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CASE REPORT

Combined Laryngocele and External Approach

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ABSTRACT

Laryngocele, a dilation of the laryngeal saccule, is an uncommon and benign air-filled lesion that expands upwards into the ventricle of Morgagni. A unilateral growth within the larynx that is in communication with the laryngeal lumen typically signals its presence. The exact cause of the disorder remains uncertain, although there are three main theories which suggest congenital reasons, increased pressure in the larynx, or mechanical obstruction of the ventricle of Morgagni. The classification for laryngoceles is based upon their location with respect to the thyrohyoid membrane, and they may be internal, external, or combined. A laryngocele, along with a gradually enlarging submandibular mass, was located in the region from below the hyoid bone to the anterior sternocleidomastoid muscle on the right side of the neck. The diagnosis of a laryngocele may be confirmed through clinical examination, endoscopic investigation and imaging tests. For cases of combined laryngocele, we recommend its removal using an external approach, which not only ensures safety and accuracy but also allows for a complete removal of the laryngocele. Additionally, this approach guarantees that the surgical intervention is carried out with maximum precision and effectiveness, as all procedures will be performed under direct visualization.

Keywords: laryngocele, thyrohyoid membrane, combined laryngocele, laryngocele treatment, external approach.

BACKGROUND

Laryngocele is an infrequent and non-malignant anomaly which refers to the abnormal expansion or protrusion of the laryngeal saccule. This particular abnormality extends upward within the false vocal fold and maintains a connection with the laryngeal lumen (1). Laryngoceles can be classified as either internal or combined, based on their association with the thyrohyoid membrane (2). Despite numerous theories, the exact cause of this condition remains poorly understood, and it may involve various congenital or acquired factors (3). Diagnosing a laryngocele involves clinical observations, endoscopic evaluation and imaging techniques (1). The management of laryngoceles is primarily influenced by the size and type of lesion as well as the surgeon's experience (2, 4). In recent years, there has been a growing preference for the endoscopic treatment using microlaryngoscopic surgery and CO_2 lasers (4, 5). However, the traditional external approach is still considered the preferred method for larger internal or external laryngoceles and most combined

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cases (2, 6). In order to address the combined laryngocele, there are three external techniques that can be employed, as described in previous studies (2, 6-8). These techniques consist of the transthyrohyoid membrane approach, thyrotomy with excision of the upper portion of the thyroid cartilage and V-shaped thyrotomy. Despite the availability of the above-mentioned procedures, managing the combined laryngocele remains a considerable surgical obstacle. The utilization of the external approach in cases of combined laryngocele offers several advantages, which we will elaborate on in the present article. Comparing with the conservative (endolaryngeal) technique, the external/open cervical approach provides a better exposure of the paraglottic space and the laryngocele, enabling more precise resection without causing any distortion to the anatomy of the larynx. Additionally, the low recurrence rate of this method is worth noting. It is more suited for combined laryngocele with large external component, guarantees complete resection of mucosa and does not require a specialized expensive infrastructure. ❑

CASE PRESENTATION

A 64-year-old female, non-alcoholic long-term smoker, presented with a history of progressive submandibular swelling. On clinical examination, the patient was found to be in fair condition with good health status of the larynx and submandibular swelling on the right side of the neck. The swelling was firm, painless and mobile anterior to the sternocleidomastoid muscle at the level of the hyoid bone. It enlarged with the Valsalva maneuver but shrank when compressed. Laryngoscopy revealed a smooth submucosal swelling above the glottis situated in the false vocal folds without affecting the surrounding mucosa. Both flexible nasopharyngolaryngoscopy and direct laryngoscopy revealed a mass filling within the laryngeal vestibule mucosa with a normal appearance (Figure 1).

A computed tomography (CT) scan of the neck confirmed the diagnosis, showing a tissue-dense mass extending into the submandibular region and the pre-epiglottic space, morphologically air-containing, with well-defined smooth limits, and a maximum transverse dimension of 3 x 2.2 cm, occupying the supraglottis, with no involvement of the subglottis (Figures 2 and 3).

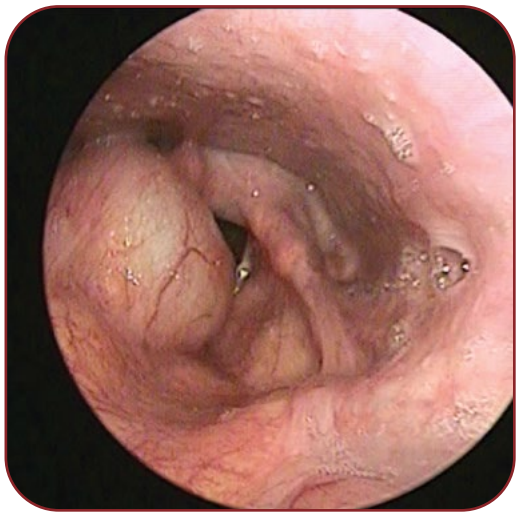

FIGURE 1.
View of a right-sided combined laryngocele during a flexible nasopharyngolaryngoscopy

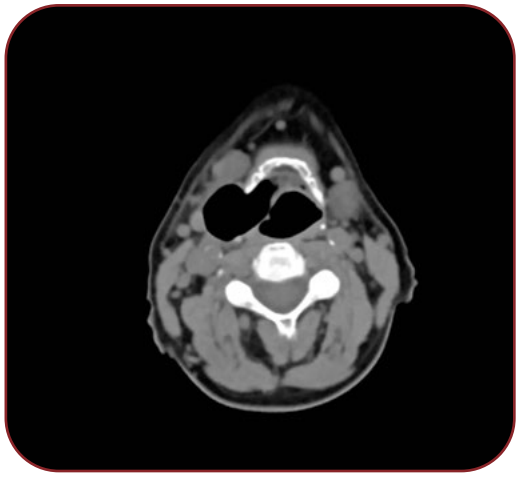

FIGURE 2.
Right-sided laryngocele revealed by the preoperative axial computed tomography image showing the soft tissues of the neck

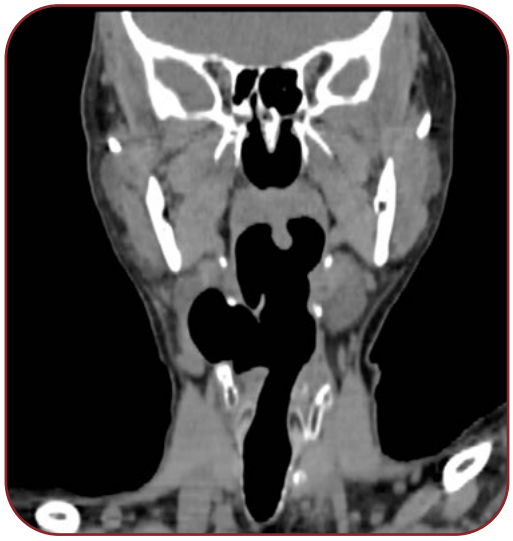

FIGURE 3.
Right-sided combined laryngocele identified by the computed tomography scan of the larynx

As a result, a diagnosis of compound laryngeal fissure was made and the patient was opera-

ted under general anesthesia through a transcervical approach via a transcervical membrane. A third-generation cephalosporin was administered preoperatively. In this procedure, the patient is placed in the supine position, with the neck hyperextended and the head facing the opposite side of the lesion. To access the site, a skin incision is made horizontally at the level of the thyroid membrane while extending from the medial border of the sternocleidomastoid muscle to the midline of the neck. The first step is to raise the flap in the subthyroid plane, reaching 1 cm above the hyoid bone and below the lower border of the thyroid cartilage. After checking for swelling, the glottis is separated until it reached the epiglottis. Next, the outer part of the capsule as well as the surrounding tissue of the glottis up to the posterior edge of the thyroid cartilage is removed. To approach the upper surface of the thyroid lamina, the sternohyoid, thyrohyoid and omohyoid muscles must first be cut and retracted from the lower edge of the hyoid bone. An incision is then made along the upper edge around the external cartilage of the thyroid gland and cut from the base of each thyroid gland to its junction with the thyroid notch. The laryngeal cleft is opened by a horizontal incision in the lateral wall to allow the dissection to proceed along the thyroid cartilage membrane.

This article describes a new approach to resecting the epiglottis through the pretyroid membrane. Identification of the superior laryngeal nerve and its internal and external branches is key to the safety of the dissection. When the incision is started from posterior to anterior along the upper border of the thyroid gland, the epiglottis is exposed more laterally. Under direct vision, continue dissecting from the epiglottic cavity towards the ventricles to ensure proximity to the epiglottic cavity (Figure 4). A ligature is applied at its lowest and deepest border before completely excising it (Figure 5). Intermittent sutures are placed on the mucosa to close the ventricles. The external perithyroid cartilage flap is repositioned, the strap muscle is fixed in its original position and the laryngeal skeleton is not deformed. The skin incision is closed in layers and a compression bandage is wrapped around the neck. The skin sutures are removed after one week.

Following the procedure, the patient was prescribed third-generation cephalosporins and steroids. Regular postoperative monitoring included visits every two weeks for the first month and then every three months, during which the

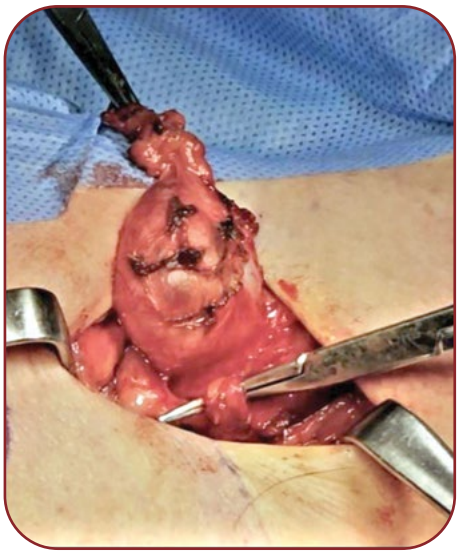

FIGURE 4.
Removal of a combined laryngocele using an external method through the transthyrohyoid membrane – intraoperative image, also showing the superior laryngeal nerve and artery anterior to the parotid forceps

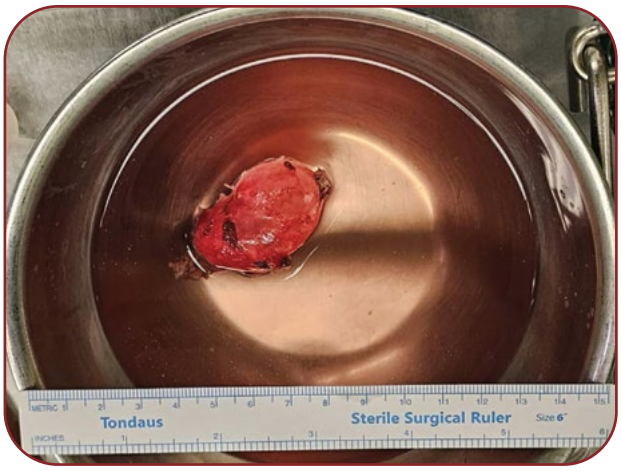

FIGURE 5.
Surgical specimen of laryngocele removed *in toto*

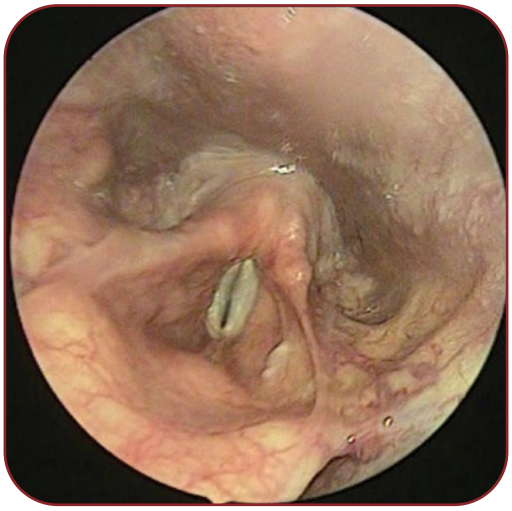

FIGURE 6.
Rigid endoscopic view of the larynx postoperatively

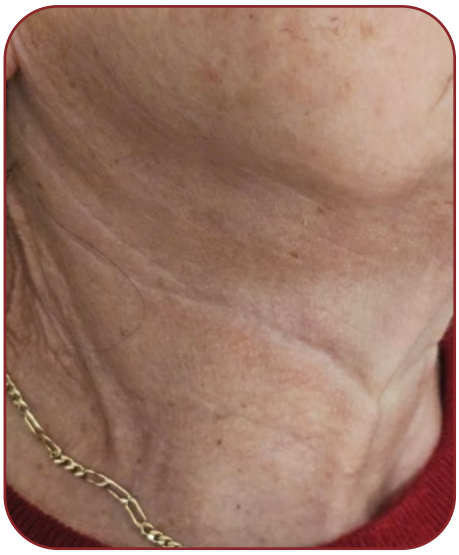

FIGURE 7.
Postoperative image showing the appearance of the patient's neck after resection of the laryngeal complex by an external approach

larynx was inspected using a flexible fiberoptic nasopharyngoscope or a 70° rigid laryngoscope. Computed tomography scans of the cervical soft tissue with and without contrast were repeated at three months postoperatively. The diagnosis of laryngocele and any suspicion of malignancy were confirmed histologically. A follow-up after six months showed a positive outcome with no signs of recurrence (Figures 6 and 7). ❑

DISCUSSION

In 1829, Larrey, a surgeon in Napoleon's army, described laryngocele as an air-filled tumor. However, it was not until 1887 that Virchow (10) developed the word "laryngocele" to describe the expansion of the laryngeal saccule, a pouch found in the upper section of the Morgagni laryngeal ventricle. In accordance with a study conducted by Holinger *et al* in 1978 (11), a definite diagnosis of laryngocele is attainable when the growth either causes symptoms, is able to be felt or can be visualized through direct or indirect laryngoscopy. Additionally, a diagnosis can be confirmed if an air-filled sac is detected through radiological imaging or during dissection and is found to extend beyond the superior border of the thyroid cartilage.

Impressively uncommon, at a rate of one in every 2.5 million persons per year (12), it has been found that laryngoceles are more widespread among men, with the highest incidence seen in those aged 60 years or older (13, 14). Moreover, according to the literature, most laryngoceles are unilateral (80-85%), without any strong predilection for either side (14). These uncomplicated laryngoceles consist of a pouch of air. In the presence of chronic inflammation or laryngeal tumors, the neck of a laryngocele can be closed, leading to its transformation into a laryngomucocele. This specific lesion is filled with mucus produced by the epithelial glands and, if a secondary infection occurs, it can become a laryngopyocele (15). Laryngocele is a benign and infrequent condition that can be internal or combined according on its location relative to the thyrohyoid membrane (2). The terms internal, external and combination laryngoceles are no longer used, since there are no wholly external laryngoceles; they all originate in the laryngeal saccule. An internal laryngocele is a form of laryngocele that remains within the larynx and does not pass through the thyrohyoid membrane. It usually only extends over the thyroid cartilage into the posterosuperior area of the false vocal fold and aryepiglottic fold. In contrast, a combined laryngocele protrudes through the thyrohyoid membrane to reach the superior laryngeal nerve and arteries in the neck (2, 16). This type of laryngocele is the most commonly encountered (17, 18).

Despite numerous theories, the underlying pathogenetic mechanisms of laryngocele remain poorly understood. Various factors, either congenital or acquired, appear to be involved. The condition may be triggered by various factors, including chronic coughing, straining, glass blowing, blowing into musical instruments and laryngeal cancer, that result in continual high pressure within the larynx and gradual expansion of the sac (11, 19). Additionally, laryngeal conditions such as cancer, chondroma, scleroderma, amyloidosis and others can also contribute to the development of laryngocele by mechanically obstructing the ventricle, increasing pressure within the larynx and facilitating the formation of the laryngocele (13). Micheau *et al* underline the close relationship between laryngocele and cancer, both when a pre-existing malignancy generates the laryngocele and when squamous cell metaplasia transforms an existing laryngocele into carcinoma (20). Thus, the appearance of a laryngocele always indicates the need for a thorough investigation of the ventricle for probable malignancy. Notably, few individuals do not have any predisposing factors, although the majority of laryngoceles occur unilaterally (7, 13, 21).

Some studies revealed a relationship between past neck surgery and the development of laryngocele. In one case, a clinically significant laryngocele progressed over several years following a tracheotomy. This was linked to local trauma sustained during the first tracheotomy implantation, which may have resulted in a weakening, defect, or mechanical obstruction in the laryngeal saccule (3). Another study found that around 3% of individuals who underwent supraglottic laryngectomy developed laryngoceles; this incidence was associated with the partial removal of the ventricle (22).

Many laryngoceles do not show any symptoms. When symptoms do occur, signs vary depending on the type of laryngocele (26). The most common manifestations of laryngocele are raucity of voice and neck swelling (12). Laryngoceles that are found inside can cause the obstruction of speech and symptoms, which may include problems in speaking, breathing, snoring, hoarseness as well as occasional blockage of the airway (7). Other symptoms include feeling like an extraneous body sense, a sore throat, and reflexive coughing. On the other hand, external laryngoceles appear as a painless and occasionally fluctuating mass in the superior anterolateral triangle of the neck, situated below the digastric muscle. The mixed form is identified by a sudden deterioration of the symptoms, particularly dyspnoea, due to the flow of air from the outside to the inside component following compression of the external component (26). The development of laryngocele might culminate in complications such as laryngopyocele, pneumonia and, in exceptional scenarios, immediate obstruction of the airways (27).

All patients with suspected laryngocele should have a diagnostic assessment consisting of: 1) an initial thorough medical history considering the length and severity of symptoms as well as any prior endoscopic or surgical procedures; 2) a thorough examination of the upper airway using a fiber optic soft nasopharyngoscope or a 70° rigid laryngoscope, which is critical in evaluating the patient's airway and ruling out upper airway blockage; also, it displays enlargement of the false vocal fold and arytenoid, which are covered with normal laryngeal mucosa; 3) the use of Valsalva manoeuvre enhances mass perception, which can be reduced by external pressure, sometimes producing fluid noises; 4) a CT of the soft tissues of the neck, with or without contrast agent, which reveals a clearly-defined gas- or fluid-filled mass connecting with the laryngeal ventricle, indicating the kind and size of laryngocele as well as the presence or absence of a carcinoma (23); it has also been demonstrated to be crucial in distinguishing laryngocele from other benign or malignant formations (7, 24); 5) a magnetic resonance imaging (MRI), which is able to provide comprehensive details regarding the borders of the laryngocele as well as its placement in relation to the thyrohyoid membrane; this allows for a clear distinction between internal, external, and mixed components. In an MRI scan, an air-filled laryngocele is usually spotted as a cystic dilatation of the low-signal cavity in the larynx ventricle (1, 18). In the case of laryngocele or laryngopyocele, MRI is the most commonly recommended imaging method due to its ability to distinguish between obstructed mucous and inflammation versus neoplastic disease (21); 6) ultrasonography of the neck is commonly utilized to initially evaluate a neck mass, mainly to differentiate its type and determine its contents and position. Ultrasonography examination performed first hand by the ENT specialist has several advantages as primary imaging solution being a non-iradiating, quick and cheap approach, which permits the serial examination of the patient and furthermore significantly contributes to the differential diagnosis of anterior neck mass, excluding for example a thyroglossal duct cyst (25).

The three most important factors in managing laryngocele include the size and type of the damage as well as the skill of the surgeon (1, 3). Smaller laryngoceles can be removed with the use of laser technology via endoscopy. On the other hand, larger laryngoceles, whether internal or external, require a more invasive exterior approach for removal. In most cases (86.2%), patients with combined laryngoceles are managed via an external approach as reported in the literature (2, 6). Cold instruments and CO2 laser microlaryngoscopy, marsupialization and robotic surgery represent the three dominant endolaryngeal procedures. Over the last few years, there have been increasing technological advancements in surgery, particularly with regard to lasers, leading to a wide acceptability of endoscopic techniques for laryngocele management (5). However, there are some disadvantages to this

approach, suchh as usage restrictions, arduousness with direct laryngoscopy and limited approximation, the probability of deficient resection necessitating additional procedures and potential trauma to the adjoining laryngeal structures, resulting in exorbitant scarring (2). When performed with the endoscopic CO_2 laser technique, the procedure can be completed in less time, with less bleeding and harm to the endolarynx, allowing for complete recovery of laryngeal functions. Nonetheless, it necessitates the use of specialized devices, extensive knowledge and carefully selected patients. In 2013, for example, the first robot-assisted surgery to remove a coupled laryngocele was performed (28).

Unlike microlaryngoscopy, a technique traditionally used to remove a laryngocele, this particular technique is believed to have many benefits. External approaches for treating laryngocele have been shown to bring along a number of advantages, including high visibility of the laryngocele, precision in procedures and low chances for recurrence. However, there are also drawbacks to these external approaches, such as scarring of the skin, longer surgery time, potential risks to the nerves in the larynx, connected morbidness and a lengthier hospital stay (1, 2, 5). For the last twenty years, three external techniques have been used to achieve this goal: through the transthyrohyoid membrane; thyrotomy in combination with the removal of the top third of the thyroid cartilage; and V-shaped thyrotomy (2, 6-8). Among these three, the transthyrohyoid membrane approach has become the most commonly employed. Although it does not require the removal of the thyroid cartilage, its drawback is that access to the paraglottic space is limited. However, less commonly used techniques include those which involve cutting off the upper 1/3 of the thyroid cartilage as well as V-shaped thyrotomy, but they offer better exposure by resecting a portion of the thyroid cartilage (2, 6, 27). ❑

CONCLUSIONS

Laryngocele is a rare condition affecting the laryngeal ventricle. As a benign process, it is critical to distinguish it from any related cancer as a causative component. A therapeutic strategy must be tailored to the size and kind of lesion, as well as the surgeon's ability, in order to minimize the danger of incomplete resection and harm to laryngeal tissues.

For the removal of a combination laryngocele, our suggestion is to opt for the external approach as a singular procedure. This approach offers excellent exposure, wide accessibility and ensures the safe dissection of the laryngocele in the paraglottic space. Most importantly, it allows for the complete resection of the whole lesion under direct visualization. After a long hospital stay, this method of treatment results in low morbidity due to minimum damage to the laryngeal structures; consequently, tracheostomy is unnecessary. The external approach remains the method of choice in combined laryngocele cases, given its endoscopic management limitations, which include restricted surgical visibility inside the larynx and potential scarring. The procedure requires experience with special instruments. The risks of incomplete removal are quite significant for sizeable or combined laryngoceles, making the endoscopic approach not applicable. ❑

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