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Giant Pleomorphic Adenoma of Submandibular Gland

Shi Nee Tan,¹ Rubinderan Muthusamy²

¹Department of Otorhinolaryngology, Head and Neck Surgery Hospital Lahad Datu, Jln Lahad Datu - Tawau, Sabah, Malaysia

²Department of Oral Maxillofacial, Hospital Lahad Datu, Jln Lahad Datu - Tawau, Sabah, Malaysia

ABSTRACT

Benign tumour of submandibular glands is rare, however amongst those which happen, pleomorphic adenoma (PA) is the most common type. Although it has been reported in several case reports, giant PA is extremely uncommon. These tumours are usually painless and slow growing but when neglected on some occasions, the long-standing size and weight can augment surgical excision. An unusual case of a giant submandibular PA in a 46-year-old male patient was reported and discussed here. Patient usually tend not to seek out for medical treatment till it became gigantic. Neglecting such tumors can cause unsightly appearance due to the gigantic mass and in a worse case scenario may even cause airway obstruction if it continues to enlarge.

Keywords: Pleomorphic adenoma, submandibular gland neoplasm, neck neoplasm



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Address for correspondence:
Dr. Rubinderan Muthusamy.
Department of Oral Maxillofacial, Hospital Lahad Datu, Jln Lahad Datu - Tawau, 91100 Lahad Datu, Sabah, Malaysia

Phone: +60193463799

E-mail: rue_d23@yahoo.com

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INTRODUCTION

Tumor of salivary gland commonly is pleomorphic adenoma (PA).^[1,2] Most of the time, benign tumor of salivary gland is painless and can grow progressively into big tumor. PA most commonly occur in the parotid gland (80%) followed by submandibular (12%), sublingual and other minor salivary glands. Pinkston JA et al., stated that about 75-85% of all PAs occur in the parotid gland with only 8% arising in the submandibular gland.^[3] The incidence rate being 2-3 cases per 100000 population. Therefore, the occurrence of this entity is considered uncommon. A unique case of an unusually long-standing tumor was discussed here to emphasize that how it can attain a gigantic size and if neglected and can cause unpleasant appearance in this patient. This giant submandibular gland swelling was completely excised without any complication.

CASE REPORT

A 46-year-old male non-Malaysian patient presented to the department of Oral Maxillofacial Clinic in Hospital Lahad Datu recently with complaint of a large mass on the left side of the neck for the past 12 years. The anterior and lateral views of the left submandibular mass are shown in Figure 1a and 1b. There was no history of difficulty in breathing, odynophagia, dysphagia, and facial disfigurement. Physical examination showed a lobulated mass, non-tender, firm in consistency from the right submandibular region. The temperature and color of the overlying skin was normal, with no ulcerations or hemorrhaging. Movement of the tumor mass can be palpated bimanually both intraorally (in the region of the left submandibular gland) and extra orally.

The patient's regional lymph nodes were not enlarged. Fine needle aspiration cytology (FNAC)

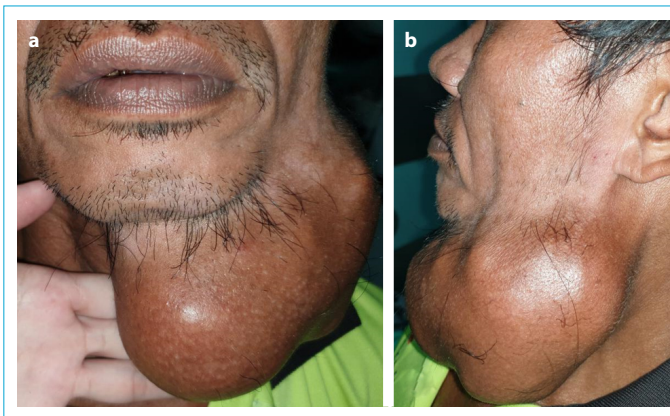


Figure 1. (a) The anterior view of the left submandibular mass. **(b)** The lateral view of the left submandibular mass.

of the mass was inconclusive. Computed tomography of the neck revealed a large mass arising from left submandibular gland measuring 9.1 cm x 8.9 cm x 9.1 cm with clear margin near the carotid sheath and sternocleidomastoid muscle. The anterior and coronal views of the mass located at left submandibular in the computed tomography scan are shown in Figure 2a and 2b.

The tumor was surgically excised. Intraoperatively, noted there are a lot of dilated vessels due to stretching of the capsule as a result of the enlargement of the mass. The mass was completely excised and weighed 450 gm. The removed mass in the operation is shown in Figure 3. Post-operative was uneventful. The patient recovered well and was discharged on day 5 post-operative. On follow-up examinations no recurrence was observed during the five months of post-surgery. Histopathological report came back as PA.

DISCUSSION

PA is the most common tumor in the salivary gland with the main site being in parotid gland.^[4,5] It has also been reported as the common tumor occurrence in submandibular region. However, giant tumor of submandibular gland is very rare.

Manish Gupta et al and Sladjana et al have reported isolated case report of giant submandibular gland presenting as a large neck swelling.^[6,7] It is commonly involved male predominance and usually at age of between 20 to 40 years.^[2,7] PA tumor usually described as a slow growing firm mass which is not aggressive. Almost all the people with these tumors are asymptomatic. Due to painless slow growing mass, patients tend to neglect the growth and present later in life. It is not uncommon for a patient with a long-standing tumor to present after more than 20 years of the tumor growth.

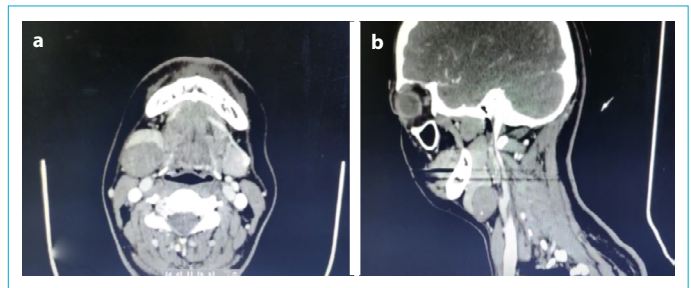


Figure 2. (a) The axial view of well-defined border of the left submandibular mass in the computed tomography scan. **(b)** The coronal view of the mass located at left submandibular in the computed tomography scan.

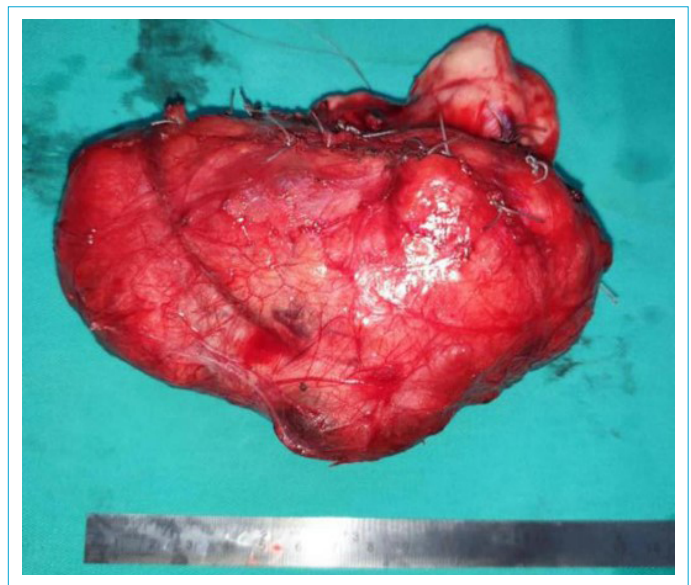


Figure 3. The removed mass in the operation.

The patient presented late to us, due to patient come from poor socio-economic background, negligence, lack of awareness and the fear of surgery. Furthermore, the patient lived alone and was away from his family. His tumor mass was very large however, it did not cause any significant compression to vital structures, and he was asymptomatic. Therefore, it did not warrant him to seek for any medical attention and hinder his reluctance.

Only a few cases have been reported on an extremely rare giant PA of the submandibular gland.^[6,8] FNAC and imaging studies such as computed tomography is good enough to provide a reasonable diagnosis. Although gigantic size, being a confirmed benign tumor feature of PA, it is usually well demarcated from the surrounding tissue by a pseudo-capsule and if this fibrous capsule is completely excised, these tumors could be cured with solely surgical excision.

Although rare malignant transformation, the incidence in PA ranges from 1.9% to 23.3%.^[5,9] This risk increases in

tumor of long history of appearance, recurrence and increased age of the patient.^[10] Great enough, our patient did not have any malignant features although long standing tumor, as the tumor only include benign epithelial and myoepithelial tissues intermixed with myxoid component which was confirmed by histopathology report.

This case highlights to us that, negligence, lack of awareness and fear of surgery among patients are the main reasons as to why patients let the tumors grow for a long period of time. Complete excision remains the main stay of treatment. Although tumor was grow slowly and painless, physicians should be aware that malignant transformation is possible in cases of huge and long-standing PAs of the salivary gland. Moreover it is also justifiable for surgical excision due to aesthetic appearance and social morbidity.

CONCLUSION

Giant PA of the submandibular glands rarely occurs, and its diagnosis should be made cautiously. Total excision is the main-stay treatment, and physicians should be aware of malignant transformation in cases of huge and long-standing PA.

Disclosures

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R.M.; Literature search – S.T., R.M.; Writing – S.T., R.M.; Critical Review – S.T., R.M.

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