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Research Article

CYSTIC LYMPHOID HYPERPLASIA OF THE PAROTID GLAND

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ABSTRACT

Lymphoid infiltrates of the salivary gland can be either reactive or neoplastic. The reactive lesion, cystic lymphoid hyperplasia may be associated with Sjogren's syndrome, human immunodeficiency virus infection,or may occur as an isolated salivary gland enlargement. The authors report a case of cystic lymphoid hyperplasia without Sjogren's syndrome. The diagnosis is mainly based on histopathological and Immunohistochemical analysis. The main differential diagnosis is a low-grade mucosa-associated lymphoid tissue type lymphoma especially that patients with cystic lymphoid hyperplasia have a particularly high risk of subsequently developing lymphoma. The distinction between parotid neoplasms and reactive lesion prior to surgery is difficult, and patients presenting with benign lymphoepitheliallesions are often misdiagnosed preoperatively and receive unnecessary surgery.

INTRODUCTION

Benignlymphoepithelial lesion (BLEL), also referred to as cystic lymphoid hyperplasia, lymphoepithelialsialadenitis (LESA), myoepithelialsialadenitis, Sjogren's-type sialadenitis [1],or autoimmune sialadenitis of the salivary glands, is characterized histologically by an extensive infiltration of chronic inflammatory cells that replace the salivary parenchymal space, which is composed of acinar and ductal cells[2]. Although BLEL-type lesions are most often associated with Sjogren's syndrome (SS) and other connective tissue diseases, they can occur as isolated salivary gland lesion in patients without associated symptoms. While patients with parotid neoplasms require surgery, patients with BLEL and similar lesions do not. However, the distinction between parotid neoplasms and BLEL prior to surgery is difficult, and patients presenting with BLEL lesions of the parotid are often misdiagnosed preoperatively and receive unnecessary surgery[1]. We report a patient who presented with a CLH without Sjogren's syndrome or HIV infection. We also discuss the differential diagnostic aspects of salivary gland lesions that are composed of lymphoid and epithelial components.

Case presentation

A previously healthy 44-year-old female presented with a five months history of a left-sidedparotid swelling that had increased slowly in size and with low pain.

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There was no associated dry mouth (xerostomia), dry eyes (xerophthalmia) or cervical lymphadenopathy. The function of facial nerve was normal. Radiological findings (CT and MRI scans) showed a poorly-defined, 5-cm cystic parotid mass located in the left gland (Fig. 1).

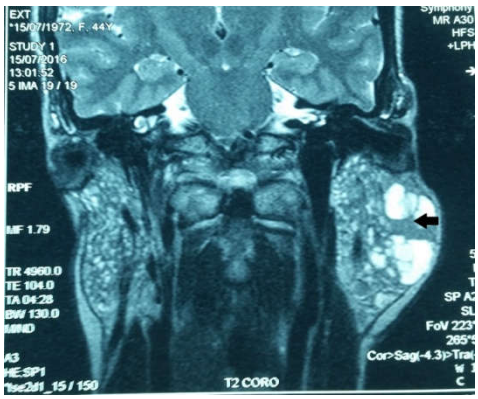

Figure 1 MRI image of the neck shows a multi-cystic lesion in the left parotid gland (arrow).

Fine needle aspiration biopsy was not performed. It yielded scattered macrophages and a few small lymphocytes, and was interpreted as cyst fluid. The patient subsequently underwent a leftparotidectomy. Intraoperatively, a poorly-circumscribed parotid lesion measuring 5.2 cm × 3.2 cm was identified. Histological examination on low power magnification revealed a solid-multicystic lesion composed of adense lymphoid infiltrate, featuring multiple lymphoid follicles with enlarged, irregular germinal centers containing tingiblebody macrophages (Fig. 2). The epithelium of the cyst was

composed of non-keratinous squamous cells overlying a dense lymphoid infiltrate.

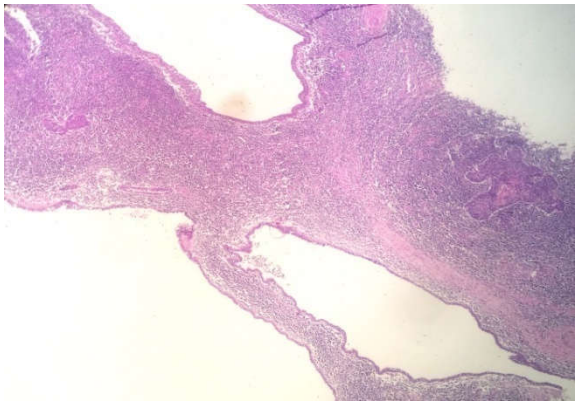

Figure 2 Photomicrograph of lymphoepithelial parotid lesion showing solid-multicystic lesion with non-keratinising squamous epithelial cyst wall (Haematoxylin& eosin staining; low power magnification ×100).

In several areas, the squamous epithelium grew down into the subjacent lymphoid component, forming anastomosing broad cords and basaloid epithelial islands with variably abundant intraepithelial lymphocytes (Fig. 3).

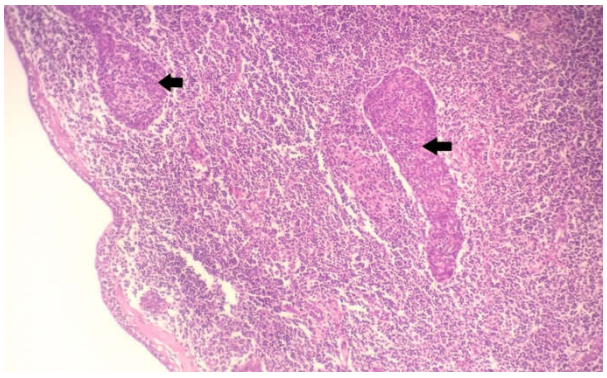

Figure 3 Lymphoepithelial islands/complexes containing nests of polygonal cells with eosinophilic cytoplasm that have an epithelial appearance (H&E; medium power magnification ×250) (arrows).

Some of these lymphoepithelial islands harbouredeosinophilic basement membrane-like material (Fig. 4).

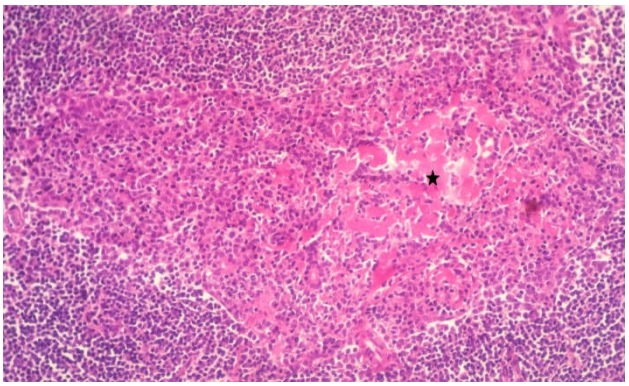

Figure 4 Lymphoepithelial islands/complexes with variable amounts of hyaline (basement membrane-like) material (H&E; high power magnification ×400) (star).

In addition, there were several variably dilated ducts where the epithelium was infiltrated by small lymphoid cells with uniform round nuclei and a moderate amount of pale cytoplasm. Immunohistochemical study showed that the lymphoid component was composed of CD20 positive B cells, and CD3 and CD5 positive T cells. The T cells were mainly identified in the interfollicular regions, but were also found scattered within the lymphoid follicles. The germinal center B cells expressed CD10, but not Bcl2. CD21 revealed preserved and focally expanded follicular dendritic meshworks. The lymphoid cells located within the epithelium of the cyst and epithelial islands were predominantly CD20positive B cells (Fig. 5), with a few scattered CD3 positive T cell.

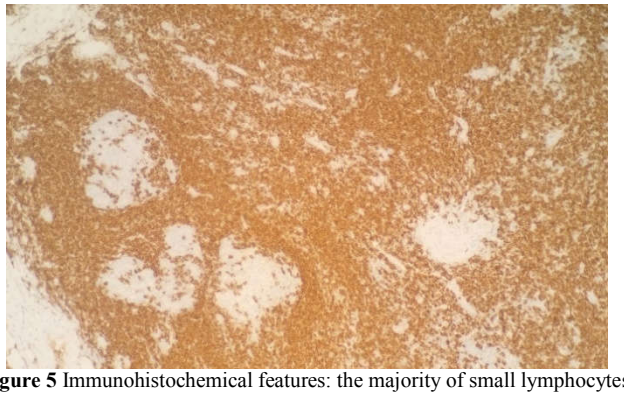

Figure 5 Immunohistochemical features: the majority of small lymphocytes represent CD20B lymphocytes (H&E ×250)

Immunohistochemistry for broad-spectrum cytokeratins (AE1/3; 5/6) highlighted the epithelial component (Fig. 6).The diagnosis of cystic lymphoid hyperplasia was retained. The patient made a good postoperative recovery and was discharged home on the 5th postoperative day. The serologic testing for HIV infection was negative.

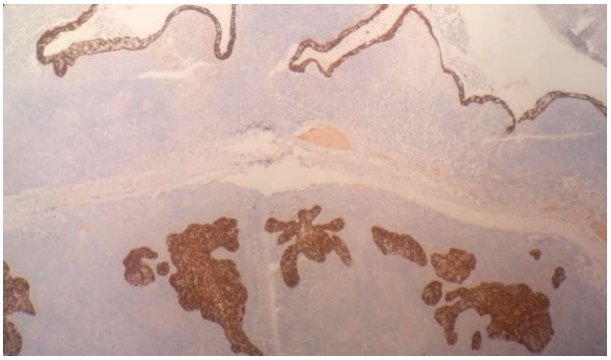

Figure 6 Immunohistochemical features: positivity of lymphoepithelial islands for CK5/6 (H&E ×100)

DISCUSSION

Of all the exocrine glands, the parotid gland is uniquely distinguished by the presence of abundant intraglandular lymphoid tissue. This feature is attributed to developmental characteristics of the gland, where at 6 ~ weeks of gestation, dichotomously branching ductal structures arising from the oral ectoderm proliferate into lymphoid-rich ectomesenchyme before the formation of the glandular capsule. This may lead to entrapment of lymphoid tissue within the parotid gland, which becomes intermixed with salivary gland acini and ducts. This intimate histologic relationship between lymphoid tissue and glandular epithelium is evident in several nonneoplastic lesions of the parotid[3].

Three different causes for the enlargement have been recognized [4]:

1. Follicular hyperplasia of deep jugular lymph nodesand to a lesser degree peri- and intraparotid lymphnodes in the setting of persistent generalized lymphadenopathy of HIV infection.

2. Lymphoepithelial lesion characterized by diffusesalivary gland enlargement due to pronounced lymphocytic infiltration, atrophy of gland parenchyma, and replacement of ducts by epimyoepithelial islands. A subset of these represents marginal zoneB-cell lymphomas of mucosa-associated lymphoidtissue (MALT lymphoma).
3. Cystic lymphoid hyperplasia showing multiple, often bilateral, epithelial-lined cysts and epimyoepithelial islands in a hyperplastic lymphoid stroma} (that reported in our case).Three to 10% of HIV-infected patients develop salivary gland lesions[5] and cystic lymphoid hyperplasia is common[6].It can occasionally be the first clinical sign and can reveal HIV infection [7].Although its presence is very indicative of HIV infection, morphologically, similar lesions have been reported in HIV-negative patients and without any features of Sjogren's disease[8].Our patient had no pathological history and HIV serology was negative.

Wu et al. reported in their study of cystic lesions of the parotid gland in 64 non-HIV-infectedpatients that 27% of patients have unilateral multilocular cystic lesions and their mean age was 52 years, unlike HIV-positive patients where the lesions develop mainly in young patient and are often bilateral[9]. Histologically, salivary gland lesions composed of both epithelial (with variable degrees of cystic change) and lymphoid components have a wide-ranging differential diagnosis, which includes various entities with different aetiopathogeneses, treatment approaches and prognoses. There is a significant clinical and histopathological overlap cystic lymphoepitheliallesions (CLEL) between HIV and non-HIV patients. However, the presence of bilateral parotid masses, lymphoepithelial complexes and multinucleated histiocytes are more commonly encountered in HIV-associated CLEL [10]. And, the unilateral involvement is more commonly found in sporadic non-HIV-related CLEL [11] (as in our case).Based onhistomorphological analysis, the most difficult differential diagnosis of CLH is lymphoepithelialsialadenitis (LESA). LESA is an autoimmune exocrinopathy that often occurs in the context of Sjögren's syndrome, which predominantly affects middle-aged or older women. The histopathologic features of LESA are similar to those of CLH, though cystic components are more commonly found in CLH. However, their absence or presence does not reliably distinguish CLH from LESA [12].Given the fact that most, if not all, cases of extranodal marginal zone B cell lymphoma (EMZBCL) of salivary glands arise from pre-existing LESA, the differential diagnosis may be difficult. EMZBCL is the most common lymphoma affecting salivary glands, accounting for approximately half of all salivary gland lymphoma cases. The earliest stage of EMZBCL is characterisedbyan expansion of monocytoid and/or centrocyte-like B cells around the lymphoepithelial complexes. The neoplastic lymphoid B cells form interconnected sheets and colonize the marginal zones. Immunohistochemically, these neoplastic B cells are typically positive for CD20, CD79a and negative for CD5, CD10, and CD23 [5]. In such instances, an epithelial neoplasm with an associated lymphoid component has to be included in the differential diagnosis. These neoplasms include lymphadenomas and lymphadenocarcinomas. Lymphadenomas are well-circumscribed tumours composed of cytologically bland neoplastic epithelial cells arranged in islands and duct-like structures, occasionally forming cysts within a dense lymphoid stroma. The cystic component may show cuboidal or columnar morphology [12]. Lymphadenocarcinoma shows varying degrees of nuclear atypia, increased mitotic activity, and most importantly, infiltrative growth. Primary lymphoepithelial carcinoma of the salivary glands is biologically and histologically analogous to nonkeratinising nasopharyngeal carcinoma [5].

CONCLUSION

Our case illustrates a cystic lymphoid hyperplasia of the parotid gland that may occur as isolated enlargement in patient without history of Sjogren's syndrome or HIV infection and the diagnosis prior to surgery is challenging. It is still sometimes difficult to distinguish between tumor and benign lymphoepithelial lesion. If the diagnosis can be made, non-neoplastic lesions will not require excision. Future studies would be useful to provide experience with atypical cases of BLEL.

Abbreviations

BLEL: Benign lymphoepithelial lesion
CLH: Cystic lymphoid hyperplasia
SS: Sjogren's syndrome
HIV: Human immunodeficiency virus
CT and MRI scans: Computed tomography and Magnetic resonance imaging
CLEL: Cystic lymphoepithelial lesions
LESA: Lymphoepithelialsialadenitis
EMZBCL: Extranodal marginal zone B cell lymphoma
H&E: Haematoxylin& eosin staining

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