction of the residual skin with improved overlying skin color in the 1-year follow-up (Fig. 6).

Discussion

Pleomorphic adenoma (PA) is a benign salivary gland tumor. It is considered the most common salivary gland tumor with a predominant site of occurrence in the parotid gland, affecting patients more frequently between the fifth and sixth decades of life [1].

An interesting finding to report in our case is the young age of the patient which is 25 years old; whereas, most of the published cases were of patients of older ages starting from the fifth decade [4, 12–15]. Few studies have reported PA in young adults [16, 17].

Despite that cases of giant PAs are uncommon, they have been described in the literature where the majority involved the parotid gland. The first reported case of giant PA of the

Fig. 1 Clinical pictures of the mass located in the right facial and neck area



parotid gland was in 1863 by Spence, who described a mixed tumor measuring > 1 kg [18]. In 1956, Short and Pullar reported another case of 2.3 kg [19]. The largest tumor has been reported in 1956 by Frylinck in a 63-year-old who weighed 26.5 kg [20]. In 1989, Schultz-Coulon reported another case of a 78-year-old lady with a tumor weight of 4.5 kg [21]. In 1998, Buenting reviewed the 10 largest PAs of parotid gland published in the literature and found an average tumor weight of 7.8 kg; nine out of 10 cases occurred in females, with a mean age of 56 years. The author also reported his own case which was the 5th largest PA reported in the literature (6.85 kg) [4]. In 2008, Takahama et al. present a case of a giant pleomorphic adenoma of parotid gland in a 78-year-old man with a history of more than 30 years which weighted 4 kg [12]. Four years later, Jain and coworkers published a case of pleomorphic adenoma of the parotid gland in a 50-year-old female patient with tumor weight of 1.8 kg [14]. In 2015, Tarsitano et al. reported the largest pleomorphic adenoma in recent literature of an 83-year-old man which weighted 7.4 kg [15]. In the same year, Sajid reported the same tumor of a weight of 1.8 kg in a 47-year-old man [22].

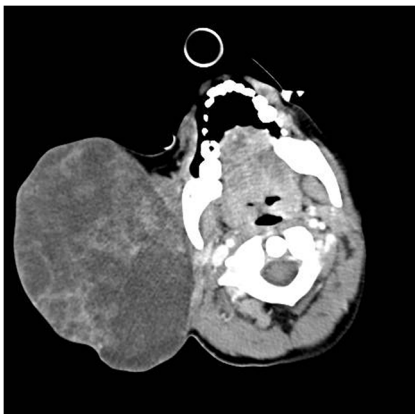


Fig. 2 Axial section of CT scan with contrast showing extent of the right lobulated heterogenous soft tissue mass

In the present case report, the patient was a young female who presented with a 10-year history of huge mass in her face and neck which weighted 1.5 kg when resected, being the largest parotid tumor to be ever reported in Saudi Arabia.

As clearly shown in the literature, all cases of giant PAs of parotid gland belong to old age group patients, which contradicts the present case where our patient is only 25 years in age with a 10-year history of evolution, which means that the tumor started growing when she was around 15 years old. In most of the cases reported in the literature, the patient's lack of information and neglect were suggestive for the tumor's long course and treatment delay. In the present case, it must be considered that the patient was mentally challenged and had a low socioeconomic status. The patient's delay to treatment could be attributed to difficulties reaching medical facility or to fear of surgery. Moreover, considering the mass was totally asymptomatic; this might have given the family sort of relive and hence delayed their visit to the hospital. Usually, mentally

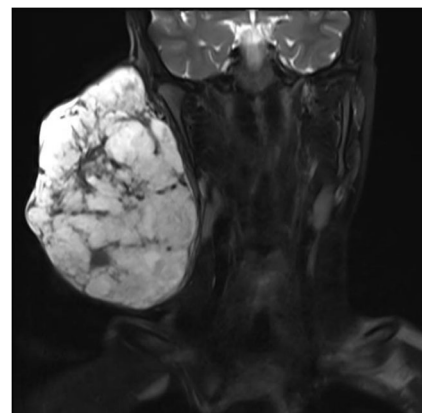
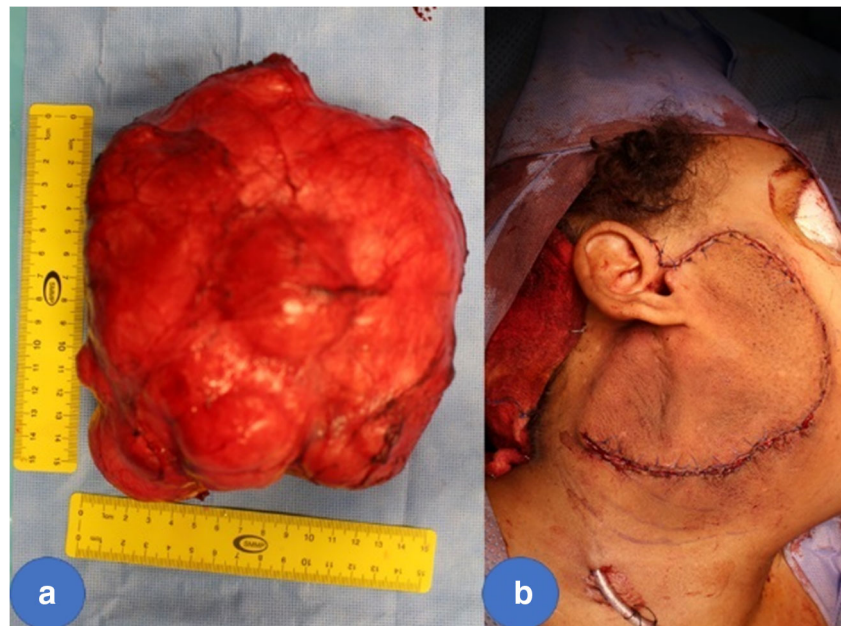


Fig. 3 Coronal section of MRI scan of neck showing a right lobulated heterogenous mixed tissues of solid and multiloculated cystic changes with enhancement to cystic wall, measuring around 12 x 10 x 12 cm in anteroposterior, transverse and vertical dimensions, and extending from the parotid area laterally without invasion to parapharyngeal spaces or retromandibular region

Fig. 4 **a** Clinical picture of the excised mass. **b** Immediate postoperative picture after wound closure



challenged individuals can be subjects to aggravation of diseases due to either their inability to complain or neglect from their guardians.

Postponing the treatment of even a benign parotid tumor carries a high risk of facial nerve injury when treatment is performed. In the current case, however, despite the large tumor size, tumor was resected carefully, and facial nerve was protected all along the procedure which resulted in no injury.

The incidence of malignant transformation in PAs usually ranges from 1.9 to 23.3% [23]. The risk of malignancy usually increases in tumors with long-standing development, recurrence, increased age of the patient, and location in a major salivary gland [24]. Some authors suggested that when the tumors have less than 5 years of growth, the risk of malignant transformation will be 1.6%, while it increases to 9.5% for those presenting for more than 15 years [25]. A case of a giant PA with malignant transformation with the classical history of

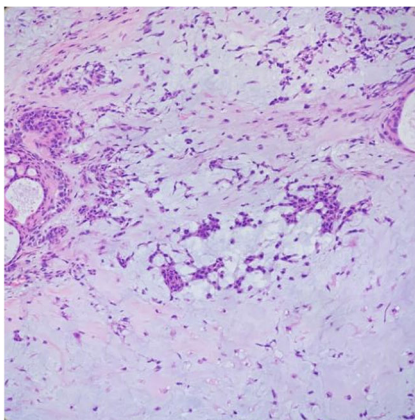


Fig. 5 Histopathology results of the right parotid tumor

a slow-growing mass for many years, with a recent rapid growth phase, was reported in 2005 in a 72-year-old lady with a slow growing parotid lesion for 20 years, with a fast increase in the last 3 months [10].

In our case, the patient did not present all the characteristics for an increased risk of malignancy, and clinically and histologically, there was no such evidence.

The treatment of choice for PAs of the parotid gland is superficial parotidectomy with preservation of the facial nerve [26] which was precisely followed in this case.

The bony, muscular, and skin deformity associated with such giant tumors is uniformly disfiguring and incapacitating, and the postoperative esthetic results might be compromised in which necessitate the need for more cosmetic intervention. In the present case, however, despite that the overlying skin of the surgical site sagged postoperatively in the 3-month follow-up, the final results after 1-year is esthetically acceptable for the patient and her family. Thus, a secondary cosmetic surgical intervention might not be indicated in this case. The family of the patient was very satisfied of the results, and they reported that it positively changed their daughter's life.

Conclusion

Nowadays, giant parotid gland tumors are rarely seen due to increased level of knowledge and the facilitation to medical institutions. Yet, they still do occur. These giant tumors are more common in females and may enlarge over a period of several decades. Patients' lack of awareness in addition to fear of surgery seems to be the main factors for such long standing lesions. Early diagnosis and treatment of parotid PAs is extremely essential to avoid any risk of malignant

Fig. 6 **a** 3-month post-operative follow up clinical picture. **b** 1-year post-operative follow up clinical picture showing good healing of the surgical site and general aesthetic improvement



transformation or possible facial nerve injury and to result in an excellent cosmetic outcome.

Compliance with Ethical Standards

Conflict of Interest The authors declare that they have no conflict of interest.

Ethical Approval Ethical approval from Research Centre of King Saud Medical City was obtained under Reference Number H2RI-17-Dec18-01.

Informed Consent Informed consent was obtained from the patient's parents.

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