

Lymphoepithelial Cyst in Jugal Mucosa

Quiste Linfoepitelial en la Mucosa Yugal

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ABSTRACT: Mouth lymphoepithelial cyst is rare, with few cases reported in literature. The aim of this article is to describe a clinical case, focusing on clinical and diagnostic aspects, treatment and prognosis. The lesion was one year old and had developed as a fibrous nodule in the jugal mucosa of a 71-year-old leucoderma patient. Considering focal inflammatory fibrous hyperplasia, fibroma and mucocele as differential diagnosis, excisional biopsy was carried out. A cystic cavity limited by pseudostratified epithelium without projections into the conjunctive tissue, with lymphoid tissue within, was microscopically identified. Without postoperative adverse events, the one-year clinical followup confirmed the favorable prognosis of this kind of lesion.

KEY WORDS: non-odontogenic cysts, lymphoepithelial cyst, ectopic lymphoid tissue.

INTRODUCTION

As it is an extremely rare lesion. There are few reports in the literature of oral lymphoepithelial cysts, with isolated cases or short series, as we notice from the reports of Bhaskar (1966) with 24 cases, Knapp (1970) with 13, Giunta & Cataldo (1973) with 21, Buchner & Hansen (1980) with 38 cases, and also Hu *et al.* (2005) with 3 cases.

Having controversial etiology, the lesion may be considered a developmental abnormality, resulting from the imprisonment of ectodermal remnants of the first and second branchial arch in the interior of the lymph nodes (Gold 1962). Other studies suggest that the entity does not have branchial origin, but represents a cystic alteration of the ectopic epithelium included in the lymph node (Bhaskar). The third etiological hypothesis is local trauma, which would stimulate epithelial cell proliferation, forming the cyst (Buchner & Hansen; Epivatianos *et al.*, 2005).

The cyst may occur at any age and in both sexes, but it is more frequent between the thirties and the fifties, in the mouth floor and tongue, although it can occur in the soft palate and in other mucosae (Buchner & Hansen). When present in the parotid gland, may be associated to cervical adenopathy,

mainly in HIV-positive patients (Mandel & Reich, 1992). Its occasional appearance in the parotid gland of patients with Sjögren syndrome is not uncommon (Chaudhry *et al.*, 1984).

The most frequent clinical aspect is an asymptomatic submucous nodule, of 0.1- 2.0 cm in its greatest diameter, of fibrous consistency on palpation and pink-yellowish in color (Epivatianos *et al.*; Chaudhry *et al.*).

The differential diagnosis involves mainly fibrous hyperplasia, fibroma, papiloma and mucocele. The definite distinction is obtained by histopathological analysis (Buchner & Hansen), which can identify a cystic cavity limited by stratified squamous epithelium without projections to the conjunctive tissue, generally parakeratinized, and with desquamated epithelial cells filling the cyst lumen. Sometimes this epithelial lining may also contain mucous cells communicating with the lining mucosa. The lymphoid tissue in the cyst wall may fill the entire cystic wall or just a part of it (Hu *et al.*).

The treatment basically consists of the surgical removal and relapses have never been reported.

CASE REPORT

A 71-year-old white man presented with submucous nodule in the left jugal mucosa. The nodule was 0.5 cm in its greatest diameter, sessile, with fibrous consistency and smooth and shiny surface. The adjacent jugal mucosa was unaffected (Fig. 1).



Fig. 1. Fibrous exophytic nodule in jugal mucosa.

Because of the clinical characteristics, the diagnostic hypotheses included focal inflammatory fibrous hyperplasia, fibroma and mucocele, the first mentioned being the clinical diagnosis.

Surgical exeresis was carried out and the specimen was examined microscopically. The

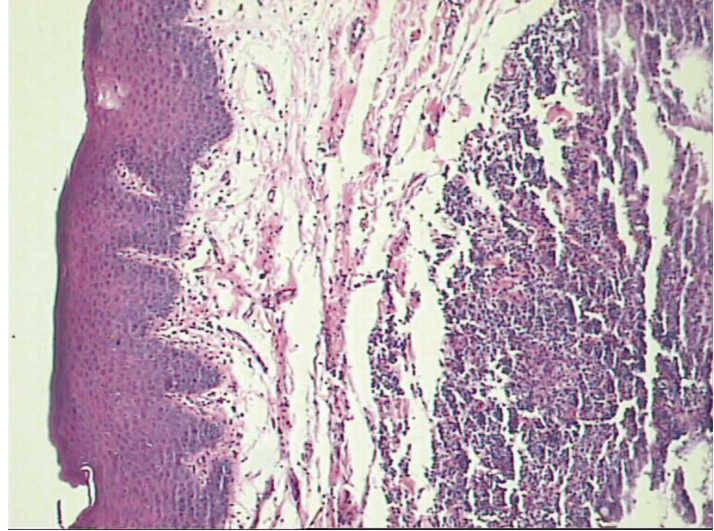


Fig. 2. Cystic cavity limited by pseudo-stratified epithelium and lymphoid tissue in the cystic wall HE. 100X.

examination revealed a cystic cavity limited by stratified squamous epithelium without projections into the conjunctive tissues (Fig. 2). The presence of lymphoid tissue was observed in the cyst wall (Fig. 3), which is compatible with oral lymphoepithelial cyst.

The one-year follow up evolved favorably, and a relapse was not observed.

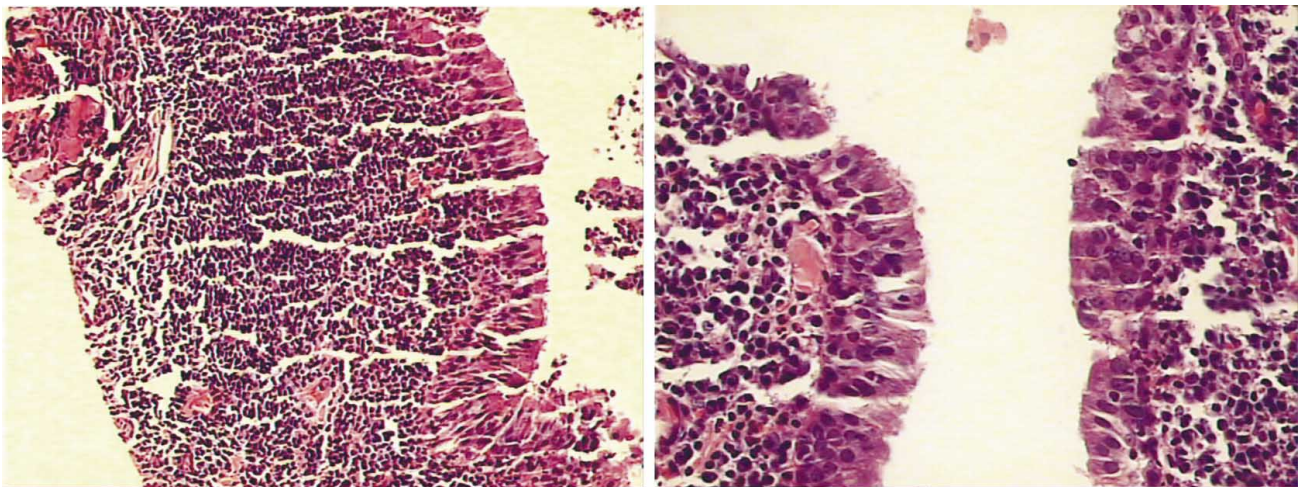


Fig.3. Lymphoid tissue in the cystic wall to the left. HE. 100X. Cystic cavity limited by pseudo-stratified epithelium without projections to the conjunctive tissue to the right. HE. 200X.

DISCUSSION

In the present report, the diagnostic hypothesis of oral lymphoepithelial cyst was not considered, due to the rare occurrence of this lesion in the mouth. Because of its rarity, there is little information available about it, hence the importance of reporting new cases.

Etiopathogenesis remains in the field of speculation, but, because of the histological characteristics of the case studied, the hypothesis of cystic alteration of the ectopic epithelium included in the lymph node can be considered, probably due to local trauma, which would stimulate the proliferation of those epithelial cells, forming the cyst (Bhaskar; Buchner & Hansen; Epivatianos *et al.*).

Regarding localization, the mouth roof has been considered the preferred site, followed by the tongue (Buchner & Hansen). However, these observations are restricted to a small number of clinical reports. No lesion

has been reported in the jugal mucosa, the site evidenced in the present report. This variation in localization sites further emphasize its origin from ectopic epithelium, since this tissue can be present in any part of the oral mucosa.

Literature reports lymphoepithelial cyst in the parotid gland (Bouquot & Nikai, 2001; Mandel & Reich), probably due to the proximity of the parotid gland to the lymphatic chains. In most cases, lesions in that site were associated to HIV-positive patients, probably due to the lymphoid tissue vulnerability to the virus (Poletti *et al.*, 1988).

The treatment administered in the present case was surgical, and relapse was not observed in the one-year postoperative period. This fact is in accordance with the literature since the observations of Giunta & Cataldo, probably because the lymphoepithelial cyst is a non-infiltrative lesion.

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RESUMEN: Un quiste linfoepitelial bucal es raro, con pocos casos reportados en la literatura. El objetivo de este artículo es describir un caso clínico, centrándose en los aspectos clínicos y diagnósticos, tratamiento y pronóstico. La lesión tuvo un año de evolución, y se había desarrollado como un nódulo fibroso en la mucosa yugal de un paciente de 71 años de edad con leucoderma. Teniendo en cuenta la hiperplasia fibrosa inflamatoria, fibroma y mucocelo como diagnósticos diferenciales, se llevó a cabo una biopsia por escisión. Una cavidad quística limitada por epitelio pseudoestratificado sin proyecciones en el tejido conjuntivo, con tejido linfoide en el interior fue identificado microscópicamente. No se observaron eventos adversos postoperatorios, y el seguimiento clínico al año confirmó el pronóstico favorable de este tipo de lesión.

PALABRAS CLAVE: quiste no odontogénico, quiste linfoepitelial, tejido linfoide ectópico.

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