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# Lymphoepithelial cyst on the tongue: case report at unusual location

## *Cisto linfoepitelial em língua: relato de caso em localização incomum*

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## ABSTRACT

Oral lymphoepithelial cyst (OLEC) is an uncommon lesion that develops in oral lymphoid tissue. The aim of the present study was to report a clinical case of OLEC in the tongue. A 22-year-old patient presented a nodular lesion, yellowish, with a softened consistency, measuring 0.5 cm in the ventral surface of the tongue. Under the clinical hypotheses of mucocele and OLEC, excisional biopsy was performed. The histopathological examination revealed a cystic lesion covered by a parakeratinized stratified squamous epithelium, which presented in its fibrous capsule a prominent lymphoid tissue. Based on the definitive diagnosis, surgical excision of the lesion was performed.

**Key words:** oral pathology; non-odontogenic cysts; treatment result.

## INTRODUCTION

Oral lymphoepithelial cyst (OLEC) is a rare and benign lesion of the oral cavity, corresponding to 0.09% to 0.18% of lesions affecting this region[1]. It is a lesion that develops within the oral lymphoid tissue, microscopically similar to the branchial cleft cyst (cervical lymphoepithelial cyst), but much smaller in size[1, 2]. It usually occurs in adults with mean age around the fourth decade of life; it is a lesion rarely found in children. The floor of the mouth is reported as the site of higher incidence (70.7%), followed by lateral border (10.7%) and ventral surface of the tongue (7.3%)[3].

Typically, OLEC appears as a small submucosal mass, generally smaller than 1 cm in diameter; in rare cases, the lesion will be greater than 1.5 cm. The cyst may be firm or soft to palpation, and the covering mucosa is soft and non-ulcerated. The lesion is white or yellow, asymptomatic and often contains keratin in its light with cheesy or creamy appearance. The cyst is asymptomatic, although occasionally some patients complain of swelling or drainage. Pain is rare, but it can occur after trauma[1,3].

Histopathologically, OLEC is characterized by the presence of a cyst covered by a parakeratinized stratified squamous epithelium and a capsule of dense fibrous connective tissue, permeated by a moderate inflammatory mononuclear infiltrate through moderate vascularization[3-5].

The treatment of OLEC consists of conservative surgical removal, and relapses are rare, leading to a good prognosis[6]. Due to its low clinical morbidity and nonspecific symptoms, diagnosis is still a challenge[1].

The present work aims to report a clinical case of OLEC in the ventral surface of the tongue, as well as to carry out the discussion on important clinical and pathological aspects and treatment of this condition.

## CASE REPORT

A male patient, 22-year-old, leucoderma, presented at the Dental clinics of the Department of Dentistry of the Universidade Federal do Rio Grande do Norte (UFRN), Natal (RN), Brazil, complaining of painless swelling in the tongue with evolution of three months. Past medical history was noncontributory, and the patient did not have human immunodeficiency virus (HIV).

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Extraoral examination of gland and neck lymph nodes showed signs of normality; the intraoral physical examination showed presence of a nodular lesion, with exophytic growth, sessile implantation, yellowish coloration and softened consistency, measuring approximately 0.5 cm in the ventral surface of the tongue (**Figure 1**). From the clinical hypotheses of mucocele or OLEC, an excisional biopsy was performed under local anesthesia. The minor salivary glands associated with the lesion were removed. Histopathological examination revealed the presence of a cystic lesion covered by parakeratinized stratified squamous epithelium surrounded by a capsule of fibrovascular connective tissue. The presence of lymphoid tissue in the cystic capsule was evidenced, besides germinative centers, and the diagnosis of OLEC was issued (**Figure 2**). After nine months of follow-up, no evidence of relapse was found.

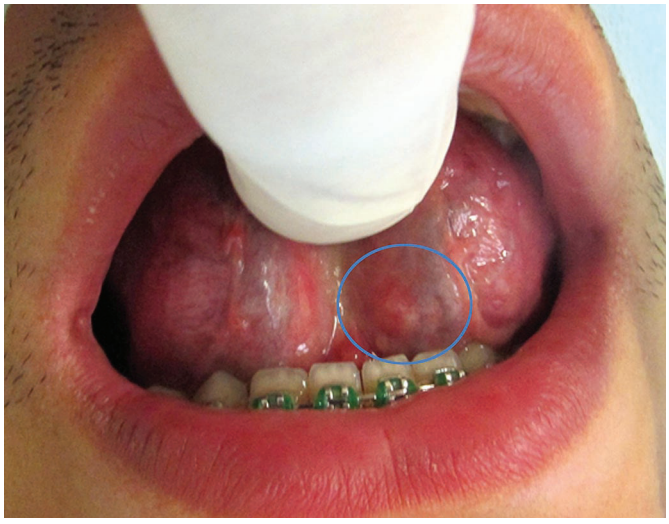

FIGURE 1 – *Clinical aspect of OLEC*

*Nodular lesion, in the ventral surface of the tongue, yellowish, slow and submucosal growing, measuring approximately 0.5 cm.*

*OLEC: oral lymphoepithelial cyst.*

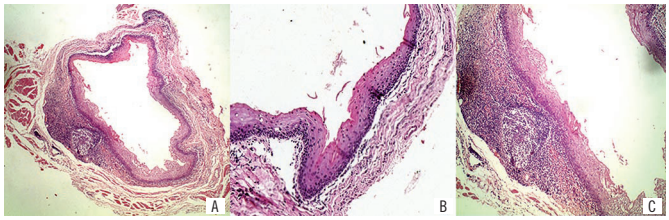


FIGURE 2 – *Histopathological aspect of OLEC*

*A and B) presence of a pathological cavity covered by parakeratinized stratified squamous epithelium surrounded by a connective fibrovascular tissue capsule (100×); C) presence of lymphoid tissue in the cystic capsule, and germinal centers (400×).*

*OLEC: oral lymphoepithelial cyst.*

## DISCUSSION

OLEC is a rare lesion in the mouth. There is variation in the literature regarding its most common location. Yang *et al*. (2012)[3] observed that the floor of the mouth is the anatomical site of higher incidence (70.7%) of OLEC, followed by the lateral border (10.7%) and the ventral surface (7.3%) of the tongue. However, Uchoa-Vasconcelos *et al*. (2014)[1] found that the tongue, followed by the floor of the mouth, are the most frequent location. The occurrence of OLEC in the ventral surface of the tongue is rare, as there are only 91 cases described in the literature[7, 8]. The present case reported is the 92nd case of OLEC in the ventral surface of the tongue.

OLEC mainly occurs in adults around the fourth decade of life with female predilection 60%-80%[1, 3], which differed from the present case, since it occurred in a male patient.

Yang *et al*. (2012)[3] performed a clinical analysis of 120 cases of OLECs and found a predilection for the female gender, with a ratio between men and women of 1:2 and ages ranging from 2 to 75 years, with mean of 44.1 years. Meanwhile, Ahamed *et al*. (2014)[9] identified that such lesion was more frequent between the second and third decades of life, corroborating the present case reported.

The etiopathogenesis of OLEC is uncertain. Some authors have hypothesized that the ectopic foci of the embryonic epithelium are trapped in the lymphoid tissue and may proliferate to form a cyst[10]. However, other authors have suggested that OLECs are the result of obstruction of normal oral tonsil crypts[2, 5, 10]. It is also possible that the traumatic implantation of epithelial cells into deeper tissue may lead to the formation of OLEC[2].

Other studies were carried out in an attempt to clarify a possible association of OLEC with HIV infection as part of diffuse infiltrative lymphocytosis syndrome, with an occurrence of 3%-10% of HIV-positive patients[9]. This hypothesis is due to the increased incidence of OLEC in HIV-positive patients, thus suggesting that it is one of the clinical manifestations of this infection[11]. However, the relationship between HIV infection and OLECs has not been fully elucidated yet.

Clinically, OLEC has features similar to other nodular lesions affecting the oral cavity. The evolution, growth rate and symptoms are not specific, therefore it is always difficult distinguishing this lesion and other benign lesions, such as mucocele, mucous retention cyst, lipoma, fibroma, sialolithiasis and dermoid cyst[2, 3, 10].

Due to its low clinical morbidity and nonspecific symptoms, the diagnosis of OLEC remains a challenge[3]. Microscopic findings are critical to conclude the diagnosis. Microscopic findings demonstrate a cystic cavity covered by stratified squamous epithelium, adjacent to

it exhibit lymphocytes masses with lymphoid follicles, and underlying a fibrous connective tissue capsule[2]. These characteristics are compatible with the lesion presented in this report.

OLEC treatment includes a conservative approach to the lesion. Decompression may be performed by aspirating the intralesional fluid, thereby reducing cystic osmotic pressure. Subsequently, definitive treatment is performed by complete enucleation of the lesion associated with excision of the involved gland[3]. This treatment was similar to that adopted in the case reported here, in which was performed the complete enucleation of the lesion associated with the excision of the accessory glands involved.

After nine months of follow-up, no signs of lesion recurrence were found, supporting the literature findings, which showed that most patients are completely recovered by excision with no lesion recurrence rates[1].

Based on the literature and in the reported case, OLEC is typically present as small asymptomatic nodules located in the oral cavity, which emphasizes the importance of a detailed clinical examination for small lesions that are often neglected. Conservative surgical excision is the treatment for OLEC, and in cases of larger lesions, marsupialization produces excellent results[12].


## RESUMO

*O cisto linfoepitelial oral (CLEO) é uma lesão incomum que se desenvolve no tecido linfoide oral. O objetivo do presente trabalho foi relatar um caso clínico de CLEO na língua. Paciente de 22 anos de idade exibia uma lesão nodular, de coloração amarelada e consistência amolecida, medindo 0,5 cm na região ventral de língua. Sob as hipóteses clínicas de mucocele e CLEO, foi realizada biópsia excisional. O exame histopatológico revelou lesão cística revestida por epitélio escamoso estratificado paraceratinizado, que apresentava em sua cápsula fibrosa tecido linfoide proeminente. Com base no diagnóstico definitivo, foi realizada a excisão cirúrgica da lesão.*



***Unitermos:*** *patologia bucal; cistos não odontogênicos; resultado de tratamento.*


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