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Traumatic laryngocoele

T. E. MITCHELL, M.A., F.R.C.S., J. M. PICKLES, F.R.C.S.

Abstract
This is the first reported case of a laryngocoele developing after laryngeal trauma. A 26-year-old man sustained a shotgun injury to his larynx. A large number of shotgun pellets was removed from his left vestibular fold. He subsequently developed a left-sided laryngocoele, probably due to fibrosis around the neck of the saccule. The laryngocoele was removed by an external approach.

Key words: Laryngeal diseases; Wounds, gunshot

Introduction

A laryngocoele is a dilatation of the laryngeal saccule above the superior margin of the thyroid cartilage. The saccule is a narrow blind pouch arising from the anterior end of the laryngeal ventricle, which extends superiorly between the vestibular fold medially and the thyroid cartilage laterally. This structure is very variable in size; it may be absent or may extend for 15 mm or more into the aryepiglottic fold (Canalis *et al.*, 1977). A laryngocoele, like the saccule from which it is derived, is lined with pseudostratified columnar epithelium containing numerous goblet cells.

Laryngocoeles may be internal, external or mixed. Internal laryngocoeles extend within the vestibular fold and may present in the vallecula. External laryngocoeles protrude through the thyrohyoid membrane adjacent to the superior laryngeal neurovascular bundle and are connected to the laryngeal ventricle by a nondilated segment of the intralaryngeal saccule. Mixed or combined laryngocoeles include both external and internal dilatations of the saccule.

Laryngocoeles communicating with the laryngeal lumen contain air. However, they may become distended with mucus, in which case some authors have called them laryngomucocoeles or saccular cysts (De Santo, 1974; Holinger *et al.*, 1978). Szwarc and Kashima (1997) suggested that enlarged saccules should only be considered to be laryngocoeles if they are symptomatic.

Case report

A 26-year-old man was admitted under the care of the ENT Department, Luton and Dunstable Hospital having sustained shotgun injuries to his right arm and neck. There was an area of skin and soft tissue loss on the lateral aspect of the right forearm. There were two small entry wounds in the right submental region with surgical emphysema on the opposite side of the neck. A lateral neck radiograph showed a large number of shotgun pellets in the region of his larynx and the presence of retropharyngeal air (Figure 1).

A tracheostomy was performed and his neck was explored through a high collar incision. Shotgun pellets were removed from the soft tissues of the neck. There was a vertical fracture of the thyroid cartilage just to the left of the midline. A laryngofissure was performed exposing a cavity in the left vestibular fold containing a large number of shotgun pellets together with pieces of fabric and feathers from the right sleeve of the patient's jacket. These

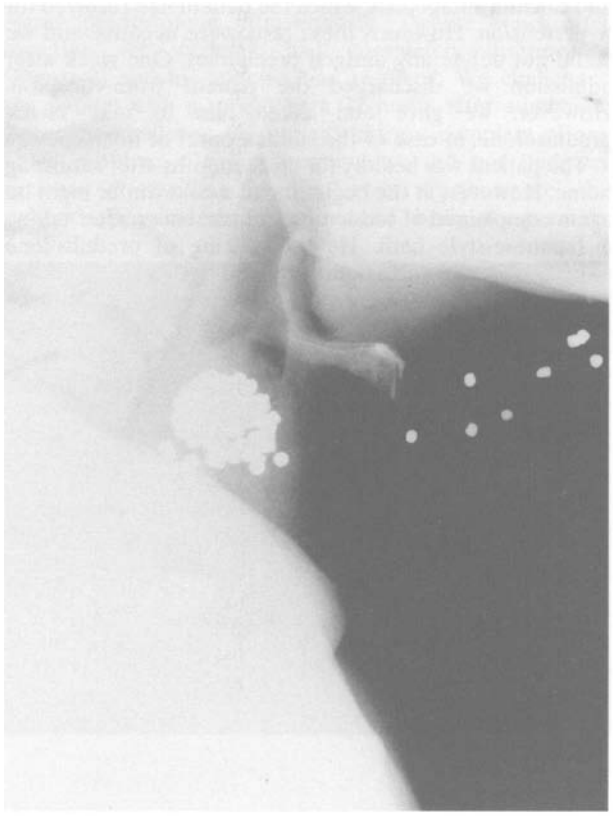

FIG. 1
Lateral neck radiograph after initial injury.

From the Department of Otolaryngology, Luton and Dunstable Hospital, Luton, UK.
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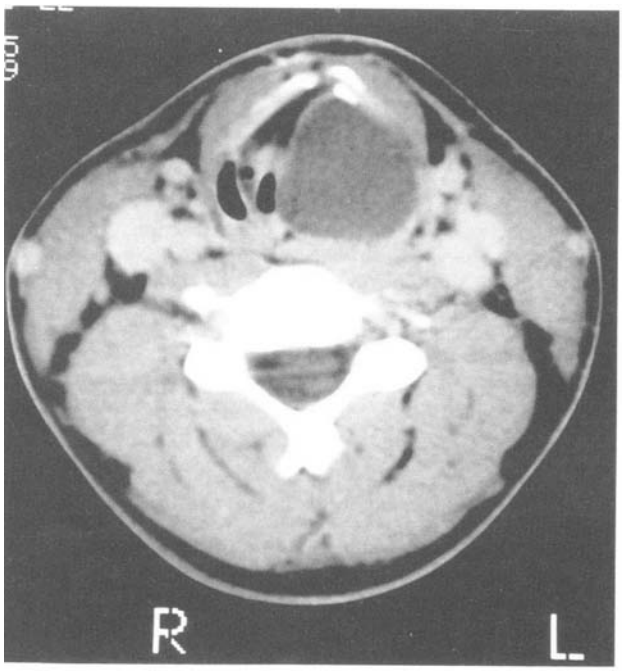

FIG. 2
CT scan showing left fluid-filled internal laryngocoele.

were removed and the wound was drained and closed. He was successfully decannulated on the 13th post-operative day and discharged from hospital the following day. At that time, he had no airway compromise and a reasonable voice. Examination of his larynx showed that his left vocal fold was immobile.

He failed to attend for outpatient review. He was not seen again until three years later when, by chance, whilst visiting a relative in the hospital, he encountered one of the authors (JMP) in a corridor. He complained of increasing hoarseness, difficulty breathing and a feeling of a lump on the left side of his throat. He was mildly stridulous at rest. Indirect laryngoscopy showed a large cystic swelling in the left supraglottis. A computed tomography (CT) scan of his larynx showed a 3 cm diameter thin-walled low density swelling in the left side of the larynx consistent with a fluid-filled internal laryngocoele, with significant narrowing of the airway (Figure 2).

A tracheostomy was performed and his larynx was explored under general anaesthetic. The upper half of the left thyroid ala was removed to allow adequate exposure and removal of the cystic swelling, which was found to communicate with the left laryngeal ventricle. Histological examination showed the cyst to be lined by respiratory epithelium, thus confirming it to be a laryngocoele rather than a pseudocyst.

He made an uneventful recovery and was successfully decannulated on the seventh post-operative day. When reviewed two weeks later, he had no airway compromise and his voice had improved considerably. Indirect laryngoscopy revealed some thickening of his left vestibular fold but otherwise his larynx appeared normal and both vocal folds were fully mobile.

Discussion

The incidence of symptomatic laryngocoeles is reported to be approximately one per 2.5 million population per year (Maran, 1997). They are five to seven times more common in men (Canalis *et al.*, 1977; Maran, 1997). They occur equally on the left and right (Melnick and Sumerson, 1987). Laryngocoeles present most commonly in the fifth and sixth decade of life, although they may occur in neonates (Chu *et al.*, 1994). However, increasing use of CT scanning and magnetic resonance imaging (MRI) to image the neck, suggests that the prevalence of asymptomatic laryngocoeles is between 4.2 per cent (Lindell *et al.*, 1978) and 12.5 per cent (Close *et al.*, 1987). This may overestimate the true prevalence as these groups were being investigated for head and neck disease. Canalis *et al.* (1977) found that 70 per cent of symptomatic laryngocoeles have an external component as compared with only 18 per cent of asymptomatic laryngocoeles.

There is an association between laryngocoeles and other laryngeal pathology, including laryngeal papillomatosis and carcinoma of the larynx. Celin *et al* (1991) identified laryngocoeles in 4.9 per cent of patients with laryngeal cancer, whilst Close *et al.* (1987) found laryngocoeles in 28.8 per cent of their series of 59 patients with laryngeal cancer. Micheau *et al.* (1978) identified laryngocoeles in 18 per cent of laryngectomy specimens removed for laryngeal cancer. From the alternative perspective, 45 per cent of 38 patients with laryngocoeles in one series (Close *et al.*, 1987) and 49 per cent of 87 patients in another series (Lindell *et al.*, 1978) also had carcinoma of the larynx.

It is likely that carcinoma of the larynx predisposes to laryngocoele formation rather than the reverse. In their series, Close *et al.* (1987) followed 18 of 21 patients who had a laryngocoele but did not have carcinoma of the larynx for between 10 and 36 months (mean – 17 months). None of them developed laryngeal cancer.

The pathogenesis of laryngocoeles is unknown. It has been suggested that a long saccule may predispose to the development of a laryngocoele (Canalis *et al.*, 1977). It is proposed that dilatation of the saccule occurs as a consequence of increased intralaryngeal pressure. Certain activities, such as glass blowing and playing wind instruments, are reported to be associated with laryngocoele formation (Holinger *et al.*, 1978; Harvey *et al.*, 1996; Szwarc and Kashima, 1997). However, a review of the literature shows that laryngocoeles have been reported in only four patients who had indulged in such activities (Maran, 1997).

It has been suggested that laryngeal tumours predispose to laryngocoele formation by blocking the neck of the saccule (Murray *et al.*, 1994; Drozd *et al.*, 1996). In 53 per cent of laryngectomy specimens removed for laryngeal carcinoma with an associated laryngocoele, invasive carcinoma was seen in the epithelium of the laryngocoele (Micheau *et al.*, 1978). Similarly, Close *et al.* (1987) found carcinoma to extend into the ventricle in six of 11 (54 per cent) laryngectomy specimens with laryngocoeles. However, of the 17 patients in their series with laryngeal cancer and laryngocoeles eight had bilateral laryngocoeles and seven had laryngocoeles on the opposite side to the tumour only. They propose that laryngocoeles develop as a result of raised intralaryngeal pressure, which may be caused by a combination of partial obstruction of the laryngeal airway, increased effort of phonation and excessive coughing associated with difficulty in clearing secretions (Close *et al.*, 1987).

It is likely that our patient developed a laryngocoele as a consequence of obstruction of the neck of the saccule caused by fibrosis. At the initial exploration of his larynx, there was a cavity within the left vestibular fold containing shotgun pellets and material from his jacket. It is not possible to determine whether the original injury or the surgical exploration was responsible for subsequent laryngocoele formation. However, we have not identified any reports in the literature of laryngocoeles developing after laryngeal surgery.

Internal laryngocoeles typically present with hoarseness or altered voice quality, dyspnoea, cough or the sensation of a lump in the throat. Indirect laryngoscopy reveals a cystic swelling of the vestibular fold. External laryngocoeles present as a soft reducible neck mass, overlying the thyrohyoid membrane, lateral to the thyroid cartilage and anterior to the sternomastoid muscle. They may enlarge with increased intralaryngeal pressure and on compression, there may be a gurgling sound, sometimes called Bryan's sign (Thawley and Bone, 1973). Ten per cent of laryngocoeles are infected at presentation. Increasingly, asymptomatic laryngocoeles are identified during the investigation of other head and neck pathology.

Plain radiography used to be the investigation of choice. It demonstrates air-filled laryngocoeles well but may not detect fluid-filled laryngocoeles, which are much better demonstrated by CT scanning. Close *et al.* (1987) found CT scanning to be more sensitive at detecting laryngocoeles than the presence of symptoms or clinical examination. MRI also demonstrates laryngocoeles well, and can distinguish between tumour and mucus in an associated laryngocoele (Celin *et al.*, 1991).

Almost half of the patients presenting with a laryngocoele will have a carcinoma of the larynx (Lindell *et al.*, 1978; Close *et al.*, 1987). Therefore, all patients with a symptomatic laryngocoele should undergo a microlaryngoscopy including close examination of the ventricles of the larynx and biopsies should be taken as indicated. Close *et al.* (1987) recommend that patients with asymptomatic laryngocoeles detected incidentally by CT scanning should be followed with regular outpatient laryngeal examination. None of the patients with asymptomatic laryngocoeles that they followed became symptomatic or developed laryngeal cancer.

The surgical management of laryngocoeles has traditionally employed an external approach. The upper half of the thyroid ala is removed or fractured downwards and subsequently replaced. The laryngocoele is excised and its neck oversewn (Maran, 1997). This approach is recommended for large internal laryngocoeles or those with an external component (Szwarc and Kashima, 1997). Small internal laryngocoeles may be uncapped using a CO_2 laser. Recurrence is prevented either as a consequence of marsupialization or fibrosis (Myssiorek and Persky, 1989).

Small internal laryngocoeles may be treated by endoscopic laser excision of the vestibular fold (Szwarc and Kashima, 1997). The laryngocoele is excised completely with reduced risk to the superior laryngeal nerve as compared to the external approach.

Conclusion

Laryngocoeles are uncommon. They are frequently asymptomatic. They may be associated with other laryngeal pathology and may herald a carcinoma of the larynx. This is the first reported case of a laryngocoele developing after laryngeal trauma.

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Address for correspondence:
Mr T. E. Mitchell,
ENT Department,
Luton and Dunstable Hospital,
Lewsey Road,
Luton LU4 0DZ.

Fax: 01582 497031