# **Clinical Case Reports International**

## 9

## Gastric Mucormycosis: A Case Report and Review of the Literature

Tormane MA, Laamiri G\*, Mroua B, Gazzah H, Beji H, Zribi S and Touinsi H

Department of General Surgery, Hospital Mohamed Taher Maamouri, Tunisia

#### Abstract

**Introduction:** Gastric mucormycosis is a rare and potentially life-threatening fungal infection that can occur in immunocompromised individuals. We present here a case of gastric mucormycosis which was revealed by abdominal pain and hematemesis that was treated by gastrectomy combined with antifungal.

**Case Report:** This report illustrates the case of a diabetic patient who presented with abdominal pain and hematemesis. Gastroesophageal fibroscopy and histopathological examination confirmed the presence of mucormycosis which was treated with fluconazole, amphotericin and gastrectomy.

**Discussion:** Mucormycosis is a rare fungal infection that primarily affects immunocompromised individuals including those with poorly controlled diabetes. The symptoms are quite diverse. Diagnosis is established by detailed microscopic and histopathological evaluation. Treatment varies from single antifungal treatment to gastrectomy. In our case the patient was treated by antifungal therapy combined with gastrectomy.

**Conclusion:** Gastric mucormycosis is a very rare entity. Clinical manifestations are not very characteristic. Antifungal treatment and surgical debridement are the pillar of treatment. Gastrectomy should be performed as soon as possible if single anti-fungal treatment and surgical debridement are unavailing.

#### Introduction

Mucormycosis is a very rare and potentially fatal infection caused by fungi of the subphylum Mucoromycotina. It can touch any organ system, but the most common are the airways, sinuses, orbit, lungs or brain [1]. Even though rare, gastrointestinal involvement in mucormycosis has also been reported [2].

### OPEN ACCESS

\*Correspondence: Ghazi Laamiri, Department of General Surgery, Hospital Mohamed Taher Maamouri, Nabeul, and University Tunis El Manar, Tunis, Tunisia Received Date: 03 Aug 2023 Accepted Date: 23 Aug 2023 Published Date: 28 Aug 2023

#### Citation:

Tormane MA, Laamiri G, Mroua B, Gazzah H, Beji H, Zribi S, et al. Gastric Mucormycosis: A Case Report and Review of the Literature. Clin Case Rep Int. 2023; 7: 1603.

**Copyright** © 2023 Laamiri G. This is an open access article distributed under the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited. It accounts for around 7% of all recorded cases and it is associated with high mortality rates particularly in immunocompromised patients with hematological malignancies, transplant recipients and poorly controlled diabetes [3]. Clinical signs generally depend on the initial site of onset.

The main route of infection is inhalation. However, for the gastrointestinal tract, ingestion of filaments is involved [4]. Early surgical management and removal of necrotic tissue, appropriate antifungal treatment and control of risk factors such as diabetes are the cornerstones of effective management of this infectious disease [2].

We present here a case of gastric mucormycosis which was revealed by profuse hematemesis and abdominal pain that successfully treated by antifungal and timely gastrectomy.

#### **Case Presentation**

A 56-year-old man, unbalanced diabetes, complained of abdominal pain, fever and nausea. He presented gradually worsening anorexia and recent profuse hematemesis.

The general physical examination revealed fever, tachycardia and laboratory investigations showed systemic inflammation and acute renal failure.

Urgent gastroesophageal fibroscopy showed an ulcerated and proliferative tumor involving the entire gastric body, sparing the antrum, with extensive deep ulcerations covered with fibrin and hard peripheral mucosal buds (Figure 1).

Biopsies were obtained and histopathological examination revealed highly active antral gastritis



**Figure 1:** Gastroesophageal fibroscopy revealing an ulcerated and proliferative tumor involving the entire gastric body.



Figure 2: Gastrectomy specimen.



Figure 3: Histopathological slide demonstrating spores and non-septate mycelial filaments of the gastric mucormycosis.

with *Helicobacter pylori* infection, areas of intestinal metaplasia, as well as the presence of mucormycosis (spores and non-septate mycelial filaments) and gastric candidiasis. the patient's renal function has been adjusted and he was treated with fluconazole, amphotericin, antibiotics targeting *H. pylori*, and a proton pump inhibitor.

The evolution was highlighted by a clinical worsening with persistent hematemesis, tachycardia and a decrease in hemoglobin despite transfusions.

We opted for emergency surgical treatment. A total gastrectomy and esophagojejunostomy was performed, the specimen was sent for histological analysis (Figure 2) and the patient was referred to intensive care. The clinical and biological course was excellent.

A brain-face-chest-abdomen and pelvis computed tomography was performed, which did not show any other location of the disease.

Anatomopathological examination of the specimen confirmed the diagnosis of gastric mucormycosis (Figure 3).

#### Discussion

We reported a successful surgical treatment for gastric mucormycosis. The main strength of our work is the choice of the surgical treatment and its timely performance in cases of single medical treatment failure.

Mucormycosis is a rare but life-threatening fungal infection that primarily affects immunocompromised individuals, including those with poorly controlled diabetes mellitus, recipients of organ transplant [5,6], or even with an intact immunity system [2,7,8].

These underlying factors not only predispose to the disease but also worsen the outcome of patients [4]. Diabetic ketoacidosis, as seen in our patient with a history of poor therapeutic compliance creates a favorable environment for fungal growth due to elevated blood glucose levels and impaired host defense mechanisms [9,10].

The transmission mechanism of mucormycosis entails ingestion or inhalation of spores, and direct inoculation of damaged mucocutaneous surfaces [11].

While pulmonary and rhinocerebral are the most common presentations [1,2], gastrointestinal mucormycosis was extremely rare [1-3].

It accounts for around 7% of all recorded cases, and has a poor prognosis, with a mortality rate of 85% [12]. The stomach is most often involved which is followed by the colon and ileum [3].

The symptoms are quite diverse, ranging from non-specific abdominal pain, fever and vomiting to hematemesis, hematochezia and even gastrointestinal perforation [2,13,14]. There are unfortunately no PCR-based or serological tests available for early diagnosis [15].

Diagnosis is established by detailed microscopic and histopathological evaluation of a tissue biopsy that shows aseptate, fungal hyphae in proximity to necrotic areas [3,16,17]. This can be obtained endoscopically or surgically [17].

The cornerstone of invasive gastric mucormycosis treatment is intravenous antifungal therapy, combined in some cases with surgical debridement or gastrectomy [2,4,17].

Rapid initiation of antifungal therapy improved patients' prognosis whereas delayed treatment has been associated with a two-fold increase in mortality rates [18]. While no consensus has been reached on the optimal antifungal treatment, amphotericin B is recommended as first-line therapy [2,3]. Polyene antifungal agents and liposomal formulations have proven to be effective due to their low nephrotoxicity, however the role of combination antifungal therapy is uncertain [1]. Single antifungal treatment is generally insufficient. Surgical debridement of necrotic tissue, sometimes leading to total gastrectomy, is necessary for the effective treatment of invasive mucormycosis [6,19,20].

Gastric mucormycosis generates tissue necrosis, which can impede the penetration of antifungal agents to the site of infection. Rapid debridement of all damaged tissue, seems appropriate in order to prevent the spread of mucormycosis to adjacent structures and to reduce the mass of infecting molds [5]. In fact, Roden et al. trial revealed that surgery was an independent predictor of improved outcomes [6]. In addition, A. Chakrabarti and al suggested that combination of surgery and amphotericin B was significantly better

#### Tormane MA, et al.,

than single antifungal therapy in terms of survival rate (79.6% *vs.* 51.7% patient survival) [10].

In our case, we opted for a total timely gastrectomy after failure of the single medical treatment.

In summary, we reported the case of a 56-year-old diabetic patient complaining of abdominal pain and hematemesis due to a gastric mucormycosis diagnosed by urgent gastroesophageal fibroscopy and histopathological examination of a tissue biopsy and treated successfully by an antifungal treatment and an early total gastrectomy.

Our case highlights that early diagnosis, prompt initiation of antifungal therapy, management of underlying conditions and a timely indication of surgical treatment, are crucial for favorable outcomes.

#### Conclusion

Gastric mucormycosis is a very rare entity that can affect patients who are usually predisposed, such as immunocompromised patients and diabetics, but also immunocompetent patients.

Clinical manifestations are not very characteristic and the confirmation of diagnosis is based on anatomopathological examination.

Early antifungal therapy and surgical debridement of all necrotic tissues are the pillar of treatment.

#### **References**

- 1. Spellberg B. Gastrointestinal Mucormycosis. Gastroenterol Hepatol. 2012;8(2):140-42.
- Sehmbey G, Malik R, Kosa D, Srinivasan I, Chuang KY, Bellapravalu S. Gastric ulcer and perforation due to mucormycosis in an immunocompetent patient. ACG Case Rep J. 2019;6(8):e00154.
- Kulkarni RV, Thakur SS. Invasive gastric mucormycosis: A case report. Indian J Surg. 2015;77(1):87-9.
- Khsiba A, Moalla M, Nechi S, Bani A, Elloumi A, Jemal S, et al. Fatal invasive gastric mucormycosis: Two case reports. Clin Case Rep. 2022;10(9):e6330.
- Skiada A, Lanternier F, Groll AH, Pagano L, Zimmerli S, Herbrecht R, et al. Diagnosis and treatment of mucormycosis in patients with hematological malignancies: Guidelines from the 3<sup>rd</sup> European Conference on Infections in Leukemia (ECIL 3). Haematologica. 2013;98(4):492-504.
- Roden MM, Zaoutis TE, Buchanan WL, Knudsen TA, Sarkisova TA, Schaufele RL, et al. Epidemiology and outcome of zygomycosis: A review of 929 reported cases. Clin Infect Dis. 2005;41(5):634-53.

- Abreu BFBB de, Duarte ML, Santos LR dos, Sementilli A, Figueiras FN. A rare case of gastric mucormycosis in an immunocompetent patient. Rev Soc Bras Med Trop. 2018;51:401-2.
- Wotiye AB, KS P, Ayele BA. Invasive intestinal mucormycosis in a 40-yearold immunocompetent patient - A rarely reported clinical phenomenon: A case report. BMC Gastroenterol. 2020;20(1):61.
- Skiada A, Pagano L, Groll A, Zimmerli S, Dupont B, Lagrou K, et al. Zygomycosis in Europe: Analysis of 230 cases accrued by the registry of the European Confederation of Medical Mycology (ECMM) Working Group on Zygomycosis between 2005 and 2007. Clin Microbiol Infect. 2011;17(12):1859-67.
- Chakrabarti A, Das A, Mandal J, Shivaprakash MR, George VK, Tarai B, et al. The rising trend of invasive zygomycosis in patients with uncontrolled diabetes mellitus. Med Mycol. 2006;44(4):335-42.
- 11. Pak J, Tucci VT, Vincent AL, Sandin RL, Greene JN. Mucormycosis in immunochallenged patients. J Emerg Trauma Shock. 2008;1(2):106-13.
- 12. Choi HL, Shin YM, Lee KM, Choe KH, Jeon HJ, Sung RH, et al. Bowel infarction due to intestinal mucormycosis in an immunocompetent patient. J Korean Surg Soc. 2012;83(5):325-9.
- Thomson SR, Bade PG, Taams M, Chrystal V. Gastrointestinal mucormycosis. BJS Br J Surg. 1991;78(8):952-4.
- Lo OSH, Law WL. Ileocolonic mucormycosis in adult immunocompromised patients: A surgeon's perspective. World J Gastroenterol. 2010;16(9):1165-70.
- 15. Spellberg B, Edwards J, Ibrahim A. Novel perspectives on mucormycosis: Pathophysiology, presentation, and management. Clin Microbiol Rev. 2005;18(3):556-69.
- 16. Gorbach SL, Bartlett JG, Blacklow NR. Infectious diseases. Lippincott Williams & Wilkins; 2004. p. 2546.
- Termos S, Othman F, Alali M, Al Bader BMS, Alkhadher T, Hassanaiah WF, et al. Total gastric necrosis due to mucormycosis: A rare case of gastric perforation. Am J Case Rep. 2018;19:527-33.
- Chamilos G, Lewis RE, Kontoyiannis DP. Delaying Amphotericin Bbased frontline therapy significantly increases mortality among patients with hematologic malignancy who have zygomycosis. Clin Infect Dis. 2008;47(4):503-9.
- Goldstein EJC, Spellberg B, Walsh TJ, Kontoyiannis DP, Edwards J Jr, Ibrahim AS. Recent advances in the management of mucormycosis: From bench to bedside. Clin Infect Dis. 2009;48(12):1743-51.
- 20. Malek A, De la Hoz A, Arduino R, Aisenberg GM. Disseminated tuberculosis and gastric mucormycosis coinfection. IDCases. 2019;18:e00595.