Bilateral mixed laryngopyocele as a rare cause of acute airway obstruction: a case report
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Introduction
Laryngocele is defined as abnormal dilatation of the saccule of the laryngeal ventricle in connection with the lumen of the larynx [1]. The exact etiology of this herniation is unknown and unclear. Some postulations suggest that there is an abnormal development of the larynx with an abnormally long saccule. The occurrence of laryngocele in certain occupations such as glass blowers and wind instrument players may be attributed to increased intraluminal pressure within the larynx [2]. Laryngoceles can be internal or external depending on the relation to the thyrohyoid membrane, and it can be combined or mixed [3]. Laryngopyocele is a rare serious complication of laryngocele and can be fatal due to acute airway obstruction [4]. We present a case of bilateral combined laryngopyocele presenting as acute airway obstruction and discuss the workup and management. The study was approved by the Alexandria University Hospital ethics committee and informed consent was obtained from the patient.

Case report
A 62-year-old male smoker presented to the Alexandria University Hospital Emergency Department with stridor. He gave a history of progressive dyspnea and odynophagia since 3 days. Past medical history included chronic obstructive pulmonary disease, type 2 diabetes mellitus, hypertension, and moderate obstructive sleep apnea (OSA). Generally, the patient was febrile (37.8°C); oxygen saturation was fluctuating between 80 and 83%. Standard ENT examination revealed tender left neck swelling with normal overlying skin. A flexible fiberoptic laryngoscopy revealed bilateral cystic swelling in the supraglottic larynx obscuring the view of vocal folds. There was no time for further investigations, so the patient was successfully intubated with a small-sized endotracheal tube. Emergency computed tomography neck revealed severe edema of the oropharynx as well as the larynx slightly supraglottic with almost total closure of the entire airway. A thin-walled peripherally enhancing cystic lesion was seen in the right supra/paraglottic, extending through the thyrohyoid membrane. Another cystic lesion with air fluid level was seen on the left side of the larynx (Fig. 1). The patient was then shifted to surgical ICU where he received broad-spectrum intravenous antibiotics (metronidazole and ceftriaxone). After the condition became stabilized, surgery was done via external cervical approach. Histopathological examination confirmed the diagnosis of laryngopyocele. The postoperative course was uneventful and the patient was discharged after 1 week.

Discussion
Laryngoceles may be congenital or acquired due to long-standing intraluminal pressure in the larynx as seen in some occupations such as glass blowers or due to chronic cough [2]. In the patient reported, chronic cough due to chronic obstructive pulmonary disease is clearly the cause of laryngocele. The association between laryngeal...
carcinoma and secondary laryngocele has been reported in several studies. Extreme caution must be exercised to rule out this possibility [5].

The true incidence of laryngoceles is underestimated as many cases are usually asymptomatic. The symptoms are variable according to the type of the laryngocele (external or internal). The symptoms include stridor, hoarseness, sore throat, cough, pain, snoring, or neck mass [6].

Laryngopyocele is a very rare but serious complication of laryngocele [4]. Forty cases had been reported in the literature and only four cases presented as acute airway obstruction [7]. Mortality due to airway obstruction secondary to laryngopyocele has been reported in the literature [4]. Computed tomography neck is the golden standard imaging method for the diagnosis of different types of laryngocoele [8].

Laryngopyocele presenting as acute airway obstruction is considered an emergency. Management initially includes securing the airway with endotracheal intubation or tracheostomy. Once the patient became stable, definitive surgical management is mandatory. In a systematic review of 23 cases of laryngopyocele, four cases were managed endoscopically, 18 cases were operated on via an external approach, and one case applied both approaches [8]. In this case, we successfully secured the airway with small ETT and then definitive surgery was done via external cervical approach. Tracheostomy was done at the beginning of surgery to secure the airway in the postoperative period. The postoperative course was uneventful and the patient was discharged after weaning from tracheostomy.

Conclusion
Laryngopyocele is a serious complication of laryngocele as it can present with acute airway obstruction. Adequate and close collaboration between the surgeon and the anesthesiologist is mandatory for securing the airway. Definitive surgical excision can be performed either endoscopically or via the external approach.

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Conflicts of interest
There are no conflicts of interest.

References
1 Marcotullio D, Paduano F, Magliulo G. Laryngopyocele: an atypical case. Am J Otolaryngol 1996; 17:345–348.
2 Amin M, Maran AGD. The aetiology of laryngocoele. Clin Otolaryngol Allied Sci 1988; 13:267–272.
3 Dray TG, Waugh PF, Hillel AD. The association of laryngoceles with ventricular phonation. J Voice 2000; 14:278–281.
4 Byard RW, Gilbert JD. Lethal laryngopyocele. J Forensic Sci 2015; 60:518–520.
5 Freeman J. Three cases of infected laryngocele. J Otolaryngol Otol 1952; 66:409–412.
6 Upile T, Jerjes W, Sipaul F, El Maaytah M, Singh S, Howard D, et al. Laryngocoele: a rare complication of surgical tracheostomy. BMC Surg 2006; 27:14–15.
7 Cassano L, Lombardo P, Marchese-Ragona R, Pastore A. Laryngopyocele: three new clinical cases and review of the literature. Eur Arch Otorhinolaryngol 2000; 257:507–511.
8 Al-Yahya SN, Baki MM, Saad SM, Azman M, Mohamad AS. Laryngopyocele: report of a rare case and systematic review. Ann Saudi Med 2016; 36:292–297.