Benign Lymphoepithelial Cyst of the parotid in immunocompetent patients

Case Report

Zakaria Toubi, Anouar Bouhlala , Reda Fadel, Dounia Kamal

Oral and maxillofacial surgery department, Hassan II University Hospital, Sidi Mohamed Ben Abdellah University, Faculty of Medicine and Pharmacy, Fes, Morocco

ABSTRACT

Benign lymphoepithelial cyst (BLC) of the parotid gland is a rare benign embryonic cystic tumor in the lateral neck. The distinctiveness of BLCs demands careful consideration in clinical practice, with their potential link to HIV infection highlighting the importance of comprehensive assessment, particularly in immunocompromised individuals. The clinical spectrum of BLCs encompasses gradual growth, palpable mobility, and their impact on patients' quality of life due to cosmetic implications. Diagnostic evaluation involves histopathological examination, imaging techniques, and cytological analysis to discern the cystic etiology, especially in cases where underlying causes may be elusive. The diverse manifestations and treatment options underscore the need for individualized approaches. Surgical excision, notably parotidectomy, yields promising outcomes, yet associated complications require careful evaluation. Advances in cosmetic reconstruction techniques offer prospects for addressing post-surgical defects. In sum, the study of BLCs enhances our understanding of salivary gland disorders, aiding clinical management and offering insights into these intricate anomalies.

Key Words : lymphoepithelial cyst; salivary gland; parotidectomy
Received: 7 March 2025**, Accepted:** 8 May 2025

Corresponding Author : *Zakaria Toubi, Adresse: N 12 Mhirez Hay Salam - Sefrou,*
Mobile: +212684234770 , **E-mail**: zakaria.toubi94@gmail.com
ISSN: 2090-097X, July 2025, Vol. 16, No. 3

INTRODUCTION

Parotid glands are susceptible to various pathological conditions, including neoplastic growths and cystic formations. Among these, benign lymphoepithelial cysts (BLCs) stand out with their distinctive histopathology and intriguing clinical implications. While historically associated with immunosuppression, particularly in HIV-infected population [1], the emergence of BLCs within immunocompetent individuals raises significant scientific curiosity due to their rarity. Generally, cystic anomalies within the parotid gland are infrequent, accounting for approximately 3% of the overall salivary gland tumor incidence. Cystic formations in salivary glands may manifest either as benign non-neoplastic entities or in correlation with both benign and malignant salivary gland tumors. It is imperative to differentiate non-neoplastic cysts of salivary glands from entities such as cystadenoma, mucoepidermoid carcinoma, and acinic carcinoma. A substantial portion of salivary gland cysts can be attributed to obstructive processes. These occurrences might arise consequent to traumatic ductal severance, partial or complete obstructions within excretory ducts, or impediments to salivary flow leading to stasis in the ducts.

 DOI: 10.21608/omx.2025.366178.1283

While ductal obstruction appears to be the etiological factor, the precise source of obstruction often remains obscured. This study aims to uncover the underlying mechanisms of BLCs in the demographic of immunocompetent patients, refining diagnostic and treatment approaches. By dissecting clinical presentations, diagnostic methods, and therapeutic considerations, this study endeavors to contribute to salivary gland pathology knowledge and the understanding of immunity-related cyst formation.

CASE PRESENTATION:

A 60-year-old male presented to our oral and maxillofacial surgery department with a chief complaint of a progressively enlarging left-sided facial swelling persisting for two years. The swelling was asymptomatic but cosmetically concerning. The patient reported no tenderness, erythema, edema, or thermal sensitivity within the mass; no earache, no facial pain, numbness, or limitation in the mouth opening. The patient had a prior history of facial trauma within the vicinity of the current swelling, during which the patient reported experiencing a hematoma in the identical anatomical region; however, this hematoma lacked radiological documentation. Clinical examination revealed an oval-shaped swelling measuring 5x4 cm over the left parotid region, characterized by firm consistency and lack of adherence to surrounding tissues (figure 1). Absence of cervical lymphadenopathy was noted, and the contralateral parotid gland appeared unremarkable. Evaluation of Stensen's duct revealed no abnormalities, with no evidence of ductal discharge upon glandular compression. Magnetic resonance imaging (MRI) demonstrated a round cystic lesion in the left parotid gland, with a maximum cross-sectional area of approximately 5cm × 4 cm with a diminished signal intensity on T1-weighted sequences and heightened signal intensity on T2-weighted sequences (figure 2). Notably, HIV enzyme-linked immunosorbent assay (ELISA) testing yielded negative results. Subsequently, the patient underwent left superficial parotidectomy under general anesthesia for excision of the swelling. Overall, the lesion appeared as a pale rounded nodule enclosed by a full capsule and connected to a small portion of parotid tissue. Its dimensions measured approximately 5 cm × 3,2 cm × 4,6 cm. The tumor's surface felt smooth and tender, with the overlying capsule showing no abnormalities in texture or color (figure 3). Histopathological examination of the excised specimen confirmed the diagnosis of a Benign Lymphoepithelial Cyst of the parotid. Follow-up evaluations at 3 months, 6 months, and 1 year postoperatively revealed no evidence of recurrence, with preservation of normal facial nerve function (figure 4).

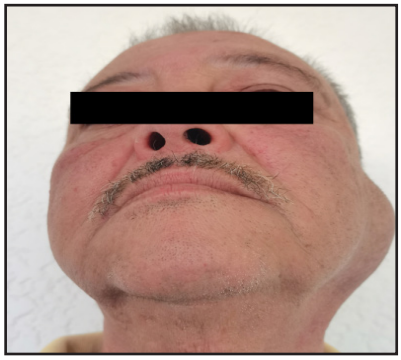

Figure 1: showing a firm, non-adherent oval swelling (5 × 4 cm) in the left parotid region detected on clinical examination.

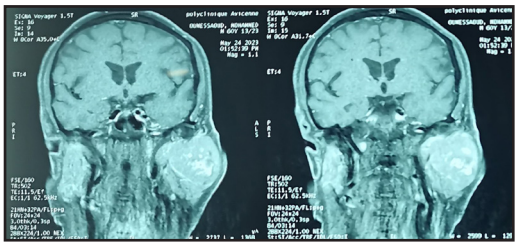

Figure 2: showing a 5 × 4 cm cystic lesion in the left parotid gland with low T1 and high T2 signal intensity on MRI.

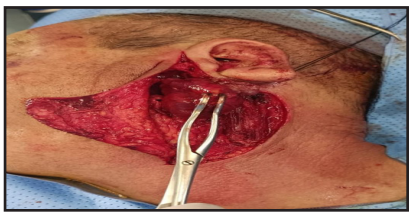

Figure 3: showing a smooth, tender tumor surface with a normal overlying capsule.

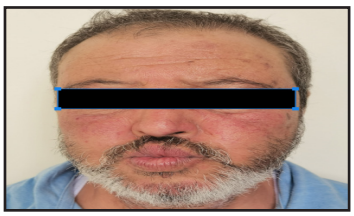

Figure 4: showing no recurrence and preserved facial nerve function at 3, 6, and 12 months postoperatively.

DISCUSSION:

Benign lymphoepithelial cysts (BLCs), characterized by rare cystic neoplasms with benign embryonic dysplasia, exhibit a notable propensity for the lateral neck region, albeit occurring less frequently in the oral cavity or the parotid gland [2,3]. The inception of BLC's recognition in the parotid gland traces back to 1895 when Hildebrant documented the first reported case. Subsequently, the literature encompasses approximately seventy reported instances of such cysts. The advent of the HIV epidemic marked a significant juncture, as it coincided with a gradual rise in the incidence of BLCs within the parotid gland. Researchers discerned a compelling correlation between BLCs and HIV infection, with the majority of cases indicating a close association, and BLCs emerging as an early clinical manifestation in this context. The prevalence of BLC symptoms is observed in approximately 3%-6% of HIV-positive adults and 1%-10% of HIV-positive children. In stark contrast, the occurrence of BLCs among non-HIV-infected patients is notably infrequent, and the precise prevalence of BLCs within this demographic remains undocumented [4-7]. The key lesson to take away is that the identification of BLCs should consistently trigger suspicion of HIV infection. When confronted with a diagnosis of BLC, clinicians must verify the patient's HIV status promptly to facilitate the timely implementation of suitable therapeutic measures. In the present case, the results of HIV tests, including antibody and nucleic acid tests, were negative. Bernier and Bhaskar [8] coined the term "lymphoepithelial cyst" with the intention of emphasizing that this lesion does not represent an embryologic remnant. They provided a specific definition, characterizing it as either a solitary or multiple cysts situated within lymph nodes in proximity to salivary glands. According to their postulation, BLCs emerge due to the degeneration of cystic salivary gland inclusions within lymph nodes. Conversely, some researchers propose the origin of BLCs to be remnants originating from the branchial arch, as evident in cases described as "branchial cysts". The pathogenesis of BLCs is intrinsically linked to the infiltration of HIV-infected cells into the lymphoid tissue of the salivary glands. This phenomenon triggers a cascade of events characterized by lymphoid hyperplasia and metaplasia within the salivary ducts, culminating in obstructive processes, ductal dilation, and subsequent cyst formation [9]. Despite the ambiguous pathogenesis of BLCs in immunocompetent patients, postulations suggest a comparable process of lymphoid hyperplasia occurring in the context of viral infections other than HIV. A noteworthy case reported by Naidoo et al. underscores this notion, where an immunocompetent patient with chronic otitis media experienced BLC development. Their analysis proposed that the longstanding ear infection prompted chronic lymphatic drainage into intracarotid lymph nodes, thereby inducing ductal obstruction and consequent cyst formation [10]. In a study carried out by Joshi et al. [5], it was revealed that the prevalence of BLC was threefold higher in males compared to females. The majority of cases were unilateral and primarily affected the right parotid gland, in contrast to the current case at hand. The average age of onset reported in the study was 44 years, whereas our case involved a 60-year-old male patient. The predominant clinical manifestation of BLCs presents as a slow growing, painless, and palpable swelling, with normal movable overlying skin. This mass has the potential to attain considerable dimensions, ultimately culminating in pronounced physical disfigurement and facial asymmetry. Notably, the presence of BLECs does not generally impede salivary gland functionality, and individuals typically maintain normal salivary secretion despite the presence of the cyst. Preoperative supplementary diagnostic techniques encompass computed tomography (CT), magnetic resonance imaging (MRI), ultrasound, and fine needle aspiration (FNA) [9]. Through a comprehensive analysis of pertinent literature [4,11], it has been ascertained that characteristic CT manifestations of BLCs encompass the presence of a singular, thin-walled round or oval cystic focus within the parotid gland, exhibiting distinct margins and demonstrating low density within the cystic cavity. During contrast-enhanced scans, discernible enhancement is observable along the cystic walls, with no concurrent enhancement detected within the cyst itself. Subsequently, MRI predominantly unveiled a solitary thin-walled cystic anomaly situated within the parotid gland, possessing a rounded or oval configuration and characterized by a well-defined boundary.

T1-weighted imaging demonstrated a diminished signal intensity inside the cyst, while T2-weighted imaging displayed a heightened signal intensity. Notably, the cyst's wall experienced marked enhancement during contrast-enhanced scans, whereas no corresponding enhancement was discerned within the cystic cavity [5].Notably, fine needle aspiration (FNA) serves as a pivotal supplementary technique for the clinical diagnosis of lateral cervical lesions. Cytological criteria for diagnosing BLC via FNA encompass the presence of viscous, yellow, pustular fluid, absence of nucleated cells, presence of keratinocytes, and the existence of squamous epithelial cells exhibiting varying degrees of maturation [4]. The conclusive identification of cystic lesions within the parotid gland relies exclusively on histopathological examination, a course of action exemplified in our own case. Under the spectrum of benign lymphoepithelial cysts (BLCs), a diversity of epithelial types has been delineated. Although squamous epithelium remains the most prevalent, instances of diverse combinations encompassing cuboidal, columnar, ciliated columnar, and mucin-producing epithelia have been documented. Furthermore, rare instances of squamous epithelium featuring sebaceous differentiation have been cataloged [5] The hallmark histopathological pattern features a cleft lined by glandular or squamous epithelium, enveloped by an abundant lymphoid tissue enriched with discernible germinal centers. Some specimens even exhibit the presence of salivary gland tissue and ducts [12]. The primary indication for addressing BLCs typically pertains to cosmetic concerns. As previously highlighted, this pathology has the potential to induce severe aesthetic distortions, resulting in social isolation and depression. Among the array of therapeutic alternatives, options encompass observation, repetitive aspiration, sclerotherapy, radiotherapy, and surgical intervention. Notably, observing asymptomatic patients reveals that recurrent aspiration therapy generally proves ineffective, as cystic reappearance transpires within a span of weeks to months. Sclerotherapy, while employed selectively, is predominantly feasible when cystic fluid can be aspirated, and radiotherapy finds greater utility in cases of BLCs associated with HIV infection [6]. While all treatment modalities yield partial responses, the sole approach consistently showcasing complete resolution devoid of recurrence is parotidectomy. Consideration of complications stemming from parotidectomy as a therapeutic avenue for BLCs parallels comparisons made with routine superficial parotidectomy for pleomorphic adenoma [13]. In the hands of skilled surgeons, parotidectomy for benign conditions has exhibited a stable paresis rate of 2.3% (pertaining to one or more facial nerve branches), while partial superficial parotidectomy has exhibited an unvarying paresis rate of 0% [13,14]. Another factor of consideration is the consequential surgical defect that arises from the excision of these substantial cysts, which can lead to a visibly deforming indentation. The rectification of parotidectomy-induced defects can be approached through a multitude of cosmetic reconstruction methods, including the utilization of cadaveric dermal matrix (Alloderm®), abdominal fat grafts, sternocleidomastoid muscle flaps, de-epithelialized radial forearm free flaps, and rectus abdominus free flaps [15]. In the case presented here, there was no requirement for reconstructive procedures, and the patient experienced favorable postoperative outcomes without any associated discomfort throughout a six-month follow-up period.

CONCLUSION:

In conclusion, the investigation into benign lymphoepithelial cysts (BLECs) within the parotid gland provides a multifaceted perspective on their clinical presentation, diagnosis, treatment, and underlying mechanisms. The distinctiveness of BLCs demands careful consideration in clinical practice, with their potential link to HIV infection highlighting the importance of comprehensive assessment, particularly in immunocompromised individuals. Despite various therapeutic options, surgery remains the preferred approach for treating BLC, as underscored by this comprehensive literature analysis and case illustration. This article aims to emphasize the importance of considering BLC as a potential differential diagnosis for parotid swellings, even in individuals without HIV infection.

REFERENCES

[1] Shanti RM, Aziz SR. HIV-associated salivary gland disease. Oral Maxillofac Surg Clin North Am. 2009 Aug;21(3):339-43.

[2]Fujibayashi T, Itoh H. Lymphoepithelial (so-called branchial) cyst within the parotid gland. Report of a case and review of the literature. Int J Oral Surg. 1981 Aug;10(4):283-92.

[3]Rahman S, Shaari R, Hassan R. Parotid lymphoepithelial cyst: A case report. Arch Orofac Sci 2006;1:71-5.

[4] Najib Z, Berrada O, Lahjaouj M, Oukessou Y, Rouadi S, Abada RA, Roubal M, Mahtar M. Cervical lymphoepithelial cyst: Case report and literature review. Ann Med Surg (Lond). 2020 Dec 28;61:185-187.

[5]Joshi J, Shah S, Agarwal D, Khasgiwal A. Benign lymphoepithelial cyst of parotid gland: Review and case report. J Oral Maxillofac Pathol. 2018 Jan;22(Suppl 1):S91-S97.

[6]Pillai S, Agarwal AC, Mangalore AB, Ramaswamy B, Shetty S. Benign Lymphoepithelial Cyst of the Parotid in HIV Negative Patient. J Clin Diagn Res. 2016 Apr;10(4):MD05-6.

[7]Dave SP, Pernas FG, Roy S. The benign lymphoepithelial cyst and a classification system for lymphocytic parotid gland enlargement in the pediatric HIV population. Laryngoscope. 2007 Jan;117(1):106-13.

[8]Bernier JL, Bhaskar SN. Lymphoepithelial lesions of salivary glands; histogenesis and classification based on 186 cases. Cancer 1958; 11:1156-79.

[9]Ahamed AS, Kannan VS, Velaven K, Sathyanarayanan GR, Roshni J, Elavarasi E. Lymphoepithelial cyst of the submandibular gland. J Pharm Bioallied Sci. 2014 Jul;6(Suppl 1):S185-7.

[10]Khadilkar MN, Prasad V, Santhoor VS, Kamath MP, Domah H. Lymphoepithelial Cyst of Parotid in an Immunocompetent Patient with Chronic Otitis Media. Case Rep Otolaryngol. 2017;2017:5169364.

[11]Glosser JW, Pires CA, Feinberg SE. Branchial cleft or cervical lymphoepithelial cysts: etiology and management. J Am Dent Assoc. 2003 Jan;134(1):81-6.

[12]Mhawej R, Richa T, Melkane AE. Benign lymphoepithelial cyst of unusual location: A case report. Acta Otolaryngol Case Rep 2016,1:110-2.

[13]Koch M, Zenk J, Iro H. Long-term results of morbidity after parotid gland surgery in benign disease. Laryngoscope. 2010 Apr;120(4):724-30.

[14]Guntinas-Lichius O, Klussmann JP, Wittekindt C, Stennert E. Parotidectomy for benign parotid disease at a university teaching hospital: outcome of 963 operations. Laryngoscope. 2006 Apr;116(4):534-40.

[15]Ghanem T. Parotid defects. Facial Plast Surg Clin North Am. 2009 May;17(2):263-9.