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Lingual Cyst with Respiratory Epithelium

An 18-month-old girl born at 37 weeks' gestation to a 37-year-old gravida 4, para 4 (G4P4) mother, with a birth weight of 3,800 g, presented with a tongue mass noted at birth. The lesion was characterized by a bluish discoloration and soft consistency but was not associated with feeding difficulties and its persistence through the months that followed prompted consult and subsequent work-up. Magnetic resonance imaging (MRI) demonstrated a simple cystic lesion measuring 2.2 × 2.2 × 2.9 cm (anteroposterior × transverse × craniocaudal) with no internal septations, solid components, restricted diffusion, or abnormal contrast enhancement. It exerted a mass effect on the mid-sublingual region and was situated just superomedial to the sublingual gland. Following clinical and radiologic evaluation, surgical excision of the lesion was recommended to the parents.

The specimen submitted for histopathologic evaluation consisted of a brown to pink tan, irregular, soft tissue fragment measuring 1.8 x 1.5 x 0.6 cm. Microsections of the entirely submitted specimen revealed a cyst lined with non-dysplastic respiratory-type epithelium consisting of ciliated, pseudostratified, columnar epithelium. *(Figures 1 and 2)* A focal area of reactive squamous metaplasia was also identified. *(Figure 3)* The cyst wall consisted of dense connective tissue lacking inflammatory cells including neutrophils and histiocytes. The tissue also showed an absence of lymphoid aggregates or follicles, dermal appendages and goblet cells. These histopathologic and clinical findings were consistent with a lingual cyst with a respiratory epithelium (LCRE).

LCRE is a rare congenital developmental cyst of the oral cavity. Since its initial description, only a limited number of cases have been reported, highlighting its uncommon occurrence and the lack of comprehensive understanding of its clinicopathologic characteristics. A LCRE predominantly affects males and pediatric patients, with cases in very young children remaining exceptionally rare. To date, only a few patients younger than four years have been documented in the literature, emphasizing the rarity of this presentation and the significance of the present case.[1–4]

LCRE most frequently arises in the anterior two-thirds of the tongue, although lesions involving the midline have also been described. Clinically, patients typically present with a painless cystic swelling causing tongue enlargement. Depending on the lesion size and patient age, symptoms may include airway compromise, feeding difficulties, impaired breastfeeding, dysphagia, and speech disturbances, underscoring the importance of early diagnosis and management.[1,3–5]

The pathogenesis of LCRE remains incompletely understood. The most widely accepted hypothesis suggests that the lesion results from the entrapment of undifferentiated foregut

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endoderm during the fourth week of embryonic development. These pluripotent cells subsequently differentiate into respiratory-type epithelium, giving rise to a cystic lesion within the tongue.[1,3,4] Despite this proposed embryologic mechanism, the histogenesis of LCRE remains controversial, making histopathological examination essential for definitive diagnosis.

Histopathologically, LCRE must be distinguished from several developmental cysts of the oral cavity, including dermoid, epidermoid, teratoid, thyroglossal duct, lymphoepithelial, mucous retention, and foregut-derived cysts.[3–5] The defining feature of LCRE is the presence of pseudostratified ciliated columnar or cuboidal respiratory epithelium lining the cyst, without evidence of gastric or intestinal mucosa. *(Figure 2)* This characteristic differentiates LCRE from lingual alimentary cysts and other foregut-derived lesions.[1,4,5] In terms of management, treatment of LCRE is similar to the other cysts and entails enucleation and/or marsupialization.[4,5]

Given the overlapping histologic features among congenital lingual cysts, descriptive histopathologic terminology has been advocated to improve diagnostic consistency. Manor and colleagues proposed classifying these lesions according to their predominant epithelial lining rather than presumed embryologic origin,[6] while subsequent authors recommended incorporating additional cyst wall components into the diagnostic description when present,[5] This descriptive approach provides a practical and reproducible framework for the classification of these uncommon lesions and remains widely accepted in the literature.[4,5]

In the present case, the diagnosis of lingual cyst with respiratory epithelium was established based on the characteristic histopathologic finding of a cyst lined predominantly by pseudostratified ciliated respiratory epithelium, consistent with the currently accepted descriptive classification. The clinical presentation is comparable to previously reported cases, particularly with respect to the patient's young age and lesion location. Given the limited number of documented cases in children younger than four years, this report contributes additional evidence to the existing literature and expands the clinicopathologic spectrum of this rare congenital entity.[2–5]

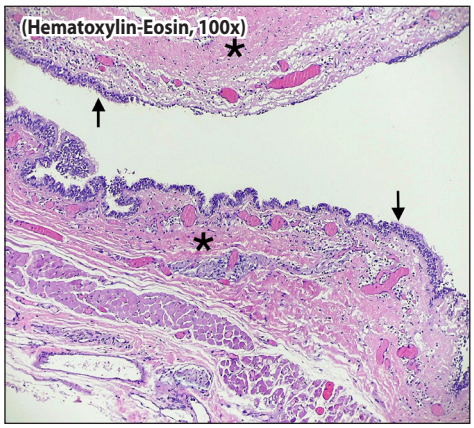


Figure 1. Hematoxylin and eosin-stained slide showing a cystic tissue lined with respiratory epithelium (arrows) and the capsule consisting of dense connective tissue (asterisks). 100x magnification.

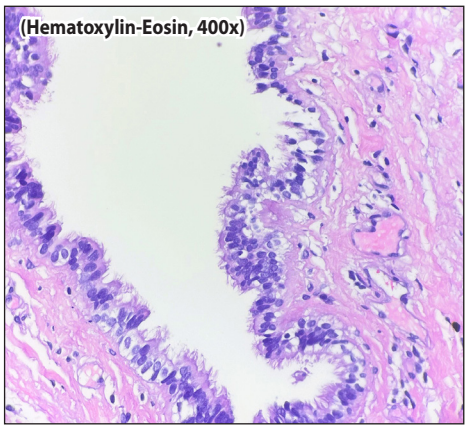


Figure 2. Hematoxylin and eosin-stained slide showing cystic tissue lined with ciliated, pseudostratified, columnar epithelium. 400x magnification.

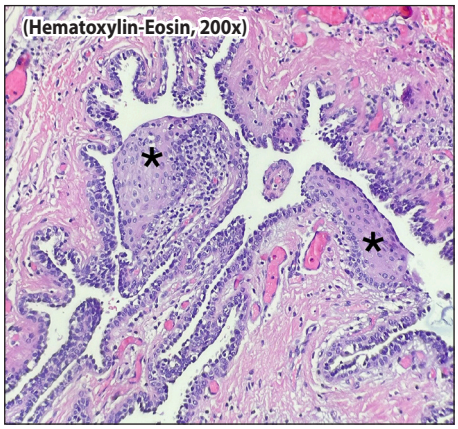


Figure 3. Hematoxylin and eosin-stained slide showing reactive squamous metaplasia (asterisks). 200x magnification.

REFERENCES

1. Azañero WD, Mazzonetto R, León JE, Vargas PA, Lopes MA, de Almeida OP. Lingual cyst with respiratory epithelium: a histopathological and immunohistochemical analysis of two cases. *Int J Oral Maxillofac Surg.* 2009 Apr;38(4):388–392. DOI: 10.1016/j.ijom.2009.01.003; PubMed PMID: 19217261.
2. Ardila CM, Álvarez-Martínez E. A case report of a lingual cyst lined by respiratory epithelium in a child. *Clin Med Insights Case Rep.* 2020 May;13:1179547620919695. DOI: 10.1177/1179547620919695; PubMed PMID: 32528226; PubMed Central PMCID: PMC7263107.
3. Cialente F, De Soccio G, Savastano V, Grasso M, Dello Spedale Venti M, Ralli M, et al. Lingual cyst with respiratory epithelium: The importance of differential diagnosis. *Bosn J Basic Med Sci.* 2021 Jun;21(3):378–382. DOI: 10.17305/bjbms.2020.4716; PubMed PMID: 32464085; PubMed Central PMCID: PMC8112560.
4. Peters SM, Park M, Perrino MA, Cohen MD. Lingual cyst with respiratory epithelium: Report of two cases and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol.* 2018 Dec;126(6):e279-e284. DOI: 10.1016/j.oooo.2018.05.011; PubMed PMID: 29929866.
5. Juneja M, Boaz K, Srikant N, Nandita KP, Vidya M. Lingual cyst lined by respiratory epithelium: A case report and review of literature. *Fetal Pediatr Pathol.*2011; 30(4):225–232. DOI: 10.3109/15513815.2011.555807; PubMed PMID: 21434827.
6. Manor Y, Buchner A, Peleg M, Taicher S. Lingual cyst with respiratory epithelium: an entity of debatable histogenesis. *J Oral Maxillofac Surg.* 1999 Feb;57(2):124-7; discussion 128-9. DOI: 10.1016/s0278-2391(99)90222-7; PubMed PMID: 9973118.