Bilateral Laryngoceles

Rachel L. Werner · Jason W. Schroeder ·
James T. Castle

Received: 10 June 2013 / Accepted: 11 July 2013 / Published online: 24 July 2013
© Springer Science+Business Media New York (outside the USA) 2013

History

A 63-year old male with a prior history of anterior cervical discectomy and fusion presented with complaints of gradual dysphagia with occasional regurgitation of undigested food and primarily a right neck swelling.

Radiographic Features

Axial computed tomography (CT) imaging (Fig. 1) demonstrated bilateral, cystic, air filled lesions extending into the soft

tissues of the neck, with the right ($4.8 \times 2.5 \times 4.5$ cm) larger than the left ($1.3 \times 1.1 \times 1.2$ cm), each fully pneumatized without fluid collection. No nodularity was associated with the walls of either and no associated mass lesion was present about the laryngeal ventricles. The right lesion showed extension through the thyrohyoid membrane with mass effect upon the supraglottic larynx, and the left showed sole placement within the soft tissues of the neck. Regionally, no additional mass lesions or adenopathies were identified. Changes of the prior C3 through C7 cervical spine fusion were observed. No mass lesion was present at either lung apex (Fig. 2).

Diagnosis

The hematoxylin and eosin (H&E) stained sections obtained from the right-sided lesion revealed a complex cystic structure of respiratory type epithelium composed of pseudostratified ciliated epithelium of varying thickness set against a background of variably dense, chronically-inflamed fibrous connective tissue (Fig. 3). The left sided lesion (Figs. 4, 5) showed prominent respiratory-type epithelium lining the cystic areas as well as adjacent mature adipose tissue, seromucous glands, and prominent vascularity observed in the periphery. The combination of imaging studies, histology and location confirmed a diagnosis of bilateral laryngoceles.

The opinions and assertions expressed herein are those of the authors and are not to be construed as official or representing the views of the Department of the Navy or the Department of Defense. I certify that all individuals who qualify as authors have been listed; each has participated in the conception and design of this work, the writing of the document, and the approval of the submission of this version; that the document represents valid work; that if we used information derived from another source, we obtained all necessary approvals to use it and made appropriate acknowledgements in the document; and that each takes public responsibility for it. We are military service members. This work was prepared as part of my official duties. Title 17 U.S.C. 105 provides that 'Copyright protection under this title is not available for any work of the United States Government.' Title 17 U.S.C. 101 defines a United States Government work as a work prepared by a military service member or employee of the United States Government as part of that person's official duties.

R. L. Werner
Department of Oral and Maxillofacial Pathology, Naval
Postgraduate Dental School, Bethesda, MD 20889, USA

J. W. Schroeder
Department of Radiology, Walter Reed National Military
Medical Center, Bethesda, MD 20889, USA

J. W. Schroeder
Uniformed Services University of the Health Sciences, Bethesda,
MD 20889, USA

J. T. Castle (✉)
Department of Anatomic Pathology, Naval Medical Center
Portsmouth, 620 John Paul Jones Circle, Portsmouth,
VA 23704, USA
e-mail: James.Castle@med.navy.mil

Fig. 1 An axial CT image with contrast shows bilateral laryngoceles with the right larger than the left. Mass effect on the supraglottic larynx is evident

Fig. 2 A coronal CT with contrast demonstrates how the *right-sided* combined laryngocele extends to both sides of the thyrohyoid membrane

Discussion

A laryngocele is defined as the abnormal dilatation of the laryngeal saccule containing air and maintaining an open communication with the laryngeal lumen [1]. There is an equal gender predilection, and most cases occur in the fifth

Fig. 3 A H&E stained microscopic slide of the *right-sided* lesion shows a large complex cyst with multiple involutions, some extending into the lumen. The epithelial lining varies in thickness throughout

Fig. 4 The *left-sided* lesion shows a smaller cystic area supported by a loosely fibrotic and hemorrhagic wall with occasional seromucous glands included

to eighth decades of life, with approximately 25 % of patients presenting with bilateral laryngoceles [1].

Three types of laryngoceles are identified: internal, external, and mixed/combined. External laryngoceles extend upward between the false vocal cord and thyroid cartilage, and laterally into the neck, while internal laryngoceles are confined to the larynx and do not pierce the thyrohyoid membrane [1, 2]. Combined laryngoceles make up the majority of cases and demonstrate both internal and external enlargements [2], with abnormal dilation of the saccule on both sides of the thyrohyoid membrane [1].

Clinically, patients may present with cough, hoarseness, dyspnea, dysphagia, and globus sensation [3]. Examination will typically reveal a lateral neck mass that is easily compressed and distends with increased intralaryngeal

Fig. 5 Both specimens demonstrated a lining of respiratory epithelium characterized by ciliated pseudostratified columnar epithelium

pressure [3]. Infected laryngoceles are termed laryngopyoceles and may present with pain and tenderness, and have on occasion resulted in death [4]. Other infrequent complications include airway obstruction, vocal cord paralysis, cutaneous emphysema [2], and obstructive sleep apnea [5].

In neonates, laryngoceles are considered remnants of lateral air sacs; in adults, they may represent either a congenital enlargement or an acquired deformity of the saccule [1]. The acquired deformity is typically associated with increased intraluminal pressure of the saccule, as may be experienced by musicians [1, 6], glassblowers [1, 2], weightlifters [1] and more uncommonly, in the presence of a tumor [2, 7]. Cases of squamous cell carcinoma [7] and amyloidosis [8] have been identified in association with laryngoceles, likely as the causative agent, with internal laryngoceles most often associated with laryngeal carcinoma [1]. Additionally, a surgical procedure in the vicinity of the larynx may be a predisposing factor in the development of a laryngocele due to the incidental, unintended creation of a one-way valve leading to saccular dilation [9]. As reported above, the current case involved a prior surgical procedure for an anterior cervical discectomy and fusion.

On CT scanning, a laryngocele appears as a well-defined lesion arising beneath the false vocal cords and communicating with the laryngeal airway [10]. The thyrohyoid membrane will limit internal laryngoceles, while both combined and external laryngoceles will lie superficial to the thyrohyoid membrane at the point of insertion of the superior laryngeal nerve and vessels [11]. Air filled laryngoceles are easily identified on CT due to low CT density [10]. Alternately, a fluid-filled external laryngocele may mimic the appearance of a branchial cleft cyst, only to be differentiated based on location [12]. Laryngopyoceles demonstrate a thick enhancing wall surrounding the

laryngocele on contrast-enhanced CT [1]. Magnetic resonance imaging is recommended for distinguishing laryngoceles from an underlying laryngeal neoplasm when fluid or debris are present within the dilated cavity [11].

Histologic findings may show a cyst lined by a variety of cell types. Respiratory type epithelium is the most common although areas of oncocytic epithelium as well as squamous epithelium may be present as a result of metaplastic change. A fibrous connective tissue wall containing mucous or seromucous glands are also typically observed. Squamous epithelium may become the predominant epithelial lining in the presence of infection [1], with an associated chronic inflammatory cell infiltrate scattered within the fibrous connective tissue, and in the case of a laryngopyocele, abundant neutrophils located within the connective tissue and adhering to the cystic epithelium [4].

The clinical diagnosis of a laryngocele requires imaging studies and laryngoscopy to determine the anatomy of the defect and to rule out the presence of an associated tumor or additional pathology [3] as the differential diagnosis would include branchial cleft cyst, cervical bronchial cyst, laryngeal cyst or oncocytic papillary cystadenoma.

Clinical presentation of bilateral branchial cleft cysts approaches a 10 % incidence. The overwhelming majority of cases show a large cystic space lined by stratified squamous epithelium, but rare cases show respiratory type epithelium exclusively or in conjunction with a stratified squamous epithelial lining. A cervical bronchial cyst is an entity that is more often encountered shortly after birth, more often found in the suprasternal notch area, thoracic cavity or mediastinum, but may also be found in the neck. Ciliated, pseudostratified columnar epithelium lines the cystic cavity in most instances, therefore; a complete clinical history is paramount to an accurate diagnosis.

Asymptomatic cases require no treatment, however, surgical excision is the treatment of choice for symptomatic cases, to include incision and drainage and antibiotic management for laryngopyoceles [1]. Traditionally, laryngoceles have been treated via an external approach for large or external cases utilizing a horizontal skin incision from the medial border of the sternocleidomastoid muscle to the midline at the superior level of the thyroid cartilage. Dissection is continued through the strap muscles to reveal the capsule of the laryngocele and is continued along the neck of the laryngocele, along the thyrohyoid membrane, extending to the surrounding paraglottic tissue until totally removed [13]. Endoscopic management is the preferred modality for internal variants. Resection is performed utilizing a CO₂ laser following protection of the false vocal fold ipsilateral to the laryngocele. Following an incision to the superior surface of the false vocal fold, the limits of the laryngocele are identified and the laser is used to dissect the lesion from the surrounding paraglottic tissue [13].

References

1. Wenig BM. Atlas of head and neck pathology. 2nd ed. Philadelphia: Saunders Elsevier; 2008.
2. Canalis RF, Maxwell DS, Hemenway WG. Laryngocele: an updated review. *J Otolaryngol*. 1977;6:191–9.
3. Verret DJ, DeFatta RJ, Sinard R. Combined laryngocele. *Ann Otol Rhinol Laryngol*. 2004;113:594–6.
4. Beautyman W, Haidak GL, Taylor M. Laryngopyocele; report of a fatal case. *N Engl J Med*. 1959;260:1025–7.
5. Araz O, Turan A, Yoruk O, et al. Laryngocele and epiglottic cyst as rare causes of obstructive sleep apnea. *Sleep Breath*. 2009;13:285–7.
6. Isaacson G, Sataloff RT. Bilateral laryngoceles in a young trumpet player: case report. *Ear Nose Throat J*. 2000;79:272–4.
7. Harney M, Patil N, Walsh R, et al. Laryngocele and squamous cell carcinoma of the larynx. *J Laryngol Otol*. 2001;115:590–2.
8. Cankaya H, Egeli E, Unal O, et al. Laryngeal amyloidosis: a rare cause of laryngocele. *Clin Imaging*. 2002;26:86–8.
9. Marom T, Roth Y, Cinamon U. Laryngocele: a long-term complication following neck surgery? *J Voice*. 2011;25:272–4.
10. Huang BY, Solle M, Weissler MC. Larynx: anatomic imaging for diagnosis and management. *Otolaryngol Clin North Am*. 2012;45:1325–61.
11. Koeller KK, Alamo L, Adair CF, et al. Congenital cystic masses of the neck: radiologic-pathologic correlation. *Radiographics*. 1999;19:121–46.
12. Glazer HS, Mauro MA, Aronberg DJ, et al. Computed tomography of laryngoceles. *AJR Am J Roentgenol*. 1983;140:549–52.
13. Dursun G, Ozgursoy OB, Beton S, et al. Current diagnosis and treatment of laryngocele in adults. *Otolaryngol Head Neck Surg*. 2007;136:211–5.