

CASE REPORT

## Cavernous lymphangioma in the laryngeal ventricle in an adult patient

Erişkin bir olguda larenks ventrikülünden kaynaklanan kavernöz lenfanjiom

Uğur ÇINAR, M.D.,<sup>1</sup> Özgür YİĞİT, M.D.,<sup>1</sup> Tülay BAŞAK, M.D.,<sup>2</sup> Burhan DADAŞ, M.D.<sup>1</sup>

Lymphangiomas of the larynx are quite rare and even rarer in adults. Isolated cavernous lymphangioma in the laryngeal ventricle was diagnosed in a 47-year-old woman. The patient presented with hoarseness with a history of three months. Direct laryngoscopy performed under general anesthesia showed a large, smooth, gray-reddish tumor above the right vocal fold, originating from the right ventricle. The tumor was removed through a micro-laryngoscopic procedure with cold instruments. Histopathologic diagnosis was cavernous lymphangioma. There was no local recurrence a year after the operation.

**Key Words:** Adult; hoarseness/etiology; laryngeal neoplasms/complications; lymphangioma/pathology/surgery.

Larenksten kaynaklanan lenfanjiomlar oldukça nadirdir; bu selim tümörlerin erişkinlerde görülmesi çok daha nadirdir. Üç aydır devam eden ses kısıklığı nedeniyle başvuran 47 yaşındaki kadın hastada sağ vokal kord superiorunda kitle belirlendi. Genel anestezi altında yapılan direkt larengoskopide, sağ ventrikülden kaynaklanan geniş, yumuşak, gri-kırmızı renkli kitle görüldü ve lezyon soğuk cerrahi ile mikrolarengoskopik olarak çıkarıldı. Histopatolojik inceleme sonucu kavernöz lenfanjiom olarak bildirildi. Hastanın bir yıllık izlemi sırasında lokal nüks saptanmadı.

**Anahtar Sözcükler:** Erişkin; ses kısıklığı/etyoloji; larenks neoplazileri/komplikasyon; lenfanjiom/patoloji/cerrahi.

Lymphangiomas are rare congenital lesions of the lymphatic system, most commonly presenting in the head and neck during infancy. Of all lymphatic malformations, 50% to 60% are present at birth, and approximately 80% to 90% become evident by the age of two years.<sup>[1]</sup> The occurrence is uncommon in the older child, and even rarer in the adulthood. Lymphangioma of the larynx is also an extremely rare incidence. Naito et al.<sup>[2]</sup> reviewed the entire English literature and documented 17 lymphangioma cases with isolated laryngeal involvement in

adults and children. Only four adult cases appeared in the English literature within 2002.<sup>[2-4]</sup>

Kennedy<sup>[5]</sup> classified lymphangiomas into four types: superficial cutaneous, cavernous, cystic hygroma, and diffuse systemic. Cavernous lymphangiomas are composed of dilated lymphatic channels with one or more endothelial layers with or without an adventitial layer.

We present an adult patient with a cavernous lymphangioma confined to the laryngeal ventricle.

◆ Departments of <sup>1</sup>Otolaryngology and <sup>2</sup>Pathology, Şişli Etfal Training Hospital, İstanbul, Turkey.

◆ Received: October 9, 2002. Accepted for publication: April 28, 2003.

◆ Correspondence: Dr. Uğur Çınar, Körkadı Sok., Turizm Sitesi, 4. Blok, No: 13, 80600 Ulus, İstanbul, Turkey.  
Tel: +90 212 - 263 18 78 Fax: +90 212 - 217 91 85  
e-mail: u\_cinar@yahoo.com

◆ Şişli Etfal Eğitim ve Araştırma Hastanesi, <sup>1</sup>Kulak Burun Boğaz Hastalıkları Kliniği, <sup>2</sup>Patoloji Kliniği, İstanbul.

◆ Dergiye geliş tarihi: 9 Ekim 2002. Yayın için kabul tarihi: 28 Nisan 2003.

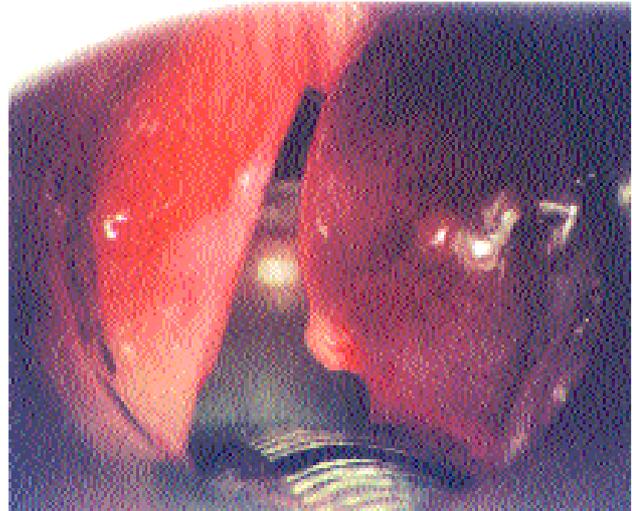
◆ İletişim adresi: Dr. Uğur Çınar, Körkadı Sok., Turizm Sitesi, 4. Blok, No: 13, 80600 Ulus, İstanbul.  
Tel: 0212 - 263 18 78 Faks: 0212 - 217 91 85  
e-posta: u\_cinar@yahoo.com

### CASE REPORT

A forty-seven-year-old female patient presented with hoarseness with a history of three months. Indirect laryngeal examination revealed a large, smooth, gray-reddish mass above the right vocal fold. Her breathing was normal. Direct laryngoscopy under general anesthesia showed a large tumor originating from the right ventricle (Fig. 1). The right true vocal cord was poorly visualized because of the extension of the mass along the right laryngeal ventricle. A red, smooth mass, 11x9x5 mm in size was removed by way of a micro-laryngoscopic procedure with cold instruments. The remaining parts of the larynx appeared normal. She was discharged on the day after surgery.

Histopathologic examination showed spaces of dilated, irregular shape and size under the thick stratum of ciliated columnar epithelium, lined by the endothelium. Some of them contained a small number of lymphocytes and a pinkish pale coagulated amorphous mass, leaving the greater parts empty. The stroma was composed chiefly of fibroblasts and focal lymphocytic aggregates near the vessels (Fig. 2, 3). The diagnosis was made as cavernous lymphangioma.

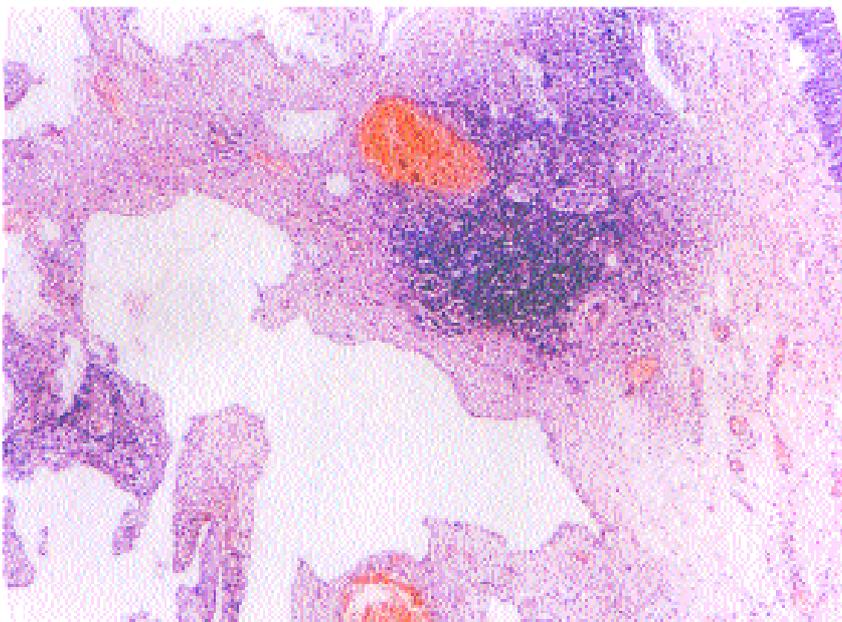
The complaints of the patient completely disappeared; her voice became full-sounding and clear. No recurrences were detected a year after surgery.



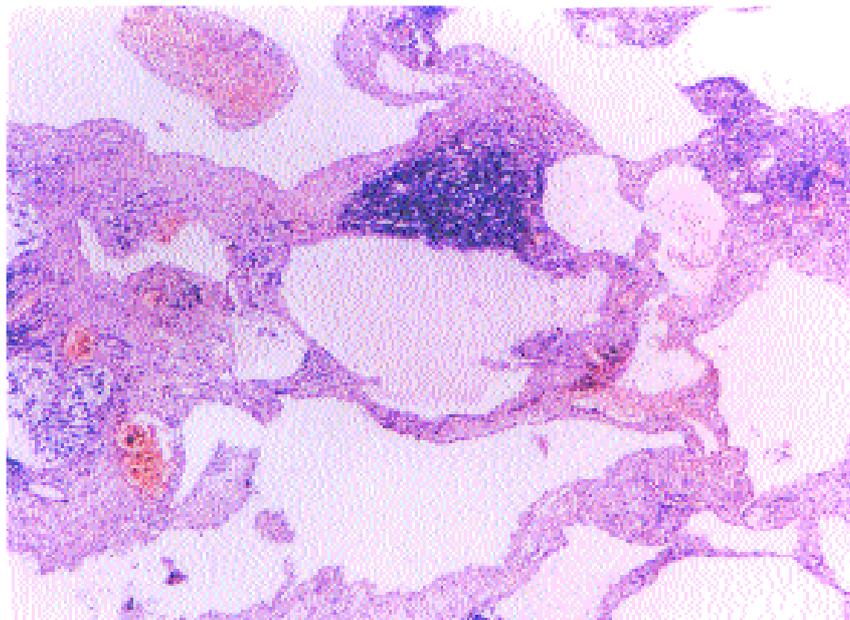
*Fig. 1 - Micro-laryngoscopic view (7x) showing a large, gray-reddish, smooth mass of the right vestibular fold. The mass was fairly solid to palpation. It was confined to the right laryngeal ventricle.*

### DISCUSSION

Lymphangioma of the larynx mainly occurs in regions where friction is caused by the passage of food and where there are numerous lymphatic vessels, such as the lingual surface of the epiglottis, aryepiglottic folds, and the arytenoid region. Its appearance is very rare in the vocal folds, in the ventricles of Morgagni, and in the subglottic area.<sup>[3]</sup> Lymphangiomas do not infiltrate the surrounding



*Fig. 2 - Under the thick stratum of ciliated columnar epithelium, there were spaces of dilated, irregular shape and size, lined by the endothelium. Some of them contained a small number of lymphocytes and a pinkish pale coagulated amorphous mass, but the greater parts were empty (H-E x 40).*



*Fig. 3 - The stroma was composed chiefly of fibroblasts and focal lymphocytic aggregates near the vessels (H-E x 40).*

tissues because of a distinct capsule. Those with a small size and location other than the vocal folds usually cause few or no symptoms. However, their growth or location in the sphere of the glottis may be associated with changes in voice and may lead to hoarseness, cough, nausea, vomiting, dyspnea, and dysphagia.<sup>[3]</sup>

Lymphangioma rarely occurs in the larynx of infants and children. Its appearance is uncommon in the older child, and even rarer in the adult. Compared to that of the adults, the lymphatic system of the larynx is better developed in infants and young children, with a greater number of channels and lymph nodes.<sup>[6]</sup> Cohen and Thompson<sup>[6]</sup> reviewed 160 pediatric patients with lymphangiomas, of whom ten had extensive laryngeal involvement accompanied by cervical, facial, and/or submandibular hygromas.

To our knowledge, there are few literature reviews giving detailed information on laryngeal lymphangioma in adults. Gerwel and Nawrocki<sup>[3]</sup> reported a 17-year-old girl with complaints of difficulty in swallowing, change in voice, and a feeling of a foreign body in the pharynx. Laryngeal examination revealed a large tumor involving the hypopharynx and the whole lingual surface of the epiglottis, which was then removed via suprahyoid pharyngotomy together with the epiglottis at

the base of the tongue. Smith and Stafford<sup>[4]</sup> reported a 69-year-old female patient with lymphangioma of the post-cricoid region. She presented with symptoms characteristic of globus pharyngeus. It was removed via pharyngoscopy. Naito et al.<sup>[2]</sup> reported a 36-year-old female patient who complained of hoarseness for several months associated with lymphangioma on her right false vocal cord. They performed tracheostomy and a laryngofissure for the removal of the neoplasm. We could not find any adult case with lymphangioma in the laryngeal ventricle in the English literature.

The recurrence of lymphangiomas ranges from 39% to 50% in pediatric patients following surgery.<sup>[7,8]</sup> The recurrence was 21% after attempted total resection as the initial procedure.<sup>[9]</sup> In our case no recurrence was detected within a year.

#### REFERENCES

1. Papsin BC, Evans JN. Isolated laryngeal lymphangioma: a rare cause of airway obstruction in infants. *J Laryngol Otol* 1996;110:969-72.
2. Naito K, Iwata S, Nishimura T, Yagisawa M, Sakurai K. Laryngeal lymphangioma-case report. *Auris Nasus Larynx* 1985;12:111-6.
3. Gerwel T, Nawrocki A. Lymphangioma of the larynx. *Arch Otolaryngol* 1960;72:251-3.
4. Smith NM, Stafford FW. Post cricoid lymphangioma. *J Laryngol Otol* 1991;105:220-1.
5. Kennedy TL. Cystic hygroma-lymphangioma: a rare

- and still unclear entity. *Laryngoscope* 1989;99(10 Pt 2 Suppl 49):1-10.
6. Cohen SR, Thompson JW. Lymphangiomas of the larynx in infants and children. A survey of pediatric lymphangioma. *Ann Otol Rhinol Laryngol Suppl* 1986; 127:1-20.
  7. Orvidas LJ, Kasperbauer JL. Pediatric lymphangiomas of the head and neck. *Ann Otol Rhinol Laryngol* 2000; 109:411-21.
  8. Ricciardelli EJ, Richardson MA. Cervicofacial cystic hygroma. Patterns of recurrence and management of the difficult case. *Arch Otolaryngol Head Neck Surg* 1991;117:546-53.
  9. Hancock BJ, St-Vil D, Luks FI, Di Lorenzo M, Blanchard H. Complications of lymphangiomas in children. *J Pediatr Surg* 1992;27:220-4.