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

Vefa Kınış¹, Ulaş Alabalık², Bülent Agüloğlu¹, Musa Özbay¹, Beyhan Yılmaz¹, İsmail Topçu¹

Departments of ¹Otorhinolaryngology, and ²Pathology, Dicle University Faculty of Medicine, Diyarbakir, Turkey.
E-mail: vefakinis@hotmail.com


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 bladder. 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. The first reports of OHGC were described by Schultz and Toyama 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 adults. It is twice as common in men. 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. 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 region. 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 infants4,5.

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 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 duct 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


