Case Report

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A Rare Case of Lympho-epithelial Cyst on Left Buccal Mucosa: Clinical Presentation and Surgical Approach

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ABSTRACT

Lymphoepithelial cysts are rare benign lesions, usually painless, yellowish nodules that arises from different parts of the body including oral cavity. They are regarded as pseudocysts of retention cysts, as the aetiopathogenesis is unclear. This article describes a case of uncommonly observed large lympho-epithelial cyst of a 64-year-old female who had presented with the history of a lump in the left buccal mucosa for 10 years. The patient underwent intraoral excisional biopsy, histopathological features shows outer peripheral part of cyst encircling lymphoid tissue, arranged in a follicular pattern of variable size with a prominent germinal cell suggestive of lympho-epithelial cyst.

Keywords: Benign cyst; lymphoepithelial cyst; salivary gland.

INTRODUCTION

Lympho-epithelial cyst (LEC) is an uncommon, soft-tissue, developmental cyst, microscopically lined by stratified squamous or pseudostratified columnar epithelium with surrounding lymphoid fibrous connective tissue.[1-3] The LECs grow in head and neck region as single or multi-loculated lesions usually presenting as part of diffuse infiltrative lymphocytosis syndrome in human immunodeficiency virus (HIV) patients.[4] It also originates from pancreas, stomach, thyroid, oesophagus, mediastinum including floor of mouth, and lateral margins of tongue.[5-7] Variable in size from 0.5 cm to 5.0 cm, it can cause significant facial distortion and malfunction. If left untreated for long time, likely to be converted to malignant conditions.[4]

CASE REPORT

A 64 years female patient presented to the department of oral and maxillofacial surgery at Lumbini Medical College and Teaching Hospital (LMCTH), Prabhas, Tansen, Palpa, Nepal with a chief complaint of swelling involving left side of lower face for the last 10 years. Patient informed that she had first noticed the swelling around 10 years back when she got full mouth extraction to receive complete denture.

On extraoral examination, there was a soft, fluctuant, compressible, non-tender swelling of size 5 cm X 6 cm on the left parotid region (Figure 1). The swelling extended from zygomatic region to the angle of mandible. Skin and mucosa over the swelling was normal and the swelling was

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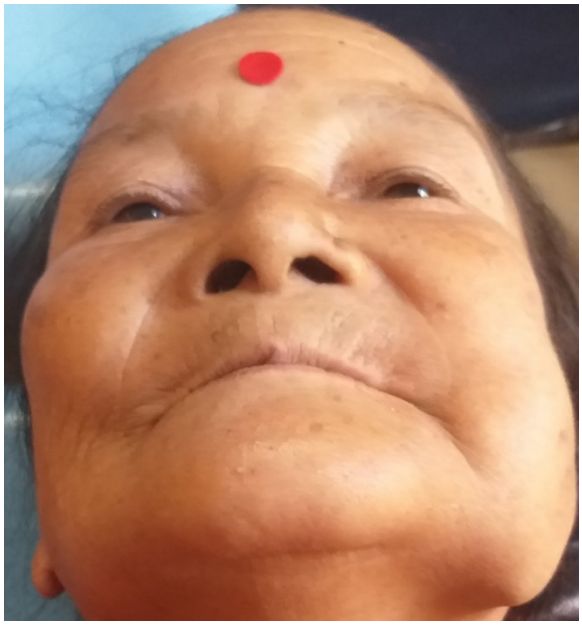

Figure 1: Preoperative clinical picture showing cyst on left side of cheek.

palpable from both intraoral and extraoral sites. No obvious dryness of mouth or tenderness along the orifice of Stenson's duct was noticed. Function of facial nerve was intact. Blood investigation of the patient was within normal limits. Medical history was suggestive of hypertension and was under medication for the last five years.

Aspiration of fluid with wide bore needle yielded dirty straw coloured fluid. The fine needle aspiration cytology (FNAC) finding revealed the presence of foamy histiocytes in mucoid background and had no epithelial component or sign of inflammation being suggestive of benign cystic lesion.

On clinical evaluation, FNAC and computed tomography (CT) finding were suggestive of benign cystic lesion so the treatment was very definitive and the patient was planned for excision of the lesion under general anaesthesia (Figure 2).

Surgical approach to the lesion was made intraorally starting about 3 mm below the opening of Stensen's duct and long buccal nerve were preserved. Cystic swelling was exposed and cystic lining and content were completely enucleated. Haemostasis was achieved and closure was done with absorbable suture. The specimen was sent for histopathological examination, which revealed a cystic lumen lined with thin stratified squamous epithelium showing few areas of epithelial degeneration. Epithelium showed absence of rete process. Lumen showed keratin deposition. Fibrous connective tissue wall lacks inflammatory cell infiltrate but few areas show well defined clusters of mature lymphocytes (Figure 3). Post-operative outcome was good immediately and at one month follow-up (Figures 4, 5).

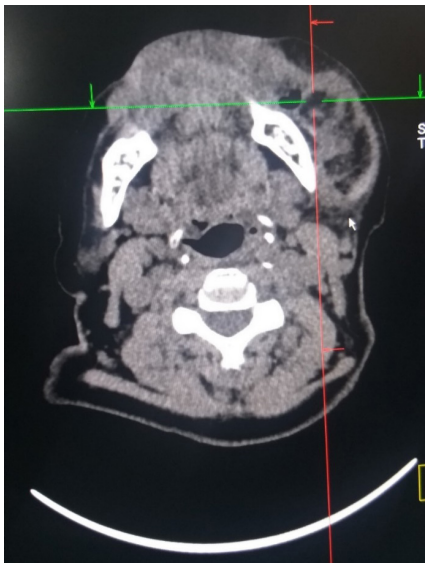

Figure 2: Computed tomography of the submandibular region showed a well encapsulated multiple mass on the left submandibular region with no bony abnormalities.

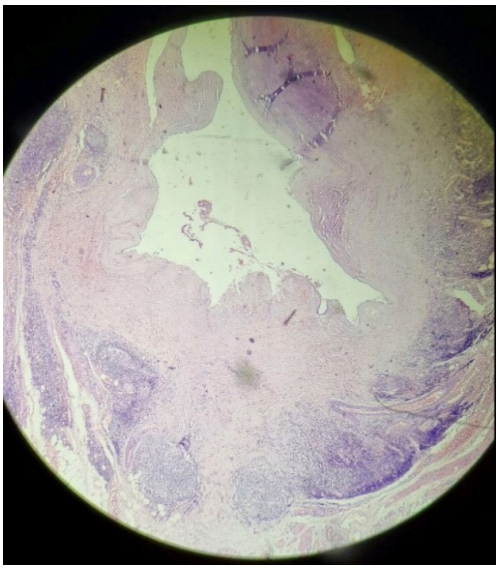

Figure 3: Histopathological picture showing mature lymphocytes.

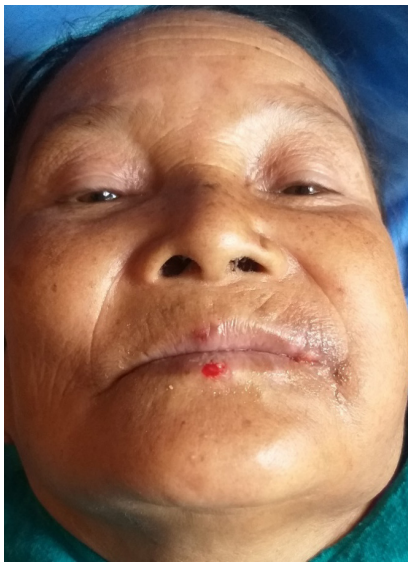

Figure 4: Immediate post-operative picture after intraoral enucleation of multiple cystic mass.

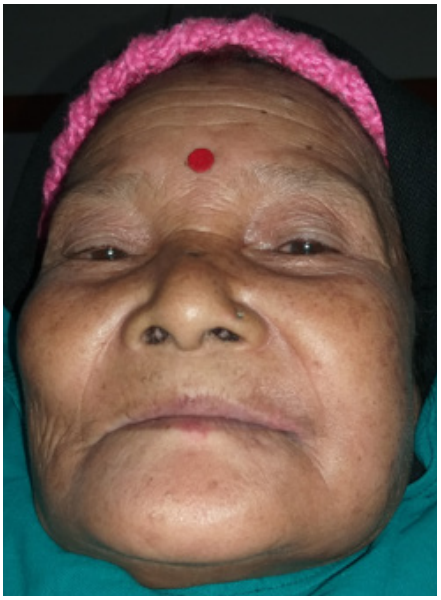

Figure 5: Post-operative picture after one month.

DISCUSSION

Lymphoepithelial cyst an uncommon lesion in oral cavity, may occur in any organ such as salivary glands, pancreas and thyroid gland.[8] Painless, benign, slow growing lesion that occurs mostly in adults with a 60-80% predilection for female, the cyst occurs mostly in second and third decade of life. There is a close association of salivary gland, especially the parotid gland with the lympho-epithelial cysts.[9] The FNAC is one of the investigation methods for therapeutic as well as a confirmatory test. For the diagnosis of Lymphoepithelial cyst, major salivary gland lesion showed the presence of proteinaceous background and a mixed population of lymphocytes, histiocytes, plasma cells, and metaplastic squamous cells.[5] Another diagnostic tool that helps in determining the nature of cyst, if it is encapsulated or breaching and invading into other structures is the CT scan. Moreover, CT plays significant role in demonstrating the coverage of lesion in all the aspect which is essential before planning surgical excision.

Other salivary gland lesions such as Warthrin's tumor, lymphoepithelial sialadenitis, chronic sclerosing sialadenitis, HIV associated salivary gland lesions, mucocele, dysgenetic polycystic disease of salivary gland or extranodal marginal zone B-cell lymphoma should be ruled out before confirming it as a lymphoepithelial cysts as those are common differential diagnosis of this condition.

The cyst should be treated as early as possible as patients with salivary gland lympho-epithelial cyst has higher chance of developing lymphoma. There are conservative as well as surgical modalities for treatment of Lympho-epithelial cyst. Decompression of the cyst is done in conservative approach, by aspirating the fluid thus reducing the cytic pressure. In immunocompromised and comorbid patient conservative approach is carried out rather than surgical, as surgery has its own risk for such patients. External radiotherapy is also one of the treatment modalities for conservative approach. Complete enucleation of the cyst along with the excision of the involved gland is its definitive surgical treatment and in most of the cases the lesion gets totally cured with no evidence of re-emergence.[10]

SUMMARY

Lympho-epithelial cysts are rare benign cyst which should be treated as early as possible as it has tendency to transform into malignancy such as adenocarcinoma, lymphoma, mucoepidermoid carcinoma. Surgical excision is the principal modality of its treatment, however, intraoral approach is quite challenging but aesthetically pleasing approach.

Conflict of interest: None.

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