

A rare sublingual cyst in an infant: oral heterotopic gastrointestinal cyst

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Oral heterotopic gastrointestinal cyst is rarely seen in the oral cavity. The tongue and floor of the mouth are the most commonly affected sites. These cysts may cause feeding and respiration problems, especially in newborns and infants. A benign mass was considered according to the physical examination and ultrasonography findings. We treated the patient with simple excision. We present a three-month-old female infant who was referred to us with a cystic mass in the sublingual region.

Key words: oral cavity, sublingual, heterotopia, gastrointestinal cyst.

Heterotopia is described as the presence of tissue or organs in an abnormal localization. It is used synonymously with choristoma. Although oral heterotopic gastrointestinal cyst (OHGC) is much more common in the gastrointestinal tract, it may also be encountered in the lungs, oral cavity, larynx, pancreas, and urinary bladder¹⁻³. OHGC is rarely seen in the oral cavity as an asymptomatic mass. In 30% of cases, the mass may grow slowly and may lead to swallowing, respiration or speech problems. It may even cause life-threatening airway obstruction in newborns and infants^{4,5}. The first reports of OHGC were described by Schultz² and Toyama³ in 1927. A three-month-old female infant with feeding and respiratory difficulties due to a mass located at the floor of her mouth is presented in this article.

Case Report

A three-month-old female infant was referred to our clinic with a history of a slow-growing mass, which was present since birth. The family reported the infant's difficulty in feeding and respiration. A properly limited, soft mass, measuring 2.5x2 cm, was observed in the sublingual region (Fig. 1). Cystic fluid was aspirated during needle aspiration. According to the benign view and limitation of the mass from surrounding tissues, computed tomography (CT) was not performed to protect the infant from exposure to the harmful

effects of the radiation. Only ultrasonographic (USG) examination was performed, which revealed a smooth-surfaced and properly limited mass. Other laboratory and physical examination findings were normal. With the approval of the family, the mass was excised under general anesthesia. The mass was easily dissected from surrounding tissues without any complications, as expected. The mass was cystic in character and limited to the sublingual space. The specimen was sent for histopathological examination, which showed a cystic structure containing gastric epithelium with gastric glands and muscle tissue in deep layers. The result was reported as OHGC (Fig. 2). All of the infant's complaints resolved after the surgery. The patient was discharged on the postoperative 2nd day.

Discussion

The presence of OHGC in the oral cavity is a very rare entity. Although OHGC is mostly seen in infants, it can be encountered in adults⁶⁻⁹. It is twice as common in men¹⁰. It is located on the tongue in 60% of patients. The floor of the mouth is the second most frequently affected region in the oral cavity^{6-9,11}. The OHGC was located in the sublingual space in our patient. The larynx, anterior neck, submandibular gland, and lips are the other possible rare localizations of OHGC in the head and neck region¹². Although these masses

are usually in cystic form, they can also be in solid form. Cystic hygroma, dermoid cysts, encephaloceles, hemangioma, neurofibroma, lymphangioma, epidermoid cyst, ranula, lingual thyroid, and hamartoma should be considered in the differential diagnosis of a mass located in the oral cavity in newborns and infants^{4,5}.

These cysts are composed of gastric, intestinal or colonic mucosa having squamous epithelial regions⁹⁻¹¹. A number of theories have been proposed about the histopathogenesis of OHGC. The most accepted one is that in the early fetal period, the tissues are entrapped and develop in an abnormal region during the migration of primitive embryonal tissues⁵⁻⁹. Gastric tissues are entrapped in the oral cavity during fusion and result in midline tongue lesions because



Fig. 1. A properly limited mass is seen in the sublingual region.

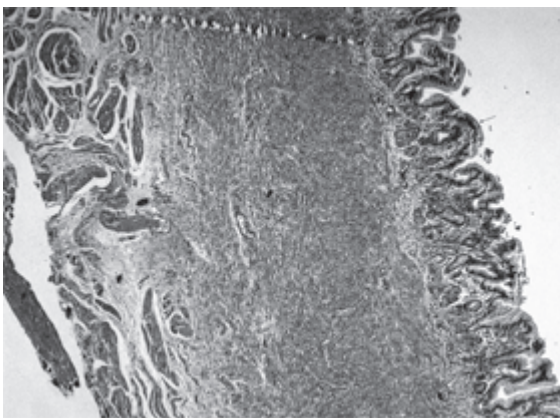


Fig. 2. Portion of the gastric cyst wall is seen. Glandular structures in the lamina propria beneath the surface epithelium (black arrow). In addition, smooth muscle bundles are seen in deep layers (hematoxylin-eosin (H&E), X25).

the primitive stomach is located in the neck in the 4th week of embryonal development. In another theory, OHGC is suggested to be derived from thyroglossal ductus cysts and salivary gland retention cysts¹¹. In another less popular theory, the histopathogenesis of OHGC is proposed based on the migration of gastric mucosa toward the floor of the mouth¹³. Although these theories have specific backgrounds, none of them can explain the histopathogenesis of OHGC completely. The discussion is ongoing.

The masses that approach huge dimensions, especially in the larynx, hypopharynx and oral cavity, may cause dysphagia and airway obstruction. Bleeding from the oral cavity due to ulceration of gastrointestinal mucosa was also reported¹⁴.

Magnetic resonance imaging (MRI) and CT are useful in the evaluation and differential diagnosis of OHGC. However, because of the difficulty in application of MRI and the harmful radioactive effects of CT, USG might be sufficient for the evaluation in some pediatric patients. Regardless of the localization of OHGC in the oral cavity, the treatment of these masses is simple excision. Furthermore, successful excision of OHGC with CO₂ laser has been reported¹⁵. The rate of recurrence is very low.

This aim of this article was to emphasize the importance of oral cavity masses as the cause of serious problems in infants and the consideration of OHGC in the differential diagnosis among these masses.

REFERENCES

1. Picard EJ, Picard JJ, Jorissen J, Jardon M. Heterotopic gastric mucosa in the epiglottis and rectum. *Am J Dig Dis* 1978; 23: 217-221.
2. Schultz L. Ciliated epithelium cyst (ecto-endodermal) of the floor of the mouth. *Arch Pathol Lab Med* 1927; 4: 359-364.
3. Toyama T. Occurrence of stomach mucosa at the base of the tongue. *J Kyoto Med Univ* 1927; 1: 13-17.
4. Fan SQ, Ou YM, Liang QC. Glial choristoma of the tongue: report of a case and review of the literature. *Pediatr Surg Int* 2008; 24: 515-519.
5. Said-Al-Naief N, Fantasia JE, Sciubba JJ, Ruggiero S, Sachs S. Heterotopic oral gastrointestinal cyst: report of 2 cases and review of the literature. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1999; 88: 80-86.

6. Ameh EA, Jimoh AO, Rafindadi AH, Shehu SM. Sublingual gastric duplication cyst causing respiratory obstruction: case report. *East Afr Med J* 2000; 77: 394-395.
7. Corić M, Seiwerth S, Bumber Z. Congenital oral gastrointestinal cyst: an immunohistochemical analysis. *Eur Arch Otorhinolaryngol* 2000; 257: 459-461.
8. Huang SF, Chuang SM, Chen WJ. Heterotopic oral gastrointestinal cyst: report of a case. *Taiwan Yi Xue Hui Za Zhi* 1989; 88: 621-623.
9. Gorlin RJ, Jirasek JE. Oral cysts containing gastric or intestinal mucosa: unusual embryologic accident or heterotopia. *J Oral Surg* 1970; 28: 9-11.
10. Morgan WE, Jones JK, Flaitz CM, Hicks JM. Congenital heterotopic gastrointestinal cyst of the oral cavity in a neonate: case report and review of literature. *Int J Pediatr Otorhinolaryngol* 1996; 36: 69-77.
11. Gruskin P, Landolfe FR. Heterotopic gastric mucosa of the tongue. *Arch Pathol* 1972; 94: 184-186.
12. Wurster CF, Ossoff RH, Rao MS, Christu PC, Sisson GA. Heterotopic gastric mucosa of the tongue. *Otolaryngol Head Neck Surg* 1985; 93: 92-95.
13. Allison PR, Johnstone AS. The esophagus lined with gastric mucous membrane. *Thorax* 1953; 8: 87-101.
14. Srinivas T, Shetty KP, Sinivas H. Mixed heterotopic gastrointestinal cyst and extranasal glial tissue of oral cavity with cleft palate. *Afr J Paediatr Surg* 2011; 8: 237-240.
15. Aydoğan B, Kiroğlu M, Soylu L, et al. Gastric cyst of the oral cavity. *Int J Pediatr Otorhinolaryngol* 1998; 45: 255-258.