

CASE REPORT

Laryngeal debridement: an alternative treatment for a laryngopyoceles presenting with severe airway obstruction

Debridement laringeo: un trattamento alternativo per un laringocele che si presenta con ostruzione severa delle vie aeree

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SUMMARY

The laryngocele is an abnormal saccular dilatation of the ventricle of Morgagni, which maintains its communication with the laryngeal vestibule. Three types of laryngoceles have been described: internal, external, and combined or mixed in relation to the position of the sac with respect to the thyrohyoid membrane. If the laryngocele becomes obstructed and infected it leads to the so-called laryngopyoceles which, although a rare disease (8% of laryngoceles), can become an emergency causing severe airway obstruction needing urgent management, even tracheostomy. An alternative method is presented of emergency management of an internal laryngopyoceles causing severe airway obstruction using a laryngeal microdebrider and avoiding tracheostomy.

KEY WORDS: Larynx • Laryngocele • Laryngopyoceles • Laryngeal debrider • Airway obstruction

RIASSUNTO

Il laringocele è una anomala, sacculare dilatazione del ventricolo del Morgagni, il quale mantiene la sua comunicazione con il vestibolo laringeo. Sono stati descritti tre tipi di laringoceli: interno, esterno e combinato o misto in relazione alla posizione del sacco rispetto alla membrana tiro-ioidea. Se un laringocele si occlude e successivamente si infetta, viene chiamato laringopiocele, il quale, nonostante sia una rara malattia (8% dei laringoceli), può diventare un'emergenza determinando una severa ostruzione delle vie aeree. Spesso necessita di un trattamento d'urgenza, a volte si rende anche necessaria la tracheotomia. Presentiamo un metodo di emergenza alternativo nel trattamento di un laringopiocele interno con ostruzione delle vie aeree, utilizzando il microdebrider laringeo ed evitando l'esecuzione della tracheotomia.

PAROLE CHIAVE: Laringe • Laringocele • Laringopiocele • Debrider laringeo • Ostruzione vie aeree

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Introduction

The term laryngocele was first introduced by Virchow, in 1867¹, to describe an abnormal dilatation of the appendix of the laryngeal ventricle of Morgagni, which maintains its communication with the laryngeal lumen. It can be present at any age, but is most common in the sixth decade of life²⁻⁴. Three types of laryngoceles have been described: internal, external, and combined or mixed in relation to the position of the sac with respect to the thyrohyoid membrane. The presenting complaint frequently includes dysphonia, dyspnoea, sore throat, globus pharyngeus, or, in severe cases, airway obstruction (presentation depends on classification, size and possible complications). If the neck of the laryngocele becomes obstructed (causes include tumours and chronic inflammation of the larynx),

the mucus produced by the mucous glands of the lining epithelium, can accumulate leading to a laryngomucocele or cystic laryngocele (virtually indistinguishable from a saccular cyst). The mucocele can secondarily become infected and it is thought that 8% do at some time⁵.

The treatment of a laryngocele is only indicated if it is symptomatic or has developed into a laryngomucocele or laryngopyoceles. Various methods have been described including complete excision of the lesion by external / endoscopic / or a combined approach. Instrumentation has included cold steel and more recently the Carbon Dioxide (CO₂) laser resection or marsupialisation^{3,4,6}

A case is described here of a laryngopyoceles presenting with severe airway obstruction treated as an emergency by endoscopic marsupialisation with a laryngeal micro-

debrider avoiding tracheostomy, to our knowledge a novel application of this instrument. The procedure was straightforward, with minimal bleeding, optimal intra-operative control of the lesion and haemostasis with prompt post-operative recovery.

Case report

A 66-year-old female presented to our department as an emergency case with a one-day history of dyspnoea and dysphagia, and a few hours of stridor. Prior to this presentation, the patient had ceased smoking for one month, having previously smoked 20 cigarettes a day, for more than 20 years. She developed a cough one week after having stopped smoking, and became hoarse following a coughing fit. Progression of her symptoms included a one-week history of right neck swelling and odynophagia, with 24 hours of dyspnoea. On presentation, the patient was stridulous and pyrexial, with an oedematous swelling over levels II and III on the right hand side of the neck. An oral examination was normal, but fiberoptic nasopharyngolaryngoscopy revealed a well circumscribed erythematous swelling of the whole hemilarynx and pyriform sinus with intact mucosa, severely obstructing her airway (Fig. 1). Initial management with intravenous steroids (dexamethasone 4 mg, 3 times daily), adrenaline nebulisers (1:5000) and intravenous broad spectrum antibiotics (cefuroxime 1.5 g, 3 times daily, metronidazole 500 mg, 3 times daily) had little effect on the patient clinically, and she became progressively exhausted with increased respiratory effort.

Although the definitive diagnosis was unclear, because a computerised tomography (CT) scan was not requested due to the unsecured and deteriorating airway, the patient was taken urgently to the operating theatre to attempt to

secure her airway. A tracheostomy was considered a possible option. However, with the significant risk that there might be an underlying ventricular malignancy leading to a possible future peristomal recurrence^{7,8}, experienced anaesthetic and otorhinolaryngology consultants awaited her with the intention of performing an awake fiberoptic nasal examination and possible intubation. Flexible endoscopy was performed after nasal anaesthesia (with 10% cocaine paste) and 6 ml of 4% lidocaine was instilled under direct vision into the pharynx, larynx and trachea. A size 5 microlaryngeal tube was rail-roaded over the scope and the patient was placed under general anaesthesia.

Surgical technique

An intra-operative workup with 0-45 degree scopes (Storz, Tutlingen, Germany) was performed along with palpation of the hemi-larynx showing a tender but fluctuating sub-mucosal swelling. In order to confirm the fluid content of the lesion, a spinal needle connected to a 10 ml syringe was used to aspirate some contents of the sac. The purulent component was confirmed and part of the pus was sent for microbiologic assessment.

The CO₂ laser was not immediately available in the emergency theatre so the surgeon opted to use a microdebrider [XPS 3000-Powered ENT System-Straightshot Magnum II Medtronic (Xomed, Minneapolis, USA) microdebrider (27.5 cm) with Tricut™ angle-tip 4 mm blade (Jacksonville, FL, USA)]. The lesion was extensively marsupialised with the debrider under endoscopic view (Fig. 2) and multiple biopsies performed. Biopsies were sent for histological evaluation from the abscess cavity and medial wall of the false vocal cord. At the end of the procedure, with a patent airway, the endo-tracheal tube was changed to a size seven and the patient was kept sedated and ven-



Fig. 1. Intra-operative view of erythematous swelling of right false vocal cord.

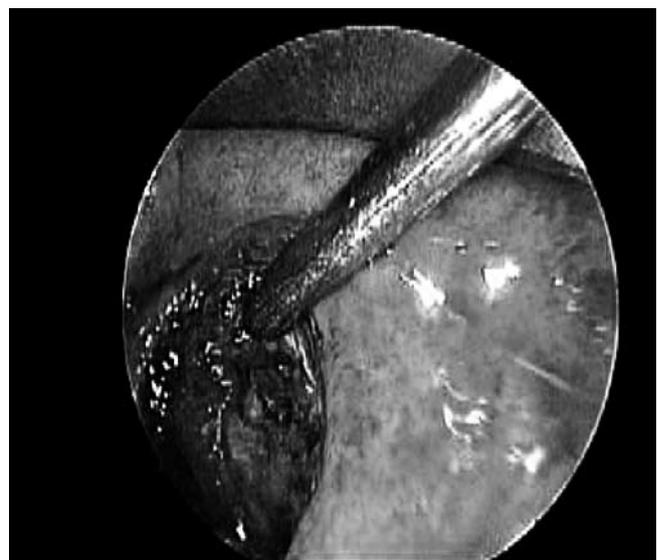


Fig. 2. Cavity following marsupialisation with the laryngeal microdebrider is clearly visible.

tilated in the Intensive Care Unit (ICU) for 28 hours with continued steroid and antibiotics. At this point, the patient had a repeat microlaryngoscopy, followed by successful extubation and subsequent discharge.

The histology results from the initial procedure revealed mildly inflamed fibrous connective tissue containing a few benign respiratory-type glands. However, the false vocal cord mucosa showed nuclear atypia with increased mitotic activity consistent with carcinoma *in situ*, with no stromal invasion.

On review, in the outpatient clinic, four weeks post-operatively, the mucosa was already well-healed, with no granulation tissue (Figs. 3, 4). Although clinical examination did not reveal any remaining lesions involving the false vocal cord, a completion CO₂ laser ventriculotomy was subsequently performed in view of the presence of the carcinoma *in situ*. The histopathology results confirmed



Fig. 3. 4 weeks post-operatively on adduction.

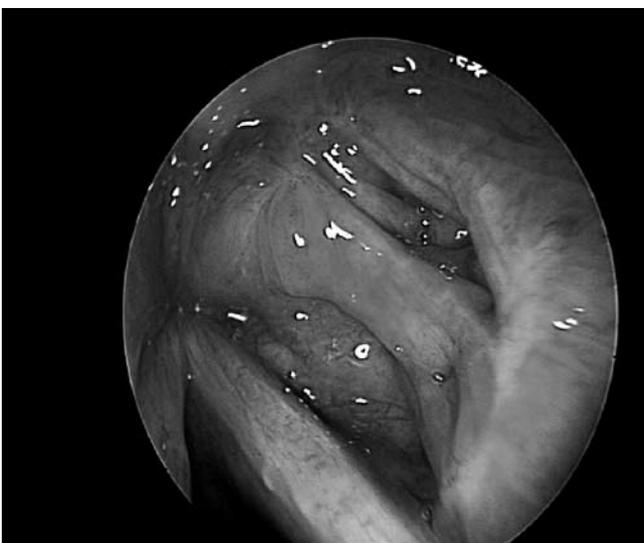


Fig. 4. 6 weeks after primary surgery at microlaryngoscopy.

that the patient was free of tumour. Close follow-up is planned.

Discussion

A laryngocele is an abnormal saccular dilatation of the appendix of the laryngeal ventricle of Morgagni, which maintains its communication with the laryngeal lumen. It is much more frequent in men than in women, having been described in the literature as between 5 to 7 times more frequent in males²⁻⁴. The condition usually presents in the sixth decade of life²⁻⁴. Unilateral presentation is more common than bilateral²⁻⁴. It is an unusual clinical entity and the majority of laryngoceles are diagnosed incidentally¹. There have been several hypotheses regarding the aetiology of laryngoceles. Some suggest that they are atavistic remnants from apes⁵, others that one has a congenital predisposition (eg. congenital long saccule) and that they are augmented by factors which increase intralaryngeal pressure, such as coughing (e.g. emphysema and/or chronic bronchitis patients)^{9,10}, straining, singing, playing wind instruments and glass blowing^{11,12}. Three types of laryngoceles have been described: internal, external and combined or mixed laryngocele¹³ which refer to the anatomical extension of the sac. The internal laryngocele is confined to the paraglottic space, medial to the thyroid cartilage and thyro-hyoid membrane. The external laryngocele is a type of laryngocele which extends and dissects superiorly through the thyrohyoid membrane. It is intimately associated with the superior laryngeal nerve. A combined or mixed laryngocele includes both internal and external components of the laryngocele.

In our case, the laryngocele was limited to the paraglottic space and we can postulate that the coughing experienced by our patient, upon cessation of smoking, was likely the causative degenerative factor in this case. Furthermore, the presence of a carcinoma *in situ* in the false vocal cord, as seen in this patient, could be considered as the obstructive factor responsible for retention of the fluid.

Laryngoceles may be associated with laryngeal malignancies, and supra-glottic laryngeal tumours are the most common¹⁻¹⁴, as in our case. The incidence, described in the literature, of laryngoceles in patients with laryngeal carcinoma is variable. Micheau et al. found 18% of laryngoceles in 546 larynx specimens after total laryngectomy¹⁵; Celin et al. reported an incidence of laryngoceles concurrent with squamous cell carcinoma of the larynx: 4.9% to 28.8%¹⁶.

The symptoms depend upon the type and size of the laryngocele. In the majority of cases, a laryngocele is asymptomatic and produces symptoms only as it enlarges or when it becomes infected¹⁷. The internal laryngocele often presents as a soft, more or less spherical sac of the mucosa arising from the ventricle or extending into the supraglottic region and gives subjective symptoms such

as dysphonia, dyspnoea, sore throat and globus pharyngeus with associated discomfort in the neck^{3,4,18}. It can, rarely, cause airway obstruction which requires emergency treatment^{3,9,19,20}. Normally, the external laryngocele is asymptomatic, but it presents as a lateral neck mass which is either ovoid or round and is, typically, soft, elastic, mobile, conducts vibration during phonation, is painless and covered by normal skin. It increases in size during the Valsalva manoeuvre and is reducible. The mixed laryngocele presents symptoms and signs of both entities¹⁰⁻¹⁸. When the connection between the laryngocele and the laryngeal lumen becomes occluded, as a result of inflammation, infection, and tumours, mucus can accumulate and a mucocele develop. A secondarily infected mucocele is called a laryngopyocele and it is thought that 8% do so at some time⁵.

A CT scan of the neck is the most important examination for correct diagnosis, moreover it permits an evaluation of the relationship between the laryngeal structures and the laryngocele²¹. The CT scan may show the simultaneous presence of other laryngeal lesions that are often associated with a laryngocele²², in particular laryngeal carcinomas^{1,14,17,23}. In our case, as a result of the severity of airway involvement, imaging was not possible before transfer to the theatre, so a vestibular carcinoma could not be excluded.

The management of a laryngocele, when symptomatic, is complete excision or marsupialisation that can be performed by endoscopic and/or external or combined approach. The choice of the approach depends on the type and size of the laryngocele. In cases of large or external laryngocele, an external approach is indicated and several techniques have been described^{24,25}; while the endoscopic approach with CO₂ laser is currently the preferred method of treatment for an internal laryngocele⁴. Treatment of the mixed laryngocele is still controversial, several authors prefer an external approach⁴⁻¹⁰, but a combined approach²⁶ and fully internal³ approach have been championed.

Endoscopic management of the laryngocele with the use of the CO₂ laser is increasing in popularity due to the advantage of a short hospital stay, good haemostasis and avoidance of an external scar^{3,4,6}. However, theatre speci-

fication requirements, limited equipment due to costs, and necessary staff training, for laser surgery, means that it may not always be rapidly available. In our case, we used the laryngeal micro-debrider as an alternative, readily available method to classical cold steel and laser.

This device was adapted from arthroscopic equipment to aid the removal of bony and soft tissue. In the field of Otolaryngology, it was used for the first time, in 1992²⁷, for endoscopic sinus surgery initially by using the arthroscopic debriders. With the success and dissemination of endoscopic sinus surgery, debrider equipment is commonly available, which can be readily converted for the treatment of laryngeal pathology with the use of additional blades. They are often used for laryngeal papillomatosis and laryngeal tumour debulking²⁸⁻³⁰, as in our Department. Given the association of malignancy with laryngoceles, all the tissue removed, by the debrider, can be easily collected, in a distal net, and sent for histological examination [Bard® Mucous Specimen Trap 40 cc (C.R. Bard, Inc. Covington, GA, USA)], or multiple biopsies performed.

Our patient had an uneventful outcome with fast, complete, epithelialisation and no granulation tissue, which is occasionally observed, after laser resection. A common preference often expressed for laser treatment is the associated improved haemostasis, but we only had minimal bleeding which was adequately controlled with topical adrenaline (1:5000 on neuropathies).

Conclusions

The Authors present a case of acute airway obstruction, caused by an internal laryngopyocele, successfully treated endoscopically with a laryngeal microdebrider, avoiding tracheostomy. As this equipment may be more readily available particularly in peripheral hospitals, it is a viable alternative to laser excision for marsupialisation of laryngoceles.

Research Integrity

Patient's consent was obtained. The Authors state they have no conflict of interest.

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