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Case Report

A rare occurrence of lymphoepithelial cyst in the palatine tonsil: a case report and discussion of the etiopathogenesis

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Abstract: Lymphoepithelial cysts are uncommon benign lesions that present as painless yellowish nodules arising from various sites in the oral cavity and other parts of the body. Their etiopathogenesis is controversial, but most authors have assumed that they develop from obstruction of crypts in oral lymphoid aggregates, thus they are not true cysts but pseudocysts of retention. This paper describes a case of a large lymphoepithelial cyst located in the tonsil of a 21-year-old man complaining of a lump in the throat for four months. The patient underwent excisional biopsy, and the histopathological features showed squamous epithelium surrounded by lymphoid tissue, which were characteristically consistent with a lymphoepithelial cyst. We discuss the etiopathogenesis of these lesions and treatment modalities, which can consist of conservative surgery or only follow-up examination.

Keywords: Lymphoepithelial cyst, palatine tonsil, pseudocysts, etiopathogenesis

Introduction

Lymphoepithelial cysts (LECs) are uncommon benign lesions arising from entrapment or proliferation of epithelium in association with lymphoid tissue [1-3] that may occur in various sites, including the pancreas [4], thyroid [5] and mediastinum [6]. In the head and neck region, LECs have been described in lateral areas of the neck (known as branchial cyst), the parotid gland, and the oral cavity [1-3].

Oral LECs present as painless yellowish submucosal nodules, usually smaller than 1 cm and located on the floor of the mouth and ventral or posterolateral surface of the tongue [7]. Demographic data are controversial. In a recent clinical analysis of 120 cases, Yang et al found female predilection, with a male-to-female ratio of 1:2 and ages ranging from 2 to 75 years with a mean of 44.1 years [8].

Microscopically, oral LECs are characterized by a cystic cavity containing keratin that is lined with a parakeratinized stratified squamous epithelium, exhibiting a smooth interface with the lamina propria. Surrounding the epithelial lining, the cyst capsule is usually thick and presents intensively infiltrated by lymphocytes arranged as germinal centers [3, 7, 9].

There is no consensus regarding etiopathogenesis of oral LECs, and Knapp's theory is the most accepted [7, 8, 10]. Supporting this classic theory, Buchner and Hansen [7] stated that oral LECs are not true cysts resulting from epithelial proliferation but rather pseudocysts originating from a dilated, obstructed crypt of the oral tonsil in which either purulent material or desquamated epithelial lining accumulates [7, 10].

The occurrence of LEC in the palatine tonsils is extremely rare, as there are few cases described in the medical literature [11]. Herein, we report a case of LEC arising from the right palatine tonsil and discuss its histogenesis.

Case presentation

An otherwise healthy 21-year-old man was referred to the Oral Medicine Center at the School of Dentistry, Federal University of Goiás, with a 4-month history of a painless lump in the

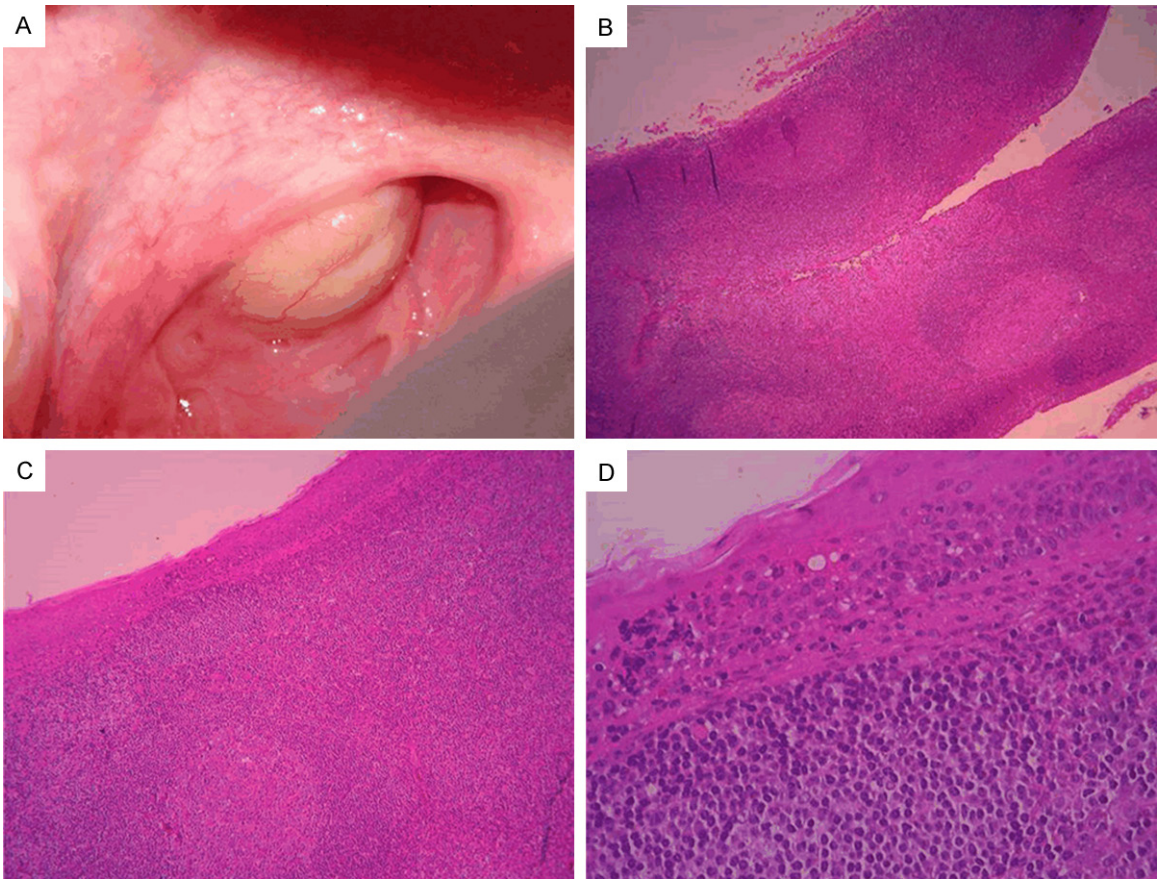

Figure 1. A. Clinical view of the lesion, arising from the right palatine tonsil and measuring approximately 1.5 cm in diameter. B. The cystic cavity contains rare epithelial cells scattered in the lumen, and the cyst wall is thick and exhibits lymphoid tissue arranged as typical germinal centers (H&E stain, 40× magnification). C. The cyst is lined by thin, parakeratinized, stratified squamous epithelium, displaying a flat interface with the underlying connective tissue and lacking rete pegs (H&E stain, 100× magnification). D. Intraepithelial infiltration of lymphocytes (H&E stain, 400× magnification).

throat. His medical and dental histories were unremarkable. There were no findings on extraoral examination. The intraoral inspection revealed a well-circumscribed, yellowish, soft nodule that measured approximately 1.5 cm in diameter arising from the right palatine tonsil (**Figure 1A**). A closer examination showed numerous small vessels over the surface of the lesion. The clinical diagnosis was LEC. Excisional biopsy was performed under local anesthesia, and no complications were observed during the surgery. The specimen obtained was sent to the Oral Pathology Laboratory of the Federal University of Goiás.

Microscopic examination showed a ruptured cystic cavity without keratin that contained rare epithelial cells scattered in the lumen (**Figure 1B**). The cavity was lined with thin, parakeratinized, stratified squamous epithelium, showing a flat interface with the underlying connective tissue and lacking rete pegs (**Figure 1C**). The subepithelial cyst wall was thick with very scarce connective tissue and densely infiltrated by lymphocytes arranged as typical germinal centers (**Figure 1B**, **1C**). In some areas, intraepithelial infiltration of lymphocytes could be seen (**Figure 1D**). The histopathological characteristics confirmed the diagnosis of LEC. The 1-week postoperative period was uneventful, but the patient was lost on subsequent follow-up.

Discussion

LECs are lesions that can be found in different parts of the body and are characterized by the presence of squamous epithelium surrounded

Table 1. Case series of lymphoepithelial cysts published in the literature

Author(s)	Year	N	Location (Number of cases)
Bhaskar & Colonel [20]	1966	24	FM (15), T (8), P (1)
Knapp [10]	1970	13	FM (7), T (2), P (4)
Acevedo & Nelson [9]	1971	9	FM (9)
Giunta & Cataldo [17]	1973	21	FM (17), P (2), BM (1), RM (1)
Buchner & Hansen [7]	1980	38	FM (19), T (14), P (4), RM (1)
Toto et al [21]	1982	6	FM (2), T (3), P (1)
Chaudhry et al [22]	1984	24	FM (24)
Nonaka et al [23]	2011	10	FM (2), T (4), P (1), OF (2)
Yang et al [8]	2012	120	FM (46), T (60), P (6), BM (3), Others (5)

FM: floor of mouth; T: Tongue, P: Palate; BM: Buccal Mucosa; RM: Retromolar area; OF: Oropharynx.

by lymphoid tissue [1-7]. Although the histopathological scenario suggests the involvement of both epithelium and lymphocytes in the genesis of these cysts, the exact mechanism of pathogenesis has not yet been completely elucidated [3, 4].

Several theories have been proposed to explain the etiopathogenesis of LEC. In their classic study, Bhaskar and Bernier sustained that LEC arose from proliferation of glandular epithelium within lymph nodes of the parotid and cervical regions [1]. Strengthening this assumption, Vickers and Von Der Muhll demonstrated cystic proliferation within lymph nodes of hamsters after surgical autogenous transplantation of bucal epithelium [12]. In addition, Adsay et al affirmed that the abdominal lymph nodes might be the source for the development of pancreatic LEC [13], in accordance with other studies [14, 15]. Although not completely rejecting the role of lymph nodes, Wu et al proposed that LECs of the parotid would start from the dilatation of ducts due to epithelial hyperplasia, which might be stimulated by lymphocytic infiltration in focal sialadenitis [2].

Regarding the etiopathogenesis of oral LECs, Knapp's theory remains the most broadly accepted. Knapp claimed that these oral cysts, actually pseudocysts, arise not from lymph nodes but from submucosal lymphoid aggregates located on the floor of the mouth, the ventral surface of the tongue, and the soft palate. Knapp coined these aggregates of ectopic lymphoid tissue of oral tonsils [10]. These "new tonsils" were added to the previously known "regular tonsils" placed at the posterior part of the oral cavity, oropharynx, and nasopharynx, which together form the lymphatic Waldeyer's ring. The ring includes the nasopharyngeal tonsil (adenoid), the paired tubal tonsils, the paired palatine (faucial) tonsils, and the lingual tonsil [7, 16].

Knapp's theory, endorsed by Buchner and Hansen [7] and Giunta and Cataldo [17], assumed that LECs, which are actually, would arise from obstruction of the crypt of an oral or palatine tonsil, resulting in a dilated cavity lined by epithelium with communication breaches with the external environment and containing keratin and desquamated cells in the lumen. However, such communication with the oral cavity has not been easily demonstrated. Acevedo and Nelson [9] do not find any continuity in nine reported cases. Giunta and Cataldo [17] and Buchner and Hansen [7] found such continuity in a few cases. Consequently, Buchner and Hansen proposed the possible existence of an additional mechanism in the pathogenesis of oral LECs [7].

It is worth noting that the great majority of intraoral LECs have been encountered on the floor of the mouth and on the ventral/posterolateral surface of the tongue [7-9, 17], which are exactly the same locations as oral tonsils [10]. In 2012, Kashima et al reported an oral tonsil in the floor of the mouth mimicking clinically a benign neoplasm or lymphoepithelial cyst. The microscopic aspect of the tonsil was very similar to that of an LEC, but without a cystic cavity. Thus, the authors postulated that oral tonsils would be earlier stages of LECs, corroborating Knapp's theory [18].

Assuming that LECs develop from oral lymphoid aggregates or tonsils, the challenge is to explain the paucity of case reports of LECs in the palatine tonsil [11] and other tonsils [19] of the lymphatic Waldeyer's ring, since all tonsils have the same histological architecture. Palatine tonsils were not cited as a location for LECs in the case

series published in the literature, as we can see in **Table 1** [7-10, 17, 20-23]. This observation makes our case report a rarity in the medical literature.

However, in 1987, Giunta reported 11 cases of yellow nodules located in the tongue and palatine tonsils. According to clinical criteria, the author categorized the lesions in two types, bacterial plugs and pseudocysts. Of the 11 cases described, 3 were presumably diagnosed as pseudocysts of the palatine tonsil and had spontaneous resolution over a period of 3 to 6 months [24]. The author stated these pseudocysts were the same type of lesions called lymphoepithelial cysts in other studies. Giunta affirmed that, in his experience, the occurrence of such pseudocysts in the palatine tonsils was not rare, but as lesions are small, they tend to undergo spontaneous regression before the patient seeks medical care. Spontaneous regression of a yellow swelling on the floor of the mouth, presumably a pseudocyst or LEC, was also reported by McDonnell [25].

In this paper, we reported a case of LEC arising from the palatine tonsil with four months of evolution and large size, considering the majority of intraoral LEC are less than 1 cm [7]. Based on Knapp, Buchner and Hansen, and Giunta, our case must be more properly diagnosed as a pseudocyst of the palatine tonsil that resulted from the obstruction of the tonsillar crypt, generating the accumulation of keratin within the lumen. Indeed, extravasation of keratin could be seen intraoperatively.

The tendency is for this type of lesion to resolve in a few months when a rupture occurs in the obstructed portion of the crypt. However, as it was a larger lesion that was causing discomfort to the patient, the minor surgery under local anesthesia produced a quicker resolution. In addition, the patient expressed some concern about his lesion, and the excisional biopsy allowed histopathology and patient reassurance.

Conclusions

Yellowish nodules located in the palatine tonsil usually represent lymphoepithelial cysts, better termed as pseudocysts with retention of keratin. Depending on the lesion size and discomfort reported by the patient, these nodules may be treated with a conservative approach, consisting only of a follow-up examination. When surgery is selected, a simple excision or marsupialization produces excellent results and a low recurrence rate.

Disclosure of conflict of interest

None.

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References

[1] Bhaskar SN, Bernier JL. Histogenesis of branchial cysts: a report of 468 cases. Am J Pathol 1959; 35: 407-23.
[2] Wu L, Cheng J, Maruyama S, Yamazaki M, Tsuneki M, Lu Y, He Z, Zheng Y, Zhou Z, Saku T. Lymphoepithelial cyst of the parotid gland: its possible histopathogenesis based on clinicopathologic analysis of 64 cases. Hum Pathol 2009; 40: 683-92.
[3] Stramandinoli-Zanicotti RT, de Castro Ávila LF, de Azevedo Izidoro AC, Izidoro FA, Schussel JL. Lymphoepithelial cysts of oral mucosa: two cases in different regions. Bull Tokyo Dent Coll 2012; 53: 17-22.
[4] Younus S, Bleibel W, Bleibel H, Hernady N. Lymphoepithelial cyst of the pancreas. Dig Dis Sci 2007; 52: 3136-39.
[5] Choi CJ, Choi SW, Cho JG, Woo JS. Bilateral lymphoepithelial cysts of the thyroid gland. Thyroid 2010; 20: 111-13.
[6] Ulku R, Yilmaz F, Eren S, Onat S. Lymphoepithelial Cyst of the Mediastinum. Tex Heart Inst J 2005; 32: 440-1.
[7] Buchner A, Hansen LS. Lymphoepithelial cysts of the oral cavity: a clinicopathologic study of thirty-eight cases. Oral Surg 1980; 50: 441-9.
[8] Yang X, Ow A, Zhang CP, Wang LZ, Yang WJ, Hu YJ, Zhong LP. Clinical analysis of 120 cases of intraoral lymphoepithelial cyst. Oral Surg Oral Med Oral Pathol Oral Radiol 2012; 113: 448-452.
[9] Acevedo A, Nelson JF. Lymphoepithelial cysts of the oral cavity. Oral Surg 1971; 31: 632-6.
[10] Knapp MJ. Pathology of oral tonsils. Oral Surg Oral Med Oral Pathol 1970; 29: 295-304.
[11] Choi J, Kisik K, Kim B. Lymphoepithelial cyst of the palatine tonsil. Ear Nose Throat J 2010; 89: 584-5.
[12] Vickers RA, Von Der Muhl OH. An investigation concerning inducibility of lymphoepithelial

cysts in hamsters by autogenous epithelial transplantation. J Dent Res 1966; 45: 1029-1032.
[13] Adsay NV, Hasteh F, Cheng JD, Bejarano PA, Lauwers GY, Batts KP, Klöppel G, Klimstra DS. Lymphoepithelial cysts of the pancreas: a report of 12 cases and a review of the literature. Mod Pathol 2002; 15: 492-501.
[14] Arai T, Kino I, Nakamura S, et al. Epidermal inclusions in abdominal lymph nodes. Report of two cases studied immunohistochemically. Acta Pathol Jpn 1992; 42:126-9.
[15] Horie Y, Taguchi K, Akagi T, Ogawa H. Lymphoepithelial cyst in peripancreatic lymph node. J Okayama Surg Pathol Assoc 1990; 27: 53-7.
[16] Perry M, Whyte A. Immunology of the tonsils. Immunol Today 1998; 19: 414-21.
[17] Giunta J, Cataldo E. Lymphoepithelial cysts of the oral mucosa. Oral Surg 1973; 35: 77-84.
[18] Kashima K, Takamori K, Igawa K, et al. Oral tonsil in the floor of mouth: Ectopic oral tonsillar tissue simulating benign neoplasms. Oral Science International 2012; 9: 29-31
[19] Hemmaoui B, Sahli M, Jahidi A, Benariba F. Nasopharyngeal branchial cyst. Eur Ann Otorhinolaryngol Head Neck Dis 2014; 131: 321-2.
[20] Bhaskar SN, Colonel L. Lymphoepithelial cysts of the oral cavity. Oral Surg Oral Med Oral Pathol 1966; 21: 120-128.
[21] Toto PD, Wortel JP, Joseph G. Lymphoepithelial cysts and associated immunoglobulins. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1982; 54: 59-65.
[22] Chaudhry AP, Yamane GM, Scharlorck SE, SunderRaj M, Jain R. A clinicopathological study of intraoral lymphoepithelial cysts. J Oral Med 1984; 39: 79-84.
[23] Nonaka CF, Henriques AC, de Matos FR, de Souza LB, Pinto LP. Nonodontogenic cysts of the oral and maxillofacial region: demographic profile in a Brazilian population over a 40-year period. Eur Arch Otorhinolaryngol 2011; 268: 917-922.
[24] Giunta JL. Bacterial plug versus pseudocyst of the tonsils. Oral Surg Oral Med Oral Pathol 1987; 63: 202-7.
[25] McDonnell D. Spontaneous regression of a yellow sublingual swelling: a case report. Pediatr Dent 1990; 12: 388-389.