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CASE REPORT

Primary Pleomorphic Adenoma of Minor Salivary Gland in the Parapharyngeal Space

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ABSTRACT

Parapharyngeal space (PPS) lesions account for only 0.5% head and neck tumors and the majority of the minor salivary gland tumors are malignant. We report a case of pleomorphic adenoma of minor salivary gland in PPS as this is of a very rare occurrence. High index of suspicion and an adequate clearance of the tumor with a cuff of surrounding dispensable normal tissues are the key to successful treatment of such tumors.

Keywords: Minor salivary gland, Parapharyngeal space, Pleomorphic adenoma.

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INTRODUCTION

Parapharyngeal space (PPS) tumors are not very frequent, accounting for some 0.5% of neoplasms of head and neck. Most of these tumors (70–80%) are benign and 40 to 50% of these originate in the salivary glands, particularly the pleomorphic adenoma.[1] Pleomorphic adenoma in the PPS can develop *de novo* or may arise from deep lobe of the parotid and extend through the stylomandibular tunnel into the PPS.[2] The origin of *de novo* pleomorphic adenoma is probably from displaced or aberrant salivary gland tissue within a lymph node.[3] However, pleomorphic adenoma arising *de novo* in the PPS is extremely rare, which made us to report this case.

CASE REPORT

A 54-year-old female presented to our ENT outpatient department with gradually progressive painless swelling of the left upper neck of 1 year duration. The symptom was associated with progressive difficulty in swallowing, gradual change in voice, and snoring. There was no relevant past medical or family history and the patient took no regular medications. Neck examination revealed a firm swelling and fullness in the upper neck involving retromandibular region on the left side. On intraoral examination, there was a smooth firm bulge of the soft palate and left lateral pharyngeal wall, pushing the palatine tonsil medially. The swelling was bimanually palpable and ballotable. Posterior nasal examination showed the extension of the swelling into the nasopharynx. There was no significant lymph node enlargement in the neck. Clinical examination did not reveal involvement of any of the cranial nerves. With a clinical diagnosis of PPS tumor, a computed tomography (CT) scan was advised, which showed homogeneously enhancing tumor measuring 5 × 5 cm in the left PPS, extending from skull base to the hyoid bone medial to ramus of mandible pushing maxilla and medial pterygoid plate anteriorly. Punctate calcification within the mass was seen with a well-defined capsule (Fig. 1). Fine needle aspiration cytology from tonsillar region was consistent with benign mixed tumor of salivary gland origin. Transcervical approach was used to gain access to the left PPS, and the tumor was completely excised. On gross examination, the lesion was single 6 × 6 cm in size with a whitish and glistening surface. Histopathological examination showed a neoplasm having an admixture of epithelial and stromal components. Ducts lined by inner epithelial and outer myoepithelial cells were seen surrounded by a chondromyxoid stroma consistent with pleomorphic adenoma (Fig. 2). Postoperative period was uneventful. Patient was then sent for radiotherapy.

DISCUSSION

Tumors of minor salivary glands account for 22% of all salivary gland neoplasms.[4] Majority of them are malignant with only 18% being benign. Among these, pleomorphic adenoma is the commonest.[4] The most common site of pleomorphic adenoma of the minor salivary glands is the palate followed by lip, buccal mucosa, floor of mouth, tongue, tonsil, pharynx, retromolar area, and nasal cavity.[4-7] Pleomorphic adenoma of the PPS is rare.[8] *De novo* occurrence of the pleomorphic adenoma in our patients can arise from displaced or aberrant salivary gland tissue within a lymph node in the PPS as suggested by Varghese et al.[3] Another possible source of such tumor is deep lobe

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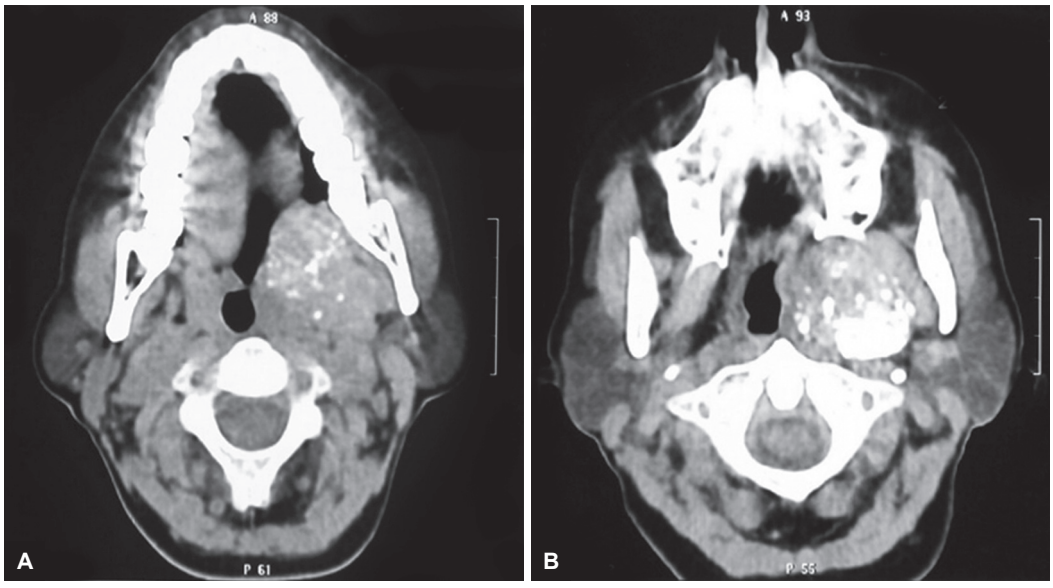

Figs 1A and B: Axial view of CT scan shows homogeneously enhancing mass in the left PPS having punctate calcification and a well-defined capsule. The mass is seen to be arising *de novo* and involving the pterygoid fossa

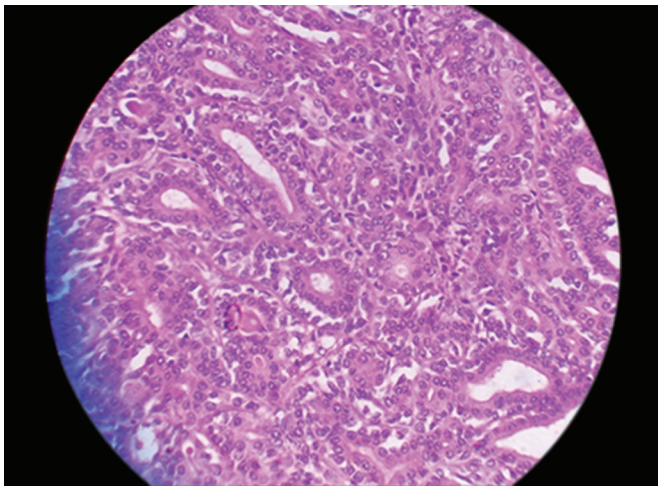

Fig. 2: Histopathology slide showing mixture of epithelial, myoepithelial, and stromal components

of parotid gland, in which case the tumor may present as a dumb bell tumor abutting the stylohyoid ligament.[9] Classical findings of benign parapharyngeal swelling are a submucosal swelling in the lateral pharyngeal wall with or without extension to retromandibular fossa or the submandibular trigone and bimanual ballot ability.[9-11] Most benign tumors of the minor salivary gland in the oral cavity present as a painless submucosal swelling,[4] and those from the PPS shows additional symptoms like otalgia, neuralgia, or palsies of ninth, tenth, or eleventh cranial nerves or trismus.

In tumors of PPS, CT scan (contrast enhanced) and magnetic resonance imaging (MRI) are important diagnostic tools.[8,12] Presence of intact fat plane helps in distinguishing benign tumors from malignant ones.[8] Extension of tumors from the deep lobe of a parotid gland is distinguishable from tumor arising *de novo* in PPS by a fine translucent line representing the compressed layer of fibroadipose tissue between the tumor and deep lobe of parotid.[13] In the investigation of PPS tumors, MRI has been shown to be superior to CT.[14-16] Pleomorphic adenoma has three malignant varieties: Carcinoma ex pleomorphic adenoma, carcinosarcoma and metastasizing pleomorphic adenoma. Carcinoma ex pleomorphic adenoma is a rare mixed tumor developing from the epithelial component of the pleomorphic adenoma.[17-19] The malignant transformation has been linked to recurrence and multiple excisions,[20] laminin and collagen IV deposition.[21]

Fine needle aspiration cytology is the modality of choice for obtaining biopsy sample for diagnosis.[2] Incision biopsy is no more advocated for salivary gland tumor due to seeding of tumor and subsequent multinodular recurrence.[2,15]

The treatment of pleomorphic adenoma is essentially surgical.[2,3,9] Though these tumors are apparently well encapsulated, resection of the tumor with an adequate margin of grossly normal surrounding tissue where possible is necessary to prevent local recurrence as these tumors are known to have microscopic pseudopod-like extension into the surrounding tissue due to "dehiscences" in the false capsule.[13] The approach of choice to the PPS to allow adequate removal of the tumor should meet two criteria: Wide intraoperative visibility for safe radical dissection and minimal functional and/or cosmetic after effects.[22] Traditionally, PPS surgery mainly uses the transcervical and parotid approaches.[9,22,23] The transoral approach is indicated for small, nonvascular tumors, as it offers poor exposition and does not give

adequate control in the event of hemorrhage.[24,25] More recently, in 1998, Goodwin and Chandler[26] considered this approach to give adequate access to the PPS, as it gives direct access to the PPS. The several kinds of mandibular osteotomies have been described in the literature to give excellent exposure.

CONCLUSION

Pleomorphic adenoma arising *de novo* in the PPS is of rare occurrence. High index of suspicion and an adequate clearance of the tumor with a cuff of surrounding dispensable normal tissues are the key to successful treatment of such tumors.

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