Case Report

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A Rare Case Requiring Emergency Tracheotomy, Isolated Tracheal Mucormycosis: It is Important to Suspect First

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Abstract

Mucormycosis is asignificant opportunistic infection, ranking as the third most common invasive fungal infection after candidiasis and aspergillosis. A 67-year-old male patient was evaluated for progressive shortness of breath. Intubation tube could not be passed through the subglottic region. An emergency tracheotomy was performed. After the tracheal incision, a gray mass and necrotic material were encountered in the tracheal lumen. The tracheal biopsy result was mucormycosis. In emergency services, situations requiring emergency airway access due to difficult intubation can often be linked to central airway obstruction. For immunocompromised diabetic patients, it is important to consider the possibility of a fungal infection in the tracheobronchial tree that may need immediate treatment. To facilitate a quick differential diagnosis, perioperative frozen examinations of the obstructive mass are recommended.

Keywords: Emergency tracheotomy, invasive fungal infection, tracheal mucormycosis, upper airway obstruction

Introduction

Situations necessitating emergency airway access due to difficult intubation or inadequate ventilation are rare but life-threatening andcricothyrotomy is the quickest method to secure airway access in emergency situations (1). However, in a 'can not intubate can not oxygenate' situaiton arising from central airway obstruction, cricothyrotomy does not bypass the central airway and an open surgical tracheotomy or bronchoscopy becomes imperative as soon as possible (2).

Mucormycosis is asignificant opportunistic infection, ranking as the third most common invasive fungal infection after candidiasis and aspergillosis but, isolated tracheal involvement is rare (3).Patients with immunosuppression, diabetes mellitus (DM), or hematological malignancy are particularly at a higher risk. The prompt diagnosis and treatment of potential invasive fungal infections in at-risk patients can be lifesaving (3).

In this article, we present a case of isolated tracheal mucormycosis that necessiated emergency tracheotomy. We discuss the management of the airway and the importance of urgent treatment of invasive fungal infection.

Case Report

A 67-year-old male patient was admitted to the nephrology department of a tertiary care institution due to chronic renal failure (CRF) and uncontrolled DM. It was discovered that the patient had been undergoing an immunosuppressive treatment for the past six months due to chronic inflammatory bowel disease. Subsequently, the patient was referred to the Otorhinolaryngology department of our hospital due to the development of progressive dysphonia and cough. Indirect laryngoscopy revealed slight edema and mucoid material on the vocal cords. Treatment for acute laryngitis commenced with third-generation Cephalosporin (1gr/day), antireflux, and antimucolytic therapy. Given the ongoing COVID-19 pandemic, we conducted the COVID-19 polymerase chain reaction (PCR) test on the patient. COVID-19 PCR on nasopharyngeal swab was negative for four times. The patient, whose shortness of breath worsened rapidly within five days, was treated with meropenem, linezolid, and colistin upon the recommendation of an infectious diseases specialist, and hemodialysis was performed due to hyperkalemia. His condition continued to deteriorate leading to clinical hypercapnic respiratory acidosis. Intubation was attempted,

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but severe resistance prevented the passage of endotracheal tube deep enough through the subglottis. Considering an intratracheal obstruction, emergency tracheotomy was performed. During the tracheotomy, as the incision made on the trachea through the intercartilaginous space, necrotic material was encountered in the tracheal lumen. Direct laryngoscopy indicated a normal laryngeal passage, but a grey mass, 2.5 cm below the vocal cords, narrowed the trachea from the anterior part. Initially suspecting a tumor or a granulomatous mass occlusion, perioperative tracheal biopsies were taken for differential diagnosis. The patient was then transferred to the intensive care unit and underwent bronchoscopy due to recurrent airway obstruction with necrotic material. During the operation, necrotic materials were removed from the tracheal lumen by fiberoptic bronchoscopy, and the main bronchi were found to be normal. Postoperatively, serum blood tests, blood culture and radiological imaging were conducted. C-reactive protein was high with accompanying leukocytosis in the hemogram. Blood culture identified Enterococcus faecium, while sputum culture and serum tests for galactomannan antigen, c-ANCA and p-ANCA were negative. Laryngeal computed tomography (CT) displayed an endotracheal mass (Figure -1) located from the inferior part of the cricoid cartilage to the third tracheal ring. Paranasal sinus CT revealed maxillary and ethmoidal inflammation findings, while thoracal and cranial CT scans revealed normal results. The tracheal biopsy result was finalized 11 days after the tracheotomy,with the pathologist reporting mucormycosis (Figure-2). Liposomal Amphotericin B (Amp B) was administered intravenously, starting with 1mg/kg/day and gradually increasing to 5 mg/kg/day. Functional endoscopic sinus surgery was conducted to investigate another potential focus of fungal infection. Nonspecific inflammation was observed during sinus surgery, and Candida nonalbicans was identified in the sinonasal biopsy culture. However, the recurrence of accumulated intratracheal necrotic material and hypercapnic respiratory acidosis persisted, ultimately resulting in the patient's demise on the twentieth day of tracheotomy. As no other focus was identified during the direct examination of the larynx with laryngoscopy and broncoscopy and normal findings were observed on laryngeal, cerebellar and thoracal CT scans, the case was classified as isolated tracheal mucormycosis.

(Permission to use the patient's data was obtained from the patient's wife and the local ethics committee of our hospital.)

Discussion

In this report, we present a male patient with DM and CRF, who could not be intubated due to tracheal obstructive pathology and underwent emergency tracheotomy. Initially,

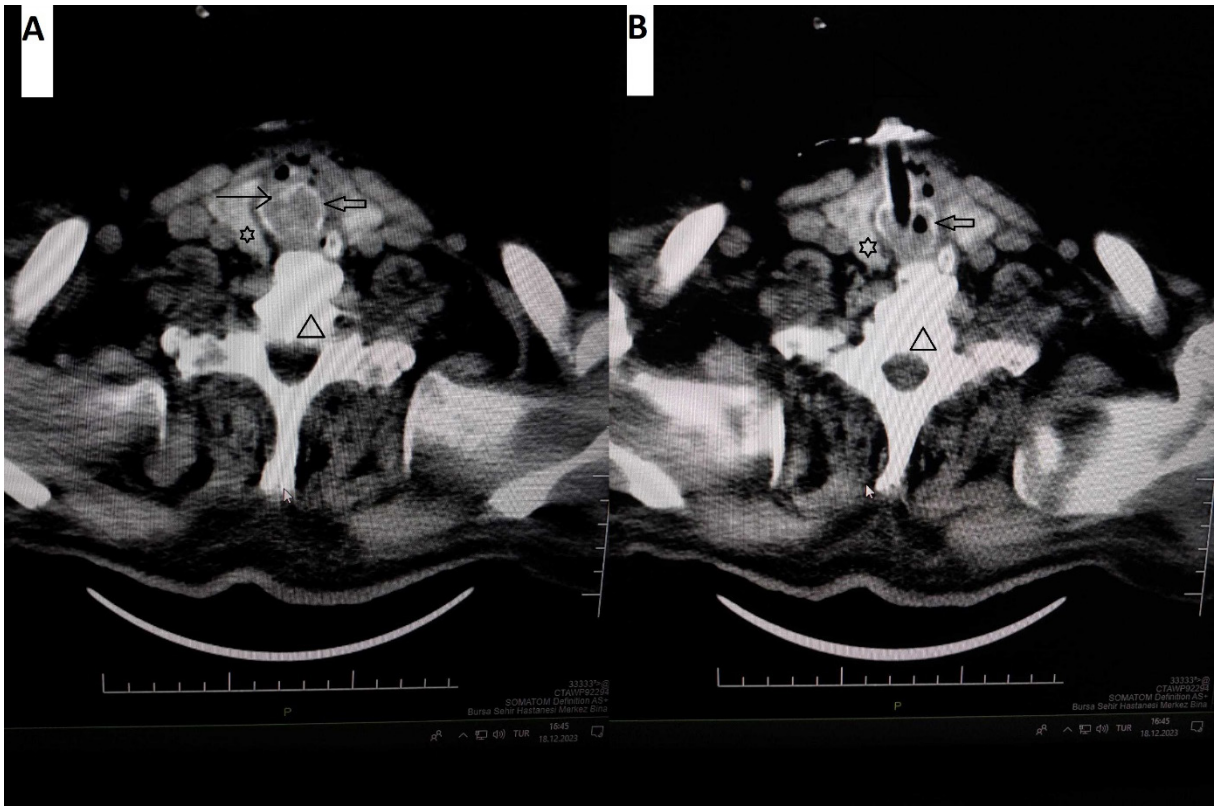

Figure 1. Contrast-enhanced axial section of neck computed tomography (Neck 5.0 Bf 37) showing the tracheal cannula positioned within the trachea. Line arrow: soft tissue mass in the anterior part of tracheal lumen. Hollow arrow: third tracheal ring. Star: right lobe of thyroid gland. Triangle: body of seventh Cervical vertebra.

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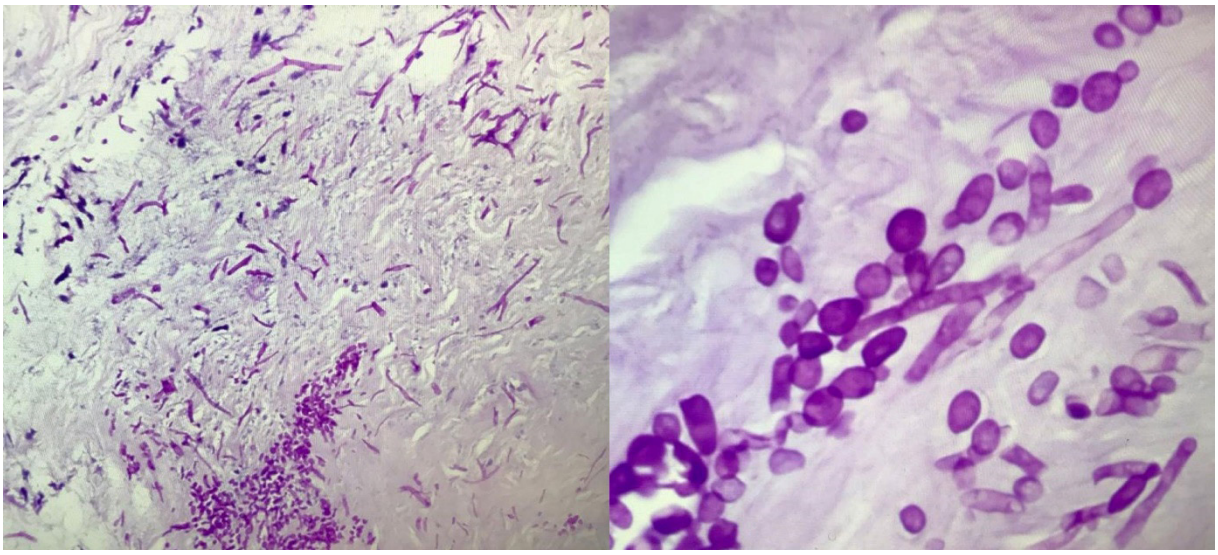

Figure 2. Photomicrographs showing large aseptate fungal hyphae in necrotic ground consistent with mucormycosis morphology (hematoxylin and eosin stain, magnification x100 and magnification x400).

it was suspected that the cause of tracheal obstruction might be a tumoral or granulomatous pathology. Perioperative biopsies were taken from the grey mass and necrotic materials in the trachea. The patient was treated with broad-spectrum antibiotics and during this period, the patient underwent repeated bronchoscopies due to the reaccumulation of obstructive necrotic material. Eleven days after tracheotomy, histopathological diagnosis of mucormycosis was reported, and antifungal treatment was initiated. Unfortunately, the patient passed away twenty days after the tracheotomy procedure. When we noticed the obstruction in the trachea, we suspected a tumor and proceeded with biopsies. Unfortunately, it took eleven days for the biopsy results to be reported, which also caused a delay in initiating the mucor treatment. Since the primary focus persisted in the trachea, the recurrence of necrotic material and airway obstruction continued. By performing a frozen section analysis and ensuring airway safety, healthcare professionals can accurately diagnose invasive infections that need prompt attention. If a mucor infection is present, it is important to surgically remove the affected areas completely and start antifungal treatment as soon as possible. This timely intervention can potentially change the course of the patient's clinical outcome. The most important thing to consider in such patients is to maintain a high level of suspicion.

Neoplasia and post-intubation tracheal stenosis are common causes of airway obstruction. CT of the neck and chest using intravenous contrast in the inspiratory and expiratory phases is the gold standard imaging for suspected tracheal obstruction (4). Causes of tracheal obstruction include granulomatous infections and, rarely, fungal infections (5). It is important to ensure airway patency immediately. Most guidelines for the management of central airway obstruction recommend awake endotracheal intubation, tracheotomy, laryngeal mask airway, or suspension laryngoscopy, with a high likelihood of requiring subsequent rigid bronchoscopy (6-8). Endotracheal intubation is particularly important due to the high risk of turning critical airway obstruction into a complete blockage during rapid sequence induction and paralysis. Additionally, airway manipulation during tracheal intubation can lead to inflammation and hemorrhage (9). However, it is possible that the expiratory positive airway pressure in non-invasive ventilation serves to stent an anatomical stenosis, preventing collapse during expiration. This mechanism may also reduce the work of breathing and enhance alveolar ventilation, facilitating the washout of hypercapnia (10).Chen et al. recommended the successful use of non-invasive ventilation for patients with distal and critical airway stenosis, particularly those experiencing hypercapnic respiratory failure. This approach allows for significant time and logistical planning before bronchoscopic intervention (2).

Tracheobronchial involvement of mucormycosis has been previously reported, and bronchoscopy is an important diagnostic method indiagnosis (11,12). Grey granulation tissue obstructing the airways can be directly visualized (3). A systematic review by He et al. documented 60 cases of mucormycosis in the tracheobronchial tree, with only 10 exhibiting tracheal involvement along with bronchial involvement (12). However, isolated tracheal mucormycosis is exceptionally rare (11,13).

Conditions that need evaluation in the differential diagnosis of focal necrotic involvement of the trachea encompass granulomatous diseases (tuberculosis, Wegener's granulomatosis), neoplastic diseases (chondrosarcoma, adenoid cystic carcinoma), and long-term intubation trauma (5). While sputum culture can serve as a preliminary diagnosis method for infectious granulomatous diseases, negative results are typically obtained and the accurate diagnosis is usually achieved through histopathology. In thecase

of mucor infections, biopsies may show tissue invasion with characteristic broadnonseptate hyphae with right-angled branching (3). In the literature, the most common comorbidity in isolated tracheal mucormycosis cases is DM, and the association with H1N1 pneumonia was reported in only one case (13). Damaraju et al. reported that among the 26 cases of isolated airway invasive mucormycosis identified in the literature, 4 cases were associated with COVID-19 infection (14). Amid the ongoing COVID-19 pandemic, we conduted a COVID-19 PCR test on our patient, yielding negative results. Patients with COVID-induced mucormycosis have been reported during the pandemic. In addition to the ongoing risks of mucormycosis, such as uncontrolled DM and immunosuppression, new factors such as excessive use of steroids and antibiotics couped with hypoxia have emerged during the pandemic. Despite both surgical and medical treatments, the mortality rate remains high (15). A multidisciplinary approach is required to prevent mucormycosis in COVID-19 patients, and the risk of mucormycosis should be consistently considered.

Early diagnosis and antifungal treatment are pivotal determinants of outcome, but above all a high degree of suspicion is required. Surgery plays a crucial role in treating the disease. Among antifungal agents, parenteral Amp B is the preferred treatment, but it can reduce the glomerular filtration rate, and hence Liposomal Amp B is recommended in cases with impaired renal function (16). In some cases, Amp B was administered through inhalation or bronchoscopic instillation (14,17,18). The placement of an airway stent and the use of a transbronchial microtube to drip Amp B have been recommended to quickly relieve airway obstruction and extend the duration of drug action, thereby enhancing the antifungal therapeutic effect (17,18). When tracheotomy or bronchoscopy was managed, the presence of necrotic material in the trachea should raise suspicion of mucormycosis, and perioperative frozen examination may be recommended for early diagnosis, especially in diabetic patients with compromised immune systems. Frozen section analysis is an effective method for promptly managing invasive fungal infections, particularly in situations where decisions need to be made during surgery (19).

Extensive endotracheal involvement is more difficult to manage, and resection and anastomosis have also been performed in the past (5). Due to the limited number of tracheal mucormycosis cases, the timing of surgery is a matter of debate. More data isneeded to decide whether surgery should be performed immediately at the time of diagnosis or whether surgical resection would be better after a course of medical treatment.

Conclusion

In the emergency service, situations necessitating emergency airway access due to difficult intubation may be releated with central airway obstruction.When emergency tracheotomy or bronchoscopy is required, it should be kept in mind that an obstructive greymass or necrotic material in the trachea may be a focus of fungal infection requiring rapid treatment. Perioperative frozen section examination may be recommended to accelerate the differential diagnosis of potential invasive fungal infection in immunocompromised diabetic patients. Early managementsupported with a confirmed frozen section may be life-saving for immunocompromised patients with fungal infections.

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