**A Rare Cause of Acute Airway Obstruction: The Mixed Laryngopyocele** M Sellami<sup>1</sup>, M Mnejja<sup>1</sup>, MA Chaabouni<sup>1</sup>, M Ben Salah<sup>2</sup>, I Charfeddine<sup>1</sup>, A Ghorbel<sup>1</sup>

# ABSTRACT

The laryngocele is an uncommon cystic dilatation of the saccule or appendix of the laryngeal ventricle, filled with air and communicating with the lumen of the larynx. Infection of laryngocele or laryngopyocele is exceptional.

Considering the rarity of the laryngoceles and the even rarer laryngopyocele, we describe an uncommon case of a mixed laryngopyocele causing acute symptoms.

Keywords: Airway obstruction, Laryngocele, Laryngopyocele

Correspondence: Dr M Sellami, Department of Otorhinolaryngology-Head and Neck Surgery Habib Bourguiba University Hospital, 3029 Sfax, Tunisia, E-mail : sellami\_moncef@yahoo.fr

West Indian Med J

From: <sup>1</sup>Department of Otorhinolaryngology-Head and Neck Surgery, Habib Bourguiba University Hospital, Sfax, Tunisia.

<sup>&</sup>lt;sup>2</sup>Department of Otorhinolaryngology-Head and Neck Surgery, Sidi Bouzid Hospital, Sidi Bouzid, Tunisia.

## INTRODUCTION

Laryngocele is an abnormal cystic dilatation of the laryngeal ventricle, filled with air and communicating with the lumen of the larynx (1).

In less than 10% of cases, the laryngocele is infected and become a laryngopyocele (2). The laryngopyocele is a life-threatening mass by leading rapid and complete obstruction of the airway (4, 3).

We report a rare case of a mixed laryngopyocele that presented as an emergency with rapidly progressive respiratory obstruction and painful neck swelling.

## **CASE REPORT**

A 63-year-old woman with a 3-years history of diabetes presented progressively worsening odynophagia lasting for 10 days initially treated by amoxicillin-clavulanic acid and dexamethasone. However, with no improvement under treatment and occurrence of a progressively worsening dyspnea, the patient was referred to our department. Upon arrival, her temperature was 38.4°C and a parenteral antibiotic therapy (ceftriaxone/ metronidazole) was started.

Physical examination revealed a patient in marked respiratory distress and a painful swelling of the anterior neck triangle at the level of the hyoid bone, extended to the right submandibular region without signs of acute inflammation (Fig. 1). On palpation, this swelling was non-fluctuant, stretched-elastic in consistency and approximately  $6 \times 4$  cm in size.

Fibre-optic laryngoscopy revealed a large, smooth right supraglottic mass involving the aryepiglottic fold and pushing the larynx towards left (Fig. 2). This mass was associated with a mild epiglottis edema and 70% airway obstruction at the level of supraglottis.

#### Sellami et al

A contrast-enhanced computed tomography (CT) scan revealed well-defined multiloculated hypodense lesion on the right side of the paralaryngeal space pushing the supraglottis towards the left side extended inferiorly until the level of the vocal cords. The mass was piercing the thyro-hyoid membrane and reaching the space between the thyroid cartilage and thyrohyoid muscle (Fig. 3).

Diagnosis of laryngopyocele was confirmed according to clinical findings, endoscopic examination of larynx, and imaging studies. During the CT scan, the respiratory condition of the patient worsened rapidly and she was taken to the operating room for the drainage of the laryngopyocele. An intubation was not achievable due to the obstruction of the upper airway and the patient was managed with an urgent surgical tracheostomy to secure the airway. The surgery consists on an external approach via a horizontal skin incision at the level hyoid bone followed by an incision of the right infrahyoid muscles between hyoid bone and the upper border of the thyroid cartilage. The laryngopyocele was drained and the pouch was resected (Fig. 4).

The pus aspirated from the cavity grew *Klebsiella pneumoniae* sensitive to amoxicillin–clavulanic acid, cefuroxime and ciprofloxacin. Postoperatively the patient progressed without complications, with full symptoms improvement and good healing on fibre-optic laryngoscopy (Fig. 5). The patient was successfully decannulated in the seventh postoperative day and discharged home three days later. The patient had no further treatment and the CT scan performed 2 months after surgery showed no recurrence.

## DISCUSSION

A laryngocele is an air-filled herniation of the saccule of the laryngeal ventricle communicating with the lumen of the larynx (1).

The incidence of laryngocele is estimated to be 1 per 2.5 million of the population per year and this condition has been reported to be five times more frequent in men, with a peak incidence in the sixth decade of life (5).

The precise aetiology of laryngocele is unknown but association with laryngeal malignancies, essentially supra-glottic laryngeal tumours, was described (6). Laryngoceles are divided into internal, external, and mixed (combined) types according to their relationship with the thyrohyoid membrane (2). Internal laryngoceles are confined to the interior of the larynx and extend postero-superiorly into the false vocal cord and the aryepiglottic fold (2). External laryngoceles develop cranially and laterally to the neck, through a continuous solution of the thyrohyoid membrane (the opening for the superior laryngeal nerve and vessels) (2). The simultaneous existence of both features is termed a combined laryngocele

By definition, a laryngocele contains air only and can be blocked by mucus and continue to enlarge, but in 10% of patients, it may become infected resulting in a laryngopyocele (3, 6).

The mucus stasis within the cyst, and altered mucociliary clearance, could lead to infection by several bacteria such as Staphylococcus aureus, Hemolytic Streptococcus B, Escherichia coli and Pseudomonas aeruginosa reaching the diseased cavity from the contiguous areas (2).

The symptoms depend upon the type and size of the laryngopyocele. The main presentation of external laryngopyocele is a neck mass and rarely presents with recurrent neck abscess (7). Internal laryngopyocele is frequently present with pharyngolaryngeal symptoms such as hoarse voice, cough, inspiratory stridor, dysphagia, and a `full' sensation in the throat (3). Mixed forms associate a neck mass with pharyngolaryngeal symptoms, as the case hereby presented.

CT examination is essential before initiation of treatment and provides the most accurate information on the extent of the laryngopyocele (3). The CT scan of the neck

permits an evaluation of the relationship between the laryngeal structures and the laryngocele (3).

The diagnosis of a laryngopyocele is primarily made on the basis of the laryngoscopy, and confirmed by CT scan (3).

The CT scan is also essential in making the differential diagnosis between laryngopyocele and other neoformations such as lipoma, paraganglioma, schwannoma, cystic lymphangioma, thyroglottic duct cysts, branchial cysts, epiglottic abscess and metastatic adenopathy (2, 4).

Securing an airway is paramount and patient are frequently managed with an urgent tracheostomy (7). The definitive management of symptomatic laryngopyocele is surgical excision including endoscopic, external and combined approach (7). Treatment of the mixed laryngocele is still controversial, several authors prefer an external approach, but a combined approach and fully internal approach have been advocated (2, 6). This approach could be performed through a horizontal neck incision, at the level of the thyro-hyoid membrane (1). Due to the important volume of the laryngopyocele, we preferred an external approach.

The laryngopyocele is a life-threatening condition by leading rapid and complete obstruction of the airway. Thus, Byard reported a case of a sudden death due to airway occlusion by a laryngopyocele (4).

## REFERENCES

- Vasileiadis I, Kapetanakis S, Petousis A, Stavrianaki A, Fiska A, Karakostas E. Internal laryngopyocele as a cause of acute airway obstruction: an extremely rare case and review of the literature. Acta Otorhinolaryngol Ital 2012; 32:58-62.
- Cassano L, Lombardo P, Marchese-Ragona R, Pastore A. Laryngopyocele: three new clinical cases and review of the literature. Eur Arch Otorhinolaryngol 2000; 257: 507-11.
- Nazaroğlu H, Ozateş M, Uyar A, Değer E, Simşek M. Laryngopyocele: signs on computed tomography. Eur J Radiol 2000; 33: 63-5.
- 4. Byard RW, Gilbert JD. Lethal laryngopyocele. J Forensic Sci 2015; 60: 518-20.
- Zelenik K, Stanikova L, Smatanova K, Cerny M, Kominek P. Treatment of Laryngoceles: what is the progress over the last two decades? Biomed Res Int 2014. 2014:819453.
- Fraser L, Pittore B, Frampton S, Brennan P, Puxeddu R. Laryngeal debridement: an alternative treatment for a laryngopyocele presenting with severe airway obstruction. Acta Otorhinolaryngol Ital 2011; 31: 113-7.
- Andreou Z, Randhawa PS, O'Flynn P, Vaz FM. Endoscopic management of an internal laryngopyocele presenting with acute airway obstruction. Case Rep Surg 2011; 2011:873613.



Fig. 1: Patient with a soft swelling approximately 6 cm in diameter located in of the anterior neck triangle extended to the right submandibular region



Fig. 2: Flexible pre-operative fibro-endoscopic assessment revealed a swelling of the right aryepiglottic fold with edematous epiglottis and airway obstruction.



Fig. 3: CT scan showing a large mixed right-sided laryngopyocele (arrow) pushing the supraglottis towards the left side extended inferiorly until the level of the vocal cords and piercing the thyro-hyoid membrane (A). The mass was reaching the space between the thyroid cartilage and thyrohyoid muscle (B)



H: hyoid bone, IHM: infrahyoid muscles, SMG: right submandibular gland Fig. 4: Intraoperative view showing the purulent material issue (arrow) after incision of the right infrahyoid muscles Sellami et al



Fig. 5: Flexible post-operative fibro-endoscopic assessment with evidence of good healing.