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Correspondence

Oral lymphoepithelial cyst at the lateral border of the tongue

KEYWORDS
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Oral lymphoepithelial cyst (LEC) is a rare cystic lesion that develops within oral lymphoid tissue.[1–5] Here, we presented an oral LEC at the right posterior lateral border of the tongue of a 42-year-old female patient.

This 42-year-old female patient came to our oral mucosal disease clinic for treatment of a small mass at the right posterior lateral border of the tongue for 2.5 months. The mass was soft, painless, and elevated, and it measured approximately 0.5 cm in greatest dimension. The clinical impression was a fibroma. Because the tumor mass was small with pink and smooth surface, the treatment plan was total surgical excision of the mass. After discussing with the patient and obtaining the signed informed consent, the small tumor mass was totally excised under local anesthesia. The removed soft tissue specimen was sent for histopathological examination. Microscopically, it showed a multicystic lesion lined by the parakeratinized stratified squamous epithelium without rete ridges (Fig. 1A–D). The cystic lumens were filled with desquamated epithelial cells and shreds of parakeratin with pyknotic epithelial cell nuclei (Fig. 1E and F). The most characteristic feature was the presence of lymphoid tissues in nearly the entire fibrous cystic wall (Fig. 1A–D) and germinal centers in the lymphoid tissues (Fig. 1G and H). The above-mentioned striking findings finally confirmed the histopathological diagnosis of an oral LEC.[1–5]

The pathogenesis of oral LEC is still not clear, but there are two possible theories that explain the formation of the oral LEC. The developmental lesion theory suggests that oral LECs may develop from the salivary or surface mucosal epithelium enclaved in the lymphoid tissue during embryogenesis. The subsequent proliferation of the entrapped epithelium in the lymphoid tissue finally leads to the formation of oral LEC. The other obstruction theory suggests that the tonsillar crypt may become obstructed or pinched off from the surface, resulting in the accumulation of the desquamated epithelial cells and keratin debris in the blind tonsillar crypt and the subsequent formation of the oral LEC.

Oral LEC is the oral counterpart of the branchial cleft cyst (cervical LEC).[1,2] Sykara et al.[2] reviewed 316 cases of oral LECs in the literature. They found that there is no gender predilection for the oral LEC patients with a female to male ratio of 1:0.9. The oral LEC patients show a broad age distribution (2–81 years), but they are more frequently in the third to fifth decades of life. The 316 oral LECs occur most commonly at the floor of mouth (160 cases, 50.63%) and the tongue (114 cases, 36.08%), followed in a descending order by the soft palate, buccal mucosa, hard palate, labial mucosa, palatoglossal fold, glossodesmus, and retromolar area.[2] Microscopically, oral LECs are predominantly lined by thin, parakeratinized stratified squamous epithelium with flattened rete pegs. However, they may be lined by orthokeratinized or non-keratinized stratified squamous epithelium or pseudostratified columnar or ciliated columnar epithelium with mucous/goblet cells. The lymphoid tissue may surround the entire oral LEC or part of the cyst and is often arranged in a follicular pattern with

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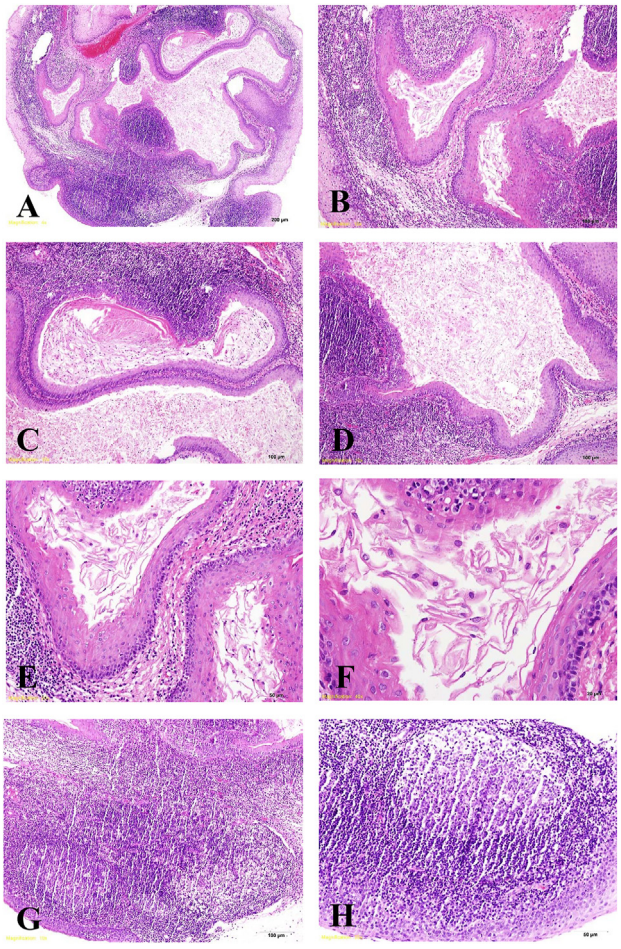

Figure 1 Histopathological photomicrographs of our case of oral lymphoepithelial cyst. (A—D) Low-power and medium-power photomicrographs exhibiting a multicystic lesion lined by the parakeratinized stratified squamous epithelium without rete ridges as well as the presence of lymphoid tissues in nearly the entire fibrous cystic wall. (E and F) High-power photomicrographs demonstrating desquamated epithelial cells and shreds of parakeratin with pyknotic epithelial cell nuclei in the cystic lumens. (G and H) Medium-power and high-power photomicrographs showing the germinal centers in the lymphoid tissues (H&E; original magnification; A, 4×; B, C, D and G, 10×; E and H, 20×; F, 40×).

prominent germinal centers.[1—5] The oral LECs are best treated by total surgical excision, and recurrence of the oral LEC has not been reported in the literature.[2]

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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None.

References

1. Neville BW, Damm DD, Allen CM, Chi AC. Developmental defects of the oral and maxillofacial region. In: Neville BW, Damm DD, Allen CM, Chi AC, eds. *Oral and maxillofacial pathology*, fourth ed. St Louis: Elsevier, 2016:33—5.
2. Sykara M, Ntovas P, Kalogirou EM, Tosios KI, Sklavounou A. Oral lymphoepithelial cyst: A clinicopathological study of 26 cases and review of the literature. *J Clin Exp Dent* 2017;9. e1035—43.
3. Wu YH, Kuo YS, Chiang CP, Lin PY. Lymphoepithelial cyst of the tongue. *J Dent Sci* 2019;14:209—10.
4. da Silva KD, Coelho LV, do Couto AM, et al. Clinicopathological and immunohistochemical features of the oral lymphoepithelial cyst: a multicenter study. *J Oral Pathol Med* 2020;49:219—26.
5. Cunha JLS, Roza ALOC, Cruz VMS, et al. Oral lymphoepithelial cyst: A collaborative clinicopathologic study of 132 cases from Brazil. *Head Neck Pathol* 2022;16:268—77.

Bing-Wei Huang[†]
Chia-Yu Li[†]
Department of Dentistry, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan

Ying-Tai Jin[**]
Department of Pathology, Taiwan Adventist Hospital, Taipei, Taiwan
Department of Pathology, National Cheng Kung University Hospital, Tainan, Taiwan

Chun-Pin Chiang[*]
Department of Dentistry, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, Hualien, Taiwan
Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan
Graduate Institute of Oral Biology, School of Dentistry, National Taiwan University, Taipei, Taiwan

**Corresponding author. Department of Pathology, Taiwan Adventist Hospital, No. 424, Section 2, Bade Road, Taipei 10556, Taiwan.
E-mail address: yingtaijin@gmail.com (Y.-T. Jin)

*Corresponding author. Department of Dentistry, Hualien Tzu Chi Hospital, Buddhist Tzu Chi Medical Foundation, No. 707, Section 3, Chung-Yang Road, Hualien 970, Taiwan.
E-mail address: cpchiang@ntu.edu.tw (C.-P. Chiang)

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[†] These two authors have equal contribution to this study.