Unusual presentation of more common disease/injury

Internal laryngocoele and referred otalgia

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Summary

A 74-year-old woman presented with severe right ear pain associated with bleeding of 2 months' duration and vertigo. She was otherwise fit and well apart from arthritis. On examination there was dried blood overlying the right ear drum and very mild inflammation of the canal but no obvious acute infection. The dried blood was removed in subsequent clinic visits. The ear drum was intact and there was no evidence of infection. However, the patient still had persistent pain in the right ear. A diagnosis of referred otalgia was made. Nose, throat, neck, temporomandibular joint, oral examination and flexible nasoendoscopic examination were normal. She underwent MRI of her neck which revealed a right supraglottic mass. Laryngoscopy was performed. Intra-operative findings revealed a smooth right supraglottic mass, which was thought to be an internal laryngocoele. The laryngocoele was de-roofed. The histology specimen showed no evidence of neoplasia.

BACKGROUND

Otalgia is defined as ear pain. Two separate and distinct types of otalgia exist: pain that originates within the ear is primary otalgia, while pain that originates outside the ear is secondary or referred otalgia.

Many remote anatomical sites share dual innervation with the ear, and noxious stimuli to these areas may be perceived as otogenic pain. By definition, referred otalgia is the sensation of ear pain originating from a source outside the

ear. To better understand referred otalgia, the physician must first be familiar with the anatomical distribution of nerves associated with the ear. Irritation of these nerves, as well as irritation of distant branches of these nerves, can cause the perception of pain within the ear (figure 1).

This case is described to increase awareness among ENT doctors in training and general practitioners of causes of referred otalgia and also to further emphasise the fact that referred otalgia is a separate entity. This case is also

Figure 1 Chart for the diagnosis of referred otalgia.

Figure 2 Internal laryngocoele during the operation

presented for its rarity. As far as we are aware, no case has been reported in the literature describing an internal laryngocoele as a cause of referred otalgia.

CASE PRESENTATION

A 74-year-old woman presented with severe right ear pain associated with bleeding of 2 months' duration and vertigo. She did not have any hearing problems. She was otherwise fit and well apart from arthritis. There was no history of swallowing problems or voice change. She had smoked about 10 cigarettes a day for the last 30 years. There was no history of alcohol intake.

On examination there was dried blood overlying the right ear drum and very mild inflammation of the canal, but no obvious acute infection. The dried blood was too painful to be removed. The patient was given β -methasone ear drops but despite this, an attempt to remove the dried blood a week later was still painful. She was then put on sodium bicarbonate ear drops for 2 weeks after which the dried blood was removed. The ear drum was intact and there was no evidence of infection. However, the patient still had persistent pain in the right ear. A diagnosis of referred otalgia was made and a search for a distant cause was initiated. Neck palpation revealed no abnormal neck swelling. There was no problem with her temporo-mandibular joints (TMJs) and no oral cause was identified. Flexible nasoendoscopic examination revealed a normal-looking pharynx and larynx.

INVESTIGATIONS

As the patient was a smoker with increased risk of occult malignancy in the head and neck, she underwent MRI of her neck for other possible causes.

MRI revealed a right supraglottic mass. Degenerative changes in the cervical spine at multiple levels were seen, but no spinal canal compromise was noted. The diagnosis was a possible right supraglottic mass.

DIFFERENTIAL DIAGNOSIS

- ▶ Pharyngitis
- ▶ Tonsillitis
- ▶ Eustachian tube dysfunction
- ▶ Pain from the teeth
- ▶ Trigeminal neuralgia
- ▶ Sinus pathology
- ▶ TMJ disease
- ▶ Parotid gland disease
- ▶ Pain from cervical spine disease
- ▶ Carcinoma of the posterior third of the tongue
- ▶ Carcinoma of the pyriform fossa or the oesophagus
- ▶ Laryngopyocoele
- ▶ Reflux disease
- ▶ Psychogenic otalgia (when no cause for ear pain can be found, suggesting a functional origin).

TREATMENT

Pharyngo-laryngoscopy and examination of the postnasal space under general anaesthetic were performed.

Intra-operative findings revealed a smooth right supraglottic mass with normal overlying mucosa, which was thought to be an internal laryngocoele (figure 2). The laryngocoele was de-roofed, thick mucus suctioned out and specimens sent for histology. Examination of the postnasal space and biopsy of the posterior pad of the right eustachian tube were also carried out.

Figure 3 Internal laryngocoele before operation.

Systematic approach to diagnose referred otalgia :

Figure 4 IMRI of internal laryngocoele.

OUTCOME AND FOLLOW-UP

Histology from the right supraglottic mass showed that it was covered by stratified squamous and columnar epithelium. Minor salivary gland and striated muscle were present over the deep aspect. There was no evidence of neoplasia. The right postnasal biopsy specimen was covered with cili-

ated columnar epithelium. Seromucinous glands were present in the stroma, showing large areas of oncocytic metaplasia in places. There was also no evidence of neoplasia. The patient was subsequently seen again in the clinic 3 months later and flexible nasoendoscopy was normal. She also reported that she had no further pain in her right ear.

This confirmed that the internal laryngocoele was indeed the cause of her referred otalgia and she was discharged from the clinic.

DISCUSSION

A laryngocoele is an abnormal enlargement of the saccule of a laryngeal ventricle. Laryngocoeles are generally filled with air, and are termed internal when limited to the interior of the larynx and external when they protrude laterally into the neck.¹

Three types of laryngocoeles have been described: internal, external and combined. The internal laryngocoele remains within the larynx, whereas the external type extends through the thyrohyoid membrane. Combined laryngocoeles, which consist of both internal and external elements, are the most common of the three types.²

Dental disorders are the most frequent causes of secondary otalgia presenting to the ENT clinic, and may account for up to 50% of referred otalgia. TMJ dysfunction syndrome is the most common dental cause of referred otalgia,³ is twice as common among women than among men and has environmental, physiological and behavioural causes.⁴ In a study of 123 patients with ear pain, the most frequent cause of referred otalgia in one group (n=72) was TMJ dysfunction (46%), while the most common cervical spine finding in a second group (n=51) was cervical spine degenerative disease (88%).⁵

The cause of laryngocoeles is felt to be either a congenital enlargement of the saccule or acquired enlargement caused by increased, sustained intralaryngeal pressure (eg, in a trumpet player). Wind and brass players are at increased risk for laryngocoeles; one report showed laryngocoeles in 52 (56%) of 93 band members studied.¹

The incidence of carcinoma in patients presenting with a laryngocoele is 2–18%. Flexible nasoendoscopy is probably the gold standard for visualisation of the pharynx and larynx in the clinical setting. This case shows that it could miss supraglottic pathology. The patient initially underwent nasoendoscopy by an ENT registrar and then later by a consultant (before her laryngoscopy under general anaesthesia).

A laryngocoele is gravity dependant, and as flexible laryngoscopy was carried out with the patient in a sitting position, the laryngocoele was not seen when this investigation was first carried out. However, when a rigid laryngoscopy was carried out with the patient in a supine position the laryngocoele fell into the field of view and was seen (figure 3).

A CT scan with contrast media would have been the first choice of investigation with a working diagnosis of laryngocoele. However, ear pain may also be the first sign of a head and neck malignancy.⁶ In view of the patient being a chronic smoker, a soft tissue malignancy had to be ruled out as soon as possible, and so we requested an MRI scan instead of a CT scan (figure 4).

Learning points

- Identifying the cause of referred otalgia is challenging.
- Clinical examination may be normal.
- Multiple repeat nasoendoscopies may be negative, and so imaging may be required.
- Laryngocoele should be included as one of the possible causes of referred otalgia.

Competing interests None.

Patient consent Obtained.

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