

Case Report

Lymphoepithelial cyst in submandibular region

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ABSTRACT

The lymphoepithelial cyst is a rare, benign, soft tissue, unilocular or multilocular developmental cyst in the head and neck region. It is seen to occur in the salivary glands (parotid>submandibular gland) and oral cavity (floor of mouth/tongue). Among various investigations, FNAC provides immediate diagnosis whereas ultrasonography and CT scan can also be used. The confirmatory diagnosis is always made postoperatively by histopathological examination. Surgery is the mainstay of treatment which includes complete enucleation of the cyst along with total excision of the involved salivary gland. This rare case has been reported to help clinicians bring lymphoepithelial cyst into their differential diagnosis for lesions affecting the submandibular region.

Keywords: Lymphoepithelial cyst, Unilocular, Multilocular

INTRODUCTION

Swelling in the head and neck region is a common clinical presentation, with the submandibular area exhibiting a diverse range of potential diagnoses. These swellings can be attributed to various aetiologies including inflammatory, infectious, traumatic, neoplastic, or congenital factors.¹ Notably, approximately 23% of swellings in the submandibular region are malignant, underscoring the importance of a thorough clinical approach.¹ Effective management necessitates comprehensive history taking, meticulous extraoral and intraoral examinations, as well as radiological and histopathological evaluations.¹ Open surgical biopsy accompanied by histopathological analysis remains the gold standard for diagnosis.¹

Lymphoepithelial cysts are rare benign developmental cysts occurring predominantly in the salivary glands and oral cavity.²⁻⁴ They are more commonly associated with the parotid gland, while involvement of the submandibular gland is uncommon.^{2,3} Clinically, they present as slow-growing, painless swellings and may

mimic other cystic lesions of the neck.³ Various imaging modalities such as ultrasonography, computed tomography (CT), and magnetic resonance imaging (MRI), along with fine needle aspiration cytology (FNAC), aid in diagnosis and surgical planning.²⁻⁵ Histopathological examination remains the definitive diagnostic method.¹⁻³ Surgical excision is considered the treatment of choice with excellent prognosis and minimal recurrence.³⁻⁵

In this paper, we report a case of submandibular swelling that ultimately resulted in the rare diagnosis of a lymphoepithelial cyst.

CASE REPORT

A 30-year-old male patient presented to the ENT department with complaints of swelling on the left side of his neck for the past 2 months. The swelling was initially small but gradually increased to its current size. There was no history of fever, pain, trauma, or previous surgical interventions. While the swelling did not cause any functional restrictions, the patient was cosmetically

concerned about it. He had consulted another doctor and received medications, but the swelling did not subside.



Figure 1: Pre-operative image showing diffuse swelling on left submandibular region.

On examination, there was a diffuse, firm, non-tender swelling measuring 7×5 cm in the left submandibular region. The swelling extended anteriorly 1 cm lateral to the midline and posteriorly up to the posterior border of the sternocleidomastoid muscle. Superiorly, it reached the ramus of the mandible and inferiorly extended to the level of the lower border of the thyroid cartilage. No abnormalities were noted during the intraoral examination.

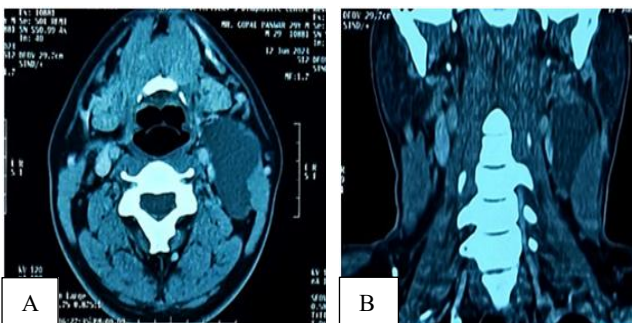


Figure 2: Contrast enhance computed tomography showing well- defined enhancing fluid density space occupying lesion in left submandibular region: (A) CECT axial cut and (B) CECT coronal cut.

An ultrasonographic examination revealed a well-defined hypoechoic cystic lesion measuring 5.3×3.6×2.3 cm in the left submandibular region extending to the angle of the mandible with no internal vascularity, suggestive of lymphangioma or branchial cyst. Fine needle aspiration yielded straw-colored fluid that was sent for cytological analysis. The report suggested a lymphoepithelial origin.²

Contrast-enhanced computed tomography (CECT) scan was conducted to assess the size, extent, and relationship to adjacent structures. The scan revealed a well-defined enhancing fluid-density space-occupying lesion measuring 6×3×4.6 cm in the left submandibular region. The lesion extended from a level slightly above the carotid bifurcation cranially to a level just below the lower border of the thyroid cartilage. It medially abutted the left carotid artery and left internal jugular vein causing slight mass effect and partial luminal narrowing of the vein.



Figure 3: Post operative image after 3 weeks.

After reviewing the findings, surgical excision of the lesion was planned. Under general anesthesia, a horizontal neck incision was made 3 cm below the angle of the mandible along the skin crease. The cyst was dissected and excised in toto.

The specimen was sent for histopathological examination, which confirmed the diagnosis of a lymphoepithelial cyst.³ The patient was followed up after

3 weeks, and the operative site appeared healthy with no swelling or discoloration.

DISCUSSION

Lymphoepithelial cysts, also referred to as branchial cysts, are benign, slow-growing lesions commonly affecting individuals during the second and third decades of life.² These cysts are usually unilateral with a predilection for the left side, while bilateral occurrence is rare.² Clinically, they present as painless, fluctuant, non-tender swellings in the cervical region.³

The majority of lymphoepithelial cysts occur in association with salivary glands, particularly the parotid gland, although the submandibular gland may rarely be involved.²⁻⁴ Scott-Brown's Otorhinolaryngology describes these lesions as developmental cysts arising from epithelial remnants trapped within lymphoid tissue.^{4,5} The most accepted theory of pathogenesis is the lymph node theory, which proposes cystic degeneration of epithelial inclusions within cervical lymph nodes.³ Histopathologically, these cysts are lined by stratified squamous or respiratory epithelium surrounded by dense lymphoid tissue with germinal centres.²

Lymphoepithelial cysts may also be associated with systemic conditions such as HIV infection, Sjögren's syndrome, Mikulicz disease, and myoepithelial sialadenitis.⁵ In HIV-positive patients, bilateral parotid lymphoepithelial cysts are particularly characteristic.⁵

FNAC is considered a valuable diagnostic tool and may provide both diagnostic and therapeutic benefit.² Imaging modalities including ultrasonography and CT scan help determine the cystic nature, extent of lesion, and relationship to surrounding structures.³ MRI may further aid in differentiating cystic lesions from vascular or neoplastic masses.⁵

The differential diagnosis includes lymphangioma, branchial cleft cyst, Warthin tumor, chronic sialadenitis, salivary duct cyst, and metastatic cystic squamous cell carcinoma in older patients.⁵ Complete surgical excision remains the treatment of choice, with recurrence being extremely uncommon following adequate removal.³ Early diagnosis and management are essential because rare malignant transformation has been reported.⁵

CONCLUSION

Lymphoepithelial cysts, though rare, should be considered in the differential diagnosis of swellings in the submandibular region, particularly when presenting as non-tender, slowly enlarging masses. While they are typically associated with the parotid gland, their occurrence in the submandibular region is uncommon. Imaging techniques such as ultrasonography, CT, and FNAC along with histopathological examination are crucial for accurate diagnosis. Surgical excision remains the primary treatment and is typically curative, with recurrence being rare following complete removal. Given their potential association with systemic conditions and the possibility of malignant transformation, early and precise diagnosis is essential for effective management.

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