

CASE REPORT

An unusual case of combined laryngocele presenting as cervical swelling

✉ Jovanović Anđelina¹, Valjarević Svetlana ², Jovanović Milan^{1,2}¹Department of Otorhinolaryngology with Maxillofacial Surgery, Clinical Hospital Center “Zemun”, Belgrade, Serbia²University of Belgrade, Faculty of Medicine, Belgrade, Serbia**Received:** 04 October 2022**Revised:** 30 October 2022**Accepted:** 03 November 2022

Check for updates

Funding information:

The authors received no specific grant from any funding agency in the public, commercial, or not-for-profit sectors.

Copyright: © 2022 Medicinska istraživanja**License:**

This is an open access article distributed under the terms of the Creative Commons Attribution License (<https://creativecommons.org/licenses/by/4.0/>), which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Competing interests:

The authors have declared that no competing interests exist

✉ **Correspondence to:**

Jovanovic Andjelina, KBC Zemun, Vukova 9,
Mob. 062/22 00 32,
andjelinakjosevski@yahoo.com

Summary

Laryngocele is a sac-like, pathological enlargement of the Morgagni's ventricle, which occurs between the true and false vocal cords. This change is filled with air and is continuous with the lumen of the larynx. It is usually asymptomatic and is accidentally detected during radiological diagnostics. We present the case of a 54-year-old patient who came for examination due to a painless swelling in her neck on the left side, which lasted for 8 months and increased upon Valsalva's maneuver. Occasionally the patient was hoarse. By clinical otorhinolaryngological examination and computed tomography of the neck and larynx, we found a cystic formation, filled with air, which extended above the larynx and spread to the side of the neck. The laryngocele was surgically removed using an external cervical approach. The neck of the laryngocele is sent through the thyroid membrane, while preserving the upper laryngeal nerve and the accompanying vascular stalk. Pathohistological examination revealed that the laryngocele consisted of respiratory layered cylindrical epithelium. Laryngocele should be taken into account in differential diagnosis of neck swelling.

Keywords: combined laryngocele, neck swelling, cervical surgical approach.

INTRODUCTION

Laryngocele is a marked expansion of Morgagni's ventricle, a part of the laryngeal duct between the true and the false vocal folds; it is filled with air and communicates with the lumen of the larynx. Laryngocele can be internal or combined (1). Most often, it is asymptomatic, but in some patients, due to its size, very difficult breathing occurs, and a tracheotomy may be required (2). The etiology is still unclear, but obstruction, increased pressure in the larynx and congenital defects are possible risk factors for its occurrence (3). The frequency of laryngocele is 1 in 2.5 million people per year. Laryngocele is 5-6 times more common in men, mostly in the sixth decade of life (4). No side bias was noted (3). Bilateral laryngoceles are extremely rare, with 6 cases published in literature so far. Laryngopyocele is also a rare clinical entity. It makes up 8% of all laryngocele cases (4).

CASE REPORT

A fifty-four-year-old woman was admitted to ENT office for examination with an eight-month-old, painless swelling on the left side of the neck, which enlarged with the Valsalva maneuver. She did not have difficulties with breathing or swallowing, but she complained of occasional hoarseness. She was not an active smoker and did not drink alcoholic beverages. Fiberoptic laryngoscopy showed floating edema of the vocal cords that were mobile during phonation and respiration, as well as a marked edema of the left ventricular fold. CT of the neck and larynx with I.V. contrast confirmed an air-filled sac-like formation extending from the supralaryngeal region to the lateral side of the neck (**Figure A**).



Figure A. CT of the neck and the larynx with i.v. contrast - sacular formation, filled with air, which extended from the supralaryngeal region to the side of the neck

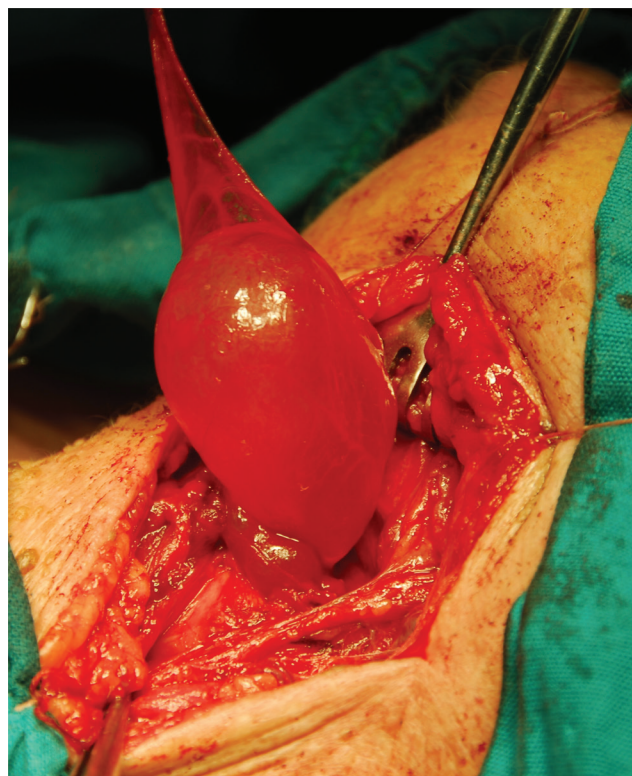


Figure B. External surgical approach

The laryngocele was approached through a horizontal cervical incision of 8 cm, from the medial edge of the sternocleidomastoid muscle (mSCM) to the medial line of the neck at the height of the upper edge of the thyroid cartilage, with lifting of the subplatysmal skin flaps. The neck of the laryngocele was followed down the thyroid membrane (figure B), with preservation of the superior laryngeal nerve and associated vascular pedicle.

When we approached the mucous membrane of the larynx, the neck of the laryngocele was ligated and the bag formation was separated. The cyst was resected, prepared and separated up to the thyrohyoid membrane and the supraglottis. Microscopic examination revealed that the laryngocele consisted of respiratory layered cylindrical epithelium.

DISCUSSION

Laryngocele is an air sac that arises from the larynx and spreads into the neck through the membrane of the thyroid gland or remains inside the larynx. It extends superiorly into the paralaryngeal space, limited medially by the ventricular fold, and laterally by the thyroid cartilage and thyrohyoid membrane. A simple laryngocele contains only air. In some cases, it may contain mucus, which is caused by the presence of mucinous glands; in that case it is called laryngomucocele. If the laryngocele becomes infected with bacterial or fungal agents, it contains pus and becomes a laryngopyocele (2,5,6).

Laryngoceles can be congenital or acquired. The etiology is not fully elucidated, but it is believed that they arise due to congenital weakness of the laryngeal wall caused by strain during coughing (6,7). Congenital defects are described as the most common cause in newborns. Despite various anatomical variations and birth defects seen in adults, it is difficult to explain all cases on these grounds. A constant increase in pressure in the lumen of the larynx can cause laryngocele even in people with a normal larynx (5). This leads to an increase in intra-abdominal pressure and a consequent increase in intraglottic pressure. Prolonged periods of increased pressure within the lumen of the larynx can cause dilatation of the laryngeal sac. This is associated with certain professions as described: weightlifters, glassblowers, singers, brass players. Chronic cough, excessive and forced cough due to chronic respiratory disorders, can also cause dilatation of the laryngeal tube and become a predisposing factor for the development of laryngocele (6,7).

Patient history, clinical otorhinolaryngological examination, endoscopic examination of the larynx and imaging of the larynx and the neck are necessary and important diagnostic steps in patients with laryngocele (2,5,6).

Laryngoceles generally have non-specific symptoms. When they are symptomatic, patients often complain of dysphonia or painless neck swelling, which increases during the Valsalva maneuver and decreases with pressure (4). Other clinical symptoms are cough, dyspnea, dysphagia, dull neck pain; globus feeling in the throat and inspiratory stridor are most often present in internal and large combined laryngoceles. In extreme cases, laryngoceles lead to upper airway obstruction and may require emergency tracheotomy. Laryngopioceles are infected laryngoceles, which, consequently, more often cause respiratory complications and even paralysis of the vocal cords and emphysema (6,7).

Radiological diagnostics, especially computed tomography (CT), is of great importance in arriving at diagnosis, in determining the size of a laryngocele, but also in detecting associated malignancies and in treatment planning. It has been noted that laryngocele can especially develop in association with supraglottic carcinoma of the larynx (3). Direct laryngoscopy is a sovereign diagnostic procedure in every case of laryngocele. Newer publications categorize laryngocele as internal or combined. The previously used classification of internal, external and combined laryngocele is being slowly abandoned. External laryngoceles cannot exist because laryn-

goceles originate from the laryngeal tube. The internal laryngocele is limited within the false vocal fold, medial to the thyrohyoid membrane, and the combined one extends upwards and protrudes through the thyrohyoid membrane to the neck (8,9). In CT, laryngocele is seen as a cyst filled with air or liquid. CT clearly differentiates a laryngocele from a saccular cyst and removes the suspicion of an occult tumor (2,5,6).

Differential diagnoses include: thyroglossal cyst, brachial cyst, saccular cyst, cystic hygroma, lymphangioma, hemangioma, ductal cyst of the submandibular gland, pharyngeal diverticulum, teratoma, dermoid cyst, lymphoma, parotid tumor, ectopic thyroid tissue, neurofibroma, lymphadenopathy or cancer. (10,11).

As laryngocele is a rare condition, there is no consensus on its surgical treatment. The operation is performed when there are symptoms. Asymptomatic cases can be monitored (11). Numerous surgical techniques are available - external, internal or combined approach and the traditional non-external one (5).

Endoscopic microsurgery and CO₂ laser application have become popular in the last two decades for internal laryngoceles (1,12). The surgical approach is related to the type and size of the formation. Most patients with combined laryngoceles are treated with an external approach (5), and that is how we operated on our patient. This approach allowed us good exposure and precision. In literature, a lower recurrence rate is registered in such cases (2). Disadvantages can be skin scars, higher morbidity, a longer duration of surgery, a longer period of hospital treatment and higher costs (2,10). Postoperative complications can be wound infections, seroma, fistula, scar, hematoma, injury to the upper laryngeal artery and nerve (13). We had no postoperative complications. Combined laryngoceles can also be treated with an endolaryngeal approach (12). We did not choose the endoscopic approach because of the size of the lesion.

Conclusion

Laryngoceles should be taken into account in differential diagnosis of neck swelling. Even with the best diagnostic and radiographic examination techniques, this differentiation cannot be clearly performed, so a direct examination of the larynx is necessary. The chosen external surgical approach provided reliable preparation and removal of the combined laryngocele, while preserving the superior laryngeal nerve and its vascular pedicle.

Literature

1. Juneja R, Arora N, Meher R, Mittal P, Passey JC, Saxena A, et al. Laryngocele: A Rare Case Report and Review of Literature. *Indian J Otolaryngol Head Neck Surg.* 2019; 71:147-151. doi: 10.1007/s12070-017-1162-x.
2. Aksoy EA, Elsürer C, Serin GM, Unal OF. Bilateral internal laryngoceles mimicking asthma. *J Res Med Sci.* 2013;18(5):453-6. PMID: 24174956.

3. Prasad N, Singh M, Nagori R, Singh S. Laryngopyocele: Presenting with pressure symptom. *Int J Appl Basic Med Res.* 2015;5(3):228-30. doi: 10.4103/2229-516X.165375.
4. Al-Yahya SN, Baki MM, Saad SM, Azman M, Mohamad AS. Laryngopyocele: report of a rare case and systematic review. *Ann Saudi Med.* 2016;36(4):292-7. doi: 10.5144/0256-4947.2016.292.
5. Dursun G, Ozgursoy OB, Beton S, Batikhan H. Current diagnosis and treatment of laryngocele in adults. *Otolaryngol Head Neck Surg.* 2007;136(2):211-5. doi: 10.1016/j.otohns.2006.09.008.
6. Keles E, Alpay HC, Orhan I, Yildirim H. Combined laryngocele: a cause of stridor and cervical swelling. *Auris Nasus Larynx.* 2010;37(1):117-20. doi: 10.1016/j.anl.2009.02.011.
7. Martinez Devesa P, Ghufoor K, Lloyd S, Howard D. Endoscopic CO2 laser management of laryngocele. *Laryngoscope.* 2002; 112(8):1426-30. doi: 10.1097/00005537-200208000-00018.
8. Biswas S, Saran M. Blunt Trauma to the Neck Presenting as Dysphonia and Dysphagia in a Healthy Young Woman; A Rare Case of Traumatic Laryngocele. *Bull Emerg Trauma.* 2020;8(2):129-131. doi: 10.30476/BEAT.2020.46455.
9. Kara İ, Kökoğlu K, Çağlı S, Yüce İ. Bilateral Laryngocele Causing Epiglottic Deformity and Upper Airway Obstruction. *Turk Arch Otorhinolaryngol.* 2019;57(2):99-101. doi: 10.5152/tao.2019.3949.
10. Thomé R, Thomé DC, De La Cortina RA. Lateral thyrotomy approach on the paraglottic space for laryngocele resection. *Laryngoscope.* 2000; 110(3):447-50. doi: 10.1097/00005537-200003000-00023.
11. Bakır S, Gül A, Kınış V, Özbay M, Özkan H. A case of mixed type laryngocele presented with deep neck infection and review of the literature. *J Clin Exp Invest.* 2012;3(3):415-9. <https://doi.org/10.5799/ahinjs.01.2012.03.0192>
12. Zelenik K, Stanikova L, Smananova K, Cerny M, Kominek P. Treatment of Laryngoceles: what is the progress over the last two decades? *Biomed Res Int.* 2014;2014:819453. doi: 10.1155/2014/819453.
13. Roediger FC, Eisele DW. Complications of Neck Surgery. In: Cummings Otolaryngology, Head and Neck Surgery, 5th Edition. Elsevier/Mosby, Philadelphia 2010; pp. 1726-1733.

KOMBINOVANA LARINGOKELA KAO REDAK UZROK OTOKA NA VRATU

Jovanovic Anđelina¹, Valjarević Svetlana^{1,2}, Jovanović Milan^{1,2}

Sažetak

Laringokela je vrećasto, patološko proširenje Morganjijevog ventrikulusa, koje nastaje između pravih i lažnih glasnica. Ova promena je ispunjena vazduhom i u kontinuitetu je sa lumenom larinksa. Obično je bez simptoma i slučajno se otkriva u toku radiološke dijagnostike. Prikazujemo slučaj pacijentkinje stare 54 godine koja se javila na pregled zbog bezbolnog otoka na vratu sa leve strane, koji je trajao 8 meseci i povećavao se naValsalvin manevar. Povremeno je pacijentkinja bila promukla. Kliničkim otorinolaringološkim pregledom i kompjuterizo-

vanom tomografijom vrata i larinksa našli smo cističnu formaciju, ispunjenu vazduhom, koja se pružala iznad larinksa i širila u bočnu stranu vrata. Laringokelu smo u celini hirurški odstranili spoljašnjim cervikalnim pristupom. Vrat laringokele je ispraćen niz tiroidnu membranu, uz očuvanje gornjeg laringealnog nerva i prateće vaskularne peteljke. Patohistološkim pregledom, utvrđeno je da se laringokela sastoji od respiratornog slojevitog cilindričnog epitela. Laringokele treba uzeti u obzir u diferencijalnoj dijagnozi otoka vrata.

Ključne reči: kombinovana laringokela, otok na vratu, spoljašnji hirurški pristup

Primljen: 04.10.2022. | **Revizija:** 24.10.2022. | **Objavljen:** 03.11.2022

Medicinska istraživanja 2022; 55(3):67-70