CASE REPORT / OLGU SUNUMU

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Co-existence of Laryngocele and Laryngopyocele: A Rare Case Report

Laringosel ve Laringopyosel Birlikteliği: Nadir Bir Olgu Sunumu

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Abstract

Laryngocele is an air-filled sac which originates from the saccule of the ventricle. It is one of the rarest lesions of the larynx. Certain occupational and chronic inflammatory diseases may play role in the formation of laryngocele, but the etiology in the majority of them is unclear. When infected, the air inside of the laryngeal saccule replaced by purulent fluid and it becomes laryngopyocele. These lesions may lead to dysphagia, roughness, and dyspnea.

There are limited number of cases in the literature. In the presented case here, laryngopyocele showed suspicious mass appearance in the radiological imaging and its diagnosis was confirmed by intraoperative direct laryngoscopy.

Laryngoceles are clinically asymptomatic unless they reach large sizes. As internal laryngoceles and laryngopyoceles may also be related to tumors originated from laryngeal ventricles, it is highly important to evaluate the lesion borders before the operation.

Key Words: Laryngocele, Laryngopyocele, Benign Laryngeal Mass

Özet

Laringosel, ventriküler sakkul kaynaklı bir hava keseciği olup larenksin çok nadir görülen iyi huylu lezyonlarından biridir. Laringosel oluşumunda belirli mesleki ve kronik enflamatuvar hastalıklar altta yatan neden olabilir, fakat büyük kısmında etiyoloji belirsizdir. Laringoseller enfekte olduklarında içerlerindeki havanın yerini pürülan mayi alır ve bu durumda laringopiyosel olarak tanımlanırlar. Bu lezyonlar disfaji, seste kabalaşma, dispne benzeri semptomlara yol açabilirler.

Literatürde sınırlı sayıda tanımlanmıştır. Sunulan olguda laringopiyosel radyolojik görüntülemede şüpheli kitle olarak raporlanmış, intraoperatif direk laringoskopi ile tanısı kesinleştirilmiştir.

Laringoseller büyük boyutlara ulaşmadıkça klinik olarak asemptomatiktirler. İnternal laringoseller ve laringopiyoseller, ventrikülden kaynaklanan tümörlere de bağlı olabileceğinden görüntüleme yöntemleriyle lezyon komşuluklarının preoperatif değerlendirilmesi büyük önem taşır.

Anahtar Kelimeler: Laringosel, Laringopyosel, İyi Huylu Larinks Kitleleri

Introduction

The laryngeal saccule or appendix of laryngeal ventricle is a pouch arising from the anterior end of the ventricle and it extends superiorly in the paralaryngeal space. It is bounded by the false vocal cords medially, thyroid cartilage laterally (1-3).

A laryngocele is defined as the abnormal dilatation of the laryngeal saccule contains air that extends superiorly within the false vocal cords. It maintains an open communication with laryngeal lumen (1,2). The simple laryngocele is a dilated or herniated saccule which contains only air. In some cases, it might be filled by mucus and become laryngomucosel. If laryngocele gets infected by bacterial or fungal agents, it contains pus and becomes a laryngopyocele (1-3).

There are different sentiments for classifying laryngoceles. Traditionally laryngoceles have been classified as internal, external or combined based on their relationship with the thyrohyoid membrane. However, some authors have been abandoned this classification since laryngoceles originated from laryngeal saccule and there is no purely external laryngocele (4).

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An internal laryngocele is confined within the false vocal cord and it is medial to thyrohyoid membrane. A combined laryngocele, which diagnosed as external laryngocele according to some authors, extends upward and protrudes through the thyrohyoid membrane to the neck. Combined laryngoceles make up the majority of cases and demonstrates both internal and external protrusions (4-6).

The estimated incidence of laryngocele is 1 per 2.5 million people per year. Laryngoceles have been reported to be 5-6 times more frequent in males with a peak incidence in 6th decade. The bilateral laryngoceles are extremely rare. There are six cases have been reported presenting bilateral larygngocele in the literature so far. Laryngopyocele is also very rare clinical entity. They constitute 8% of all laryngocele cases. Only 61 articles about laryngopyoceles have been reported since 1952 in the literature (6).

Herein, we presented a female patient who has suffered from both laryngocele and laryngopyocele on her left and right ventricular bands respectively.

Case Report

Sixty-four years old female patient referred to our clinic with hoarseness of voice which gradually increased since last year, dysphagia and blunt pain in the throat. She had diagnosed with asthma 11 years ago and operated from vocal cords because of Reinke's edema in 2013. She was a heavy smoker with at least 1 pack for more than 40 years. She had normal otorhinolaryngologic examination and no palpable mass on neck. In the laryngostroboscopic examination of the patient, a mass lesion with smooth surface was observed in the supraglottic region bilaterally on the superior and anterior segments of the band ventricles (Figure 1). The mass lesion on the right ventricular band increased its fullness with trumpet maneuver while the lesion on the left ventricular band did not. The other supraglottic structures was normal. There was no prominent space occupying lesion on bilateral vocal cords however minimal irregularities on free edges of bands and grade 1 Reinke's edema were seen in the glottic region. Vocal cord movements were natural.

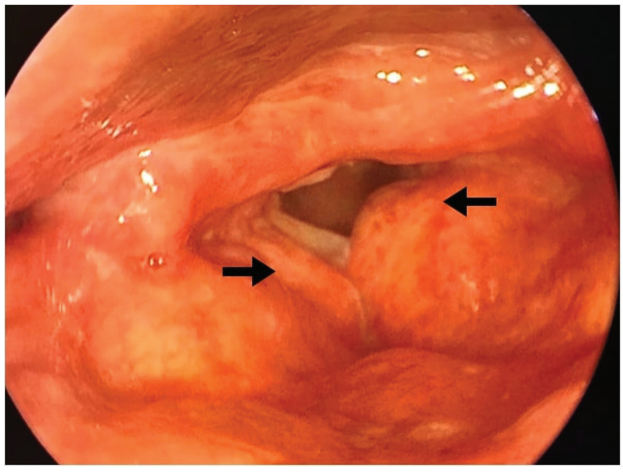

Figure 1: The laryngostroboscopic examination of the patient, a mass lesion with smooth surface was observed in the supraglottic region bilaterally on the superior and anterior segments of the band ventricles.

Because of the suspicious appearance of the lesion on the right ventricular band the larynx magnetic resonance imaging (MRI) has been obtained. The larynx MRI-T1 weighted images showed that low signal cystic dilatation of the laryngeal ventricle on the right side. On the left side MRI-T1 weighted images revealed that mass of intermediate signal on the unenhanced scan (Figure 2). Direct laryngos copy was performed under general anesthesia since the larynx MRI report suggested the histological diagnosis. When the lesion on the left band was punctured, purulent material discharge from the lesion was observed (Figure 3). Approximately 1.5 cc purulent material was aspirated. The wall of the cystic mass, which was accepted as a pyocele, was taken out by using micro scissor and forceps and it sent to the pathology for histological diagnosis. Subsequently, the mass that was reported as laryngocele on the right ventricle was marsupialized with the help of neodymium-doped yttrium aluminum garnet (Nd: YAG) laser. There was no complication encountered during and after surgery. In the postoperative

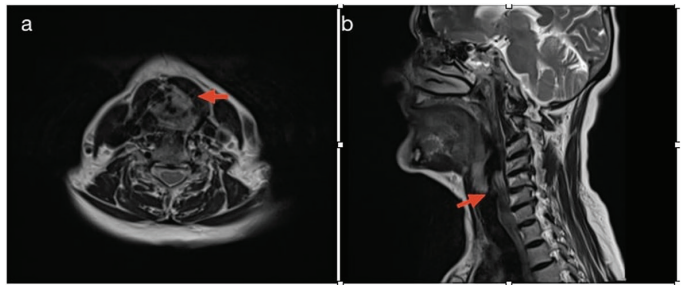

Figure 2: Red arrow shows the intermediate signal increase of the left sided mass lesion on the unenhanced 71 weighted axial image. Note the MRI T1 weighted image also showed that low signal cystic dilation of the latyngeal ventricle on the right side **(a)**. On the T1 weighted unenhanced axial image revealed that there is a narrowing on the glottic region due to the mass lesion located on band ventricle **(b)**

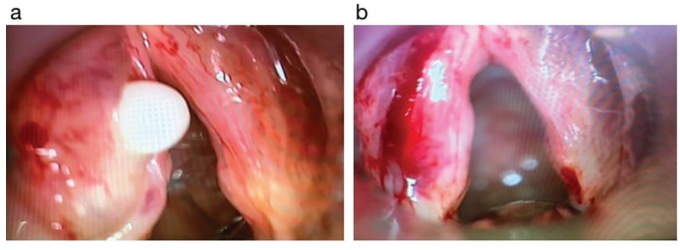

Figure 3: The intra operative direct laryngoscopic Picture of the lession on the left side. Purulent ma-terial discharge was observed when the lesion on the left band punctured **(a)**. Intraoperative direct laryngoscopic picture of the band ventricles just after the approximately 1.5 cc purulent material has been discharged **(b)**

first month evaluation patient's symptoms were resolves and laryngostoroboscopy was not showed any swelling in both ventricules (Figure 4).

Figure 4: Post operative 1-month laryngostroboscopic result of the patient

Discussion

Laryngocele is a rare condition characterized by cystic dilatation of laryngeal saccule filled with air that opens up into the larynx lumen. However, how it gets infected and turns into laryngopyocele is not clear. Tumor obstruction or mucous gland hypersecretion result in mucus retention and may be invaded by bacteries during respiratory tract infection.

Laryngoceles are mostly asymptomatic and often detected incidentally when neck radiographs have been obtained because of the unrelated symptoms. When they are symptomatic; laryngoceles commonly present with dysphonia or neck swelling. During trumpet maneuver which is basically forced expiration during nasopharynx and mouth is closed, the size of the mass may be increase by the air and the swelling becomes more prominent (7).

Other clinical symptoms of patients are cough, dyspnea, dysphagia, globus sensation in the throat and inspiratory stridor. On the other hand, laryngopyoceles are mostly symptomatic since they cause blunt pain in neck region and tenderness. Laryngopyoceles are relatively dangerous than laryngoceles since they can cause sudden airway obstruction. Infrequently Vocal cord paralysis and cutenous emphysema may seen as well.

Laryngoceles can be congenital or acquired. There is no direct causative agent or etiologic factor related with their development. They are more common in glass blowers, singers, wind instrument players or in people with congenitally having large saccule. Prolonged periods of increased pressure within the laryngeal lumen may cause the dilatation of laryngeal saccule. So, excessive and forced cough because of the chronic respiratory disorders may also cause dilatation of laryngeal saccule and become a predisposing factor for development of laryngocele (3,5). In our case, our patient had asthma for more than 20 years and getting treatment with some nonspecific bronchodilatator agents.

In 2019, Saravanam et al. (8) has reported the first laryngopyocele case with the bilateral laryngocele as an airway emergency. This is the second case in the literature a patient with laryngocele has also suffered from laryngopyocele as our current knowledge.

Laryngopyoceles may associate with laryngeal tumors. Micheau et al. (9) found 18% of laryngoceles in 546 laryngectomy specimens. Close et al. (10) reported that the incidence of asymptomatic laryngoceles was 12.5% in larynx carcinoma. Therefore; any malignancy of larynx should be ruled out when a laryngopyocele case has been clinically encountered.

History of patient, clinical findings, endoscopic examination of larynx and imaging studies are the diagnostic steps for patients with laryngoceles. Computed tomography (CT) scan of the neck provides definitive diagnosis of laryngocele with its well defined borders. CT also shows the communication of lesion with laryngeal airway. Furthermore, CT scan is necessary to differentiate the laryngocele from saccular cyst and not to lose any evidence of occult tumor. Indeed, MRI is superior to CT in its ability to evaluate soft tissue structures and lesions. The thyrohyoid membrane, paralaryngeal structures, false vocal cords are clearly visualized on MRI (1-3).

Laryngoceles are air filled and typically appears as low signal cystic dilatations of laryngeal ventricles in MRI.

There is no consensus regarding the surgical treatments of laryngoceles. Various modalities of treatment methods have been discussed so far.

Traditionally external approach for the resection of laryngoceles is the method of choice for large laryngoceles while endolaryngeal resection is preferred for pure internal and/ or small cases. Dursun et al. (1) have used endoscopic resection with CO_2 laser and stated that the operation time dramatically reduced with this technique. This technique is considered by many authors to be quick, precise and safe alternative to external approach with fewer complications; resulting in speedier rehabilitation for both patients and their voice. Endoscopic management provides a reliable and cost-effective management for laryngoceles nevertheless it has some handicaps such as providing limited surgical exposure, causing endolaryngeal scar, requiring experience with special instruments (11). We used Nd-YAG laser for larygocele and cold instruments for laryngopyocele and did not have a significant scarring in follow-up.

In conclusion, laryngoceles and laryngopyoceles are rare diseases of laryngeal ventricle. Despite their benign nature they can be co-existed with laryngeal cancers or they can mimic the laryngeal cancer in radiological studies. So, they need to be

diagnosed accurately. Even with the best imaging techniques, this differentiation cannot be made clearly and direct laryngeal examination may be needed as seen as in our case. Our case is very unique one in the literature since the lesion on left side of the ventricular band was laryngocele and the right side was laryngopyocele. According to our current knowledge this is the second case in the literature both laryngocele and laryngopyocele have been diagnosed and treated in one patient simultaneously.

Ethics

Informed Consent: Informed consent has taken from the patient who had this operation.

Peer-review: Externally peer-reviewed.

Authorship Contributions

Surgical and Medical Practices: G.G., A.T., Concept: G.G., A.T., Design: G.G., A.T., Data Collection or Processing: G.G., A.T., Analysis or Interpretation: G.G., A.T., E.A., Literature Search: G.G., A.T., E.A., Writing: G.G.

Conflict of Interest: No conflict of interest was declared by the authors.

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