CASE REPORT

Laryngopycocele presenting with acute airway obstruction

J I Raine,¹ D Allin,² D Golding-Wood²

¹Emergency Department, Tweed Heads Hospital, Tweed Heads, New South Wales, Australia

²Department of ENT, Princess Royal University Hospital, Orpington, Kent, UK

Correspondence to

Dr Juliet Isobel Raine,
juliet.raine@doctors.org.uk

Accepted 1 July 2014

SUMMARY

A 66-year-old woman was transferred to the emergency department with a left-sided neck lump and fever. She was stridulous with reduced consciousness level and required immediate intubation to protect her airway. CT imaging showed a fluid-filled and air-filled lesion in the left side of the neck; the diagnosis was later confirmed as an infected laryngocoele. She underwent ultrasound guided drainage of the lesion with successful reduction of the neck lump. Following elective tracheostomy she was weaned from ventilation. Subsequent direct endoscopy and biopsy showed no evidence of malignancy or other lesion. She was discharged with permanent tracheostomy to be sustained until definitive surgical resection of the laryngocoele.

BACKGROUND

This case describes the management of a patient with an acutely threatened airway. It revises the importance of prioritising the maintenance of a patent airway during resuscitation; this may necessitate intubation or insertion of a surgical airway. We describe the use of ultrasound-guided drainage to rapidly give symptomatic relief of a fluid-filled lesion causing airway obstruction; this emphasises the value of radiological intervention in such cases. The underlying aetiology was a pus-filled laryngocoele (laryngopycocele). This is a rare phenomenon with, to our knowledge, 44 documented cases in the literature. Laryngopycoceles presenting with acute airway obstruction are rare still; this case report describes the 11th documented case of a laryngopycocele presenting in this way.^{1–9} The scarcity of such cases and their management makes this a valuable case to report.

CASE PRESENTATION

A 66-year-old woman with sudden onset neck swelling was referred by her general practitioner to the ENT (ear, nose and throat) team for urgent assessment in the accident and emergency department (A&E department) of a district general hospital. She elected not to attend A&E department that day. Two days later she was found unconscious by her husband and was transferred to the A&E department by ambulance.

On initial assessment she was severely obtunded with a Glasgow Coma Score of 6 and was noted to have 'noisy' breathing. She had a 5-day history of fever, shortness of breath and dysphagia; this was preceded by a 2-week history of a progressive lump in the left side of her neck.

Her medical history was unremarkable beyond chronic obstructive pulmonary disease (COPD) controlled with salbutamol and a history of lifelong heavy smoking.

INVESTIGATIONS

On admission she was acidotic (pH 6.95) and in hypercapnic respiratory failure. Her white cell count was $14.9 \times 10^9/L$ (89.6% neutrophils) with a C reactive protein of 37 mg/L.

A CT of the head, neck and chest on admission showed a fluid-filled and air-filled lesion in the left neck. It had two contiguous components: one within and one lateral to the larynx (figure 1).

DIFFERENTIAL DIAGNOSIS

The differential diagnosis for a neck mass is extensive; they may be subdivided into those of congenital, inflammatory and neoplastic origin. In order to refine the differential one must take into consideration the findings on history and examination. In our case the patient had an acute history and was septic on presentation. Neck masses which may progress rapidly and bring about sepsis include an infected congenital lesion of the neck, for example a branchial cleft cyst or laryngocoele.

Radiological investigation will be pivotal in determining the correct diagnosis of a neck mass. In our case the CT findings described above were consistent with a mixed laryngocoele.

Importantly, our patient was 66 years old. All patients with congenital neck masses which present in adulthood must be carefully investigated for underlying malignancy. Hence, the tissue histology and aspirate cytology were examined in the search for any malignant cells.

TREATMENT

The patient was intubated in the A&E department resuscitation room and transferred to the intensive care unit where she was treated with broad-spectrum antibiotics. The CT images were reviewed by a radiologist with special interest in neck anatomy to confirm the diagnosis of laryngocoele. On the second day of admission, once the safety for radiologically guided drainage had been established, she underwent ultrasound-guided aspiration which yielded 11 mL of pus. She had an elective tracheostomy and following this was successfully weaned from ventilation.

OUTCOME AND FOLLOW-UP

Direct endoscopy and biopsy showed no evidence of malignancy or other lesion. Imaging before

CrossMark

To cite: Raine JI, Allin D, Golding-Wood D. *BMJ Case Rep* Published online: [please include Day Month Year] doi:10.1136/bcr-2014-204102

Figure 1 Axial CT of the neck (contrast) showing laryngocoele (internal component: large arrow, external component: small arrow).

discharge showed diminution of the laryngocoele. She was discharged with a permanent tracheostomy with a view to definitive resection of the laryngocoele as an elective procedure at a tertiary centre.

DISCUSSION

This case reminds us of one of the fundamental principles of resuscitation: securing the threatened airway must be a priority. This is relevant for all clinicians, whether treating an obtunded patient on the ward or a patient with traumatic facial injury brought into the emergency department. An adequate airway is crucial for facilitating the perfusion of the vital organs, allowing oxygenation of the myocardium and prevention of secondary hypoxic damage.

This case details the infected 'laryngocoele' as a cause of upper airway obstruction with concurrent sepsis. A laryngocoele is an abnormal dilation of the laryngeal sacculae. The incidence of symptomatic laryngocoeles in the UK is approximately 1/2 500 000 per annum.¹⁰ Laryngocoeles are rare conditions, with only several hundred case reports documented in the world literature.

Laryngocoeles are defined according to anatomical location and constituents. If the extent remains within the larynx it is deemed to be 'internal' whereas 'external' laryngocoeles extend laterally through the thyrohyoid membrane. If internal and external extension is present then it is classed as 'combined'. Laryngocoeles are air filled when in continuity with the larynx, however, they may obstruct, usually secondary to infection or malignancy. A pus-filled laryngocoele becomes a laryngopyocoele; this occurs in an estimated 8% of cases.³

The aetiology of laryngocoeles was historically attributed to raised intraglottic pressure. There is infact little evidence for this; a review of 139 cases found only one such case.³ The patient in our case had a history of heavy smoking and COPD;

chronic cough may have contributed to the development of the laryngocoele. There is an association with laryngeal carcinoma and for this reason all cases must have careful surveillance for malignancy.^{11 12}

Laryngocoeles may present with dysphagia, dysphonia, neck pain or swelling. Internal laryngocoeles can extend superiorly into the aryepiglottic fold, invading the airway. Fluid-filled laryngocoeles may rapidly increase in size leading to acute upper airway obstruction. In these cases the priority is maintenance of a patent airway; emergency intubation or cricothyroidotomy may be required. It is best to forestall these scenarios where possible, hence early assessment of neck lumps with appropriate intervention by an ENT specialist is vital.

Radiological intervention may assist in the management of fluid-filled lesions. Ultrasound-guided drainage can rapidly alleviate symptoms. The merits of this intervention have been discussed in other case series.⁹ The definitive management of laryngocoeles is open or endoscopic surgical excision of the structural anomaly.

Learning points

- ▶ Patients with sudden onset neck swellings, particularly those who are symptomatic, should be reviewed by an ENT (ear, nose and throat) specialist on an urgent basis. This promotes timely alleviation of symptoms.
- ▶ Laryngocoeles are a rare but important cause of neck lumps. They may become infected and be complicated by coincidental presentation with sepsis. Malignancy must be excluded as an underlying aetiology.
- ▶ Laryngocoeles, like other neck masses, may present with acute upper airway obstruction; this case emphasises the importance of securing the threatened airway.
- ▶ Radiological assessment is integral in the diagnosis and management of neck lumps. Interventional radiology is a minimally invasive tool which may be used to rapidly alleviate symptomatic fluid-filled lesions.

Contributors DA and DG-W assisted with detail, direction and editing of manuscript. Dr Kathryn C assisted with obtaining patient consent.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Weissler MC, Fried MP, Kelly JH. Laryngopyocoele as a cause of airway obstruction. *Laryngoscope* 1985;95:1348–51.
- 2 Fredrickson KL, D'Angelo AJ. Internal laryngopyocoele presenting as acute airway obstruction. *Ear Nose Throat J* 2007;86:104–6.
- 3 Thawley SE, Bone RC. Laryngopyocoele. *Laryngoscope* 1973;83:362–8.
- 4 Vasileiadis I, Kapetanakis S, Petousis A, et al. Internal laryngopyocoele as a cause of acute airway obstruction: an extremely rare case and review of the literature. *Acta Otorhinolaryngol Ital* 2012;32:58–62.
- 5 Illum P, Nehen AM. Laryngopyocoele with a report of two cases. *J Laryngol Otol* 1980;94:211–18.
- 6 Andreou Z, Randhawa PS, O'Flynn P, et al. Endoscopic management of an internal laryngopyocoele presenting with acute airway obstruction. *Case Rep Surg* 2011;2011:873613.
- 7 Özcan C, Vayisoglu Y, Güner N, et al. External laryngopyocoele: a rare cause of upper airway obstruction. *J Craniofac Surg* 2010;21:2022–4.

- 8 Fraser L, Pittore B, Frampton S, *et al.* Laryngeal debridement: an alternative treatment for a laryngopyoceles presenting with severe airway obstruction. *Acta Otorhinolaryngol Ital* 2011;31:113–17.
- 9 Mace AT, Ravichandran S, Dewar G, *et al.* Laryngopyoceles: simple management of an acute airway crisis. *J Laryngol Otol* 2009;123:248–9.
- 10 Stell PM, Maran AG. Laryngocoele. *J Laryngol Otol* 1975;89:915–24.
- 11 Close LG, Merkel M, Burns DK, *et al.* Asymptomatic laryngocele: incidence and association with laryngeal cancer. *Ann Otol Rhinol Laryngol* 1987;96:393–9.
- 12 Harvey RT, Ibrahim H, Yousem DM, *et al.* Radiologic findings in a carcinoma-associated laryngocele. *Ann Otol Rhinol Laryngol* 1996;105:405–8.

Copyright 2014 BMJ Publishing Group. All rights reserved. For permission to reuse any of this content visit <http://group.bmj.com/group/rights-licensing/permissions>.

BMJ Case Report Fellows may re-use this article for personal use and teaching without any further permission.

Become a Fellow of BMJ Case Reports today and you can:

- Submit as many cases as you like
- Enjoy fast sympathetic peer review and rapid publication of accepted articles
- Access all the published articles
- Re-use any of the published material for personal use and teaching without further permission

For information on Institutional Fellowships contact consortiasales@bmjgroup.com

Visit casereports.bmj.com for more articles like this and to become a Fellow