## RARE CASES OF MIXED LARYNGOCELE

J. Olszewski, J. Mitoński, P. Pietkiewicz, H. Zielińska-Bliźniewska Medical University of Lodz, Lodz, Poland Institute of Physiology and Pathology of Hearing, Warsaw, Poland

The causes of laryngocele formation are still difficult to determine. The aim of the study was to present two cases of mixed laryngocele in patients hospitalized at the Department of Otolaryngology, Laryngological Oncology, Audiology and Phoniatrics, Medical University of Lodz. Laryngocele is a lesion that can coexist with other inflammatory and proliferative diseases, especially with malignant tumors, and therefore it often requires extended diagnostics in hospital setting.

**Keywords:** laryngocele, laryngoscopy, videolaryngostroboscopy, thyrotomy.

#### Introduction

Laryngocele is a rare entity characterized by cystic dilatation of the laryngeal saccule, an appendix which ends blindly in the 1/3 of the anterior part of the laryngeal ventricle bottom. The laryngeal saccule extends upward between the vestibular fold and the inner surface of the thyroid cartilage to its upper edge or higher [1, 2].

The incidence is estimated to be 1 per 2.5 million of the population per year and is about 5 times more frequently seen in men than in women, usually at the age of 40-59 years [1,3,4]. The etiology still remains unclear, but it seems that congenital and acquired factors associated with elevated intralaryngeal pressure could play a role [3].

Usually laryngocele is asymptomatic, although cases were reported presenting with swelling of the neck, hoarseness, cough, dysphagia, foreign body sensation in the throat or dyspnea. The presentation of symptoms depends on the size and type of laryngocele [3]. The most common content is air (pneumocele) less often mucus (mucocele). In some cases, there may come to superinfection and formation of laryngopyocele with symptoms of increasing painful swelling of the neck with airway obstruction [5].

Depending on the location in relation to the thyrohyoid membrane, there are distinguished [1, 3, 4]:

- Internal laryngocele confined to the interior of the laryngeal cavity, hoarseness is a usual symptom as well as sometimes cough, dyspnea, dysphagia, sore throat or stridor. The change can be seen on indirect and direct laryngoscopy as a bulge in the epiglottic space.
- External laryngocele usually seen as a nodular change of the neck, located at the level of the hyoid bone forward from the sternocleidomastoid muscle. The lesion increases during the Valsalva maneuver and decreases as a result of palpation [3].
- Mixed laryngocele most frequent and it is the combination of both internal and external laryngocele. The diagnosis is based on the medical history, indirect and direct laryngoscopy, videolaryngostroboscopy and neck imaging ultrasound, contrast-enhanced CT or MRI.

The aim of the study was to present two cases of mixed laryngocele in patients hospitalized at the Department of Otolaryngology, Laryngological

Oncology, Audiology and Phoniatrics, Medical University of Lodz.

Case report 1

A 59-year-old female (A.Z) admitted to the Department of Otolaryngology, Laryngological Oncology, Audiology and Phoniatrics, Military Medical Academy Memorial Teaching Hospital -Central Veterans' Hospital for the diagnostics of nodular lesion located in the left angle of the mandible. The patient reported hoarseness, periodic dysphagia, heartburn. She also explained that the lesion was growing imperceptibly and was periodically painful. She first noticed the swelling about a year ago. She admitted occasional smoking and no alcohol abuse. Until admission to the Department she had not been diagnosed for this reason. On examination she was found to have a lesion of approximately 4 cm in diameter, located in the left angle of the mandible, which was fluctuant and painless on palpation. The swelling was covered with normal skin, soft to the touch, of normal temperature. Neck nodes were not enlarged on palpation. Indirect laryngoscopy revealed the following deviations: left vocal fold invisible – hidden under the thickened left vestibular Videolaryngostroboscopy: the epiglottis unchanged. The right true vocal cord is smooth and mobile. The left true vocal cord invisible, completely obscured by hyperemic, hypertrophied left false vocal cord. Slight thickening of the right false vocal cord. Phonation by false vocal cords. Symmetrical arytenoids with bilateral swelling, hyperemic, of normal mobility. Interarytenoid mucosa appeared edematous and hyperemic – reflux-related lesions. Wide rima glottides. Flattening of the left piriform fossa, right – normal.

Conclusions: left false vocal cord hypertrophy. Suspicion of a tumor (cyst) of the laryngeal ventricle on the left side (fig. 1)

Neck ultrasound: no specific lesion is visible in the place of the thickening on the left side, the features of subcutaneous emphysema on the 2 cm section are seen. In the lower pole of the left parotid gland, a poorly contoured hypoechoic focus of 10mm in diameter is visible.

BACC of the described lesion: the obtained salivary gland lobe specimens showed no cytological features of atypia

Neck 2-phase CT (fig. 2 and 3): pathological mass on the left side of the neck is the air reservoir (of about 47x34 mm) connected most probably to

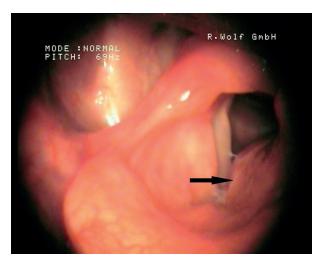


Fig. 1. – Videolaryngostroboscopy:visible left true vocal cord obscured by the false vocal cord



Fig.2. - CT frontal view: visible laryngocele

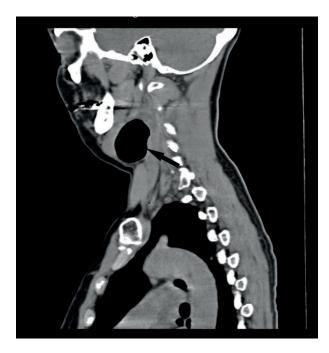


Fig.2. – CT sagittal view: visible laryngocele

the front part of the glottis compressing the left piriform fossa and dislocating left submandibular gland to the front. (PWZ: 8568478 – Department of

Diagnostics and Radiological and Isotope Therapy, Military Medical Academy Memorial Teaching Hospital – Central Veterans' Hospital in Lodz).

On the basis of the medical history, physical examination and additional tests, in particular the computed tomography examination, the patient was diagnosed with laryngocele and qualified for surgical treatment.

Case Report 2

A 74-year-old female patient (D.T.) admitted to the Department in a planned mode due to left sided neck soft nodular swelling and voice change for past 1,5 year. In the medical history the patient reported that the swelling gradually increased. The patient also noticed that during the speech the lesion increases in size. She did not report any other complaints at the time of the admission. Chronic diseases: chronic obstructive pulmonary disease (COPD), arterial hypertension, history of tuberculosis.

The otolaryngological physical examination revealed in the submandibular region a soft nodular lesion, about 5 cm in size, prominent during speech was found. The skin covering the swelling was normal. Lymph nodes available for palpation examination were not enlarged. No significant deviations from normal were observed in ears, nose and throat.

Larynx videolaryngostroboscopy (fig. 4): the epiglottis has regular configuration, mucous membrane is pink. The protruded left false vocal cord obstructs the left true vocal cord, pronounced partial features of left laryngeal ventricular prolapse in the anterior part. The right true vocal cord is smooth, mobile, with the remaining mucosal wave. The left true vocal cord in its visible part is mobile and smooth. Wide rima glottidis. Piriform sinuses unaffected, symmetrical, without residue.



Fig. 4. – Videolaryngostroboscopy: visible left true vocal cord obscured by the false vocal cord.

Neck CT: A 2.7 x 4.7 x 2.2 cm irregular airfilled cavity within the anterior triangle of the neck. It extends between the thyroid cartilage and the hyoid bone. This lesion displaces the infrahyoid muscles and causes neck soft tissue swelling on the left side. Medially, communication is seen with the intermediate laryngeal cavity through the left laryngeal ventricle. CT scan indicates laryngocele (fig. 5 and 6).



Fig. 5. CT frontal view: visible laryngocele



Fig.6. CT sagittal view: visible laryngocele

Results of laboratory tests: RBC 4.21 [\*10′6/ $\mu$ L], HGB 12.8[g/dl], HCT 38.3 [%], PLT 199 [\*10′3/ $\mu$ L], WBC 8.76[\*10′3/ $\mu$ L], CRP 0.9 [mg/l]. Ionogram, glucose and coagulogram in normal range. Chest X-ray: prominent peribronchial markings. No focal lung lesions. Diaphragm normal, angles clear. Heart size within normal limits.

Basing on the clinical picture, videolaryngostroboscopy and CT scan of the neck, the following was diagnosed: Left-sided external laryngocele with partial left laryngeal ventricular prolapse (mixed laryngocele) and the patient was qualified for surgical treatment.

The surgical treatment and postoperative period without complications. The patient was discharged from the Department in good general condition. Histopathological diagnosis No. 4734 / 2018HIST: Cystic tumor of 2x1.8x1.4 cm in size - laryngocele.

## Discussion

The causes of laryngocele formation are still difficult to determine. The risk group includes people exposed to repeated high pressure in the larynx, e.g. related to the profession of: singers, trumpeters, glassblowers, or to disease entities, i.e. chronic cough, constipation, prolonged use of laryngeal masks in anesthetic patients [1,6]. There have been also described cases of laryngocele resulting from mechanical obstruction (laryngeal or lung cancers, amyloidosis) and those following neck surgery [7,8]. In our case, no factors favoring laryngocele formation were found. Markowski et al. [2] describe that mixed laryngocele often causes voice disorders and promotes the occurrence of laryngeal vestibule phonation. Pathologically excessive intralaryngeal pressure affects the functions of the vestibular folds. Surgical treatment aiming at restoring both laryngeal pressure and anatomy, results in return of the normal quality of voice. In the patient described by us, symptoms of chronic hoarseness were caused by improper phonation of the vestibular folds and the hypertrophy of the left false vocal cord as confirmed by laryngeal videostroboscopy.

Apart from phoniatric symptoms, the patient presented a nodular lesion in the neck. Laryngeal and neck symptoms are compatible with the characteristics of the mixed laryngocele. Dysphagia described by the patient is also identified with the presence of laryngocele and does not have to be a symptom of gastro-esophageal reflux, which was attributed to the patient. Intermittent painfulness of the lesion was an important information. On palpation the nodular lesion seemed to be filled with fluid. Then, it should be suspected that the lesion can be infected.

Wójtowicz et al. [5] described a case of a patient with similar symptoms with additional dyspnea and palpation pain of the nodular neck lesion. It should be remembered that the symptoms depend on both the size and type of laryngocele/pyomucocele, which include: hoarseness, shortness of breath, sore throat, foreign body sensation in the throat with dysphagia, painful swelling of the neck or fever.

The tumor aspiration biopsy allows identification of the pathogen and thus the implementation of appropriate antibiotic therapy. In contrast to uninfected, asymptomatic laryngocele, pyomucocele requires faster surgical intervention [5].

The coexistence of laryngocele with malignant tumors is also described. In the literature, these values range from 5% [9] to 15% of cases [5]. Laryngeal cancer is the most frequent comorbidity. However, other neoplastic changes cannot be ruled out. In the presented case during the ultrasound examination of the neck, a hypoechogenic lesion was detected in the left parotid gland during the ultrasound examination. Anyway, BACC showed normal salivary gland formation without the features of atypia. Surgery is the treatment of choice. Small internal laryngoceles are removed by direct microlaryngoscopy using a CO<sub>2</sub> laser. This technique is considered a fast, precise and safe alternative to the surgical method from an external approach. There is also a smaller number of complications which allow patients to recover more quickly [6,10]. Unfortunately, the limited operating field favors incomplete resection of the lesion. That is why the external approach surgery is preferred for external or mixed laryngocele. There have been described three methods over the period of about 20 years — transthyrohyoid membrane approach, thyrotomy with resection of upper 1/3 of thyroid cartilage and V thyrotomy [6, 10]. The first method was the most commonly used, in 68% of cases [6]. The advantage of this method is that the thyroid

cartilage remains intact, although it limits access to periglottic space.

#### **Conclusion**

Laryngocele is a lesion that can coexist with other inflammatory and proliferative diseases, especially with malignant tumors, and therefore it often requires extended diagnostics in hospital setting.

# References

- 1. Pajor A, Maciaszczyk K, Józefowicz-Korczyńska M. Laryngocele mieszane opis przypadku i przegląd piśmiennictwa. *Otorynolaryngologia*. 2012;11(2):79-82.
- 2. Markowski J, Gierek T, Paluch J, Witkowska M, Jura-Szołtys E. Laryngocele mieszane opis przypadku i aspekty foniatryczne. *Otorynolaryngologia*. 2009;8(3):142-145.
- 3. DynaMed Plus [Internet]. Ipswich (MA): EBSCO Information Services. 1995 Record No. 114851, Laryngocele. Available from: http://www.dynamed.com/login.aspx?direct=true&site=DynaMed&id=114851.
- 4. Balasubramanian T. Laryngocele a review. *Otolaryngology online journal*. 2015;5(1):5.
- 5. Wójtowicz T, Bruzgielewicz A, Osuch-Wójcikiewicz E. Piomukocele mieszane jako przyczyna duszności. *Polski Przegląd Otorynolaryngologiczny*. 2013;2:117-120.

- 6. Zelnik K, Stanikova L, Smatanova K, Cerny M, Kominek P. Treatment of Laryngoceles: What is the progress over the last two decades? *BioMed Research International*. 2014. Article ID: 819453. doi: 10.1155/2014/819453.
- 7. Keles E, Alpay HC, Orhan I, Yildrim H. Combined laryngocele: A cause of stridor and cervical swelling. *AurisNasus Larynx*. 2010;37:117-120.
- 8. Marom T, Roth Y, Cinamon U. Laryngocele. A rare long-term complication following neck surgery? *Journal of Voice*. 2011;25(3):272-274.
- 9. Celin SE, Johnson J, Curtin H, Barnes L. The association of laryngoceles with squamosus cell carcionoma of the larynx. *Laryngoscope*. 1991;101:529-536.
- 10. Dursun G, Ozgursoy OB, Beton S, Batikhan H. Current diagnosis and treatment of laryngocele in adults. *Otorynolaryngology Head and Neck Surgery*. 2007;136:211-215.

# РЕДКИЕ СЛУЧАИ СМЕШАННОГО ЛАРИНГОЦЕЛЕ

Ольшевски Ю., Милоньски Я., Питкевич П., Зелиньска-Ближневска X. Медицинский университет Лодзи, Лодзь, Польша Институт физиологии и патологии слуха, Варшава, Польша

Причины образования ларингоцеле все еще трудно определить. Цель исследования – представить два случая смешанного ларингоцеле у пациентов, госпитализированных в отделение отоларингологии, ларингологической онкологии, аудиологии и фониатрии Медицинского университета Лодзи. Ларингоцеле – это поражение, которое может сосуществовать с другими воспалительными и пролиферативными заболеваниями, особенно со злокачественными опухолями, поэтому часто требуется расширенная диагностика в условиях стационара.

Ключевые слова: ларингоцеле, ларингоскопия, видеоларингостробоскопия, тиреотомия.

Поступила: 18.05.2018 Отрецензирована: 12.06.2018