

Oral foregut cyst in the ventral tongue: a case report

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Abstract (J Korean Assoc Oral Maxillofac Surg 2014;40:313-315)

An oral foregut cyst is a rare congenital choristoma lined by the respiratory and/or gastrointestinal epithelium. The exact etiology has not been fully identified, but it is thought to arise from misplaced primitive foregut. This lesion develops asymptotically but sometimes causes difficulty in swallowing and pronunciation depending on its size. Thus, the first choice of treatment is surgical excision. Surgeons associated with head and neck pathology should include the oral foregut cyst in the differential diagnosis for ranula, dermoid cyst, thyroglossal duct cyst and lymphangioma in cases of pediatric head and neck lesions.

Key words: Oral foregut cyst, Foregut cyst, Lingual cyst, Lingual cyst with respiratory epithelium

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I. Introduction

Foregut cysts develop from embryonic rests of foregut epithelium, and are usually observed in the abdomen and thorax¹. Oral foregut cysts are rare but may occur on the tongue, floor of the mouth, and pharynx² and occasionally cause airway obstruction or feeding difficulty³. This report is about a case of an oral foregut cyst that developed on the ventral tongue.

II. Case Report

A 2-year-old female patient presented with swelling on the ventral side of the tongue without any swallowing or feeding problems. Her parents had known about the existence of the cystic lesion, which had remained relatively unchanged in size, but they postponed the operation because the patient was too young. A fluctuant mass was palpated on the mid-ventral side of the tongue, which was covered by normal textured

mucosa. Magnetic resonance imaging (MRI) demonstrated a unicystic oval-shaped mass in the tongue with 1.88×1.84×2.41 cm dimension.(Fig. 1)

The surgical procedure proceeded as follows. Under general anesthesia with nasotracheal intubation, the patient was placed on the operating table in supine position. Intraoral irrigation, extraoral preparation, and draping were done in the usual manner. After local injection with 2% lidocaine on the ventral tongue, a vertical incision was performed along the lingual frenulum, and dissection of the mass was carefully performed. The dissection process was fairly straightforward because the cystic wall was relatively firm compared with other odontogenic cysts, and an oval-shaped mass was removed.(Fig. 2) The specimen was sent to the department of oral pathology. It was found to consist of ciliated pseudostratified columnar epithelium, the origin of which was considered to be respiratory epithelium.(Fig. 3. A) The healing process in this case was very good and without complications such as infection and wound dehiscence.

III. Discussion

In 1895, the German literature first referred to foregut cysts as “congenital ranula”⁴. Oral cavity cysts lined by gastrointestinal mucosa were first described in 1927 by Toyama⁵. Cystic lesions in the floor of the mouth that show homogenous high T2 signal and variable T1 signal⁶ can be diagnosed as ranula, dermoid cyst, thyroglossal duct cyst, and lymphangioma.

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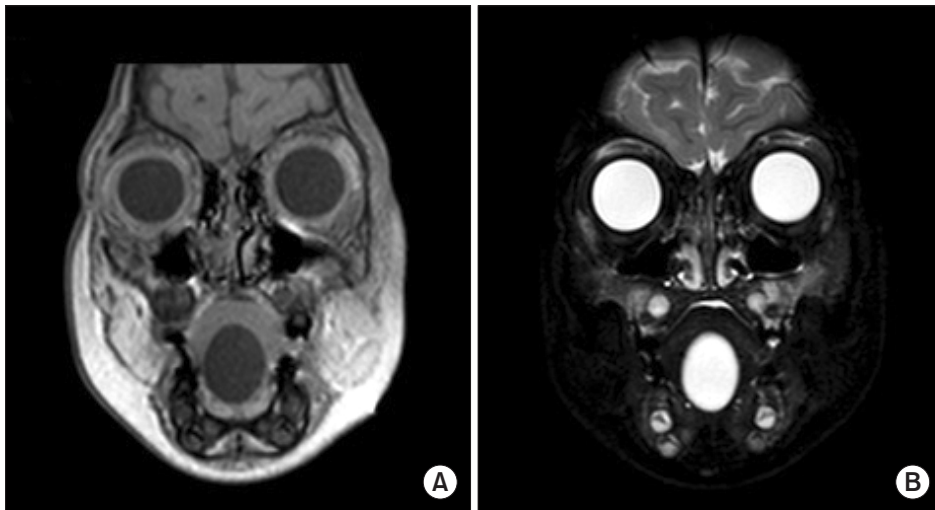


Fig. 1. Magnetic resonance (MR) images. A. Coronal MR image showed a low attenuated on T1-weighted in the ventral tongue. B. A high attenuated on T2-weighted. Thus, we could conclude this lesion is consisted with fluid accumulation.

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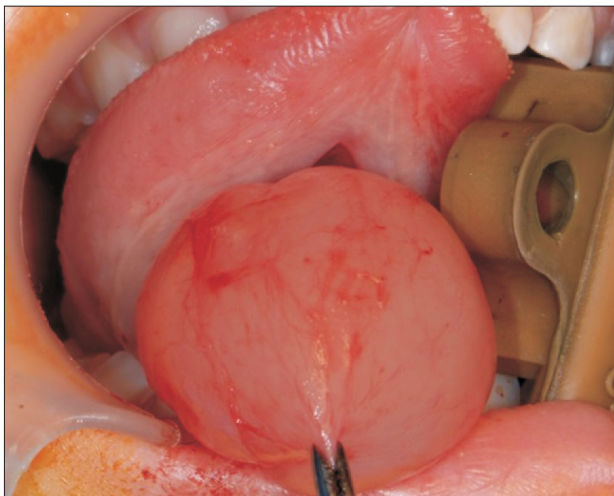


Fig. 2. The cystic mass was dissected and removed (oval shaped with 2 cm diameter).

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A ranula is a pseudocyst related to the function of salivation; it shows a low-to-intermediate T1 signal of a thin unilocular wall and high T2 signal of a round lesion⁷. A dermoid cyst is a genuine cystic lesion lined by squamous epithelium, and develops due to unusual ectodermal differentiation. It is located in the midline of the floor of the mouth, and has a variable T1 and high T2 signal⁸. Dermoid cysts consist of hair follicles and sebaceous glands, so they are generally whitish or pale yellow in color⁶, which can be used as evidence in ruling out dermoid cysts. In the case of thyroglossal duct cysts, a well-demarcated cystic wall and a variable T1 image are observed depending on the composition of cystic fluid⁶. Lymphatic malformation may be present as a variable signal on T1-weighted images and high signal on T2-weighted im-

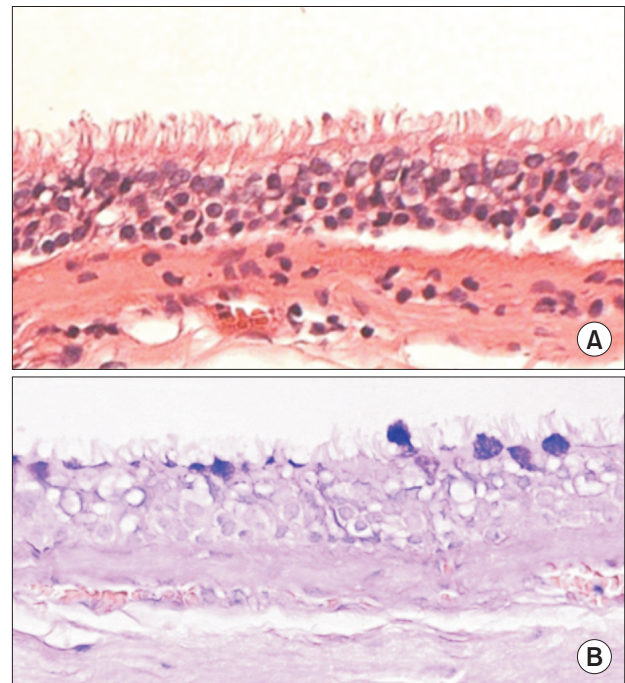


Fig. 3. A. Ciliated columnar epithelium was seen (H&E staining, $\times 200$). B. Goblet cell responded positively to a periodic acid Schiff (PAS) staining ($\times 400$).

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ages. However, several septations and multicystic lesions are common findings⁹. Therefore, the initial diagnosis with these findings can include ranula, not only because of the consistency of the lesion but also the information from high T2 and low T1 signals on MRI.

In our case, the final pathologic result was diagnosed as a foregut cyst that originated from respiratory epithelium based on the ciliated columnar epithelium in H&E stain (Fig. 3. A)

and the positive reaction of goblet cells in periodic acid Schiff (PAS) stain.(Fig. 3. B) A positive result from PAS stain indicates continuous mucin secretion; therefore, delay of surgical intervention is improper because of possible increasing size.

In several articles, most cases of foregut cysts were located in the dorsal area of the tongue rather than the ventral^{3,10,11}. The size of the cyst varied from 1.0 to 6.5 cm with the mean being 2.3 cm. Manor et al.¹¹ reported that out of the total 52 cases of foregut cyst, 12 cases were lined with respiratory epithelium only, 21 cases with gastric epithelium and 3 cases with mostly intestinal epithelium. One was gastric and intestinal epithelium combined, and the remaining 15 cases contained respiratory, intestinal and gastric epithelium.

Foregut cyst is a rare congenital choristoma, and may occur from misplacement of undifferentiated cells. The foregut goes through a transformation during differentiation in the third week: the respiratory tract originates from the ventral portion, and the proximal gastrointestinal tract, including the esophagus, stomach and duodenum, develops from the dorsal. The pharyngeal arches are close to the primitive foregut, so the embryonal rests can be entrapped¹². These rests differentiate to their own characteristic epithelium within the developing tongue, which leads to development of a foregut cyst, such as the one in this report. Early diagnosis of foregut cyst is important because it can obstruct the airway and impede swallowing and feeding¹³. Most of these lesions can be simply treated by surgical excision with good postoperative healing and low recurrence rate.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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