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Case Report

Metastasizing Pleomorphic Adenoma of the Parapharyngeal Space to the Cervical Lymph Node: A Case Report

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Abstract

Background: Pleomorphic adenoma (PA) is the most common benign salivary gland tumor. Despite its benign histological appearance, rare cases demonstrate metastatic potential without malignant transformation, termed *metastasizing pleomorphic adenoma (MPA)*. ***Case Presentation:*** Here in we report a case of a 32-year-old female with no comorbidities, presenting with a left parapharyngeal mass. MRI revealed a well-circumscribed, lobulated lesion inseparable from the deep lobe of the parotid, with heterogeneous enhancement and cystic/necrotic components. Histo pathological examination of the resected mass revealed pleomorphic adenoma. Interestingly, metastatic deposits with identical histology were identified within the excised cervical lymph node. ***Conclusion:*** This case highlights the paradoxical behavior of pleomorphic adenoma, which, although histologically benign, may metastasize. Awareness of MPA is crucial for clinicians to ensure long-term surveillance following the excision of pleomorphic adenomas.

Keywords: Pleomorphic adenoma, Metastasizing pleomorphic adenoma, Parapharyngeal space, Salivary gland tumor, Lymph node metastasis.

INTRODUCTION

Pleomorphic adenoma (PA) is the most common benign salivary gland neoplasm, accounting for 70–80% of parotid gland tumors followed by submandibular gland tumors. It is typically slow-growing, encapsulated, and composed of epithelial and myoepithelial elements within a mesenchymal-like stroma.

A rare but well-documented phenomenon is *metastasizing pleomorphic adenoma (MPA)*, defined by the World Health Organization as a histologically benign PA that metastasizes to distant or regional sites, most commonly bone, lung, and lymph nodes. 42 patients (19 men and 23 women) were described in the selected papers. Its pathogenesis is debated, with hypotheses including hematogenous dissemination, lymphatic spread, or iatrogenic implantation during prior surgery.

Here in, we present a case of pleomorphic adenoma in a young female who presented with a parapharyngeal mass in addition to an occult metastatic deposit within the excised regional cervical lymph node.

CASE PRESENTATION

Patient: 32-year-old female with no history of medical illness or prior head and neck surgery.

Clinical Presentation: She reported a progressively enlarging, painless swelling in the left oropharyngeal region associated with a sensation of airway narrowing. No dysphagia, odynophagia, weight loss, or systemic symptoms were reported.

Imaging: MRI of the neck with contrast demonstrated a well-circumscribed lobulated soft tissue mass centered in the left parapharyngeal space, extending into the ipsilateral submandibular space and medially into the soft palate. It measured approximately 3.7 × 3.8 × 4.9 cm. The lesion was isointense on T1-weighted imaging, heterogeneously hyper- and hypointense on T2, with thick peripheral enhancement and central non-enhancing necrotic/cystic components (figure;1). The mass was inseparable from the deep lobe of the parotid gland, displaced the medial pterygoid muscle, and abutted the base of the tongue and longus colli muscle. Moderate narrowing of the oropharyngeal airway was noted. No

carotid encasement or perineural spread was observed. Asymmetric prominent cervical lymph nodes were present but initially considered reactive.

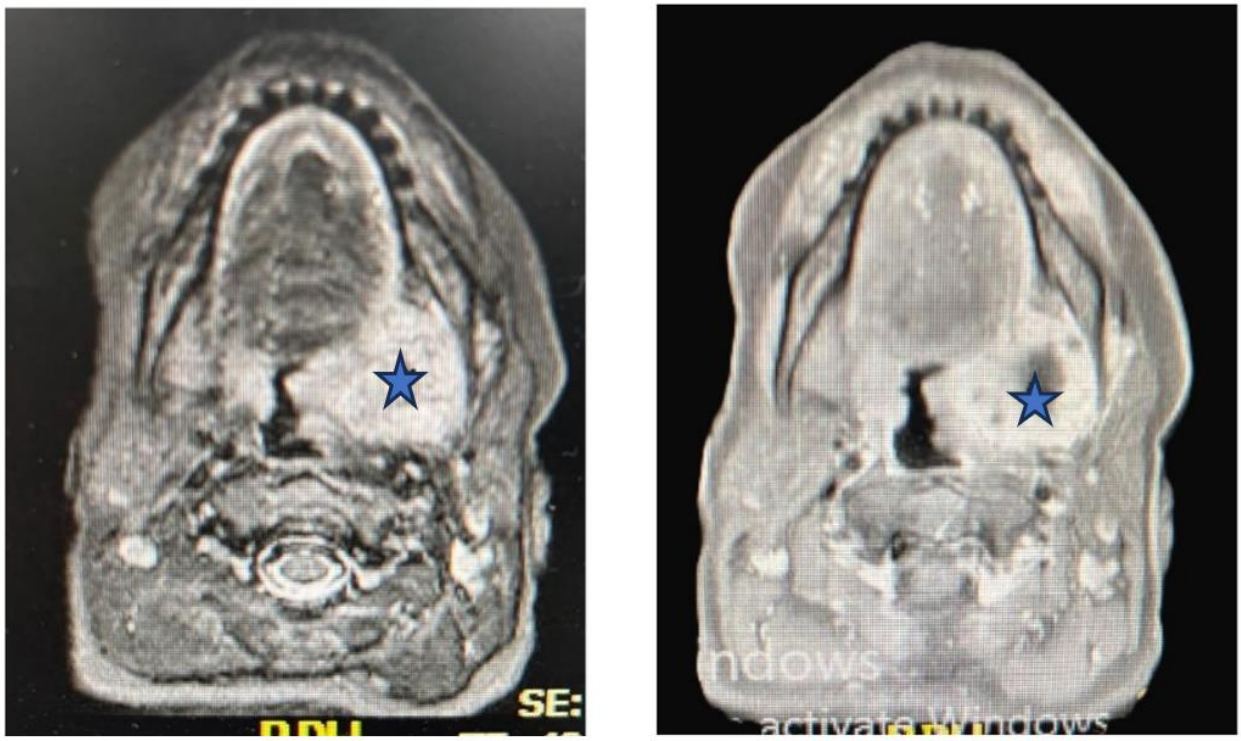

Figure 1: MRI pictures of parapharyngeal mass (highlighted by star)

Management: The patient underwent marginal surgical excision of the mass and the involved lymph node. She is under close clinical and radiological surveillance, given the potential for recurrence or further metastasis.

Histopathology: Grossly we received lobulated white lesion measures 4.5 cm in maximum diameter (figure; 2). Another container with left cervical lymph nodes consists of multiple lymph nodes, the largest measures 1.5 cm in maximum diameter.

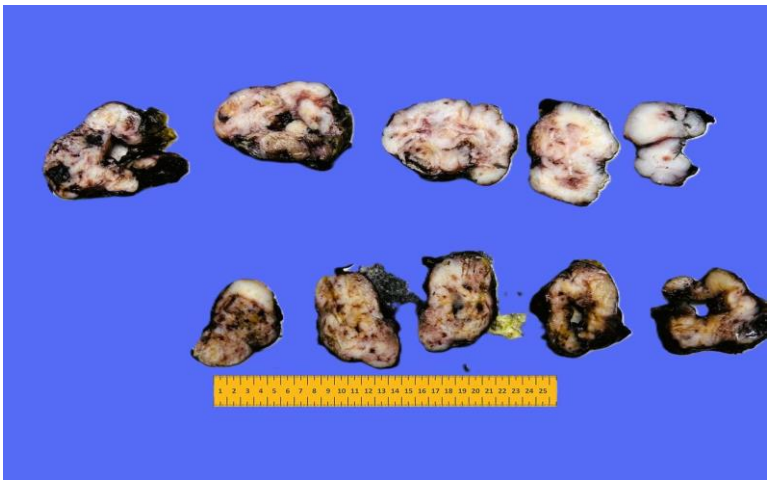

Figure 2: Gross picture of parapharyngeal lesion, show white homogenous cut surface with focal area of hemorrhage

- Microscopically, examined sections of the mass revealed a well circumscribed lesion, composed of epithelial and myoepithelial cells in a fibromyxoid stroma, demonstrating focal minimal to mild cytological atypia, rare mitoses, without extracapsular extension, necrosis or features of malignant transformation (figure; 3).

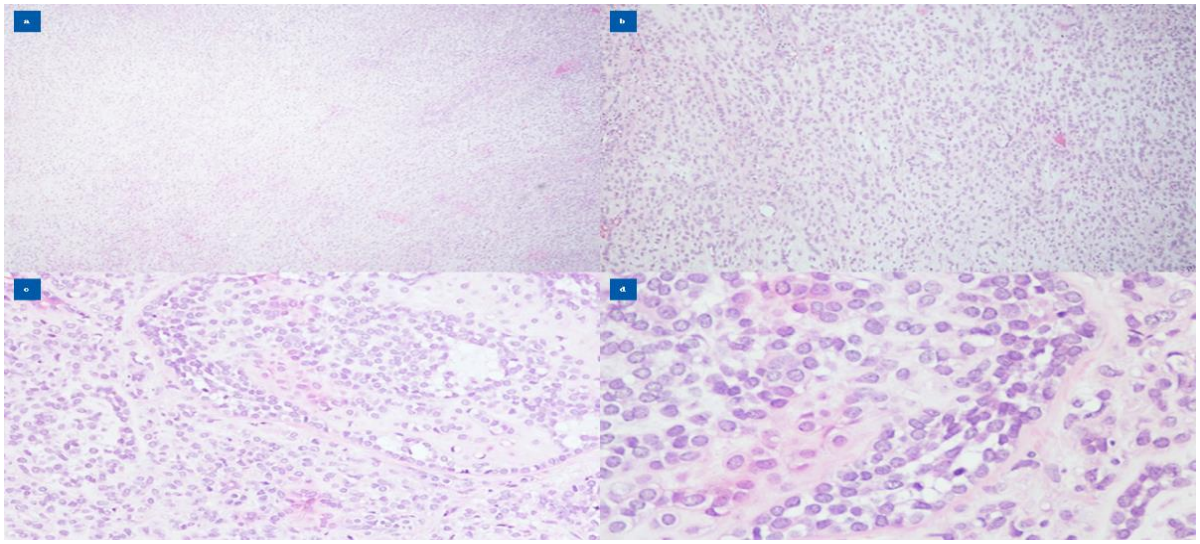

Figure 3: Microscopic picutres of (a &b) show epithelial and myoepithelial cells, in a chondromyxoid stroma (100x). (c & d) higher magnification showing hypercellularity mainly myoepithelial (200x &400x)

IHC findings (figure; 4):

- **Pan-Cytokeratin (CK):** Positive in epithelial tumor cells, confirming epithelial component.
- **p63:** Nuclear positivity in myoepithelial cells, supporting dual cell population.
- **GFAP:** Strong cytoplasmic staining in stromal/myoepithelial elements, confirming myoepithelial differentiation.

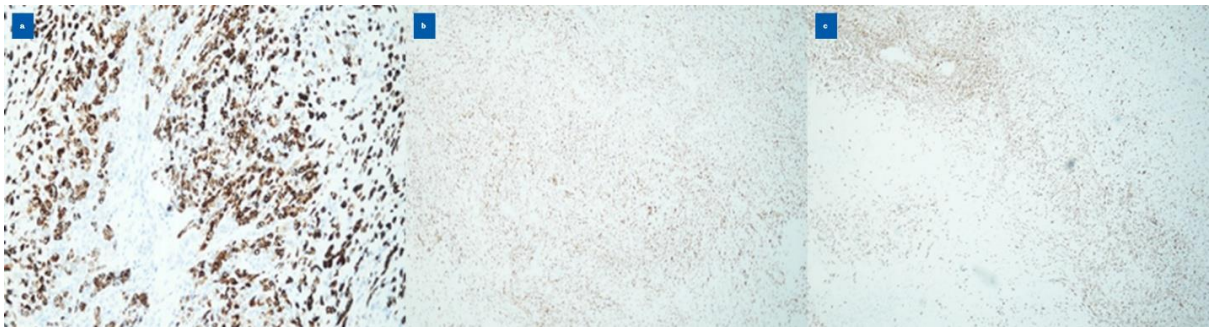

Figure 4: Immunohistochemical stains with positive staining: (a) GFAP, (b) P63 and (c) Pan (CK)

- A section from the resected cervical lymph node features metastatic deposit of pleomorphic adenoma that is morphologically identical to the original lesion of the parapharyngeal mass, this confirms the diagnosis of metastasizing pleomorphic adenoma (figure; 5).

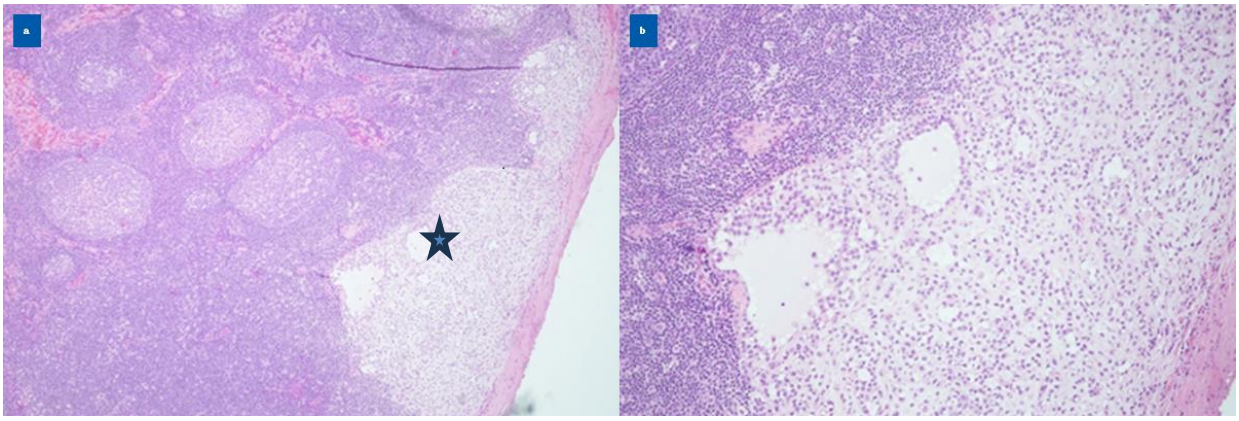

Figure 5: Microscopic picture of cervical lymph node metastatic deposite (highlited by star)

Diagnosis: Metastasizing pleomorphic adenoma (MPA) of the parapharyngeal space with regional lymph node involvement.

DISCUSSION

Metastasizing pleomorphic adenoma is a rare clinical entity, first described by Foote and Frazell in 1953. Despite its histologically benign morphology, MPA exhibits malignant clinical behavior.

Epidemiology & Sites:

MPA most often arises in patients with a history of recurrent PA, but primary de novo cases, such as ours, are also reported. Common metastatic sites include bone (45%), lungs (36%), and lymph nodes (20%).

The mean patient age at presentation of benign metastasizing PA is 49.5 years, and the mean interval between primary tumour diagnosis and metastatic disease is 14.9 years (range: 0–51 years). It often occurs after multiple recurrences.

Pathogenesis: Theories include:

1. Vascular invasion of tumor cells.
2. Surgical implantation during prior excision.
3. Underlying molecular or genetic alterations predisposing to metastasis.

Differential Diagnosis:

- **Carcinoma ex pleomorphic adenoma (Ca ex PA):** shows malignant transformation with cytological atypia, mitoses, and infiltrative growth (not seen here).
- **Other salivary gland malignancies:** e.g., mucoepidermoid carcinoma or polymorphous adenocarcinoma.

Management: Complete surgical excision of the primary and metastatic sites remains the treatment of choice. Radiotherapy may be considered for unresectable disease or multiple metastases. Long-term follow-up is mandatory, as recurrence and late metastasis have been reported even decades after initial surgery.

CONCLUSION

Our case report illustrated that pleomorphic adenoma of the parapharyngeal space may show cervical lymph node metastasis. This case emphasizes that even histologically benign pleomorphic adenomas can metastasize. Clinicians should remain vigilant, and patients require long-term surveillance following excision of PA, especially in deep-seated or recurrent cases.

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