

Synchronous pleomorphic adenoma in parotid gland and parapharyngeal space: a case report and literature review

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Abstract

Synchronous pleomorphic adenoma in the parotid gland and parapharyngeal space are rare. We report a case of simultaneous pleomorphic adenoma in parotid gland and parapharyngeal space in a 65-year-old male presenting to the ENT outpatient department at Northwest General Hospital, Peshawar. The patient presented with a left parotid lump, intra oral examination of which revealed the left palatine tonsil to have been pushed medially. CT of the neck showed a completely separate lump in the left parapharyngeal space, and FNAC of the parotid lump was suggestive of mucoepidermoid carcinoma. The superficial parotid lump was excised first, and the parapharyngeal growth was accessed intraorally and excised. Histopathology revealed both the growths to be pleomorphic adenomas. It is important to raise awareness of synchronous salivary gland tumours, which are a rare occurrence, to highlight its optimal investigation to ensure complete surgical excision, and appropriate management.

Keywords: Parapharyngeal space, parotid gland, pleomorphic adenoma, salivary glands, synchronous.

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Introduction

Primary tumours of the parapharyngeal space are generally very uncommon, and account for only 0.5% of all head and neck cancers,¹ while synchronous tumours are even rarer. The most frequent occurrence is bilateral parotid tumours, followed by tumours in the parotid and submandibular glands.² Tumours in the parapharyngeal space may arise de novo or be an extension of a parotid gland tumour. The commonest pathological type arising in the parapharyngeal space is pleomorphic adenoma,³ 80% of which are benign in nature.¹ The following report

presents the case of a patient with synchronous tumours arising in the parotid gland and minor salivary gland in the parapharyngeal space. Literature review revealed only two previous reports of ipsilateral synchronous pleomorphic adenoma involving the parotid gland and the parapharyngeal space.^{4,5} The objective of this case report is to highlight the fact that although synchronous salivary gland tumours are rare, it is important that they be fully investigated to ensure optimal management and complete surgical excision.

Case Report

A 65-year-old male patient presented to the ENT outpatient department at Northwest General Hospital and Research Centre, Peshawar, in September 2020 with a painless swelling just over the left angle of the mandible. Apart from an increase in size over the few months since it was noticed, there were no additional symptoms. Physical examination revealed a lump on the left side of the neck measuring approx. 4 x 4 cm in size, which was non-tender and mobile. The lump appeared to be arising from the left parotid gland. Intraoral examination showed the left palatine tonsil and soft palate to have been shifted medially, although the patient denied any upper aerodigestive tract symptoms. No facial palsy was noted on examination.

An initial ultrasound of the neck confirmed a lump in the superficial part of the parotid gland. No comment was made on any deep lobe involvement. A fine needle aspiration cytology (FNAC) of the parotid mass showed the possibility of but did not confirm a muco-epidermoid carcinoma. CT of the neck was requested for further workup of the mass, which interestingly revealed two separate masses, i.e., one in the superficial part of the parotid gland, and another arising from the parapharyngeal space (Figure). The CT report showed that the two pathologies were separate entities and the parotid lump involved the superficial part of the gland with no deep lobe involvement. The intraoral growth seemed to be arising from the left parapharyngeal space. Based on the FNAC report and CT finding, surgical intervention was discussed with the patient who provided his informed consent.

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Figure: CT scan coronal and axial views showing the synchronous left parotid and parapharyngeal tumours in the same patient.

The surgical plan was to start with excising the superficial parotid lump first with macroscopic clear margins, while preserving the facial nerve. During surgery, care was taken to ensure that there was indeed no deep lobe involvement. After a successful superficial parotidectomy, the left parapharyngeal growth was accessed trans-orally. A cruciate incision was made on the left soft palate at the point of maximum prominence of the tumour. The growth was removed in its entirety. There was a breach in the capsule, however, the surgical team ensured that all pathologic tissue associated with the lump was cleared. A drain was put in the neck for the superficial swelling, while the intraoral incision was closed with Vicryl sutures.

The patient's post-operative recovery was uneventful with an intact and functioning facial nerve. He was discharged on the second post-operative day, when he

was able to manage fluids and soft diet. On histopathology, both these growths were of the same benign nature, i.e. pleomorphic adenoma. The superficial lump measured 6.3 x 5.5 x 3.5 cm and had been excised with clear margins. The patient was followed-up in the ENT clinic after three months and there was no sign of recurrence of the disease on either site and no evidence of facial weakness. Verbal consent was taken from patient for writing of this case report at the time of follow-up.

Discussion

Multiple primary salivary gland tumours are a rare occurrence, observed in only 3.4% of the cases with the commonest of them being Warthin tumour, followed by pleomorphic adenoma. The most prevalent multiple salivary gland tumours are bilateral parotid tumours.⁶ Primary tumours of the parapharyngeal space account for less than 0.5% of all head and neck neoplasms.¹ The prevalence of primary tumours in major and minor salivary glands was reported to be 94% and 6% respectively,⁷ making this an exceptionally rare case, presenting with synchronous pleomorphic adenomas in the parotid gland and minor salivary gland in the parapharyngeal space.

To the best of our knowledge, there have only been two previous reports of such cases, with synchronous pleomorphic adenomas arising in these precise locations. Kim et al⁴, in 2001, reported a similar case occurring in a 64-year-old female, while in 2009 Kefeli et al⁵ reported a case of simultaneous pleomorphic adenomas of the parotid and parapharyngeal space in a 58-year-old male patient. Pleomorphic adenoma in minor salivary glands are a rare entity. Other case reports mention pleomorphic adenomas, albeit not synchronous, arising in minor salivary glands in the palate,⁸ the upper lip,⁹ and the nasopharyngeal space.¹⁰ Approximately 70-80% of parapharyngeal space neoplasms are benign, and the most common of them are pleomorphic adenomas.³

Evaluation of tumours in the parapharyngeal space require a CT scan or MRI to ascertain the size and extent of spread of the tumour. Chances of recurrence and missing clinically unapparent tumours are higher in cases where preoperative CT and/or MRI evaluation of the tumour hasn't been carried out. Currently, FNAC is useful to obtain biopsy specimens in cases where malignancy is suspected, but its extensive use is contentious, and should be done only after imaging to rule out a vascular lesion. Although, if done with image-guidance, it proves to be a safe and easy tool, with an accuracy of 90-95%, and may aid the surgeon in making vital decisions regarding treatment options. Surgical excision is the treatment of choice of pleomorphic adenoma. Excision should include a wide margin of normal

surrounding tissue to prevent its local recurrence.¹¹

Conclusion

This case is a reminder for the occurrence of ipsilateral synchronous tumours in patients with salivary gland masses is which not out of the realm of possibility, and as such, the patients should be examined and investigated carefully and completely to ensure entire surgical removal

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Conflict of Interest: None.

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References

1. Fan S, Lin S geng, Zhang H qing, Li Q xing, Chen W xiong, Wang Y yuan, et al. A comparative study of the endoscopy-assisted transoral approach versus external approaches for the resection of large benign parapharyngeal space tumors. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2017; 123:157-62. doi: 10.1016/j.oooo.2016.09.010.
2. Singh A, Handa AC, Sachdev R. Synchronous parotid and homolateral submandibular gland pleomorphic adenoma. *Iran J Otorhinolaryngol.* 2019; 31:185-90.
3. Locketz GD, Horowitz G, Abu-Ghanem S, Wasserzug O, Abergel A, Yehuda M, et al. Histopathologic classification of parapharyngeal space tumours: a case series and review of the literature. *Eur Arch Otorhinolaryngol.* 2016; 273:727-34. doi: 10.1007/s00405-015-3545-5.
4. Kim SH. Simultaneous primary pleomorphic adenoma in parotid gland and minor salivary gland in the parapharyngeal space. *J Laryngol Otol.* 2001; 115:829-30. doi: 10.1258/0022215011909080.
5. Kefeli M, Akpolat I, Cengel S, Karagoz F, Kandemir B. Two synchronous and different salivary gland tumors located in the parotid gland and parapharyngeal region: a case report. *Turkish J Pathol.* 2009; 25:41-3.
6. Colangeli W, Kapitonov A, Facchini V, Zappalà M, Belli E. Synchronous Ipsilateral Pleomorphic Adenomas of Parotid and Submandibular Glands: An Unusual Finding. *Case Rep Otolaryngol.* 2020; 2020:1-5. doi: 10.1155/2020/8887867.
7. Mengi E. Salivary gland tumors: a 15-year experience of a university hospital in Turkey. *North Clin Istanbul.* 2020; 7:366-71. doi: 10.14744/nci.2020.57767.
8. Tandon A, Jaiswal R, Siddiqui S, Bordoloi B. Keratinizing pleomorphic adenoma: An unusual case report. *J Oral Maxillofac Pathol.* 2018; 22:S69-72. doi: 10.4103/jomfp.JOMFP_200_17.
9. Shah BA, Singh AP, Sherwani AY, Ahmad SM. Pleomorphic adenoma of the upper lip: A rare case report. *Natl J Maxillofac Surg.* 2020; 11:289-91.
10. Celik S, Kilic O, Zenginkinet T, Kalcioğlu MT. Nasopharyngeal Pleomorphic Adenoma: A Rare Case Report and Review of the Literature. *Case Rep Otolaryngol.* 2018; 2018:1-4. doi: 10.1155/2018/2481370.
11. López F, Suárez C, Vander Poorten V, Mäkitie A, Nixon IJ, Stojan P, et al. Contemporary management of primary parapharyngeal space tumors. *Head Neck.* 2019; 41:522-35. doi: 10.1002/hed.25439.