Kulak Burun Bogaz Ihtis Derg 2008;18(4):260-262

CASE REPORT

Lingual cystic lymphangioma in an elderly patient

Yaşlı bir hastada lingual kistik lenfanjiyom

Vural FİDAN, M.D.,[1] Yavuz SÜTBEYAZ, M.D.[2]

A 67-year-old man presented with a multilobulated, soft lesion at the tip of the tongue, causing articulation problem. The patient stated that it had been present for more than 30 years. Oral examination showed a multilobular mass with a large base, 15×15×10 mm in size. Magnetic resonance imaging showed a multilocular cystic tumor. The patient underwent total excision. After surgery, tongue movement and sensation were normal. Histopathologic diagnosis was cystic lymphangioma.

Key Words: Adult; lymphangioma, cystic/pathology/surgery; tongue/pathology; tongue neoplasms/pathology.

Altmış yedi yaşında erkek hasta, dilin ucunda 30 yılı aşkın süredir var olan ve konuşma sorunu yaratan multilobüler, yumuşak bir kitle yakınmasıyla başvurdu. Ağız incelemesinde 15x15x10 mm boyutlarında, geniş tabanlı multilobüler bir kitle görüldü. Manyetik rezonans görüntülemede de multiloküler kistik tümör gözlendi. Kistik lezyon total eksizyon ile çıkarıldı. Ameliyattan sonra dilin hareketi ve duyumu normaldi. Histopatolojik tanı kistik lenfanjiyom olarak kondu.

Anahtar Sözcükler: Erişkin; lenfanjiyom, kistik/patoloji/cerrahi; dil/patoloji; dil neoplazileri.

Lymphangioma is generally known as a disease of childhood when there is active lymphatic growth. Lymphangiomas can occur in the first two years of life.[1] There are very few adult cases of cystic lymphangioma.[1] The most common location of this malformation is the head and neck region, but it can also occur in the mediastinum, chest wall, abdomen, inguinal region, arm, and sometimes in the leg.[1] Within the head and neck region, the posterior triangle appears to be the most common site, and another common site is the dorsum of the tongue. It is rarely found on the tip of the tongue.

In this article, we presented an adult patient with a cystic lymphangioma that developed on the tip of the tongue.

CASE REPORT

A 67-year-old man was referred to our department for a multilobulated soft lesion at the tip of the tongue causing articulation problem. The patient stated that the lesion had been present for more than 30 years, during which time he had not sought medical treatment. The lesion showed a slow progression but did not cause difficulty in eating and speaking. The patient did not have any other health problem, only had a history of local trauma. On examination, oral hygiene was very poor, there was a multilobular mass 15x15x10 mm in size, with a large base on the tip of the tongue (Fig. 1). The tongue had normal color, texture, and movement. The soft mass had an elastic consistency, and its surface was covered by

◆ *[1]Department of Otolaryngology, Erzurum Numune Hospital (Erzurum Numune Hastanesi Kulak Burun Boğaz Hastalıkları Kliniği), Erzurum; [2]Department of Otolaryngology, Medicine Faculty of Atatürk University (Atatürk Üniversitesi Tıp Fakültesi Kulak Burun Boğaz Hastalıkları Anabilim Dalı), Erzurum, Turkey.*

◆ *Received - September 5, 2006 (Dergiye geliş tarihi - 5 Eylül 2006). Request for revision - June 12, 2007 (Düzeltme isteği - 12 Haziran 2007). Accepted for publication - July 2, 2007 (Yayın için kabul tarihi - 2 Temmuz 2007).*

◆ *Correspondence (İletişim adresi): Dr. Vural Fidan. Erzurum Numune Hastanesi Kulak Burun Boğaz Hastalıkları Kliniği, 25200 Erzurum, Turkey. Tel: +90 442 - 234 85 10 Fax (Faks): +90 442 - 232 13 90 e-mail (e-posta): vuralf@mynet.com*

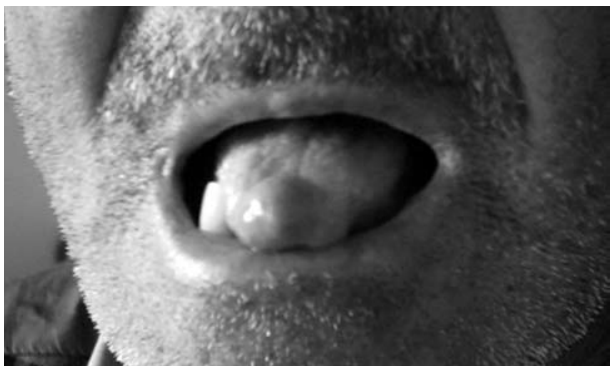

Fig. 1. *Clinical appearance of the lesion on the tip of the tongue.*

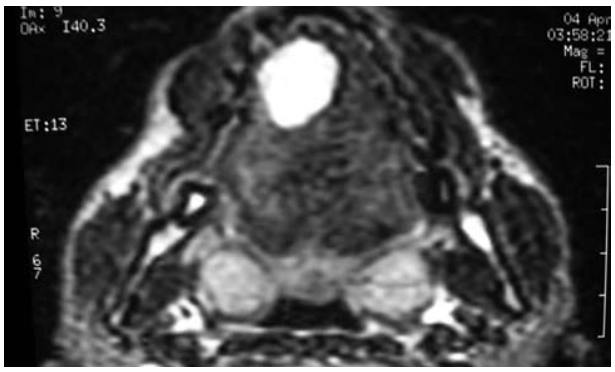

Fig. 2. *Magnetic resonance image of the tongue.*

epithelium. There was no neurosensory disturbance. Laboratory data were all normal. Magnetic resonance imaging showed a 1.5x1.5x1-cm multilocular tumor at the tip of the tongue (Fig. 2). Examination and ultrasound imaging of the head and neck region showed no other mass lesion.

The patient underwent total excision of the lesion suggestive of a benign neoplasm. A parallel incision was made to the mucosal line of the lesion and the mass was excised from the tongue with sharp dissection leaving about a 5-mm safety margin. The wound was primarily closed. The surgical specimen seemed well-demarcated with some cystic components. After surgery, tongue movement and sensation were normal.

Histopathologically, the lesion was composed of multiple, irregular-shaped lymphatic channels which were lined by flat endothelial cells. An incomplete layer of smooth muscle often lined the walls of these malformed channels (Fig 3a). Lingual tissue sections were stained immunohistochemically with CD34 and vimentin (Fig 3b, c).

DISCUSSION

The lymphatic system arises from five primitive buds of the venous system.[2] These five buds give rise to paired jugular and posterior sacs, and a single retroperitoneal sac. Lymphangiomas predominantly develop in the head and neck region because of the anatomic location of these sacs.[3] However, the etiology of lymphangioma in the adult may be different from that in the infant. In our case, there was no obvious history, but local trauma. Lymphangioma and cystic lymphangioma may be found together in neighboring regions.[4]

Histologically, lymphangiomas are classified in three groups: *(i)* lymphangioma simplex consists of thin-walled lymphatics; *(ii)* cavernous lymphangioma is composed of dilated lymphatic vessels with surrounding adventitia; and *(iii)* cystic lymphangioma (hygroma).[4] Lingual lymphangiomas are often simple or cavernous.[4] In our case, lymphangioma was of cystic type with large cysts in gross pathology.

Lymphangiomas of the tongue show macroglossia with translucent vesicles of lymphatic origin. Cystic lingual lymphangiomas usually occur on the dorsum of the tongue, and the tip of the tongue is a rare location.[4]

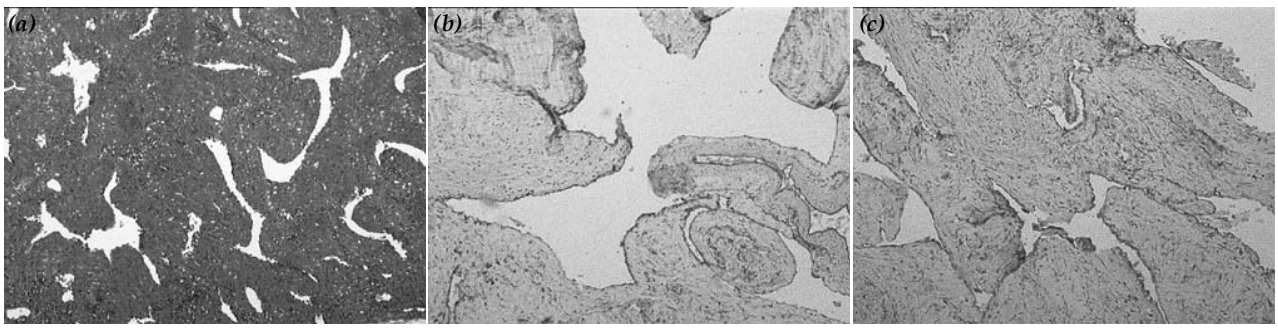

Fig. 3. ***(a)*** *Numerous irregularly dilated lymphatics in loose connective tissue (H-E x 200). Staining of lymphangioma with* ***(b)*** *CD34 and* ***(c)*** *vimentin.*

Lymphangioma is readily diagnosed on hematoxylin-eosin stained sections. Cystic lymphangiomas have multiple loculated, dilated, lymphatic channels lined by a single layer of endothelium. The differential diagnosis should include other neoplasms and epithelium-lined cysts.[4] In our case, there was a well-defined epithelial lining suggestive of lymphangioma. Positive staining of the cyst wall for CD34 and vimentin confirmed the diagnosis.[4]

Magnetic resonance imaging shows a multilocular pattern of low signal intensity on T_1-weighted, and high signal intensity on T_2-weighted images.[5]

The occurrence of cystic lymphangiomas in adults is very rare.[6] In our case, the patient was 67 years old and cystic lymphangioma had existed on the tip of the tongue for more than 30 years. The majority of clinical presentations in adults include an asymptomatic mass in the posterior triangle of the neck, with a rapid growth over weeks,[7] usually after local trauma or a respiratory infection.[8]

Many treatment alternatives exist for lymphangiomas, including surgical excision, laser surgery, cryotherapy, electrocautery, steroid administration, sclerotherapy, embolization, and radiation therapy,[8] but surgical excision is the most preferred option.[2,9,10] We performed total surgical excision. Long-term follow-up of lymphangiomas is required due to high recurrence rate (21%).[7]

REFERENCES

1. Cystic hygroma. Lancet 1990;335:511-2.
2. Kennedy TL, Whitaker M, Pellitteri P, Wood WE. Cystic hygroma/lymphangioma: a rational approach to management. Laryngoscope 2001;111:1929-37.
3. Charabi B, Bretlau P, Bille M, Holmelund M. Cystic hygroma of the head and neck - a long-term follow-up of 44 cases. Acta Otolaryngol Suppl 2000;543:248-50.
4. Ikeda H, Fujita S, Nonaka M, Uehara M, Tobita T, Inokuchi T. Cystic lymphangioma arising in the tip of the tongue in an adult. Int J Oral Maxillofac Surg 2006;35:274-6.
5. Yonetsu K, Nakayama E, Miwa K, Tanaka T, Araki K, Kanda S, et al. Magnetic resonance imaging of oral and maxillofacial angiomas. Oral Surg Oral Med Oral Pathol 1993;76:783-9.
6. Sherman BE, Kendall K. A unique case of the rapid onset of a large cystic hygroma in the adult. Am J Otolaryngol 2001;22:206-10.
7. Schefter RP, Olsen KD, Gaffey TA. Cervical lymphangioma in the adult. Otolaryngol Head Neck Surg 1985;93:65-9.
8. Livesey JR, Soames JV. Cystic lymphangioma in the adult parotid. J Laryngol Otol 1992;106:566-8.
9. Clymer MA, Fortune DS, Reinisch L, Toriumi DM, Werkhaven JA, Ries WR. Interstitial Nd:YAG photocoagulation for vascular malformations and hemangiomas in childhood. Arch Otolaryngol Head Neck Surg 1998;124:431-6.
10. Wiggs WJ Jr, Sismanis A. Cystic hygroma in the adult: two case reports. Otolaryngol Head Neck Surg 1994;110:239-41.