CASE REPORT

LARYNGEAL MUCORMYCOSIS IN AN UNCONTROLLED DIABETIC PATIENT: A CASE REPORT

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Background: With rising conditions in immunocompromised states, the world is facing the challenge of fungal infections with mucormycosis which was rare previously. With the rise in diabetic patients, COVID-19-related immunosuppression, and steroid use, along with an increasing number of transplant and chemotherapy patients, there has been a notable surge in mucorales infections. Although patients with rhino-orbit-cerebral mucormycosis are the most common type of pulmonary rare ones. **Methods**: Here, we are reporting 1st case of laryngeal mucormycosis in a 55-year-old male presented with respiratory distress in the ENT department of Shaikh Zayed Hospital, Lahore. He had a 4-year history of poorly controlled diabetes, with no other identifiable risk factors. An emergency tracheostomy was performed, pus was aspirated, and necrotic tissue was debrided to relieve the airway obstruction. **Conclusion:** Laryngeal mucormycosis was diagnosed in the subglottic region via multiple biopsies through direct laryngoscopy under anesthesia. The emergence of new variants of mucormycosis is calling for vigilant identification and meticulous control of predisposing risks. Concomitant debridement and intravenous amphotericin B are recommended under current guidelines.

Keywords: Covid-19; Larynx; Mucormycosis

Citation: Shahzadi E, Shayan A, Qazi ZUS, Latif S, Malik SA, Chaudhary U. Laryngeal mucormycosis in an uncontrolled diabetic patient: A case report. J Ayub Med Coll Abbottabad 2024;36(3):678–80.

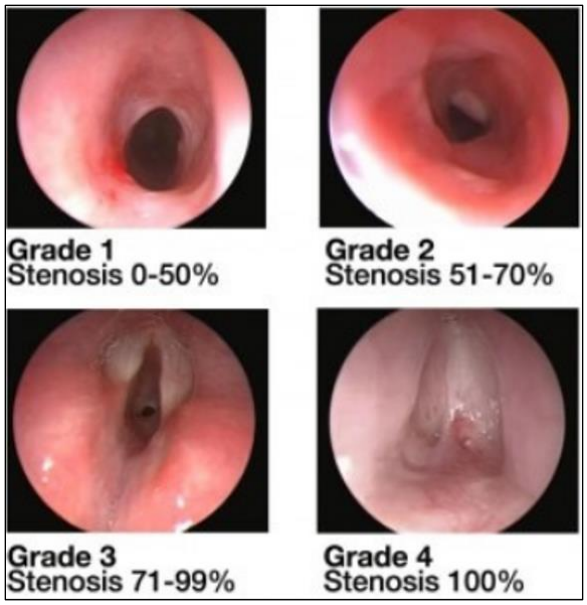
DOI: 10.55519/JAMC-03-13598

INTRODUCTION

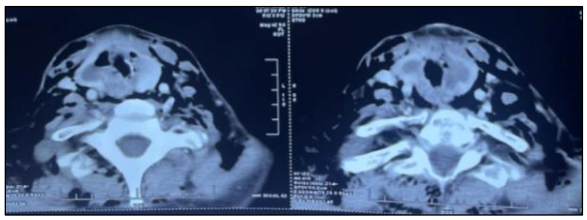
Mucormycosis, a rare fungal infection globally, has seen a rise in immunocompromised diabetic patients.[1] Pulmonary mucormycosis is seen in cases of transplant recipients and hematological malignancies.[2] The increased use of high-dose corticosteroids in the management of COVID-19 disease enhances the risk of mucormycosis.[3] Locally, this case of laryngeal mucormycosis represents the first reported instance of the infection confined to the subglottic area, making it a significant finding.

CASE REPORT

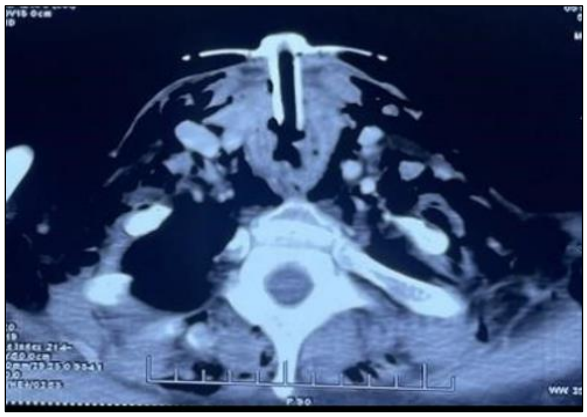
A 55-year-old male with a history of dysphagia and dyspnea was diagnosed with ongoing stridor. He was diabetic for four years and taking oral hypoglycemics. Physical examination revealed respiratory distress in a conscious patient. Vital signs noted were: blood pressure 125/80 mmHg, heart rate 96 beats/minute, temperature 37.6 °C, respiratory rate 26 breaths/minute, and no pulmonary crepitations. 70-degree endoscopic vocal cord examination revealed a circumferential growth in the supraglottic region with bilateral adducted cords with a narrow airway. An urgent tracheostomy was performed, removing a large amount of pus and necrotic tissue, indicating granulomatous disease in the subglottic region. Laboratory results showed poor glycemic control (fasting sugar 260mg/dl) and a leucocyte count of 16x10^9/L. He was started on intravenous antifungal and nebulization with ipratropium and Ventolin after which his condition improved. A computed tomography scan with intravenous contrast from the skull base to the diaphragm revealed circumferential edema, while the CT chest scan showed no significant findings. Endoscopic 70-degree camera evaluation revealed an edematous supraglottic region with reduced vocal cord mobility. A direct laryngoscopy under GA revealed subglottic growth and swollen vocal cord findings. A biopsy was taken from the subglottic site, showing fibroconnective tissue with squamous mucosa and broad aseptate fungal hyphae, indicating invasive fungal infection-favoring mucormycosis. (Figure) A repeat biopsy was performed via direct laryngoscopy from the deeper subglottic region and tracheostomy site due to the rare presentation of mucormycosis in the larynx. Moderate chronic nonspecific inflammation, fibrosis, and invasive fungal disease with aseptate hyphae consistent with mucormycosis were confirmed. Additional workup for chronic granulomatous infection, including chest X-ray, antinuclear antibody test, p-ANCA, and c-ANCA, was negative. A diagnosis of laryngeal mucormycosis was confirmed. The patient was advised to start intravenous liposomal amphotericin B, 200 mg twice daily, to complete a 2g dose, but he requested discharge and was lost to follow-up. Thus, recovery details remain unknown.



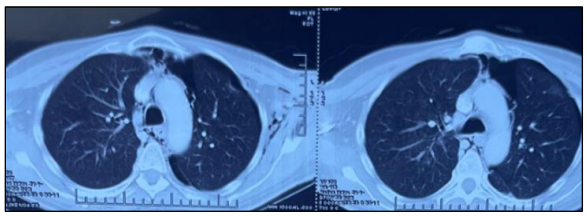
70-Degree examination; circumferential thickening and edematous the supraglottic region with bilateral vocal cord shows reduced movement affecting more on the right side.



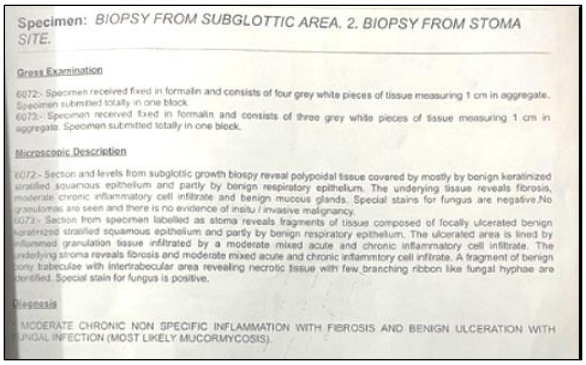
The axial view of the CT scan neck. There is hypodense soft tissue thickening surrounding the trachea extending to adjacent planes at the subglottic level



Tracheostomy tube in place



Unremarkable CT Chest



Specimen: BIOPSY FROM SUBGLOTTIC AREA. 2. BIOPSY FROM STOMA SITE.

Gross Examination

6072:- Specimen received fixed in formalin and consists of four grey white pieces of tissue measuring 1 cm in aggregate. Specimen submitted totally in one block.
6073:- Specimen received fixed in formalin and consists of three grey white pieces of tissue measuring 1 cm in aggregate. Specimen submitted totally in one block.

Microscopic Description

6072:- Section and levels from subglottic growth biospy reveal polypoidal tissue covered by mostly by benign keratinized stratified squamous epithelium and partly by benign respiratory epithelium. The underying tissue reveals fibrosis, moderate chronic inflammatory cell infiltrate and benign mucous glands. Special stains for fungus are negative.No granulomas are seen and there is no evidence of insitu / invasive malignancy.
6073:- Section from specimen labelled as stoma reveals fragments of tissue composed of focally ulcerated benign keratinized stratified squamous epithelium and partly by benign respiratory epithelium. The ulcerated area is lined by inflammed granulation tissue infiltrated by a moderate mixed acute and chronic inflammatory cell infiltrate. The underlying stroma reveals fibrosis and moderate mixed acute and chronic inflammtory cell infiltrate. A fragment of benign bony trabeculae with intertrabecular area revealing necrotic tissue with few branching ribbon like fungal hyphae are identified. Special stain for fungus is positive.

Diagnosis

1. MODERATE CHRONIC NON SPECIFIC INFLAMMATION WITH FIBROSIS AND BENIGN ULCERATION WITH FUNGAL INFECTION (MOST LIKELY MUCORMYCOSIS).

Histopathology: The tissue specimen of the patient was cross-verified by 2 laboratories which confirmed mucormycosis

Figure 1: Graphical Details of the case

DISCUSSION

This case of mucormycosis in the subglottic area is particularly rare, with few reports describing such a presentation. While mucormycosis is known to affect respiratory epithelial sites such as the nose, sinuses, and larynx, involvement of the subglottic region has been noted in only isolated cases.[4–6] Both medical and surgical interventions are currently recommended for the management of this disease.[7] Antifungal drug of choice is liposomal amphotericin B.[6,8] Only good glycemic control and antifungal therapy is a viable management option in mucormycosis.[9] The unique finding of isolated subglottic mucormycosis in our patient, without orbital, cerebral, or pulmonary spread, may be explained by the patient's underlying immunocompromised state due to diabetes.

Ethics approval and consent to participate:
The project received institutional research ethics board approval before the beginning of the study from the Shaikh Zayed Hospital Complex Ethics Board.

Consent for publication: NA.

Conflict of interest: The authors declare that they have no competing interests.

Data availability: NA.

Funding: None.

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Submitted: July 15, 2024	*Revised: September 26, 2024*	*Accepted: September 27, 2024*

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