Journal of Dental Sciences (2019) 14, 209–210

Available online at www.sciencedirect.com

ScienceDirect

journal homepage: www.e-jds.com

Correspondence

Lymphoepithelial cyst of the tongue

KEYWORDS
Lymphoepithelial cyst;
Tongue;
Oral cavity

Oral lymphoepithelial cysts are uncommon developmental cysts that occur most frequently in the ventral surface and lateral borders of the tongue and the floor of mouth.[1] Here, we presented a case of lymphoepithelial cyst at the right ventral surface of the tongue of a 64-year-old male patient.

This 64-year-old male patient came to oral mucosal disease clinic in the dental department of Far Eastern Memorial Hospital for treatment of a leukoplakia lesion at the right lower labial mucosa and a small soft tissue tumor at the right ventral surface of the tongue near the lingual frenum area (Fig. 1A). The soft tissue tumor was asymptomatic and present for 3 years. Clinical examination showed a yellowish and slightly firm submucosal mass measuring about $0.6 \times 0.5 \times 0.3$ cm in size (Fig. 1A). The oral leukoplakia lesion was treated by cryotherapy with liquid nitrogen. The lingual soft tissue tumor was removed under local anesthesia and sent for histopathological examination. Microscopically, it showed a lymphoepithelial cyst lined by parakeratinized stratified squamous epithelium and few keratin shreds in the cystic lumen. There was a prominent lymphocytic infiltrate in the subepithelial connective tissue of the fibrous cystic wall (Fig. 1B–D). However, no lymphoid follicles were noted in the marked lymphocytic infiltrate.

The lymphoepithelial cyst is a rare developmental cyst. When the lymphoepithelial cyst is found in the lateral neck region, it is called as branchial cleft cyst. Sykara et al. reported 26 cases of oral lymphoepithelial cysts. These 26 oral lymphoepithelial cysts occur in 14 women and 11 men (one 32-year-old female patient with two identical lymphoepithelial cysts located at the bilateral posterior lateral borders of the tongue). The mean age of the 25 patients was 33 ± 10 years. Oral lymphoepithelial cysts are usually firm (76%) or soft (24%) and covered by normal to yellow (80%) or white oral mucosa (20%). The majority of lesions measure less than 1 cm in greatest dimension (18 cases, 78%; three cases with unknown size). Most cases are located at the tongue (18 cases, 69%) with 9 at the ventral surface, 8 at the lateral borders, and one at the tongue tip. The remaining 8 cases (31%) are at the floor of mouth. All lesions are asymptomatic and surgically excised under local anesthesia. Microscopically, 24 cases show parakeratinized epithelial lining and all 26 cases have lymphoid tissues in the fibrous cystic walls with 14 cases demonstrating a follicular pattern with prominent germinal centers. The oral lymphoepithelial cyst can be easily diagnosed without the help of immunohistochemical staining to identify the tumor or cell origin.[2–5] The pathogenesis of oral lymphoepithelial cyst remains obscure. Total excision is the treatment of choice for all oral lymphoepithelial cysts and recurrence of the lesion has not been reported in the literature.[1]

https://doi.org/10.1016/j.jds.2019.02.003

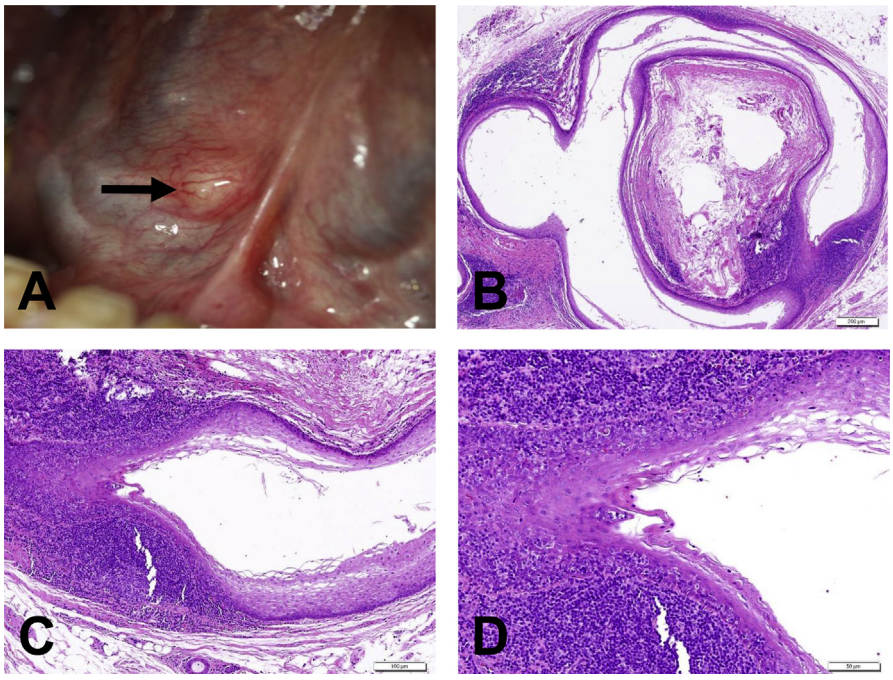

Figure 1 Clinical and histological photographs of our case of oral lymphoepithelial cyst. (A) The clinical photograph showed a yellowish submucosal mass at the right ventral surface of the tongue near the lingual frenum (arrow). (B) Low-power microphotograph demonstrated a cystic cavity with the epithelial lining and bluish fibrous cystic wall (hematoxylin and eosin stain; original magnification, 4×). (C and D) Medium-power (C; original magnification, 10×) and high-power microphotographs (D; original magnification, 20×) showed a lymphoepithelial cyst lined by parakeratinized stratified squamous epithelium and few keratin shreds in the cystic lumen. There was a prominent lymphocytic infiltrate in the subepithelial connective tissue of the fibrous cystic wall (hematoxylin and eosin stain).

Conflicts of interest

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

References

1. 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.
2. Wu YH, Chang JYF, Wang YP, Chiang CP. Langerhans cells in plexiform ameloblastoma. *J Dent Sci* 2017;12:195—7.
3. Chang CH, Wu YC, Wu YH, Sun A, Kuo YS, Chiang CP. S100 protein-positive Langerhans cells in 80 dentigerous cysts. *J Dent Sci* 2017;12:405—12.
4. Kuo YS, Wu YH, Sun A, Chiang CP. Burkitt's lymphoma of the mandible. *J Dent Sci* 2017;12:421—3.
5. Kuo YS, Wu YH, Sun A, Chiang CP. Eosinophilic granuloma of the mandible mimicking a periapical lesion. *J Dent Sci* 2017;12:424—5.

Yu-Hsueh Wu
Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan
Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, Taiwan

Ying-Shiung Kuo
Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan
Department of Dentistry, National Taiwan University Hospital, College of Medicine, National Taiwan University, Taipei, Taiwan

Chun-Pin Chiang
Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan
Graduate Institute of Clinical Dentistry, School of Dentistry, National Taiwan University, Taipei, 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

Ping-Yi Lin*
Department of Dentistry, Far Eastern Memorial Hospital, New Taipei City, Taiwan

*Corresponding author. Department of Dentistry, Far Eastern Memorial Hospital, No. 21, Section 2, Nanya South Road, Banciao District, New Taipei City 220, Taiwan.
E-mail address: rhyne.tw@yahoo.com.tw

Received 10 February 2019
Available online 20 March 2019