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CASE REPORT

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SUMMARY: Lingual duplication cysts are rare in the pediatric population and exceedingly rare in adults. Imaging is an important part of the evaluation of lingual lesions and is helpful in defining the location and extent for surgical planning. We present a lingual duplication cyst and discuss the imaging findings and radiologic differential diagnosis.

Migrational anomalies during fetal life can result in histologically normal tissue that arises in unexpected locations. When these anomalies result in mass lesions, they are called choristomas. Foregut duplication cysts are a form of choristoma in which gastrointestinal or respiratory epithelium is found in a mass arising in the neck or chest. Foregut duplications constitute approximately one-third of gastrointestinal duplication cysts, and they are usually divided into bronchogenic, esophageal, and neurenteric forms. However, foregut duplication cysts will occasionally arise in locations that do not correspond to this classification scheme. For example, 0.3% of enteric duplications occur in the oral cavity.¹ One of the rarest locations for a foregut duplication cyst is within the anterior tongue. Although this type of duplication is occasionally seen in children, it almost never presents after the third decade of life.²⁻⁵ The differential diagnosis of an adult with an anterior tongue lesion includes benign and malignant neoplasms, venolymphatic malformation, dermoid cyst, and foregut duplication cyst. In these patients, cross-sectional imaging plays a critical role in narrowing the differential diagnosis and providing surgical guidance. We present a unique case of a 45-year-old patient with a lingual duplication cyst.

Case Report

A 45-year-old man presented to the emergency department with a mass in his tongue that had acutely increased in size for 24 hours. The lesion had been present since childhood but had been about the size of a quarter until the previous day, when it enlarged to the size of a golf ball. He complained of difficulty in swallowing and in controlling secretions, as well as severe pain. His speech was impaired, but not his breathing. A contrast-enhanced CT of the neck failed to demonstrate the lesion due to artifacts from dental amalgam. MR imaging of the oral cavity showed a 5.1 × 3.7 cm well-circumscribed mass within the midline of the anterior tongue (Fig 1). The mass displayed intermediate signal intensity on T1, high signal intensity on T2, and partial rim enhancement on enhanced sequences. The differential diagnosis included lingual duplication cyst, dermoid cyst, and lymphatic malformation. The patient was treated with antibiotics and scheduled for surgery. Before surgery, the mass spontaneously discharged a small amount of brown fluid. At the time of surgery, the mass had decreased to 2.5 × 1.0 × 1.3 cm and was found to be a deep smoothly lined unilocular cyst in the submucosa. Histopathologic examination confirmed the mass as a foregut duplication cyst lined with squamous,

ciliated columnar, and gastric mucosa. The patient was discharged on postoperative day 5 and recovered without further incident. There was no evidence of recurrence on clinical follow-up.

Discussion

Embryologically, the primitive foregut gives rise to the pharynx, the respiratory tract, the esophagus, the stomach, and the duodenum to the level of the ampulla of Vater. There is no single theory that fully explains the embryogenesis of foregut duplications, but notochordal abnormalities appear to have a role.⁶ Lingual duplications may alternately reflect abnormal cellular migration because the primitive foregut and the pharyngeal arches (containing the developing tongue) are in close proximity.² Foregut duplications are considered choristomas, with a wide range of ectopic tissues. In a prior review of 52 pediatric oral cavity choristomas, gastrointestinal epithelium lined most cysts, and the anterior two-thirds of the tongue was most frequently affected.²

The clinical presentation of any foregut duplication cyst is a reflection of its location, its mass effect, and complications of the ectopic mucosal lining.⁶ Oral cavity duplication cysts are usually asymptomatic, but there is a potential for respiratory and/or feeding problems in the infant. While mass effect is a common presentation in younger patients, degeneration into malignancy is a possible outcome in the adult, indicating that early surgical removal is preferred.⁷ The clinical differential diagnosis of a mass in the anterior tongue includes dermoid cyst, hamartoma, neurofibroma, teratoma, lymphoepithelial cyst, squamous cell carcinoma, and glandular neoplasms. Although radiologic findings usually cannot provide a definitive diagnosis, imaging has an important role in the evaluation of these patients, both for narrowing the differential diagnosis and for surgical planning. Complete excision of the cyst is the treatment of choice for foregut duplications, with no recurrences reported.⁸

MR imaging is the technique of choice for oral cavity lesions, providing excellent contrast resolution of soft tissues with multiple pulse sequences, but enhanced CT may also be useful in the absence of dental amalgam. Lingual duplications typically appear as nonenhancing or thinly rim-enhancing cystic lesions. On T1-weighted images, they may be of variable intensity, depending on the proteinaceous content. On T2-weighted images, they appear uniformly hyperintense.⁸ CT typically reveals a well-defined homogeneous cystic mass, but loculation is possible.⁹ Prenatally, sonography can also be of diagnostic use, followed by MR imaging for confirmation.¹

The radiologic differential diagnosis of a cystic anterior tongue mass includes dermoid cyst, venolymphatic malformation, and foregut duplication cyst. Because dermoid cysts

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Fig 1. MR imaging of an adult lingual foregut duplication cyst. *A*, Sagittal T1-weighted image shows the location of the cyst within the intrinsic muscles of the tongue. The cyst is of intermediate T1 intensity due to its proteinaceous content. *B*, Axial T2-weighted image shows the high T2 signal intensity. *C*, Fat-suppressed contrast-enhanced coronal T1-weighted image shows partial areas of rim enhancement (arrows) around the cyst.

have various skin elements in their epithelial lining, they appear on CT as cystlike masses with variable (or even mixed) densities. On MR imaging, they appear as nonenhancing cystic lesions, which may have increased T1 signal intensity from fat content. The presence of proteinaceous fluid in duplication cysts produces similar signal intensities on T1 and T2 sequences, so these lesions may be difficult to distinguish.⁸ Mucocele of the tongue demonstrates the typical findings of a cystic lesion, with low signal intensity on T1 and high signal intensity on T2.¹⁰ Venolymphatic malformations display similar signal intensities on T1 and T2 sequences, with a heterogeneous and often infiltrative appearance.^{11,12} Thus, differentiating among these various lesions radiographically can be problematic, and excisional biopsy is needed for the final diagnosis.

In summary, lingual duplication cysts are uncommon in the pediatric population, and the diagnosis is exceedingly rare in adults. Nevertheless, this diagnosis must be considered even in middle-aged patients presenting with an anterior tongue mass.

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