Laryngocoele presenting as acute airway obstruction

Leong S C L, Badran K, McCormick M S

ABSTRACT

A laryngocoele is an abnormal dilatation of the laryngeal saccule. Symptomatic laryngocoeles can present as rapidly developing airway obstruction. In this case report of a 37-year-old man with a laryngocoele, we discuss the management dilemma presented by his repeated flight from the hospital prior to definitive treatment. This resulted in repeated admissions with life-threatening airway symptoms, culminating in emergency tracheostomies. Surgical excision of the laryngocoele was not achieved until the third presentation. We recommend early surgical intervention in patients who are potentially non-compliant to treatment or follow-up. A brief literature review of laryngocoele, as well as management of the difficult airway, are also discussed.

Keywords: airway obstruction, emergency tracheotomy, laryngocoele, tracheostomy

Singapore Med J 2007; 48(3):e84-e86

INTRODUCTION

This is a case report of laryngocoele presenting as acute airway obstruction. The patient is unique as a management dilemma as he frequently absconded from hospital. Airway management, with particular focus on awake fibreoptic intubation, is discussed.

CASE REPORT

A 37-year-old man presented to the casualty department with a three-day history of worsening dyspnoea, dysphagia and dysphonia. His symptoms were preceded by the appearance of an enlarging left neck mass over two days. He was a heavy cigarette smoker but otherwise had no significant medical history. We were suspicious that he was a drug-dealer, as he was well known to several patients in the ward who were intravenous drug abusers.

On examination, he was systemically well with no evidence of drug intravenous neck injections. There was a 5-cm horizontal scar in the midline neck, approximately two finger-breadths above the sternal



Fig. I Sagital CT image of the neck shows the internal component of the laryngomucocoele (L), which significantly obstructs the upper trachea (T).

notch. He was able to maintain a patent airway but had stridor, trimus and harsh dysphonia. The trachea was visibly deviated to the right. There was an 8 cm large diffuse mass in the left submandibular region, extending into the anterior triangle in levels I, II and III. Fibreoptic endoscopy revealed an oedematous epiglottis and a mass in the aryepiglottic fold, which was obstructing the laryngeal inlet.

Urgent computed tomography (CT) showed a laryngomucocoele at the level of the hyoid bone (Fig. 1). The airway was significantly narrowed and displaced to the right (Fig. 2). His symptoms continued to deteriorate progressively, despite maximum medical treatment of intravenous antibiotics and steroids. He was also given supplemental humidified oxygen with nebulised adrenaline. He underwent emergency tracheostomy and excision of the cyst later that day. General anaesthesia was accomplished via awake fibreoptic intubation. An external surgical approach was undertaken. The cyst was dissected down to the thyrohyoid membrane and into the supraglottis. The neck of the cyst was then transfixed and the sac excised (Fig. 3).

When he was able to give a history and the old case notes were available, it was revealed that the patient had

Department of Otolaryngology, Royal Liverpool University Hospital, Prescot Road, Liverpool L7 8XP, The United Kingdom

Leong SCL, MRCSE, DO-HNS Senior House Officer

Badran K, AFRCSI Senior House Officer

McCormick MS FRCS Consultant

Correspondence to: Dr Samuel C Leong 34A, Upper Parliament Street, Liverpool L8 1TE, The United Kingdom Tel: (44) 7887 507 519 Fax: (44) 1332 254 879 Email: lcheel@ doctors.org.uk



Fig. 2 Axial CT image of the neck shows the narrowed airway (P), which is displaced to the right. The fluid level within the laryngomucocele is clearly demonstrated (P: pharynx, l: internal, E: external).



Fig. 3 Operative photograph shows the laryngocoele stretched out to reveal the narrow neck of the sac (arrow).

two similar presentations of symptomatic laryngocoele in the preceding 12-month period. He had required urgent tracheostomy within hours of each admission due to impending total airway obstruction. Both times, the patient presented late in the day when an experienced surgeon was not available to excise the laryngocoele. On both occasions, he had surreptitiously left the hospital the following day and failed to attend followup clinic appointments. He admitted to removing the tracheotomy tube on his own accord several days after discharge when he had felt better.

The patient admitted to feeling increasingly frustrated at the need for repeated hospital attendance. He was, therefore, happy to comply with management on this admission. Culture of the mucus revealed *Streptococcus pneumonia* and *Haemophilus influenzae*. Histopathology of the excised sac was consistent with that of laryngocoele. The tracheostomy tube was removed two days after surgery and he was discharged from hospital the next day.

DISCUSSION

A laryngocoele is an abnormal dilatation of the laryngeal saccule.⁽¹⁾ The saccule arises from the laryngeal ventricle and extends into the paralaryngeal space. It was first described in 1829 by Dominique Larrey while serving as a surgeon in Napoleon's army in Egypt. The incidence is unknown as laryngocoeles are usually asymptomatic. Amin and Maran found enlarged laryngeal saccules in 21.5% of cadavers.⁽²⁾ The incidence of symptomatic laryngocoele, however, was estimated to be one in 2.5 million population per annum in the United Kingdom.⁽³⁾

A laryngocoele can be congenital or acquired. It may be due to prolonged periods of increased pressure within the laryngeal lumen as observed in wind instrument players.⁽⁴⁾ Laryngocoeles have also been associated with laryngeal carcinoma.⁽⁵⁾ It was hypothesised that distortion of the saccule neck by carcinoma may create a one-way valve which increases intraluminal pressure. Laryngocoele has also been described in an intravenous drug abuser who injected into blood vessels in the neck.⁽⁶⁾

Laryngocoeles are classified into three types: internal, external and mixed. Internal laryngocoeles are confined within the larynx and do not penetrate the thyrohyoid membrane, but extend postero-superiorly into the aryepiglottic fold and false vocal folds. External laryngocoeles extend outside the thyrohyoid membrane, through the opening for the superior laryngeal nerves and vessels, to appear in the lateral neck. Our patient presented with a mixed laryngocoele (a combination of internal and external laryngocoele), which is the most common presenting type. Diagnosis of laryngocoele can be obtained with CT. Symptomatic laryngocoeles usually present as a neck mass associated with hoarseness, stridor, foreign body sensation, sore throat, snoring or cough. Respiratory distress can develop rapidly when the air-filled pouch becomes blocked by mucus and continues to enlarge, and if the mucus becomes infected, a laryngopyocoele develops. While maintenance of a safe and secure airway remains the most important immediate therapeutic goal, airway management can be a challenge to both anaesthetist and otolaryngologist.

Although tracheotomy is the gold standard in airway management, the problem faced in these situations is how to induce anaesthesia and achieve intubation without loss of the airway. Endotracheal intubation is often difficult, and direct laryngoscopy may precipitate laryngeal spasm and acute airway collapse. Induction of anaesthesia can also be particularly dangerous, as neither successful intubation nor ventilation of the lungs can be guaranteed. Awake fibreoptic intubation is a recommended technique which is gaining popularity in the management of difficult airway, including deep neck space infections.⁽⁷⁾ Although an experienced anaesthetist is required, this technique ensures a patent airway in a controlled situation where the patient receives adequate oxygenation. Once intubated, the surgical procedure can be performed calmly to better explore an area which is often anatomically distorted by mass effect and oedema.

This patient presents a management dilemma uncommon to general otorhinolaryngological practice. A review of the case notes revealed that his previous tracheostomies were performed out-of-hours with excision of the laryngocoele planned as a later procedure after the infection and airway oedema had improved. While it can be argued that his worsening symptoms should have been recognised earlier in order that the entire procedure could be performed with more experienced surgeons present, he never attended hospital until late evening. Furthermore, he repeatedly absconded from hospital, only to reappear following deterioration in his symptoms to the point where he could no longer breathe properly. This resulted in lifethreatening readmissions due to impending airway obstruction.

There is currently no consensus on the timing of surgery, although laryngocoeles are conventionally

excised following initial management of airway obstruction. In our experience, excision can take place after the infection and oedema has improved although this may entails a second anaesthetic. This will allow a better overall surgical field and ensures completeness of excision. Various other surgical technique have been described, including deroofing of the cyst and laser excision.⁽⁸⁾ In cases where patients may be potentially noncompliant to treatment or follow-up, we recommend expeditious surgical intervention where excision of the laryngocoele can be performed by an experienced surgeon in the same single procedure should a trachestomy be required. Awake fibreoptic intubation should be considered in cases of difficult airway.

REFERENCES

- DeSanto LW. Laryngocele, laryngele mucocele, large sacculas, and laryngeal saccular cysts: a developmental spectrum. Laryngoscope 1974; 84:1291-6.
- Amin M, Maran AGD. The aetiology of laryngocele. Clin Otolaryngol Allied Sci 1988; 13:267-72.
- Stell PM, Maran AGD. Larynogocoele. J Laryngol Otol 1975; 89:915-24.
- Isaacson G, Sataloff RT. Bilateral laryngoceles in a young trumpet player: case report. Ear Nose Throat J 2000; 79:272-4.
- Harney M, Patil N, Walsh R, Brennan P, Walsh M. Laryngocele and squamous cell carcinoma of the larynx. J Laryngol Otol 2001; 115:590-2.
- Jahendran J, Sani A, Rajan P, Mann GS, Appoo B. Intravenous neck injections in a drug abuser resulting in infection of a laryngocele. Asian J Surg 2005; 28:41-4.
- Ovassapian A, Tuncbilek M, Weitzel EK, Joshi CW. Airway management in adult patients with deep neck infections: a case series and review of the literature. Anesth Analg 2005; 100:585-9.
- Martinez Devesa P, Ghufoor K, Lloyd S, Howard D. Endoscopic CO2 laser management of laryngocele. Laryngoscope 2002; 112:1426-30.