Extensive gastric mucormycosis: a rare case presentation and literature review

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European Journal of Medical Case Reports

Volume XX(XX):01–06 DOI: XXXX





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ABSTRACT

Background: Rarely seen, gastric mucormycosis mostly occurs in people with weakened immune systems. Among whom diabetes mellitus often plays a key role, and rare case reports exist.

Case Presentation: We describe a 31-year-old woman with poorly controlled type 1 diabetes mellitus who had abdominal pain with shock with tachycardia leading to Intensive Care Unit admission. The endoscopic exam revealed wide-ranging involvement of the stomach from the fundus to the antrum, and also involving the GE junction and distal esophagus. Such extensive involvement has not been reported yet. Intensive treatment with antifungal drugs and supportive therapy failed to stop the severe sepsis, and the patient passed away after 20 days in the hospital.

Conclusion: Extensive gastric mucormycosis is marked by its aggressiveness and difficulty in diagnosis, so health professionals should be especially careful in identifying patients at risk. Different studies on early recognition and effective approaches for management are discussed.

Keywords: Gastric mucormycosis, diabetes mellitus, fungal infection, antifungal therapy, gastrointestinal necrosis.

Type of Article: CASE REPORT Specialty: Gastroenterology

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 Received:
 24 May 2025

 Revised (1):
 31 July 2025

 Accepted:
 05 August 2025

Introduction

The fungal infection mucormycosis is brought on by the order Mucormycetes order (Mucorales) and spreads quickly [1]. Early on, mucormycosis primarily leads to infection in the sinuses and brain, although infection of the stomach and gut is often put off and is lethal in immunocompromised people, those with diabetes and HIV being most at risk [2]. An increasing number of reports from people with normal immune systems go against the current patterns [3,4]. A major sign of this infection is that it spreads rapidly within blood vessels, which can result in clotting, dying tissue, and serious problems such as perforation [5,6].

The symptoms of gastric mucormycosis are not specific, so diagnosis is difficult and requires tissue diagnosis. Despite antifungal therapy, mortality is still high, so surgery is often required [7,8]. This case describes an extensive degree of involvement in the esophagus and stomach

in a type 1 diabetic, adding a new form to the list of symptoms this disease produces.

Case Description

A 31-year-old female patient who has had type 1 diabetes mellitus for 10 years and is treated with insulin presented to the emergency department with a sudden, sharp pain across her whole upper abdomen. During the week before hospitalization, she felt progressively sick, with vomiting, decreased appetite, and dysphagia. Her diabetes was not well controlled as she was non compliant with insulin doses Hemoglobin A1c was 10.2, done 2 weeks back. Apart from uncontrolled diabetes, there is no history of corticosteroid use, organ transplant, or any other causes of immunosuppression. During hospitalization, the person was seriously ill and their vital signs were unstable. Her blood pressure was 88/60, PR was 112/minute, respiratory rate was 22, and her oxygen level was 94% on room air. On abdominal examination abdomen was tender with

maximum tenderness in the upper third, without obvious signs of peritonitis. Blood tests found high blood glucose, increased white blood cells, neutrophils predominant, increased C-reactive protein and procalcitonin levels, and metabolic acidosis on arterial blood gas.

In view of an acute abdomen with unstable vitals, she was hospitalized in the intensive care unit (ICU). Intravenous (IV) fluid as per sepsis protocol and empirical broad-spectrum antibiotics were started. With insulin and IV fluids, diabetic control was made. UGI endoscopy was done, which revealed ulcers starting from the distal esophagus till the fundus, body, and antrum with areas of hyperemia and necrosis. Such extensive involvement has not been seen before with gastric mucormycosis cases (Figure 1).

Because of the massive infiltration in a poorly controlled diabetic aggressive invasive fungal infection was suspected, and multiple biopsies were taken via endoscope. A histology sample showed wide, angular-shaped hyphae with common right-angle branching expanding into the wall layers, thus proving mucormycosis (Figure 2).

IV liposomal Amphotericin B (Amp B) was started at a dose of 5 mg/kg/day, with IV fluid and insulin therapy. Despite using every possible treatment—vasopressors, assistance with breathing, and antifungal drugs her overall condition deteriorated with severe sepsis with multiorgan dysfunction, with acute kidney injury and ongoing low blood pressure. Surgical intervention could not be planned due to the poor performance status of the patient.

After 20 days of ICU care, the patient succumbed to overwhelming sepsis. This tragic outcome underscores the fulminant nature of extensive gastric mucormycosis, especially when compounded by poorly controlled type 1 diabetes mellitus.

Discussion

Gastric mucormycosis is very rare, most often occurring in people with low immunity, mainly those with diabetes. We find our case to be remarkably significant because it includes the entire stomach (fundus, body, and antrum) with the gastroesophageal junction and esophagus, all without interruption. So far, no case of this severe invasion of both the stomach and esophagus has been found in medical publications, as described in Table 1.

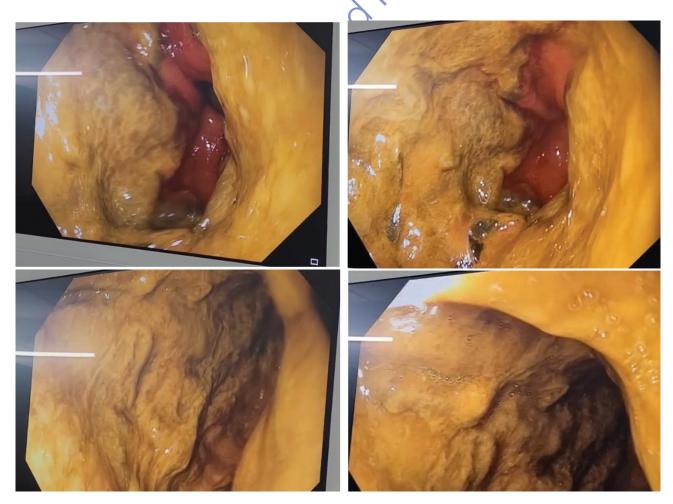


Figure 1. Visualization of the entire stomach affected with Mucormycosis (areas of ulcers and necrosis with hyperemia fundus, body, and antrum).

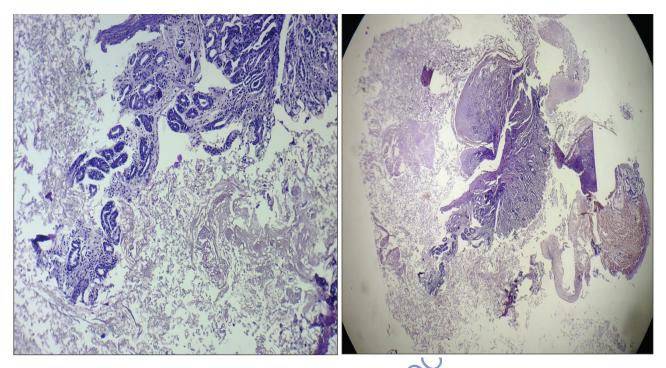


Figure 2. Visualization of broad, non-septate, ribbon-like hyphae with right-angle branching infiltrating the gastric wall, suggestive of mucormycosis.

 Table 1. Rare gastric mucormycosis cases: clinical features, management, and outcomes.

| REFERENCE | AGE/ GENDER | RISK FACTORS | CLINICAL PRESENTATION | SITE OF INVOLVEMENT | DIAGNOSTIC METHOD | TREATMENT | OUTCOME |
|-----------------------|------------------|-------------------------------------------------------------------------------|-------------------------------------------------|----------------------------------------------------------------------------------|-------------------------------|----------------------------------------------|---------------------------------|
| Current case | 31/F | Type 1 diabetes (10 years), poorly controlled (HbA1c 10.5%) | Severe abdominal pain, hypotension, vomiting | Entire stomach (fundus, body, antrum), GE junction, distal esophagus | Endoscopy + Histopathology | IV Liposomal Amphotericin B + ICU care | Fatal (sepsis after 20 days) |
| Kgomo et al. [1] | 45/M | Diabetes mellitus | Abdominal pain, vomiting | Gastric body and antrum | Endoscopy + Histopathology | Amphotericin B + Surgery | Survival |
| Tormane et al. [2] | 60/F | Immuno suppression (transplant) | Gastrointestinal bleeding | Gastric fundus | Endoscopy + Culture | Amphotericin B | Survival |
| Alfano et al. [7] | 54/M | Liver and kidney transplant | Abdominal pain, Gastrointestinal bleeding | Gastric mucosa | Histopathology | Amphotericin B + Surgery | Survival |
| Janakiram et al. [8] | 55/M | Alcohol use disorder (non- diabetic) | Abdominal pain | Gastric antrum | Endoscopy + Biopsy | Medical management only | Survival |
| Termos et al. [5] | 58/F | Diabetes mellitus | Gastric perforation, abdominal pain | Entire stomach (total stomach necrosis) | Imaging + Histopathology | Surgery + Amphotericin B | Fatal |
| Rivero et al. [14] | 50/M and 36/F | Diabetes mellitus (poor control) | Abdominal pain, vomiting | Gastric body and antrum | Histopathology + Culture | Amphotericin B | One survival, one fatal |
| Khsiba et al. [15] | 39/F and 45/M | Diabetes mellitus | Severe abdominal pain, vomiting | Gastric body and antrum | Endoscopy + Histopathology | Amphotericin B + Surgery | |

Two cases in a case report described gastric mucormycosis, which only led to local necrosis and did not involve the esophagus [9]. In the same manner, two gastric mucormycosis cases described by Khsiba et al. [10] had diffuse gastric ulcers, yet neither affected the esophagus nor the entryway of the stomach. In 2018, Kgomo et al.

[1] described a man aged 45, diabetic involving the gastric body and antrum, and he recovered from the disease after Amp B treatment and surgery. The authors describe how an immunosuppressed transplant recipient with gastric fundus disease survived only by using antifungal drugs, indicating that prompt identification and prompt treatment are essential for an improved outcome [2]. In the same way, Alfano et al. [7] highlighted that gastric mucormycosis in transplant recipients often leads to worse outcomes and usually requires both surgery and medication. Our case study points out that severe mucormycosis may happen in young diabetics who are non-compliant to insulin therapy, leading to low immunity.

In the clinical setting, severe abdominal pain, low BP, tachycardia, tachypnea, and hyperglycemia are attributed to the systemic inflammatory response [1,9]. High blood sugar and acidosis decrease neutrophil chemotaxis and phagocytosis, which encourages Mucorales to enter blood vessels [6,7]. When there are large, non-septate hyphae that branch at right angles, seen on biopsy, this is the primary means to diagnose [1,2].

Unlike previous reports, which had spotty or limited involvement in the stomach, our patient experienced necrosis extending over the whole stomach, the gastroesophageal junction, and the esophagus. Because of such extensive involvement with poor performance status, surgical therapy was not feasible.

Our patient died after 20 days, even though both early liposomal Amp B and good blood sugar control were started. When the disease is widespread and the person seeks care late, the situation often results in death, unlike successful cases treated by antifungal drugs, sometimes with surgery [2,8]. Another group of cases found that many patients with diffuse gastric mucormycosis did not survive, suggesting that early recognition is very important [9-11]. We have added a new area by showing that mucormycosis may also affect the gastroesophageal junction and esophagus, alongside the stomach.

Few cases of gastric mucormycosis have been reported and show important information on who these patients are, what risks they face, how the disease shows up, how it is detected, treated, and what the outcomes are (Table 1). Once again, diabetes mellitus and immunosuppression are the major risk factors, as shown in our case by poorly controlled diabetes. Presentation may be nonspecific abdominal signs or more serious complications, including gastric perforation. Early diagnosis by endoscopy and examination under a microscope is vital for applying early drugs against fungal infections. Most individuals receive Amp B given by liposomes, and surgery is only performed when the disease is severe. Responses are unpredictable, so it is important to identify the infection early and give specific treatment to help the patient.

Conclusion

A young female patient, 31 years old with type 1 diabetes mellitus, with widespread gastric mucormycosis, such a rare infection can be deadly. We see that when mucosal infection is severe, the person deteriorates fast and often has a poor outcome, even when treated with antifungals and highly supportive care. Recognizing the disease quickly by closely assessing the symptoms, performing an endoscopy with biopsies can make a big difference in the patient's outcome. Due to the high number of deaths linked to diffuse gastric mucormycosis, mostly in those with diabetes, new research is required to improve rapid diagnosis, extra therapeutic approaches, and standard management plans. Clinicians should maintain a high index of suspicion for invasive gastric mucormycosis in diabetic patients with unexplained abdominal pain and systemic instability.

What is new?

The case study describes the unprecedented extent of gastric involvement, spanning from the fundus to the antrum and affecting the gastroesophageal junction and distal esophagus, a scenario not previously reported. This rare presentation in a 31-year-old woman with poorly controlled type 1 diabetes mellitus, coupled with the failure of antifungal therapy, underscores the severity and aggressive nature of the disease. The case contributes to the literature by highlighting the challenges in early diagnosis and the management of extensive gastric mucormycosis, offering valuable insights for clinicians.

List of Abbreviations

ABG Arterial blood gas
Amp B Amphotericin B
BID Bis in die
CRP C-reactive protein

F/F Female/Male (used for gender specification

in cases)

GI Gastrointestinal
HbA1c Hemoglobin A1c
ICU Intensive Care Unit
IV Intravenous

Mucorales Mucormycetes Order

Conflicts of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

Funding

None.

Consent for publication

Informed consent was obtained from the study participants, briefing on the purpose of the study.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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Summary of the case

| 1 | Patient (gender, age) | 31 years, female | |
|---|-----------------------|---------------------------------------------------------------------------|--|
| 2 | Final Diagnosis | Extensive Gastric Mucormycosis in a uncontrolled Type 1 Diabetes Mellitus | |
| 3 | Symptoms | Pain, Vomiting, Lethargy | |
| 4 | Medications | Insulin, Liposomal Amphotericin B, Broad Spectrum Antibiotics | |
| 5 | Clinical Procedure | Endoscopy | |
| 6 | Specialty | Gastroenterology | |