

CASE REPORT



Laryngocele associated with laryngeal carcinoma

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Abstract

Laryngocele is an abnormal dilatation of Morgagni ventricle (saccule) in direct communication with the laryngeal lumen. Symptoms are not characteristic: hoarseness, dyspnea, foreign body sensation and cough. Sometimes it is presented as cervical swelling causing airway obstruction and need an emergency tracheotomy. In this paper, we report a case of upper airway obstruction due to laryngocele associated with a laryngeal carcinoma who was treated by emergency tracheotomy and, finally by total laryngectomy. A review of the literature is also presented.

Keywords: laryngocele, emergency tracheotomy, laryngeal carcinoma.

■ Introduction

Laryngocele is a rare cystic dilatation of the laryngeal saccule, which arises from the laryngeal ventricle and extends into paralaryngeal space [1]. The etiology is unknown but it is probably related to both congenital and acquired factors.

Larrey, Napoleon's surgeon, in 1829, was the first to report a series of laryngoceles that he observed in men who would hourly chant the Koran from the minarets [2, 3]. Burke EN and Golden JL were the first ones who analyzed this disease; in an article published in 1958, they considered a Morgagni ventricle that extended beyond the superior border of the thyroid cartilage to qualify as a laryngocoeles [4]. In an autopsy review of 100 normal larynxes, Broyles EN [5] observed that 7% of them had saccules of sufficient length that they would be considered laryngoceles based on the early criteria. Consequently, the definition was changed: a laryngocoeles is a large Morgagni ventricle (saccule) that is symptomatic and palpable [6].

Most authors accept Burke and Golden's original description and recognize both symptomatic and asymptomatic laryngoceles [7].

There are three types of laryngoceles. An internal laryngocoele is confined to the interior of the larynx and extends postero-superiorly into the ventricular fold and the aryepiglottic fold; this type appears on laryngoscopy as a smooth swelling of the supraglottis. An external laryngocoele extends superiorly to appear laterally in the neck through the opening in the thyrohyoid membrane for the superior laryngeal nerve and vessels; these

clinically present as a swelling in the neck at the level of hyoid bone anterior to sternocleidomastoid muscle. The simultaneous existence of both features is termed a mixed (combined) laryngocoeles.

■ Patient, Methods and Results

Our 60-year-old man, R.D., from rural environment, was transferred from Urological Department (he underwent a TURP procedure for BPH) in ENT Clinic Craiova (O.F. 23887/2007) with a 4-month history of dysphonia and dysphagia. For last two weeks, patient present moderate dyspnea. The medical history of the patient included working in polluted air and smoking (20 cigarettes per day for 20 years). The indirect laryngoscopy showed a round compressible-soft mass on the right ventricular fold which changes its size during the phonation and Valsalva manoeuvre and a vegetative tumoral mass which involved the left vestibule and vocal fold, anterior to commissure and subglottis. The left half of the larynx had low mobility.

A plain lateral neck radiograph (2218/29.09.2003) revealed a round and air-containing mass laterocervically and submandibular, 5/4 cm in diameter and a soft tissue intensity opacity, 3/2 cm in diameter, located in the left subglottis. The lateral cervical echography showed right micro-nodular adenopathy of 8.2 cm in diameter, right above the place where the common carotid artery divided.

CT scans showed a large well-defined air-containing lesion displacing and narrowing the laryngeal lumen (Figure 1).



Figure 1 – Sagittal CT scan of laryngocoele.

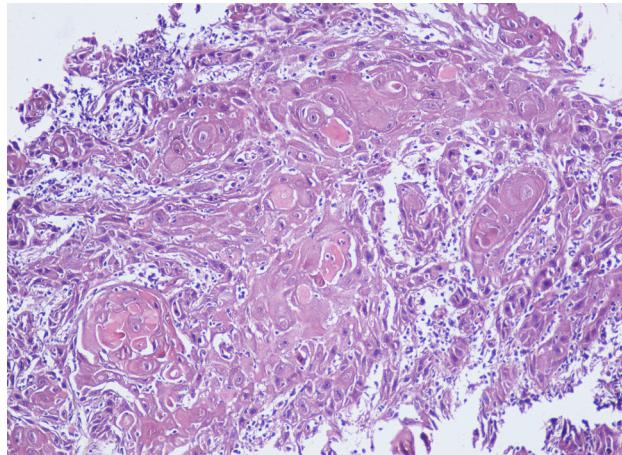


Figure 2 – Well-differentiated squamous cell carcinoma (HE stain, $\times 100$).

During hospitalization, dyspnea became more important and we decided to perform an emergency tracheotomy prior biopsy. The histopathologic examination of the tumoral mass (355/08.05.2007) revealed larynx carcinoma (Figures 2–5).

Final decision was to perform a total laryngectomy (S.R.784/16.05.2007).

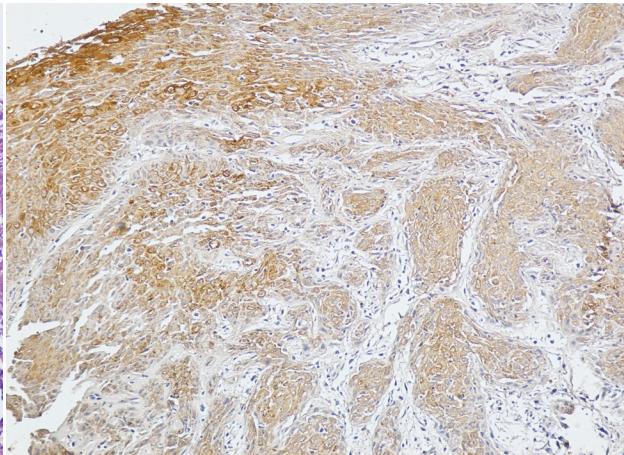


Figure 3 – Squamous cell carcinoma: diffuse strongly positive cytoplasmic immunomarkers for AE1/AE3 cytokeratin (LSAB technique, $\times 100$).

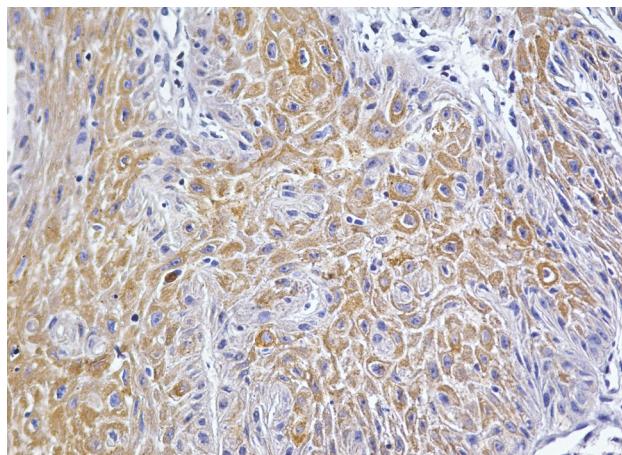


Figure 4 – Squamous cell carcinoma: strongly positive cytoplasmic immunomarkers for tumor cytokeratin 34 β E12 (LSAB technique, $\times 200$).

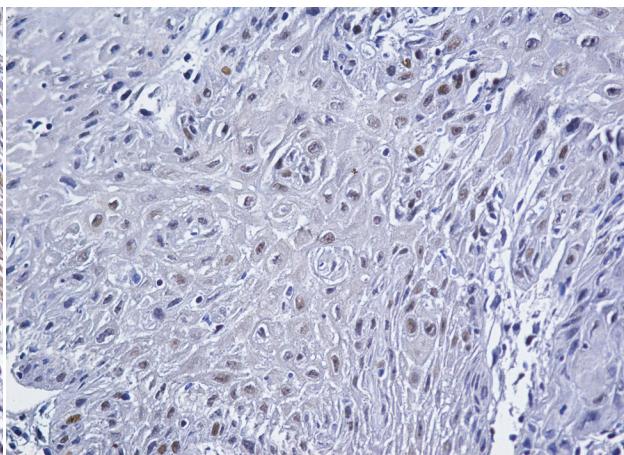


Figure 5 – Squamous cell carcinoma: weak positive immunomarkers for nuclear p63-protein in tumor cells (LSAB technique, $\times 200$).

Discussion

Laryngocoele is a rare disease (5% of benign laryngeal lesions), whose etiology is still unknown. It may be due to prolonged periods of increased pressure within the laryngeal lumen [8, 9] as observed in wind instrument. Different articles published in specialty reviews try to associate laryngocoeles with different laryngeal pathology: laryngeal amyloidosis [10], laryngeal chondroma [11], history of a tracheotomy [12], papillomatosis in children [13] or extralaryngeal pathology – ankylosing spondylitis [14] that would cause an increased intra-thoracic pressure, oncocytic cysts [15]. Many studies [16–18] made a connection between the presence of the larynx carcinoma and the appearance of laryngocoeles (the distortion of the saccule neck by carcinoma may create a one-way valve which increases intraluminal pressure).

In a study on 139 laryngocoeles, Stell PM and Maran AGD showed that the sex incidence is 5:1 in favor of men, and the maximum age incidence is in the sixth decade [8]; about 8% become infected and present as laryngopyocele.

Patients with an internal laryngocoele usually complain of hoarseness, dyspnea, foreign body sensation and cough. Laryngoscopy shows a swelling of the false cord on one side. The external laryngocoele presents as a swelling in the neck.

Laryngocoeles are lined by pseudostratified, columnar, ciliated epithelium with occasional foci of stratified squamous epithelium and a mixture of submucosal serous and mucous glands [8, 19]. This composition distinguishes these lesions from laryngeal cysts, which are lined entirely by squamous epithelium [8].

In some cases, the clinical presentation is not that

typical, so that computed tomography can be helpful. CT scan shows an air or mucus-containing tumor at the level of the false cord. Internal laryngoceles may proceed into an external one, which presents as a swelling at the level of the hyoid bone.

Uncomplicated laryngoceles appear on CT as air-filled structures lying in the paralaryngeal space (internal), lateral neck (external) or in both locations (mixed). Obstruction of the neck of the laryngcele by either tumor or chronic inflammation can result in a fluid-filled structure, producing on CT a well-circumscribed mass of either near water or soft-tissue density, depending on its composition.

In our case, there is an association between laryngcele and laryngeal carcinoma. First symptoms that occurred were dysphagia and dysphonia, rapidly followed by dyspnea, which required tracheotomy and finally total laryngectomy. Post-operative recovery was free from complications.

Conclusions

Laryngoceles is not a common illness. Its diagnosis is usually difficult and requires complex explorations. There is a rare association of laryngcele with laryngeal carcinoma. Thus, it is important to carry out an investigation in patients with laryngcele aiming at ruling out any associated malignancy.

References

- [1] Pruszewicz A, Obrebowksi A, Maciejewska B, *Bilateral internal laryngcele with open nasality – report of a case*, Otolaryngol Pol, 2006, 60(6):935–938.
- [2] Holinger LD, Barnes DR, Smid LJ, Holinger PH, *Laryngcele and saccular cysts*, Ann Otol Laryngol, 1978, 87(5 Pt 1):675–685.
- [3] Koeller KK, Alamo L, Adair CF, Smirniotopoulos JG, *Congenital cystic masses of the neck: radiologic-pathologic correlation*, Radiographics, 1999, 19(1):121–146; quiz 152–153.
- [4] Burke EN, Golden JL, *External ventricular laryngcele*, Am J Roentgenol Radium Ther Nucl Med, 1958, 80(1):49–53.
- [5] Broyles EN, *Anatomic observations concerning the laryngeal appendix*, Ann Otol Rhinol Laryngol, 1959, 68(2):461–470.
- [6] DeSanto LW, *Laryngcele, laryngeal mucocele, large saccules, and laryngeal saccular cysts: a developmental spectrum*, Laryngoscope, 1974, 84(8):1291–1296.
- [7] Close LG, Merkel M, Burns DK, Deaton CW Jr, Schaefer SD, *Asymptomatic laryngcele: incidence and association with laryngeal cancer*, Ann Otol Rhinol Laryngol, 1987, 96(4):393–399.
- [8] Stell PM, Maran AGD, *Laryngcele*, J Laryngol Otol, 1975, 89(9):915–924.
- [9] Amin M, Maran AGD, *The aetiology of laryngcele*, Clin Otolaryngol Allied Sci, 1988, 13(4):267–272.
- [10] Cankaya H, Egeli E, Unal O, Kiris M, *Laryngeal amyloidosis: a rare cause of laryngcele*, Clin Imaging, 2002, 26(2):86–88.
- [11] Papila I, Acioğlu E, Karaman E, Akman C, *Laryngeal chondroma presenting as a laryngopyocele*, Eur Arch Otorhinolaryngol, 2005, 262(6):473–476.
- [12] Upile T, Jerjes W, Sipaul F, El Maaytah M, Singh S, Howard D, Hopper C, Wright A, *Laryngcele: a rare complication of surgical tracheostomy*, BMC Surg, 2006, 6:14.
- [13] Altamar-Ríos J, Morales Rozo O, *Laryngcele and pyolaryngcele*, An Otorrinolaringol Ibero Am, 1992, 19(4):393–399.
- [14] Erdogmus B, Yazici B, Ozturk O, Ataoglu S, Yazici S, *Laryngcele in association with ankylosing spondylitis*, Wien Klin Wochenschr, 2005, 117(19–20):718–720.
- [15] McDonald SE, Pinder DK, Sen C, Birchall MA, *Oncocytic cyst presenting as laryngcele with surgical emphysema*, Eur Arch Otorhinolaryngol, 2006, 263(3):237–240.
- [16] Akbas Y, Unal M, Pata YS, *Asymptomatic bilateral mixed-type laryngcele and laryngeal carcinoma*, Eur Arch Otorhinolaryngol, 2004, 261(6):307–309.
- [17] Uğuz MZ, Onal K, Karagöz S, Gökcé AH, Firat U, *Coexistence of laryngeal cancer and laryngcele: a radiologic and pathologic evaluation*, Kulak Burun Bogaz İhtis Derg, 2002, 9(1):46–52.
- [18] Harney M, Patil N, Walsh R, Brennan P, Walsh M, *Laryngcele and squamous cell carcinoma of the larynx*, J Laryngol Otol, 2001, 115(7):590–592.
- [19] Chu L, Gussack GS, Orr JB, Hood D, *Neonatal laryngoceles. A cause of airway obstruction*, Arch Otolaryngol Head Neck Surg, 1994, 120(4):454–458.

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