

Oral Foregut Duplicate cyst presenting as tongue-tie in a pediatric patient: a clinicopathologic case report

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Case Report

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Figures are available in the Supplementary Files section.

Figure 1F and 2A,B are not available with this version

Oral Foregut Duplicate cyst presenting as tongue-tie in a pediatric patient: a clinicopathologic case report

Abstract

Oral foregut duplication cysts (OFDCs) are rare congenital anomalies arising from abnormal foregut development during embryogenesis, characterized by ectopic gastrointestinal or respiratory epithelium within the oral cavity. We present the case of a 6-year-old boy referred to with a presumptive diagnosis of ankyloglossia ("tongue-tie") who was found to have a well-circumscribed midline cystic mass within the intrinsic muscles of the tongue. MRI demonstrated a homogeneous T2-hyperintense, T1-isointense lesion with increased diffusivity and minimal wall enhancement. The mass was excised via a ventral tongue approach and delivered intact. Histopathologic examination revealed a cyst wall lined by ciliated respiratory-type epithelium with an underlying smooth muscular layer, consistent with an oral foregut duplication cyst. The differential diagnosis, including ranula, leiomyoma, rhabdomyoma, and malignant peripheral nerve sheath tumor, is discussed. This case highlights the importance of considering foregut duplication cysts in the differential diagnosis of congenital tongue masses, particularly when clinical presentation mimics ankyloglossia. Complete surgical excision is curative, with a low risk of recurrence.

Keywords: Foregut duplication cyst; tongue mass; ankyloglossia; congenital cyst; respiratory epithelium; pediatric oral pathology

Introduction

Foregut duplication cysts are benign developmental anomalies that occur along the foregut-derived alimentary tract. As defined by Rickham et al., foregut duplications must (1) contain intramural smooth muscle, (2) be attached to a structure formed by the foregut, and (3) contain epithelium and/or mucosa derived from the foregut.[1][2] These lesions may contain various types of epithelium including squamous, gastric, respiratory, and cuboidal, and their nomenclature varies accordingly — they have been called lingual choristoma, enterogenous cyst, bronchial duplication cyst, and alimentary tract cyst, among other terms.[2]

Foregut duplication cysts have an overall incidence of approximately 1 in 4,500 and most commonly arise in the thorax and abdomen, with only 0.3% of cases presenting in the head and neck.[2] The oral cavity, specifically the oral tongue, accounts for approximately 60% of head and neck cases.[2][3] The majority of these are asymptomatic at presentation, with a smaller percentage presenting with feeding difficulties, speech abnormalities, and rarely, respiratory distress.[2][3]

We present a case of an oral foregut duplication cyst in a 6-year-old boy that was initially misdiagnosed as ankyloglossia, discuss the clinical, radiographic, and histopathologic features, and review the differential diagnosis of midline tongue masses in the pediatric population.

Case Report

A 6-year-old otherwise healthy boy was referred to the oral and maxillofacial surgery clinic with a presumptive clinical diagnosis of "tongue-tie." The patient's mother reported that at birth, a laser frenectomy procedure was recommended. The child had no difficulty breastfeeding or bottle feeding. At six months of age, a frenectomy was again recommended, but the family declined as the patient had no difficulty eating or drinking and was following a normal growth curve. At preschool age, the patient was referred to speech therapy for difficulty articulating certain vowels. He continued to have no difficulties eating, drinking, or breathing and maintained normal growth. The mother recently noted snoring at night but no daytime sleepiness or apnea. She reported no perceived changes in tongue size and no difficulties with food based on texture or consistency.

On clinical examination, the child appeared in no distress but had somewhat altered speech with imprecise articulation. Head and neck examination revealed no facial asymmetry, no neck masses or lymphadenopathy, and full range of motion of the neck. The skin of the face and neck appeared normal.

Intraoral examination revealed normal mouth opening with mixed dentition in appropriate position without diastemas. The tongue appeared displaced superiorly and posteriorly by a "rubbery" submucosal mass palpable on the ventral aspect of the tongue. The lingual frenum was not enlarged. The floor of the mouth was effaced by the mass bilaterally. The posterior extent of the mass could not be assessed due to tongue displacement and limited patient cooperation. The overlying mucosa and dorsal aspect of the tongue were freely movable over the mass and of normal color and texture. Tongue mobility was limited due to displacement by the mass (Fig. 1A). The remainder of the oral cavity was without mucosal lesions or abnormalities.

MRI of the face with and without contrast demonstrated a circumscribed midline cystic mass within the anterior aspect of the intrinsic muscles of the tongue. The mass demonstrated homogeneous T2 hyperintensity (Fig. 1B) and homogeneous T1 isointensity (Fig. 1C). Diffusion-weighted images and apparent diffusion coefficient (ADC) maps demonstrated increased diffusivity (Fig. 1D, E). The walls of the lesion were mildly thick but smooth with minimal enhancement (Fig. 1F). There was no communication with adjacent structures or spaces, no inflammatory changes, and no lymphadenopathy.

Given the clinical and radiographic findings, the decision was made to proceed with excision under general anesthesia. The mass was approached via a vertical incision along the ventral tongue (Fig. 1G). The orifices of the Wharton ducts bilaterally were identified and found not to be obstructed by the mass. Intraoperatively, the tongue musculature and lingual nerves bilaterally were displaced laterally by the mass. The mass was easily separated from surrounding tissues with blunt dissection and was delivered intact for histopathologic examination (Fig. 1H).

Gross examination revealed a well-circumscribed, smooth-walled cystic mass. Hematoxylin and eosin (H&E)-stained sections demonstrated a cyst wall lined by ciliated respiratory-type epithelium (Fig. 2A, B) with a smooth muscular layer underlying the epithelial lining. Some

chronic subepithelial inflammation was present. No cartilage or seromucinous glands were identified. The histopathologic findings were diagnostic of an oral foregut duplication cyst.

Discussion

Oral foregut duplication cysts are rare congenital anomalies believed to result from disruptions in foregut development during embryogenesis, particularly between the fourth and seventh weeks of gestation.[1][2][4] During this critical period, the foregut differentiates into structures including the pharynx, esophagus, and portions of the respiratory tract. When epithelial separation is incomplete or embryological tissue persists abnormally, duplication cysts may form.[1][4] While no definitive environmental or genetic cause has been identified, some cases have been linked to syndromes such as VACTERL association, suggesting a multifactorial etiology.[1]

The present case is notable for the prolonged misdiagnosis as ankyloglossia. The restricted tongue mobility caused by the submucosal mass mimicked the clinical presentation of tongue-tie, leading to repeated recommendations for frenectomy over a six-year period. This diagnostic pitfall underscores the importance of careful palpation of the tongue in any child presenting with restricted lingual mobility, as a submucosal mass may not be immediately apparent on visual inspection alone.

The differential diagnosis of a well-circumscribed midline tongue mass in a pediatric patient includes several entities:

Ranula. A ranula is a mucous retention cyst arising from the sublingual gland, commonly located on the floor of the mouth. It presents as a bluish, dome-shaped, fluctuant swelling. Histologically, ranulas are pseudocysts lacking an epithelial lining, with mucin surrounded by granulation tissue.[5] In the present case, the intralingual location, the presence of a true epithelial lining, and the smooth muscle wall excluded this diagnosis.

Leiomyoma. Leiomyomas are benign tumors of smooth muscle origin that are rare in the oral cavity, usually presenting as slow-growing, firm, painless nodules. Histologically, they are composed of bundles of spindle-shaped smooth muscle cells with elongated nuclei.[6] The cystic nature of the present lesion and the respiratory epithelial lining were inconsistent with leiomyoma.

Rhabdomyoma. Extracardiac rhabdomyomas occur in the head and neck region and present as well-circumscribed, painless masses. Histologically, they show large polygonal cells with abundant eosinophilic cytoplasm and cross-striations.[7] The cystic architecture and epithelial lining of the present lesion excluded this diagnosis.

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3. Conflict of interest: The authors declare that they have no conflict of interest
4. Ethical approval: All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards
5. Informed Consent for publication was obtained by patients parent as patient was a minor.
6. Data availability : No datasets were generated or analyzed during the current study.
7. Author contribution: Manuscript written by Dr. Kolokythas, Ryan, Van Ess. Images and image descriptions by Dr. Niec.

References

1. The Evolution of Multidisciplinary Head and Neck Cancer Treatment. Contrera KJ, Reddy PD, Helou V, et al. The Laryngoscope. 2025;135(7):2255-2260. doi:10.1002/lary.32064.
2. Reemergence of the Natural History of Otolaryngologic Infections: Lessons Learned From 2 American Presidents. Naples J, Schwartz M, Eisen M. Otolaryngology--Head and Neck Surgery : Official Journal of American Academy of Otolaryngology-Head and Neck Surgery. 2017;157(3):462-465. doi:10.1177/0194599817709437.
3. Introduction: Reimagining Epidemics. Fissell ME, Greene JA, Packard RM, Schafer JA. Bulletin of the History of Medicine. 2020;94(4):543-561. doi:10.1353/bhm.2020.0081.
4. Death in the White House: President William Henry Harrison's Atypical Pneumonia. McHugh J, Mackowiak PA. Clinical Infectious Diseases : An Official Publication of the Infectious Diseases Society of America. 2014;59(7):990-5. doi:10.1093/cid/ciu470.
5. George Washington's Recurrent Health Problems and Fatal Infection Re-Examined. Mackowiak PA. Clinical Infectious Diseases : An Official Publication of the Infectious Diseases Society of America. 2021;72(10):1850-1853. doi:10.1093/cid/ciaa1324.
6. Endocrine and Autoimmune Aspects of the Health History of John F. Kennedy. Mandel LR. Annals of Internal Medicine. 2009;151(5):350-4. doi:10.7326/0003-4819-151-5-200909010-00011.
7. A History of Innovations in the Diagnosis and Treatment of Oral and Head and Neck Cancer. Polverini PJ, Lingen MW. Journal of Dental Research. 2019;98(5):489-497. doi:10.1177/0022034519833645.

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