

Case Report

A wandering mystery: freely mobile ectopic pleomorphic adenoma clinically mimicking cervical lymphadenopathy

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ABSTRACT

Ectopic pleomorphic adenoma is a rare entity. It usually originates from heterotopic salivary gland tissue. During an excision via surgery, the tumor ought to be excised completely with an intact capsule. We present a case of ectopic pleomorphic adenoma of the parotid gland in the neck. It was in the right jugulo-diaphragmatic region and freely mobile. Cervical lymphadenopathy was the first clinical diagnosis. But, on investigations, it turned out to be pleomorphic adenoma. This case emphasizes that ectopic pleomorphic adenoma can be a differential diagnosis of neck swellings and needs meticulous surgical excision to prevent recurrence and malignant change.

Keywords: Neck, Ectopic pleomorphic adenoma, Heterotopic salivary gland tissue, Lymph node

INTRODUCTION

When encountering a swelling in the lateral aspect of the upper neck, the clinical differential diagnosis often includes lymphadenopathy or unilateral thyroid swelling. This case report presents a pleomorphic adenoma located ectopically in the neck, highlighting its diagnostic and therapeutic implications.

Pleomorphic adenomas are the leading cause of benign salivary gland tumors. The bulk of these originate in the main salivary glands (85%), followed by minor salivary glands (10%) and ectopic locations (5%). Willis was the first to develop the term "pleomorphic adenoma".

Ectopic regions after the salivary glands are primarily limited to the lips, followed by the oral cavity, neck, and nasal cavity.

Often occurs throughout a range of age brackets, but is more prevalent in people aged forty to sixty, and males are more inclined to it than females.¹⁻³

Ultrasonography (USG) is the preliminary imaging technique recommended for children as well as pregnant women, while computed tomography (CT) scans are the suggested diagnosing technique for inflammatory disorders, and abscesses, in addition to calculi that pertain to salivary duct dilation. Magnetic resonance imaging (MRI) is frequently preferred when diagnosing conditions with oncogenic potential or when a CT scan is not recommended.⁴

These tumors frequently manifest as a single, relatively painless swelling that may be mobile. The conventional treatment for this benign condition is complete tumor resection with an intact capsule; inadequate resection or a breach in the tissue capsule results in recurrence. Tumors

that cannot be completely wiped out have a greater chance of becoming malignant.¹⁻³

CASE REPORT

A 23-year-old gentleman came to the outpatient department with a complaint of swelling in the upper neck on the right side, close to the angle of his mandible (Figure 1) over three years. The patient had no other complaints.

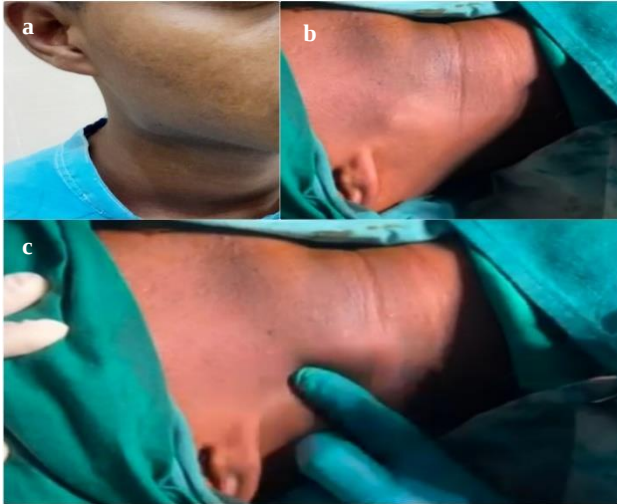


Figure 1 (a-c): Freely mobile swelling in the neck in the right jugulo-digastric region.

The 3×2 cm sized firm swelling was present in right jugulo-digastric region without signs of inflammation and was freely mobile.

All blood investigations were within normal limits. Ultrasound of the neck and right parotid area shows a hetero-echoic lesion with hypo-echoic margins below the superficial lobe of the right parotid, with maximal dimensions measuring 1.5×2.7×2.8 centimeters.

Ultrasonography-guided fine needle aspiration cytology testing was used, which revealed benign-looking ductal epithelium cells, plasmacytoid myoepithelial cells, and several oncocyctic cells suggesting differential diagnosis of cellular pleomorphic adenoma and oncocytoma class Milan IVa (benign).

MRI of the right parotid suggested a hyperechoic lesion inferior to the superficial lobe (Figure 2). No indication of diffusion limitation or blooming is visible which is mostly suggestive of a benign pleomorphic adenoma.

An open surgical approach was planned. An upper cervical incision was made over the right upper neck parallel and slightly beyond the jawline of the mandible, allowing the swelling to be excised without a breach in the capsule and safeguarding the greater auricular nerve and the specimen (Figure 3). The swelling had no connection with the parotid gland intraoperatively.

The patient's surgical recovery was uneventful, with discharge on day three, suture removal on day six, and antimicrobial agents for two more days.

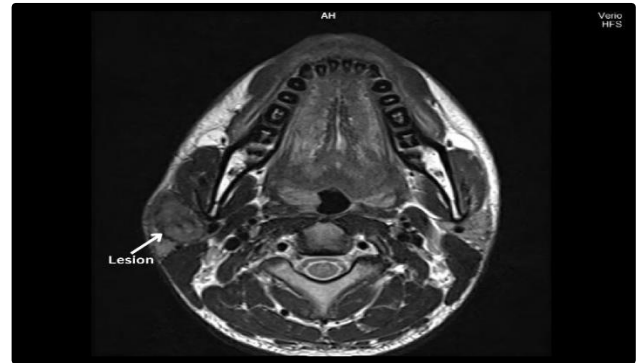


Figure 2: Cut section of MRI showing the lesion just inferior to the superficial lobe of the parotid gland.

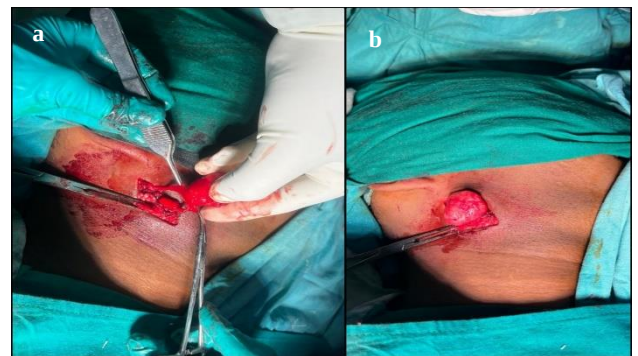


Figure 3 (a and b): Intraoperative image of swelling attached to the greater auricular nerve and showing specimen.

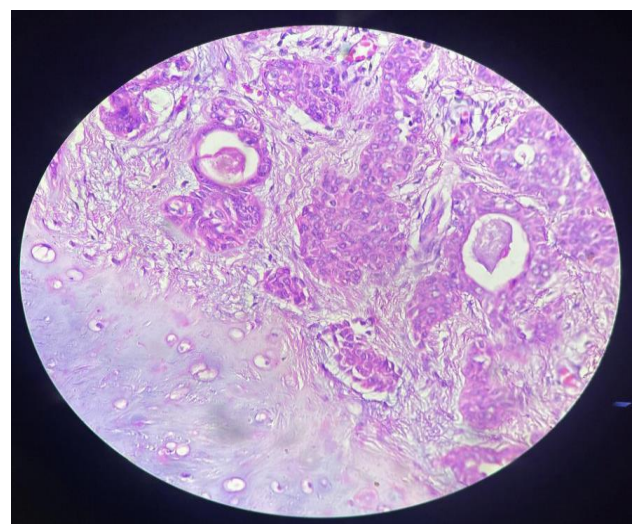


Figure 4: Tubules and sheets of epithelial cells with myxoid stroma suggesting pleomorphic adenoma.

According to the patient's histology report, the tumor was composed of chondro-myxoid stroma, with epithelium and myoepithelial components distributed throughout (Figure

4). There was no evidence of necrosis, squamous metaplasia, abnormal mitosis, atypia, or malignancy in the present sections. The findings mentioned above indicate a benign pleomorphic adenoma.

A one-year postoperative follow-up demonstrated the absence of recurrence.

DISCUSSION

Pleomorphic adenoma is the most common benign tumor affecting the salivary glands. The cells of origin are thought to be the intercalated ducts reserve cells. The majority of those affected are women in their forties and sixties.

Rarely patient may present with neoplasm of the ectopic salivary gland. Ectopic salivary gland tissue located in areas external to the minor, major, and auxiliary glands is often called "heterotopic salivary gland tissue (HSGT)".

These ectopic locations of tissue might be seen in several sites, notably the temporal bone, thyroglossal duct, pituitary gland, mandible, and, in rare instances, the neck, where they are frequently limited to the anterior triangle.⁵

Several justifications have been given for the organization of HSGT, among them the existence of epithelium remnants of the brachial apparatus. Warthin's is perhaps the most prevalent tumor of the HSGT, then follows pleomorphic adenoma.⁶

The primary mode of assessment for cervical swelling is USG, followed by a CT scan or MRI, according to the suspected reason. Because of this unusual location, USG-guided FNAC testing is essential before surgery. MRI will be particularly successful in contrasting tumor types.

Regardless of the favorable prognosis, operative removal of the lesion with an intact capsule is a preferred therapeutic modality to mitigate the incidence of regional recurrence post resection and cancerous transformation into "carcinoma ex pleomorphic adenoma (CEPA)" or "carcino-sarcoma (true malignant mixed tumor)". The percentage of cancerous transformations was confined to "3-15%".^{7,8} Intra-operative factors that might lead to regional recurrence include pseudo capsular breaches, tumor spillage during surgical removal, and tight margins. Female sex, early onset, and hypo cellular tumors are also strong predictors for recurrence. The rate of recurrence ranges from 10 to 45 years after the operative procedure.^{9,10}

CONCLUSION

This case emphasizes the need to take ectopic pleomorphic adenoma into account when making a differential

diagnosis of lateral neck swellings. It is uncommon entity but noteworthy in view of the possibility of recurrence and malignant change if not removed with an intact capsule. Proper diagnostic imaging and surgical methods are critical for a good outcome.

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