

CASE REPORT

Isolated laryngeal Mucormycosis requiring laryngectomy

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Abstract

We report a case of isolated laryngeal mucormycosis in a patient who presented in diabetic ketoacidosis (DKA). The patient was managed with antifungal therapy and eventual total laryngectomy. To our knowledge, this is the first case presented of mucormycosis with isolated laryngeal involvement.

KEYWORDS

laryngectomy, laryngotracheal, Mucor

1 | INTRODUCTION

Mucormycosis, previously known as Zygomycosis, is a highly aggressive invasive fungal disease that primarily affects immunosuppressed and diabetic patients. The disease commonly affects the sinuses and lungs.¹ Laryngotracheal involvement is rare. On review of existing literature, there have been limited published cases of mucormycosis with laryngotracheal involvement.²⁻⁵ Treatment involves reversal of underlying medical conditions and antifungal therapy.¹ Mortality remains high even with aggressive management. The necessity of debridement and/or definitive surgical management is not well defined. Herein, we describe the case of a woman with diabetes mellitus (DM) with isolated laryngeal mucormycosis managed with antifungal therapy and eventual total laryngectomy. To our knowledge, this is the first case presented of mucormycosis with isolated laryngeal involvement.

2 | CASE REPORT

A 66-year-old woman with type II DM was admitted to a community hospital for diabetic ketoacidosis (DKA) without coma after 3 days of nausea, vomiting, altered mental status, and difficulty swallowing. No infection was suspected at the time; the patient was started on a DKA treatment protocol.

On hospitalization day 3, the patient developed stridor, prompting ENT consultation and flexible laryngoscopy revealing diffuse laryngeal erythema with hyperkeratotic areas and bilateral vocal fold hypomobility. These findings prompted concern for fungal laryngeal infection. Computed tomography (CT) was performed, which was unremarkable. The patient was started on IV solumedrol, racemic epinephrine, and oral fluconazole.

Her respiratory status deteriorated leading to intubation on hospitalization day 9. Subsequent direct laryngoscopy

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with biopsies revealed necrotic tissue and acute inflammation with negative fungal cultures. Antibiotics were initiated for positive cultures for klebsiella. Tracheostomy and gastrostomy tubes were placed at that time.

Repeat imaging was performed on hospitalization day 23 due to lack of clinical improvement, which demonstrated a progression of inflammation and fluid collection surrounding the larynx. The patient subsequently underwent surgical debridement with pathology consistent with invasive mucormycosis. The patient was started on amphotericin B and underwent a repeat endoscopic debridement a week later. The patient was transferred to a tertiary referral center for Laryngology and infectious disease management.

After transfer, the patient was continued on liposomal amphotericin B and started on isavuconazole and clindamycin. Repeat CT imaging revealed progressive erosive changes of the cricoid cartilage and likely involvement of the thyroid cartilage with infiltrative soft tissue surrounding the left cricoarytenoid joint and collapse of the subglottic trachea (Figure 1). Direct microlaryngoscopy (MDL) demonstrated irregular granulated tissue of the posterior glottis extending to the infraglottis (Figure 2). MDL with debridement was performed three times, with lack of resolution of abnormal tissue and concern for persistent disease though organisms were not found on pathology. At this time, the patient examination had clinically stabilized with bilateral vocal fold immobility, aphonia, and dysphagia consistent with a non-functional larynx. After extensive discussions with her care team, it was predicted her clinical examination would not significantly improve, and with concern for persistent fungal infection, the patient underwent a total laryngectomy on hospitalization day 67. Fungal organisms were noted on the final pathology report (Figures 3 and 4). The patient's post-operative course was complicated by a small salivary leak which resolved with conservative antibiotic management. The patient completed an additional 4 months (6-month total) course of isavuconazole upon discharge.

Limited follow-up was available due to patient's moving immediately upon discharge. On telephone follow-up 3 months and 1 year after hospitalization, she was doing well, with tracheoesophageal prosthesis (TEP) placed for voice rehabilitation.

3 | DISCUSSION

Mucormycosis is a life-threatening opportunistic infection most commonly affecting patients with conditions causing impaired host resistance. Poorly controlled DM is the most common predisposing factor, but other factors include cancer, AIDS, chemotherapy, organ transplantation, and iron chelation treatment.¹ Laryngotracheal involvement is rare and has significant morbidity and mortality. Mucormycosis isolated to the larynx has yet to be reported.

This case illustrates some of the difficulties involved in the treatment of this disease including the time-consuming nature of definitive diagnosis and rapid progression of the disease. Definitive medical treatment of our patient was delayed by the low yield of fungal cultures in the initial stages of the disease. By the time positive biopsies were obtained, the predicted return of functionality to the patient's larynx was guarded with the collapse of the laryngeal framework. Regardless of the antifungal coverage, early laryngoscopy and biopsy in the stridulous patient with associated risk factors is essential.

Surgical debridement is an adjuvant treatment used in rhinocerebral mucormycosis to maintain disease control as the underlying immunocompromise is reversed. Its utility and necessity are unclear in laryngotracheal disease. A review of the literature reveals 8 cases of local disease to the pharynx, subglottis, or trachea.^{2–6} Seven of eight cases underwent surgical debridement or resection, of which 5 survived. In the two cases in which the patient did not survive, they died of disseminated disease with underlying neutropenia with comorbidities of HIV and

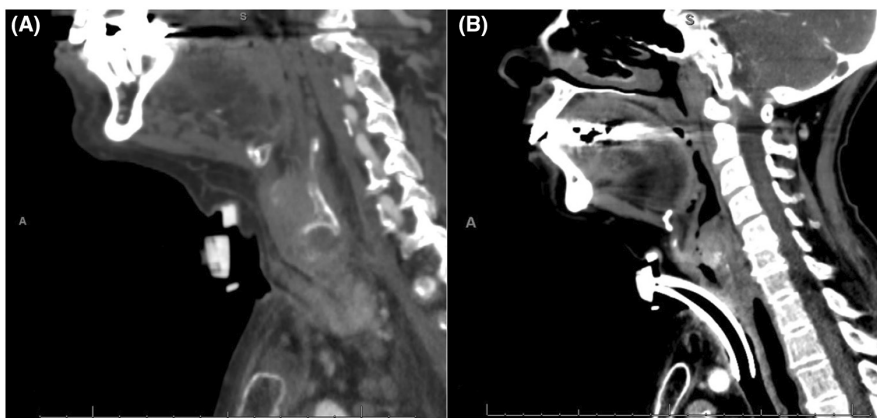


FIGURE 1 Sagittal CT neck imaging with (A) erosive changes of the cricoid and thyroid cartilage seen on bone-window, and (B) infiltrative soft tissue surrounding the left cricoarytenoid joint on soft-tissue window.

FIGURE 2 Endoscopic views from MDL demonstrating an (A) edematous supraglottis, and (B) irregular granulated tissue of the posterior glottis extending to the infraglottis.

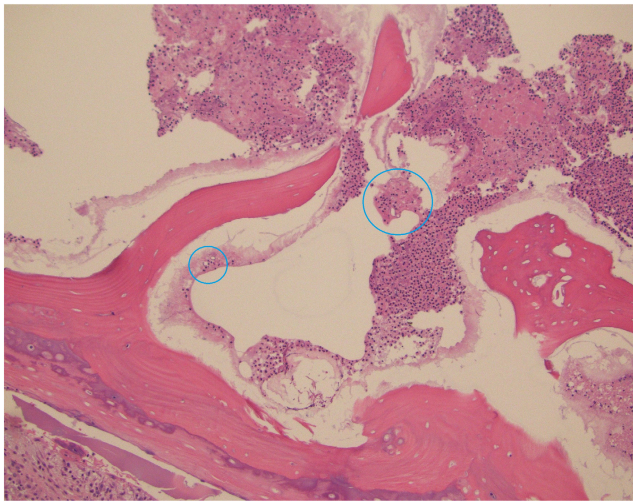
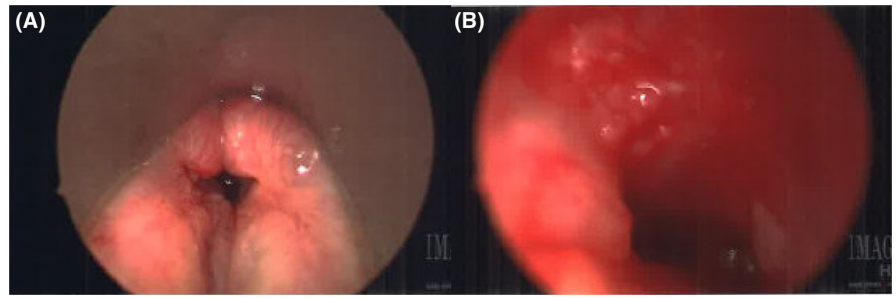


FIGURE 3 Hematoxylin and Eosin (H&E) stain 200x, Non-viable ossified tracheal cartilage with infiltrative neutrophilic response. Fungal hyphae are few in number (blue circle).

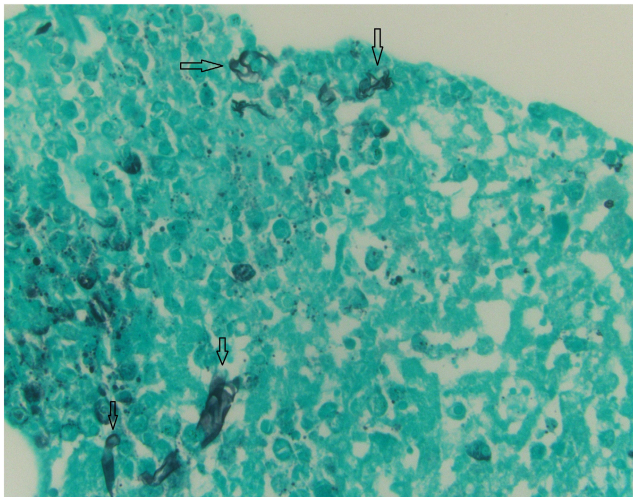


FIGURE 4 Grocott Methenamine Silver (GMS) stain 400x, Fibrinopurulent debris with large hyphal structures demonstrating a ribbon-like appearance (black arrows), consistent with Zygomycetes-type fungi.

Burkitt's lymphoma³ and the other did not undergo surgical treatment with underlying DM.⁴ Of note, 6 of the 8 cases had an underlying DM diagnosis.^{2,3,5}

In contrast to rhinocerebral mucormycosis, surgical resection after the initial diagnosis may be indicated in laryngotracheal disease. As presented in this case, surgery provides two roles in confirming clearance of any residual disease that was hidden from superficial biopsy and may provide improved functionality and quality of life. In parallel, Mohnidra et al. described debridement of necrotic tissue to clear disease and provide a patent airway for voice rehabilitation.⁵ This suggests diligent monitoring of abnormal tissue and considering definitive resection if abnormal tissue persists is critical as persistent disease was present in this case.

4 | CONCLUSION

Laryngeal mucormycosis is an extremely rare and exceedingly morbid condition. Clinical threshold for direct laryngoscopy and biopsy should be low for patients with associated risk factors who develop stridor. Early diagnosis and initiation of amphotericin are the most important factors for lowering morbidity and mortality. Despite antifungal therapy, surgical resection including laryngectomy may be indicated to provide better quality of life and definitive treatment in this population.

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None.

AUTHOR CONTRIBUTIONS

Taylor G. Lackey was involved in conception and design, acquisition of data, interpretation of data, manuscript preparation, and presentation of research. James R. Duffy was involved acquisition of data and manuscript preparation. Carrie Marshall was involved in acquisition of data and manuscript preparation. Daniel S. Fink was involved in conception and design, acquisition of data, interpretation of data, and manuscript preparation.

CONFLICT OF INTEREST

The authors declare no conflict of interest, financial relationships or support relevant to this manuscript. This research received no specific grant from any

funding agency in the public, commercial, or not-for-profit sectors.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

CONSENT

The Colorado Multiple Institutional Review Board (COMIRB) does not require single patient case studies to be submitted for review because it is not considered human subject research (<https://research.cuanschutz.edu/comirb/home/get-help/human-subject-research>). Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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