

Case Report

Malays. j. med. biol. res.



Parapharyngeal Space Pleomorphic Adenoma: Common Tumor Type at an Uncommon Site

Shalina Kaur^{1*}, Noor Dina Hashim², Afiza Izura M Mohammad Sofi³, Gurdev Singh Naraman Singh⁴,
Khairil Amir Sayuti⁵

¹Pusat Perubatan, Universiti Kebangsaan Malaysia, MALAYSIA

²Doktor Pakar, Otorinolaringologi, Surgeri Kepala & Leher, Pusat Perubatan, Universiti Kebangsaan Malaysia, MALAYSIA

³Department of Otorhinolaryngology, Hospital Raja Perempuan Zainab II, Kota Bharu, Kelantan, MALAYSIA

⁴Universiti Sains Malaysia, Kubang Kerian, Kelantan, MALAYSIA

⁵Department of Radiology, Hospital Universiti Sains Malaysia, & School of Medical Sciences, Universiti Sains Malaysia, Kubang Kerian, Kelantan, MALAYSIA

*Email for Correspondence: shalinakaurgill86@gmail.com

ABSTRACT

Pleomorphic adenoma provides as much as 40-70% of tumors in the minor salivary gland with palate being the most frequent area engaged. Head and neck tumors in parapharyngeal space amounts to less than 1%. We share a case of pleomorphic adenoma primarily arising in parapharyngeal space. This report highlights clinical features, pathology, radiological findings, and treatment of this tumor.

Keywords: Imaging, parapharyngeal space tumor, Pleomorphic adenoma, uncommon site

Manuscript Received: 13 October 2020

Revised: 16 December 2020

Accepted: 10 January 2020

This article is licensed under a Creative Commons Attribution-NonCommercial 4.0 International License.

Attribution-NonCommercial (CC BY-NC) license lets others remix, tweak, and build upon work non-commercially, and although the new works must also acknowledge and be non-commercial.



INTRODUCTION

The most usual benign tumor of salivary glands is pleomorphic adenoma. It comprises of about 3–10% of all the tumors of head and neck region and 50% of the major and minor salivary gland neoplasms. Intraorally, the most common presenting site is hard palate (42.8–68.8%) followed by upper lip (15–20%), and cheek (5.5%). Other sites consist of throat (2.5%), retromolar region (0.7%) and small percentage from floor of mouth and alveolar mucosa. Other ectopic presentations are along the stenson's duct, accessory parotid tissue, and parapharyngeal space (Sharma et al., 2016).

Parapharyngeal space (PPS) pleomorphic adenoma can appear primarily or may appear from inner lobe of the parotid gland and extend into the PPS (Rodríguez-Ciurana et al., 2000). PPS bear a resemblance to an inverted cone shape with concave faces. Anteriorly this space is placed posterior to infratemporal fossa, medially bounded by nasopharynx and the lateral pharyngeal wall, posterior boundary is the vertebral column, and laterally is ramus of the mandible (Akin et al., 2014).

Frequently there are delay in diagnosing PPS tumor because it enlarges internally within the space without any visible neck swelling. Therefore, the tumors tend to be very large and may cause compression symptoms. Managing these tumors represents a challenge to surgeons because of the adjacent vital neurovascular structures (Akin et al., 2014).

CASE REPORT

A 37-year-old female presented with slow growing painless intraoral swelling for a week duration. She complains of mild discomfort during swallowing with no symptoms of airway obstruction. There was no prior history of fever and sore throat. Neck examination revealed no neck nodes.

On intraoral examination, there was a firm bulge with a smooth overlying mucosa at right peritonsillar area crossing the midline, pushing the uvula to contralateral side. Flexible nasopharyngolaryngoscopy (FNLPs) showed a medialised right lateral pharyngeal wall extending from nasopharynx to base of tongue. No associated cranial nerves palsy observed.

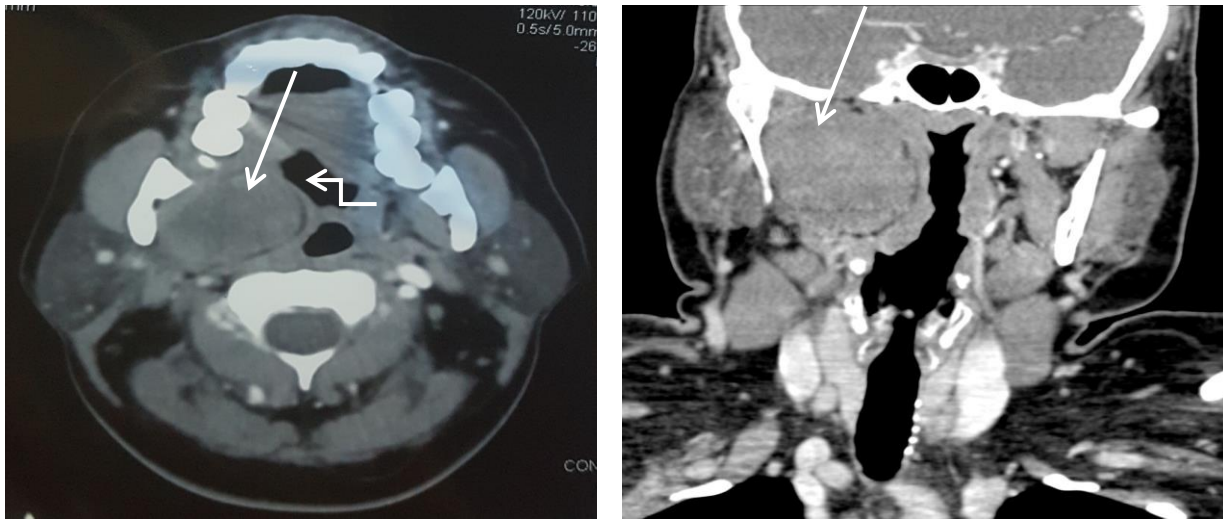


Figure 1 & 2: CT imaging of head and neck revealed a lesion arising from right parapharyngeal space (white arrow) measuring 3.7cm (AP) x 4.4 cm (W) x 4.8cm (CC). Medially the mass causes narrowing of oropharynx (crooked arrow), obliterates the right Rosenmüller fossa and Eustachian tube with bulging of buccal mucosa medially. No clear fat plane was seen with deep lobe of right parotid gland which may suggest involvement (white arrow head).

An intraoral biopsy was performed. Histopathological examination (HPE) revealed a pleomorphic adenoma.

Superficial parotidectomy and excision of parapharyngeal mass via modified bailey and cervical incision was planned. Intraoperative findings showed an isolated encapsulated parapharyngeal mass extending towards the submandibular space. The submandibular gland was removed to provide access. As the tumor did not infiltrate the deep lobe of parotid, thus it was spared.



Figure 3: On gross examination the dimension of the tumor was 6 x 4 x 3 cm with a lobulated appearance and weighs 38gram.

Histopathology established the diagnosis as pleomorphic adenoma. Patient was discharged well post operation. No recurrence was observed at 1 year post-operative follow up.

DISCUSSION

Head and neck neoplasm constitute less than 0.5% by PPS tumors. Of these, pleomorphic adenoma is the commonest benign tumor (40%) followed by Schwannoma and paraganglioma (Varghese et al., 2003). Pleomorphic adenoma may emerge from the parotid gland, deep lobe part and enlarge into the PPS (Ladeinde et al., 2004).

It is very rare to diagnose a pleomorphic adenoma arising primarily in PPS (Varghese et al., 2003). The emergence of it is probably from displaced or abnormal salivary gland tissue in a lymph node. Frequently they present with a large tumor as they may be misdiagnosed when being small or patient have been asymptomatic.

Patients with PPS tumor may have trismus, otalgia, neuralgia, palsies of 9th, 10th or 11th cranial nerves although generally painless submucosal swelling may be the only complaint from a patient with benign minor salivary gland tumor of the buccal cavity (Varghese et al., 2003).

Imaging is essential to diagnose PPS tumors, as it's difficult clinically. In radiological point of view, Computed Tomography (CT) scan can be performed to evaluate the lesion and relation to the surrounding structure (Izzo et al., 2006).

Lesions in PPS can arise within the space itself or from its surrounding structures. CT scan aids localizing this mass by establishing the relationship of the lesion with the parapharyngeal fat. A parotid area lesion displaces the fat anteromedially, masticator area lesion displaces the fat posteromedially, carotid area lesions displaces the fat anteriorly, mucosal area lesions displaces it posterolaterally and retropharyngeal area lesions displaces it anterolaterally. It can be very difficult to say if the lesion is arising from the deep lobe of the parotid gland or the parapharyngeal space itself. The best way could be the identification of fat plane, between the mass and the deep lobe. Deep lobe parotid mass usually causes expansion of the stylomandibular tunnel with displacement of the styloid process posteromedially (Rajagopal et al., 2008). CT and Magnetic resonance imaging (MRI) have equal benefits in identifying the lesion to the pre or poststyloid space however MRI is superior to CT in its ability to ascertain the soft tissue characteristics of PPS tumors.

Magnetic resonance angiography (MRA) could be done for supportive evidence if high vascularity tumor is demonstrated on CT scan, as MRA will be able to correlate the tumor to the vessels (Izzo et al., 2006). MRI with DWI (Diffusion weighted imaging) and ADC (Apparent diffusion coefficient) sequences can help to differentiate histological type of common parotid masses and characterize it. It's important to provide further imaging for the surgeon in the inconclusive cases in order to help them with their surgical approach thus reducing morbidity (Terra et al., 2017).

Surgery is the preferred modality for pleomorphic adenoma (Varghese et al., 2003). There are numerous surgical techniques for removal of parapharyngeal tumors such as transcervical, transparotid, transoral, or combination of these techniques. Mandibulotomy approach is reserved for more immense tumors as it delivers ample access to the PPS and its contents. However, the consequences depend on how immense the incision and manipulation. Benign tumors in this area are usually well encapsulated and with keeping mandibulotomy as a backup can always be resorted if needed. Most frequently excision of PPS tumors is via the transcervical approach (Papadogeorgakis et al., 2010). This approach gives adequate control over the neurovascular bundle in the neck and provides a better entry to the PPS. Larger tumors emerging from the deep parotid lobe require partial parotidectomy with preservation of mandibular nerve when possible (Basaran et al., 2014).

Submandibular gland may be excised or superiorly reflected to give better entry into the anteroinferior aspect of PPS. This approach is called cervico-submaxillary, where the submandibular gland is either removed or superiorly reflected to give better entry to the anteroinferior aspect of PPS.

After resection of tumor, it's vital to have adequate normal surrounding tissue margin to avoid local recurrence. Wide surgical resection margins for tumor's arising from PPS, might not be possible because of its close location to surrounding vital structures (Varghese et al., 2003).

In this patient, transparotid-transcervical approach was used to remove the tumor. This is a practical way to fully eradicate giant parapharyngeal space tumors with minimizing the risk of tumor cell spillage.

CONCLUSION

There shouldn't be a delay in diagnosing PPS tumor as patient may present with obstructive symptoms and this would be a challenge to surgeon's management. Pleomorphic Adenoma of PPS is rare and its management must start with meticulous preoperative diagnostic plan. Surgical approach should be well plan after imaging such as CT scan or MRI is done to provide guidance to surgeons. The tumor should be completely removed by selecting surgical method which is safe with least complications and low risk of recurrence.

REFERENCES

- Akın, I., Karagöz, T., Mutlu, M., Şahan, M., Önder, E. (2014). Pleomorphic Adenomas of the Parapharyngeal Space. *Case Reports in Otolaryngology*, vol. 2014, Article ID 168401. <https://doi.org/10.1155/2014/168401>
- Basaran, B., Polat, B., Unsaler, S., Uluhan, M., Aslan, I., & Hafiz, G. (2014). Parapharyngeal space tumors: the efficiency of a transcervical approach without mandibulotomy through review of 44 cases. *Acta otorhinolaryngologica Italica*, 34(5), 310–316. <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC4299156/>
- Izzo, L., Casullo, A., Caputo, M., Costi, U., Guerrisi, A., Stasolla, A., Basso, L., Marini, M., & De Toma, G. (2006). Space occupying lesions of parotid gland. Comparative diagnostic imaging and pathological analysis of echo color/power Doppler and of magnetic resonance imaging. *Acta otorhinolaryngologica Italica*, 26(3), 147–153. <https://www.ncbi.nlm.nih.gov/pmc/articles/PMC2639964/>
- Ladeinde, A. L., Adeyemo, W. L., Bamgbose, B. O., Ogunlewe, M. O. & Ajayi, F. O. (2004). Concurrent pleomorphic adenoma in parapharyngeal space and submandibular gland. *World Journal of Surgical Oncology*, 2(1), 1-5. <https://doi.org/10.1186/1477-7819-2-6>
- Papadogeorgakis, N., Petsinis, V., Goutzakis, L., Kostakis, G., Alexandridis, C. (2010). Parapharyngeal space tumors: surgical approaches in a series of 13 cases. *International Journal of Oral and Maxillofacial Surgery*, 39(3), 243-250. <https://doi.org/10.1016/j.ijom.2009.11.011>
- Rajagopal, K., Ramesh, A., Sreepathi, S., Shetty, C. (2008). CT Evaluation of Parapharyngeal Masses: Pictorial Essay. *The Internet Journal of Radiology*. 10(2), 1-12. <https://ispub.com/IJRA/10/2/3794>
- Rodríguez-Ciurana, J., Rodado, C., Sáez, M., & Bassas, C. (2000). Giant parotid pleomorphic adenoma involving the parapharyngeal space: report of a case. *J Oral Maxillofac Surg*. 58(10), 1184-1187. <https://doi.org/10.1053/joms.2000.9587>
- Sharma, N. K., Singh, A. K., Pandey, A., Verma, V. (2016). Pleomorphic adenoma involving soft tissue overlying the anterior border of ramus of the mandible: A rare ectopic presentation. *Journal of Oral Biology and Craniofacial Research*, 6(Supplement 1), S62-S64, <https://doi.org/10.1016/j.jobcr.2015.09.008>.
- Terra, G. T., Oliveira, J. X., Hernandez, A., Lourenço, S. V., Arita, E. S., & Cortes, A. R. (2017). Diffusion-weighted MRI for differentiation between sialadenitis and pleomorphic adenoma. *Dento maxillo facial radiology*, 46(1), 20160257. <https://doi.org/10.1259/dmfr.20160257>
- Varghese, B. T., Sebastian, P., Abraham, E. K., & Mathews, A. (2003). Pleomorphic adenoma of minor salivary gland in the parapharyngeal space. *World Journal of Surgical Oncology*, 1(2), 1-3. <https://doi.org/10.1186/1477-7819-1-2>