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Case Report

Benign Lymphoepithelial Cyst: An Unusual Cause of Parotid Swelling in Two Immunocompetent Patients

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Abstract

BACKGROUND: Lymphoepithelial cysts, which are benign and slow-growing tumours, usually involve the head and neck regions. Benign lymphoepithelial cysts (BLECs) are the most common cause of parotid swelling in human immunodeficiency virus (HIV)-positive patients and are less common in immunocompetent patients.

CASE PRESENTATION: Here, we present two cases of immunocompetent patients with long-standing, progressively enlarging parotid swelling. Postoperative histopathological examination of these patients revealed features of BLEC.

CONCLUSION: Wide surgical excision is the gold standard for treatment and recurrences is rare. These cases are of particular interest because of the rarity of BLEC in HIV-negative patients and highlight an important differential diagnosis of parotid swelling.

Introduction

Benign lymphoepithelial cysts (BLECs) are benign, slow-growing, uninoculated or multiloculated lesions that are usually associated with the salivary glands in the head and neck regions. BLECs are more commonly observed in up to 60% – 80% of female patients [1]. They are pathognomonic for human immunodeficiency virus (HIV) infection and can occasionally manifest as the first symptoms of retroviral infection [2].

BLECs originate from epithelial remnants retained in the lymphoid tissue, which are trapped in the parotid gland during embryogenesis [3]. Here we report two cases of HIV-negative patients with BLEC of the parotid.

Case Reports

Case 1

A 45-year-old female presented to our clinic with a 1-year history of swelling in her left cheek, which was slowly increasing in size and causing discomfort and cosmetic impairment. She had no other comorbidities or family history of head and neck tumours. Physical examination revealed a soft non-tender mass, measuring 4 cm × 4 cm, over the left parotid gland. Intraoral examination was unremarkable, and facial nerves were intact. These findings were confirmed via computed tomography (CT), which revealed a solitary well-marginated rim-enhancing cystic lesion, measuring 3.2 cm × 3.3 cm, within the superficial lobe of the left parotid gland.

Although fine-needle aspiration cytology (FNAC) was performed, the sample obtained was inadequate and unsatisfactory. Left superficial parotidectomy was performed without complications, and there was no recurrence at 6-month follow-up.

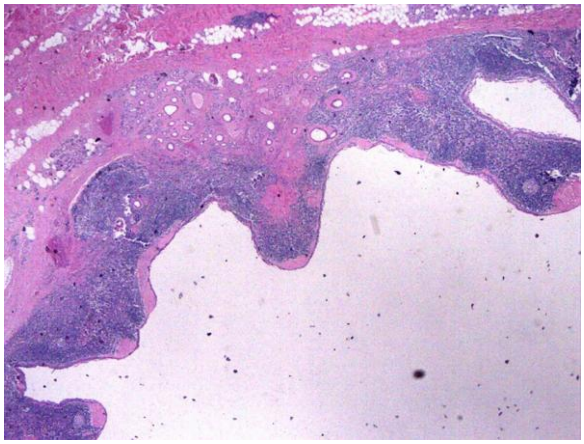

Figure 1: Haematoxylin and eosin staining of parotid cyst showing subepithelial stroma with reactive lymphoid follicles and germinal centres (Magnification x 20)

Gross specimen revealed a grey/ brown encapsulated cystic lesion, measuring 3.0 cm × 4.5 cm × 2.5 cm, filled with brownish mucinous fluid. Microscopic examination revealed subepithelial stroma with reactive lymphoid follicles and germinal centres. Adjacent normal salivary glands and fatty tissue appeared normal, and there was no oncocytic cell lining or neoplastic tissue (Figure 1 and 2). A final diagnosis of BLEC of the left parotid gland was made based on the radiological and histomorphological findings. HIV antibodies were analysed using ELISA, and a negative result was obtained.

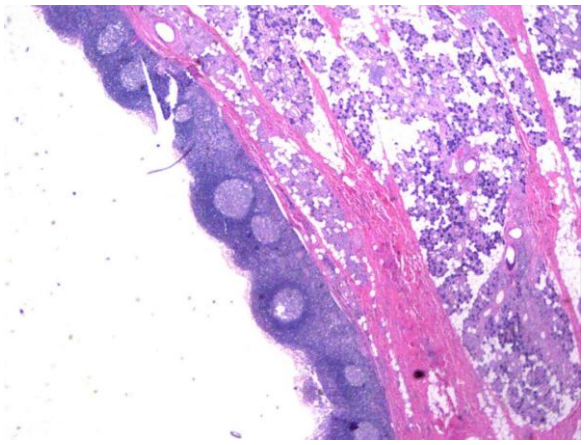

Figure 2: of parotid cyst showing reactive lymphoid follicles and germinal centres with adjacent normal salivary glands and fatty tissue (Magnification x 40)

Case 2

A 48-year-old female presented to our ear, nose, and throat outpatient clinic with a history of persistent swelling of the left cheek for the past 11 years, which had gradually progressed to the current size of 4 cm × 5 cm. The swelling was not associated with pain but was cosmetically disfiguring. Clinical examination revealed a swelling over the left parotid region, measuring 4 cm × 5 cm, which was mobile, soft, and not adherent to the underlying skin. There was no cervical lymphadenopathy, Stenson's duct was normal with no purulent discharge, and facial nerves were intact. CT revealed a non-enhancing, homogenous lesion involving the superficial lobe of the left parotid gland (Figure 3 and 4).

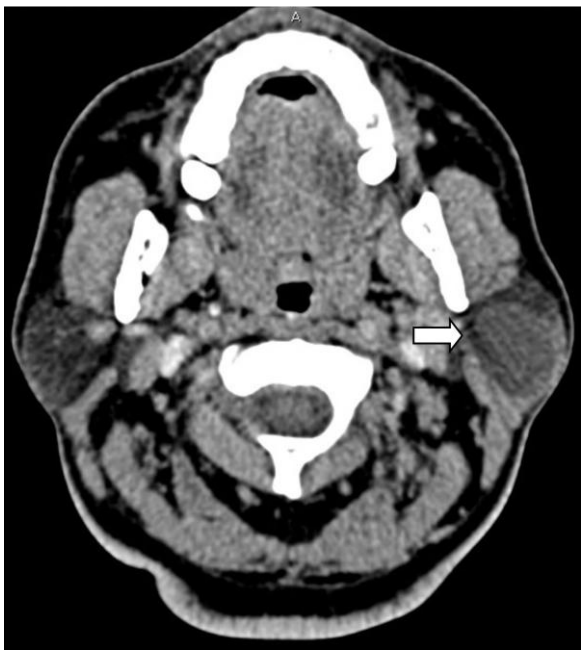

Figure 3: Preoperative CT scan neck, axial view. Cyst like lesion seen in the left parotid gland (Arrow)

FNAC was inconclusive. She underwent left superficial parotidectomy under general anaesthesia. Excised parotid mass measuring 45 mm x 25 mm x 20 mm.

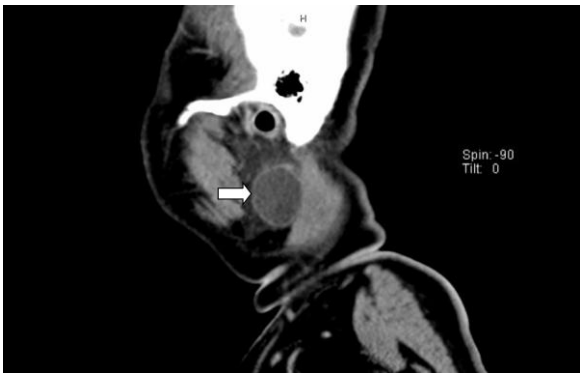

Figure 4: Preoperative CT scan neck sagittal view, Cyst like lesion of the left parotid gland (Arrow)

Cut surface shows a large cystic area containing clear fluid (Figure 5). Histopathological analysis revealed cystic structures comprising dense

polymorphous and polyclonal lymphoid tissue forming scattered reactive follicles, closely associated with the glandular lining epithelium of the cyst. A final diagnosis of BLEC was made. HIV antibodies were analysed using ELISA, and a negative result was obtained.

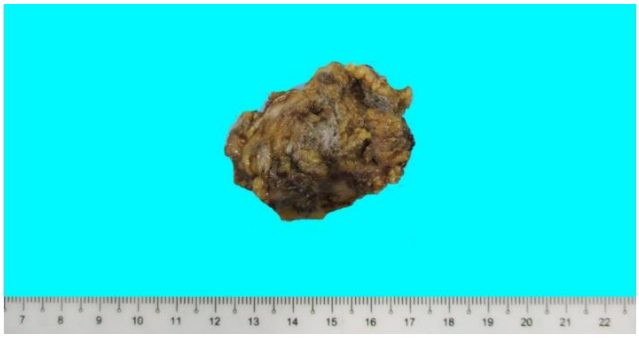

Figure 5: Post-operative specimen (left parotid cyst measuring 4.5 x 2.5 cm)

Discussion

BLECs are single or multiple cysts occurring in the lymph nodes within the salivary gland. They are usually slow-growing and are commonly observed in HIV-positive patients in whom cysts often bilaterally present and are accompanied by cervical lymphadenopathy [1]. The pathogenesis of BLECs is associated with the migration of HIV-infected cells into the lymphoid tissue of the salivary glands, leading to lymphoid hyperplasia and metaplasia in the salivary duct and ultimately resulting in ductal obstruction, dilatation, and cyst formation [1], [4]. BLECs are less common in immunocompetent patients and may be associated with Sjögren's syndrome [5]. Although the pathogenesis of BLEC in immunocompetent patients is unclear, it is postulated to have a similar process of lymphoid hyperplasia in viral infections other than HIV. Naidoo et al. reported a case of BLEC in an immunocompetent patient with chronic otitis media and concluded that a long-standing ear infection caused chronic lymphatic drainage into the intracarotid lymph nodes, leading to ductal obstruction and cyst formation [6].

BLECs are diagnosed based on history, physical examination, and FNAC. Histopathological findings, including the presence of proteinaceous background with mixed infiltration of lymphocytes, histiocytes, plasma cells, and metaplastic squamous cells, are usually suggestive of BLEC [7]. CT and magnetic resonance imaging typically show the bilateral and multicystic appearance of the lymph nodes in immunocompromised patients and unilateral involvement in immunocompetent patients [4]. Radiological examinations are useful tools to aid in the diagnosis of BLECs and access the borders of the mass to evaluate the involvement of the surrounding structures.

Although the definitive treatment is surgical excision, other treatment modalities include conservative observation, cyst aspiration, sclerotherapy, radiotherapy, and highly active antiretroviral therapy in immunocompromised patients [3]. Superficial parotidectomy was performed in both patients in the present case, and the 3- and 6-month follow-up revealed no signs of local recurrence of the tumour. Close monitoring is required in immunocompromised patients with BLECs because of the high risk of developing lymphomas [1].

In conclusion, it is important to be aware that lymphoepithelial lesions of the salivary glands can manifest as a cystic lesion in immunocompetent patients. Histopathological and radiological examinations are the gold standard for a definitive diagnosis of BLECs, and surgical excision is the first-line treatment in such cases, as demonstrated in our two cases.

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Declaration of Patient Consent

The authors certify that both the patients have signed the appropriate patient consent form. In the form, the patient has given his consent for his images and other clinical information to be reported in the journal. The patient and his parents understand that his name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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