Cutaneous mucormycosis involving a colostomy site

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Abstract

Cutaneous mucormycosis is a rapidly advancing fungal infection that most commonly occurs due to airborne spread or direct inoculation and requires early detection and prompt treatment for optimal survival. Major risk factors include diabetes, transplantations, malignancies, surgical procedures, and HIV. Diagnostic criteria are based on microscopy and culture. We present an immunocompromised patient with cutaneous mucormycosis that developed in a peristomal ulcer following a hemicolectomy procedure. Histopathologic evaluation was indicative of mucormycosis. Intravenous posaconazole treatment was initiated, but unfortunately, the patient's condition deteriorated and he passed away.

Keywords: colostomy, cutaneous mucormycosis, Mucorales, zygomycosis

Introduction

Cutaneous mucormycosis, previously called zygomycosis, is a potentially deadly fungal infection belonging to the order Mucorales that is usually acquired via direct inoculation caused by trauma, which includes surgeries, burns, and injections [1,2]. Individuals who are diabetic, neutropenic, or immunocompromised have an elevated risk of acquiring mucormycosis [3]. Hematological malignancies have also been identified as a risk factor [2].

The general clinical presentation of mucormycosis is an indurated, erythematous to violaceous plaque or ulceration, with necrotic eschars in later stages [4]. Areas commonly affected include the arms and legs, but the face, chest, and back can also be affected [2]. Rapid dissemination can occur in mucormycosis, underscoring urgent treatment to improve outcomes [1]. This report presents a clinical case of cutaneous mucormycosis at a colostomy site in the context of various comorbidities with dermatologic and histopathologic analysis.

Case Synopsis

A 53-year-old man with a previous history of alcoholic cirrhosis, congestive heart failure, and recent left hemicolectomy for treatment of adenocarcinoma of the sigmoid colon presented with a violaceous ulcerated plaque adjacent to a colostomy site. Following the colostomy procedure, the patient had experienced multiple complications including anastomotic leak.

Physical examination demonstrated an ulcer adjacent to the stoma, along with violaceous patches and plaques surrounding the colostomy site (**Figure 1**). Necrotic tissue at the base and superficial eschar formation was also noted.

An abdominal skin biopsy of the peristomal ulcer was obtained, demonstrating deep fungal elements diagnostic of mucormycosis. Histopathology of the ulcer edge demonstrated granulomatous inflammation with bile deposition along with neutrophils and plasma cells on H&E stain (**Figure 2A**). Broad ribbon-like hyaline pauci-septated hyphae with right-angle branching were also seen, which was consistent with findings of the order

Figure 1. *Peristomal ulcer with violaceous plaques and eschar formation.*

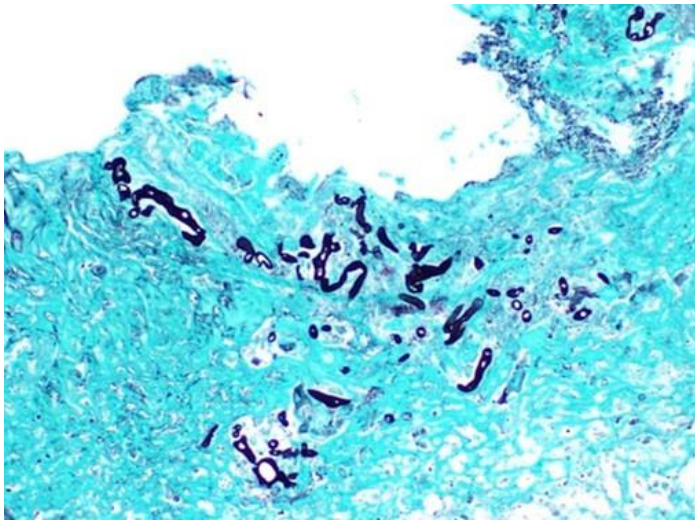

Figure 3. *Grocott methenamine silver (GMS) stain highlighting fungus, 200×.*

Mucorales (**Figure 2B**). These findings were highlighted with Grocott methenamine silver stain (**Figure 3**). Angioinvasion was not seen. Tissue culture failed to grow any fungal organisms. Serological testing for (1,3)-Beta-D-Glucan was negative. Unfortunately, the patient's condition worsened; he experienced multiorgan failure and he passed away.

Case Discussion

Mucormycosis has a variety of subtypes, including pulmonary, gastrointestinal, and cutaneous, with the rhino-orbital-cerebral form being the most common [1]. Involvement of an ostomy site is a rare clinical presentation. One report identified two cases of mucormycosis related to ostomy bags [5]. Another case of ostomy site infection by mucormycosis has been reported [6]. These cases are detailed in **Table 1**.

The classic clinical finding of cutaneous mucormycosis is a necrotic eschar with an erythematous, indurated background [7]. Diagnosis of cutaneous mucormycosis is usually dependent on histopathologic findings and culture [1]. Broad, ribbon-like hyphae with right-angle branching are a characteristic histological finding [8]. Other findings

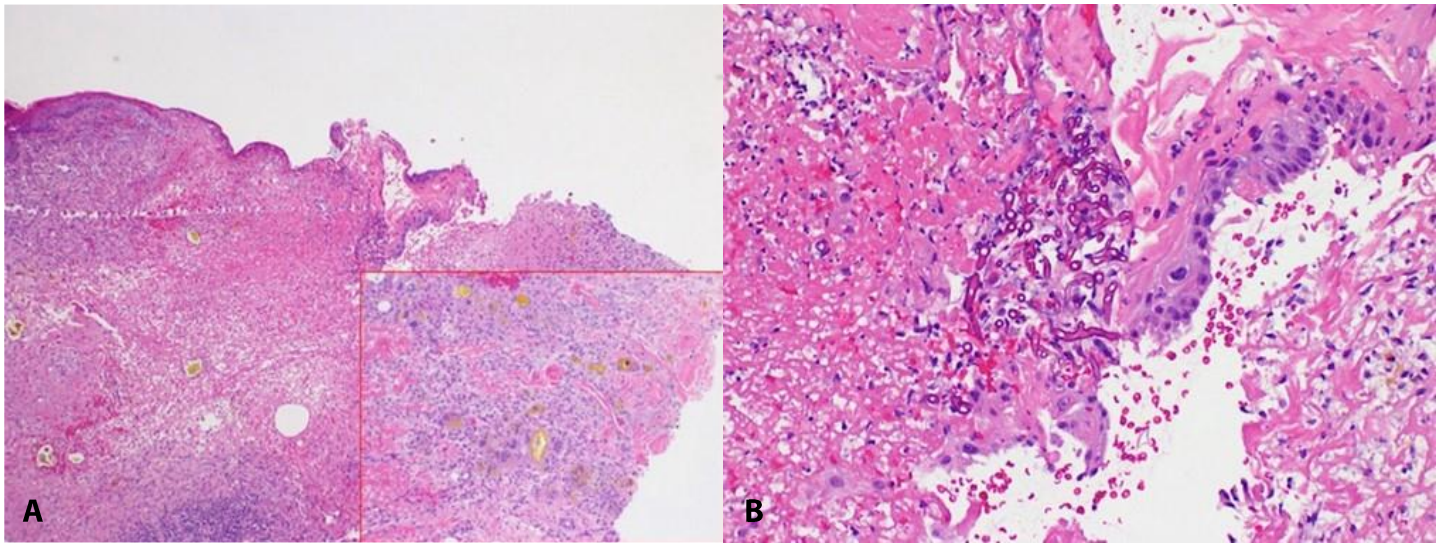

Figure 2. *H&E histopathology.* ***A)*** *Ulcer edge with a dermal infiltrate of neutrophils and histiocytes with bile deposition, 40×. Inset: bile in giant cells. Bile pigment is yellow.* ***B)*** *Fungus with broad ribbon-like hyaline pauci-septate hyphae. Right angle branching is noted, 200×.*

suggestive of cutaneous mucormycosis include dermal infiltration by neutrophils, tissue necrosis, and in chronic lesions, suppurative granulomatous inflammation with multinucleate giant cells [8]. Treatment often involves surgical debridement and antifungal therapy, such as amphotericin B or posaconazole [9].

An important condition in the differential diagnosis for ulceration involving a stoma site is peristomal pyoderma gangrenosum (PPG). Peristomal pyoderma gangrenosum typically presents as a well-demarcated ulceration with an undermined, violaceous border and is mostly seen in patients with inflammatory bowel disease [10]. A superficial eschar would not be expected in PPG, in contrast to the typical presentation of cutaneous mucormycosis. Histopathologic findings can also distinguish PPG from mucormycosis. Peristomal pyoderma gangrenosum typically demonstrates a dense dermal infiltrate of neutrophils [10,11]. Granulomatous inflammation and tissue necrosis are not dominant features in PPG. Importantly, special stains will fail to reveal any fungal organisms in biopsies of PPG.

Conclusion

Mucormycosis is a fungal infection that typically affects diabetic and immunocompromised individuals. Involvement of ostomy sites is rare. Treatment of cutaneous mucormycosis is difficult due to its progressive nature. Therefore, early identification and treatment is crucial.

Potential conflicts of interest

The authors declare no conflicts of interest.

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Table 1. *Cases of mucormycosis involving ostomy sites.*

Patient	Age	Sex (M/F)	Race	Comorbidities	Location	Clinical Presentation	Pathology: H&E	Special Stains	Other testing/ Culture	Treatment	Follow-Up
1 [5]	47	M	N/A	N/A	Ileostomy	Chest pain, severe ischemia in LE, hepatic necrosis, and ARF s/p CABG surgery. Ischemic bowel requiring resection and ileostomy	N/A	N/A	Mucorales from culture Isolate grew *R. arrhizus*	Originally received BSA and caspofungin, later switched to amphotericin B	Death via multiorgan failure
2 [5]	76	F	N/A	N/A	Ileostomy	Mucoid diarrhea and abdominal pain. Colonoscopy revealed focal colitis c/b perforated cecum, requiring abdominal colectomy with ileostomy. Necrotic stoma	N/A	N/A	Mucorales from culture. Isolate grew *R. arrhizus*	Debridement was performed and amphotericin B was given	Patient recovered
3 [6]	48	F	Caucasian	Congenital malrotation, juvenile diabetes, prior small bowel and pancreas transplant c/b chronic rejection and renal failure	Ileostomy	Acute hypotension, fever, and lethargy s/p hemodialysis	Ribbon-like branched structures	GMS-stained fungal organisms	Gram-negative rods were seen on culture. Culture grew *Rhizopus,* confirmed by pathology following surgical debridement	Abdominal wall debridement, amphotericin B, voriconazole, and isavuconazole	Death via multiorgan failure
4	53	M	Caucasian	Alcoholic cirrhosis, CHF, and adenocarcinoma of the sigmoid colon	Ileostomy	Erythematous peristomal ulcer s/p colostomy procedure c/b multiple infections while	Ribbon-like pauciseptated hyphae with right-angle branching. Bile	GMS-stained fungal organisms	No fungal organisms were grown on culture. Skin biopsy of ulcer had fungal	IV posaconazole and IV cefepime	Death via multiorgan failure

						in the hospital. Anastomotic leak also occurred following a hemicolectomy	deposition and infiltration of neutrophils and plasma cells were also seen		elements characteristic of mucormycosis		

Patient 4 is from our case report.
ARF, acute renal failure; BSA, broad-spectrum antibiotics; CABG, coronary artery bypass graft; CHF, congestive heart failure.; GMS, Grocott methenamine silver; H&E, hematoxylin and eosin; IV, intravenous; LE, lower extremities.