A laryngocele revealing a small cell lung carcinoma

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Abstract. A laryngocele revealing a small cell lung carcinoma. Background: Laryngocele is defined as an abnormal dilatation of the laryngeal ventricle. It is a very rare entity, and the exact underlying mechanism is still unclear. Laryngoceles are associated with larynx carcinoma but not yet lung cancer.

Case presentation: A 46-year-old man presented with stridor, wheezing, dyspnea at rest, hoarseness evolving over two months, and cyanosis secondary to cervical swelling. His medical history included a 72 pack-year smoking habit and chronic obstructive pulmonary disease for 10 years. Airway management included a surgical tracheotomy for respiratory distress. A neck CT revealed laryngocele. A chest X-ray showed a left basal pulmonary opacity. Bronchoscopic exploration found an infiltrated and narrow left stem bronchus with complete stenosis of the lingula. Pathology revealed a small cell lung carcinoma.

Conclusion: Our case shows the possible association of laryngocele and lung carcinoma. The pathophysiology was explained by the long history of tobacco smoking and the underlying chronic obstructive pulmonary disease with chronic cough.

Introduction

Laryngocele, as described by Virchow in 1867,¹ is an abnormal dilatation of the laryngeal sacculae, which develops between the ventricular folds, the base of the epiglottis, and the inner surface of the thyroid cartilage. The first case of laryngocele was reported in 1829 by a French military surgeon named Dominique Larrey,² but the exact etiology behind its occurrence remains unclear. However, some congenital and acquired factors have been implicated in the condition’s development. The clinical presentation is often asymptomatic, and the presence of the disorder is usually discovered during autopsy. In some cases, laryngocele may present as a chronic history of altered voice or a cervical mass, which sometimes blocks the airway and requires urgent care. We report a case of laryngocele causing airway obstruction, requiring an urgent tracheotomy, and revealing a small cell lung carcinoma.

Case report

A 46-year-old man with a 72 pack-year cigarette habit, chronic bronchitis over the last 10 years, and dyspnea on exercise was treated 3 years prior with salbutamol on an “as needed” basis. The patient’s past medical history revealed no prior laryngeal problems or alcohol habits, and he presented with acute stridor, wheezing, dyspnea at rest, hoarseness evolving over two months, and cyanosis secondary to cervical swelling. Oxygen saturation was 86% under 6 liters of oxygen, and the respiratory rate was 36 cycles/min with 110/70 mmHg of blood pressure. A physical examination found a significant tumefaction measuring 10 cm from his right carotid triangle to the sub-mandibular region. This tumefaction was soft, mobile, and easily reduced with pressure but no tenderness during deep palpation, and it became more pronounced during cough. A left basal consolidation was found upon chest examination. The patient was transferred to the emergency department because of neurological deterioration and suffocation due to acute respiratory distress requiring an urgent tracheotomy. A 95% oxygen saturation of the room air improved these signs. Laryngoscopy was not possible in our emergency department, so it was not performed before the tracheotomy. A neck CT scan (Figure 1) revealed a large bilateral cystic lesion filled with air and communicating with the laryngeal lumen, confirming the diagnosis of laryngocele. A rhinolaryngoscopy was normal. The chest X-ray (Figure 2) revealed a left para-hilar opacity associ-
ated with a homolateral pleural effusion. The pleural puncture released 8 ml of a clear exudative liquid containing 35 g/l of protein. No germs or neoplastic cells were found. The pleural biopsy was non-specific. A bronchoscopy procedure introduced through the tracheotomy cannula showed an infiltrated and narrow left stem bronchus with complete stenosis of the lingula and a thick feature of the left interlobar area. Multiple biopsy specimens were obtained from these endobronchial lesions by brushing and aspiration. The pathological exam, along with a positive immunohistochemical staining for chromogranin, neuron secretory enolase (NSE), and synaptophysin, confirmed the presence of small cell lung carci-

nomia. After announcing the diagnosis to the patient and discussing the benefits and possible side-effects of the various treatments, he refused the treatment proposal and was referred to a palliative care center.

Discussion

Laryngocele is an abnormal dilatation of the laryngeal sacculae. It is a very rare entity, with an estimated incidence of 1 case per 2.5 million people per year and is five- to seven-fold more frequent in males during their fifth or sixth decades of life. The 46-year-old age of our patient is similar to the average reported by Dursun et al. The exact cause of the appearance of laryngocele remains obscure. However, many hypotheses have been put forth: congenital, acquired, or both. Congenital factors have been implicated to explain the existence of laryngocele in newborns or adults having a defect or anatomical variation of the laryngeal sacculus in which it is exposed. Acquired factors include exposition due to an elevated intrapharyngeal pressure, especially among divers and players of musical instruments that require blowing. The presence of pharyngeal or laryngeal carcinoma has also been described in many cases, suggesting a direct relationship between the two diseases. Therefore, it is important to search for a local throat neoplasm in patients with laryngoceles, especially laryngeal carcinoma. Laryngeal carcinoma can provoke a laryngocele via several mechanisms, including increased intraluminal pressure, an obstruction of the upper airways, speech effort, excessive cough, and local mechanical conditions. In our

Figure 1

a: Axial CT scan with contrast showing a bilateral laryngocele. b: Frontal reconstruction showing the external spread of the laryngocele.

Figure 2

Chest X-ray showing a para-hilar and basal opacity of the left lung with pleural effusion.
patient, no local carcinoma was found by CT or laryngoscopy. Chronic bronchitis in smokers or chronic obstructive pulmonary disease (COPD) can also lead to laryngocele. Obviously, chronic cough increases intralaryngeal pressure and can lead the development of laryngocele in the elongated appendix, and smoking can cause local alterations and histological modifications, resulting in not only laryngocele, but even more often to throat carcinoma which is itself a factor of laryngocele. Thomé et al. described nine cases of laryngoceles in 1995, among which were two smokers with chronic cough. In our case, chronic bronchitis, a long history of smoking, and lung carcinoma are probably the most influential factors in the pathogenesis of the disease. Lung carcinoma would be responsible for the laryngocele by worsening the chronic cough. Amyloidosis has also been reported as an acquired factor. Different clinical forms of laryngoceles have been distinguished: unilateral or bilateral, and internal or external (mixed). An internal laryngocele remains in the paralaryngeal space and lies medially on the thyroid membrane; it is usually seen as a swelling of the false vocal cord and the aryepiglottic fold, whereas an external laryngocele extends to the upper zone and penetrates through the thyrohyoidian membrane. In our case, the patient had an external bilateral laryngocele. Clinically, most laryngoceles are asymptomatic. When symptomatic, patients commonly complain of hoarseness or a swelling of the neck. Typically, this swelling becomes more prominent during Valsalva maneuver. Other symptoms related to laryngoceles include dyspnea, cough (due to the encroachment of the laryngeal space), inspiratory stridor, dysphagia, and the sensation of a foreign body in the throat. A CT scan or MRI differentiates between cysts filled with air, liquid, or mixed and can also proved the exact localization, whether it is an internal or external. The principal differential diagnosis includes a saccular cyst, bronchial cyst, neck abscess, and lymph nodes. Complications of laryngocele include infection, known as laryngopyocele, laterolarynx infection, bronchitis or pneumonia by aspiration, upper airway obstruction, and rupture. Laryngocele treatment depends on its size and local repercussions. Small internal laryngoceles can be removed endoscopically or trans-endoscopically excised by laser, causing less edema and less postoperative adherence compared to the conventional method. Small and recurrent internal laryngoceles, which may be associated with malignancy and large internal or external laryngoceles, are removed by an external approach. Small and asymptomatic laryngoceles are followed and only removed when they are not tolerated, they become more symptomatic, or cause some esthetic discomfort.

Conclusion

Laryngocele seems to be a benign disorder; however, it can cause respiratory obstruction that may threaten the patient's life. Patients with laryngocele must be fully investigated to find a potential cause. Laryngocele revealing a lung carcinoma is extremely rare and, to the best of our knowledge, has not been previously reported. In this case, smoking with COPD and chronic cough may be the determining factors in the pathogenesis of the disease.

References


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