CASE REPORT

An alternative surgical approach for sublingual dermoid cysts: a case report

Sublingual dermoid kistler için alternatif bir cerrahi yaklaşım: Olgu sunumu

Mehmet EKEN, M.D., Cenk EVREN, M.D., Arif ŞANLI, M.D., Ziya BOZKURT, M.D.

A 52-year-old woman presented with a painless swelling in the floor of the mouth. She stated that it had existed there for 40 years and had undergone a gradual enlargement within the past year. Examination showed a mass, 5x4 cm in size. An aspiration from the lesion revealed a cyst. The lesion was removed by a U-shaped superior based flap. Histopathological examination showed stratified squamous epithelium lining the cystic lumen. This technique can be an alternative approach in such lesions because it protects the oral mucosa by providing good exposure, and prevents injury to the lingual nerve and submandibular duct.

Key Words: Dermoid cyst/surgery; mouth floor; mouth neoplasms/surgery. Kırk iki yaşında kadın hasta ağız tabanında ağrısız kitle yakınmasıyla kliniğimize başvurdu. Hasta, kitlenin 40 yıldır var olduğunu ve son bir yıldır büyüdüğünü belirtti. Muayenede yaklaşık 5x4 cm boyutlarında kitle görüldü. Şişlikten yapılan aspirasyon materyalinin incelemesi sonucu lezyonun kist olduğu anlaşıldı. Süperior tabanlı "U" şeklinde flep ile kistik yapı çıkarıldı. Histopatolojik incelemede kist duvarının çok katlı yassı epitelle döşendiği görüldü. İyi görüş sağladığı için oral mukozayı koruyan ve lingual sinir ve submandibular duktusa zarar vermeyen bu yöntem alternatif bir cerrahi yaklaşım olabilir.

Anahtar Sözcükler: Dermoid kist/cerrahi; ağız tabanı; ağız neoplazileri/cerrahi.

Dermoid cysts of the floor of the mouth are quite uncommon. In fact, dermoid cysts occur primarily in the testes and ovaries and the most common location in the head and neck region is the external third of the eyebrow. Only 1.6% of dermoid cysts are located in the floor of the mouth.^[1]

The pathogenesis of midline cysts of the floor of the mouth is not well-established, and dysontogenetic and traumatic etiologies and thyroglossal abnormalities have been implicated. According to the anatomic relationship between the cyst and the muscles of the floor of the mouth, it is possible to distinguish three different types of dermoid cysts: median genioglossal, median geniohyoid, and lateral.

Dermoid cysts generally exhibit a slow and progressive growth, and the diagnosis of even congenital cysts is commonly delayed to the second or third decade of life.^[2] Clinically, they are associated with dysphagia, dysphonia, and dyspnea. The treatment of dermoid cysts of the floor of the mouth is surgical and may be intraoral or extraoral depending on the localization and the size of the mass.

Department of Otolaryngology, Dr. Lütfi Kırdar Kartal Training and Research Hospital (Dr. Lütfi Kırdar Kartal Eğitim ve Araştırma Hastanesi 2. Kulak Burun Boğaz Hastalıkları Kliniği), İstanbul, Turkey.

Received - August 20, 2005 (Dergiye geliş tarihi - 20 Ağustos 2005). Request for revision - March 30, 2006 (Düzeltme isteği - 30 Mart 2006). Accepted for publication - September 5, 2006 (Yayın için kabul tarihi - 5 Eylül 2006).

Correspondence (İletişim adresi): Dr. Mehmet Eken. Dr. Lütfi Kırdar Kartal Eğitim ve Araştırma Hastanesi 2. Kulak Burun Boğaz Hastalıkları Kliniği, 34865 Kartal, İstanbul, Turkey. Tel: +90 216 - 441 39 00 / 1531 Fax (Faks): +90 216 - 352 00 83 e-mail (e-posta): memedeken@yahoo.com

We present a case of dermoid cyst that existed over a 40-year period in the floor of the mouth and its enucleation with an alternative approach.

CASE REPORT

A 52-year-old woman presented with a complaint of painless swelling in the floor of the mouth. The swelling was the size of a peanut when it was first noted 40 years ago and showed a gradual enlargement within the past year. Examination showed a swelling of firm consistency in the floor of the mouth, measuring 5x4 cm (Fig. 1a). There was no pain on palpation. A computed tomography scan revealed a large radiolucent area in the floor of the mouth. An aspiration biopsy from the lesion revealed a cyst. The clinical and radiologic findings were suggestive of a dermoid cyst. It was decided to enucleate the lesion. A U-shaped incision was made along the mucosa of the floor of the mouth with a distance of 2 mm to the mandibular ridge crest. The U-shaped superior based flap was elevated above the cyst wall with blunt dissection. After flap elevation, in order not to injure the submandibular duct and lingual nerve, the cyst was carefully removed by finger dissection from the surrounding tissues and was enucleated (Fig. 1b). This approach provided us a wide exposure of the whole cyst and important structures. The lesion healed uneventfully. Microscopic examination of the specimen showed stratified squamous epithelium lining a cystic lumen. No signs of recurrence were observed during a follow-up of one year.

DISCUSSION

The development of dermoid cysts in the floor of the mouth is a rare condition. Typically, intraoral dermoid cysts present as non-tender, slow-growing masses in the sublingual, submental, or submandibular region. They are most often seen in young adults and can become unusually large with few symptoms. In our case, the cyst underwent a slow, expansive growth in the floor of the mouth throughout a 40-year period. To our knowledge, this is the longest existence of a dermoid cyst in the floor of the mouth.

Dermoid cysts may be congenital or acquired without any clinical or histological differences between the two types. Congenital cysts are dysembryogenetic lesions that arise from ectodermic elements entrapped during the midline fusion of the first and second branchial arches between the third and fourth weeks of intrauterine life.^[2] Alternatively, they may arise from the tuberculum impar of His, which, with each mandibular arch, forms the floor of the mouth and the body of the tongue.^[1] Acquired cysts derive from traumatic or iatrogenic inclusion of epithelial cells or from the occlusion of a sebaceous gland duct. Some authors also proposed that midline cysts might represent a variant of thyroglossal duct cyst.^[3]

Anatomic classification according to the relation of the cyst to the muscles of the floor of the mouth forms three groups: sublingual or median genioglossal cysts located above the geniohyoid muscles; median geniohyoid cysts located in the submandibular region between the geniohyoid and mylohyoid muscles; and lateral cysts located in the submaxillary region.^[4]

Diagnostic imaging of the lesion includes computed tomography, magnetic resonance imaging, and ultrasonography.^[5] Differential diagnosis includes the sublingual and minor salivary glands neoplasms, thyroglossal cyst, cystic hygroma, acute infection, neurofibroma, hemangioma, and lymphangioma.^[6]

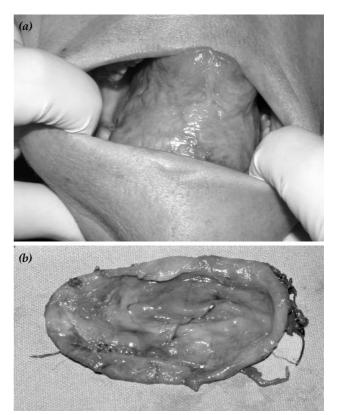


Fig. **1** - *(a) The appearance of the dermoid cyst located in the floor of the mouth. (b) Surgical specimen of the cyst.*

Surgical enucleation is the only effective treatment for these cystic lesions. Several intraoral approaches have been described including a midline vertical, mucosal incision for small cysts along the ventral surface of the tongue^[3] or a bilateral incision along the mandibular ridge crest,^[7] midline glossotomy^[8] or modified midline glossotomy associated with partial evacuation of the cyst.^[9] We performed a different approach using a U-shaped incision along the mucosa of the floor of the mouth with a distance of 2 mm to the mandibular ridge crest. The Ushaped superior based flap was elevated above the cyst wall and the cyst was enucleated by finger dissection. This approach enables protection of the oral mucosa and provides a wide exposure for distinguishing critical structures such as the submandibular ductus and the lingual nerve.

REFERENCES

1. Howell CJ. The sublingual dermoid cyst. Report of five cases and review of the literature. Oral Surg Oral Med

Oral Pathol 1985;59:578-80.

- 2. Zachariades N, Skoura-Kafoussia C. A life-threatening epidermoid cyst of the floor of the mouth: report of a case. J Oral Maxillofac Surg 1990;48:400-3.
- Walstad WR, Solomon JM, Schow SR, Ochs MW. Midline cystic lesion of the floor of the mouth. J Oral Maxillofac Surg 1998;56:70-4.
- Longo F, Maremonti P, Mangone GM, De Maria G, Califano L. Midline (dermoid) cysts of the floor of the mouth: report of 16 cases and review of surgical techniques. Plast Reconstr Surg 2003;112:1560-5.
- Mathews J, Lancaster J, O'Sullivan G. True lateral dermoid cyst of the floor of the mouth. J Laryngol Otol 2001;115:333-5.
- 6. Miles LP, Naidoo LC, Reddy J. Congenital dermoid cyst of the tongue. J Laryngol Otol 1997;111:1179-82.
- 7. Lowry RE, Tempero RM, Davis LF. Epidermoid cyst of the floor of the mouth. J Oral Surg 1979;37:271-3.
- Brusati R, Galioto S, Tullio A, Moscato G. The midline sagittal glossotomy for treatment of dermoid cysts of the mouth floor. J Oral Maxillofac Surg 1991;49:875-8.
- Di Francesco A, Chiapasco M, Biglioli F, Ancona D. Intraoral approach to large dermoid cysts of the floor of the mouth: a technical note. Int J Oral Maxillofac Surg 1995;24:233-5.