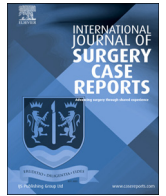




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Gastrointestinal mucormycosis after abdominal aortic aneurysm repair and prolonged hospitalization: A case report and review of the literature

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ABSTRACT

INTRODUCTION: Mucormycosis is a rare fungal infection typically affecting immunocompromised hosts. One form of the disease affects the gastrointestinal tract.

PRESENTATION OF CASE: We present the case of a 70-year old patient with no recognized risk factors that developed gastrointestinal mucormycosis after urgent abdominal aortic aneurysm repair.

DISCUSSION: There are several risk factors for this infection, such as hematological malignancies, solid organ or stem cell transplants and diabetes. The infectious agent causes thrombosis and necrosis of involved tissues and organs and carries a high mortality rate.

CONCLUSION: Mucormycosis is an opportunistic infection which can sometimes affect the gastrointestinal tract. A high index of suspicion is necessary in order to make an early diagnosis and promptly start an appropriate treatment regimen.

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1. Introduction

Mucormycosis is a rare fungal infection caused by the *Mucorales* species that usually presents in immunocompromised patients. This fungus is relatively aggressive in nature when compared to other systemic fungal infections. Various forms of the disease exist, including gastrointestinal mucormycosis [1,2]. This article discusses the case of a patient who underwent ruptured abdominal aortic aneurysm (AAA) repair without classic mucormycosis risk factors that went on to develop a complicated abdominal presentation of the infection.

2. Presentation of case

A 70-year old female patient known for a 4.6 cm AAA presented to the emergency department with increasing abdominal pain over the previous three days. Her previous medical history was positive for chronic obstructive pulmonary disease (COPD), smoking, hypertension, dyslipidemia and osteoporosis. She had several previous surgeries, such as appendectomy in her youth, left partial mastec-

tomy for breast cancer 13 years prior and right femoral hernia repair one year prior. The patient had no fever and was hemodynamically stable on presentation. Computed tomography (CT) of the abdomen showed a ruptured AAA with a retroperitoneal hematoma.

She was transferred to the operating room for urgent AAA repair with an infra-renal aortic prosthesis. The inferior mesenteric artery was ligated as is common during this procedure. Although present at the end of the surgery, femoral pulses were lost bilaterally in the immediate post-operative period. The patient was brought back to the operating room where she was found to have bilateral iliac thrombosis. An aorto-bifemoral bypass was then performed and the patient was transferred to the intensive care unit (ICU).

On the first post-operative day, a non ST-elevating myocardial infarction (NSTEMI) was diagnosed based on electrocardiographic changes and a rise in troponins. A coronary angiography demonstrated a non-revascularizable occluded circumflex artery. It was decided to treat the condition medically. One day after coronary angiography, the patient developed a right brachial artery thrombosis at the site of the catheter insertion. She was brought to the operating room once again for right brachial embolectomy.

The following three weeks were uneventful albeit for post-operative ileus. Twenty-four days after her initial AAA repair, the patient developed diffuse abdominal pain and her bloodwork showed new onset leukocytosis. She remained afebrile and hemodynamically stable. There was no clinical evidence of peritonitis. CT of her abdomen showed air in the left retroperitoneum and possible free air above the bladder. The CT was repeated with oral and

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rectal contrast and showed descending colon wall thickening and contrast extravasation compatible with left colic perforation secondary to ischemic colitis. The patient was consented for urgent exploratory laparotomy.

Intra-operatively, there was evidence of ischemia of a short segment of small bowel, the descending and sigmoid colon as well as the rectum. The ischemic segment of small bowel was resected with primary anastomosis and a left colectomy with sigmoidectomy and proctectomy was performed. A transverse terminal colostomy was created and two Jackson-Pratt (JP) drains were left in place. The patient remained stable and was transferred to the ICU where the two following weeks were marked by two failed extubations in the context of exacerbated COPD and volume overload.

Fourteen days after the exploratory laparotomy another abdominal CT demonstrated infarction of the inferior pole of the spleen, left renal artery thrombosis and a non-enhancing left kidney with possible air bubbles in the peri-renal tissue suggestive of infection. There were also two small intra-abdominal fluid collections in which the JP drains were well positioned.

The patient was brought back to the operating room for another laparotomy. The small bowel proximal to the anastomosis was dilated and 60 cm of small bowel including the anastomosis was deemed non-viable and was resected with primary anastomosis. The transverse colostomy was taken down to gain better access to the left kidney and spleen. There was necrosis of the inferior pole of the spleen and the entire left kidney. Splenectomy and left nephrectomy were performed, another terminal transverse colostomy was fashioned and a percutaneous feeding tube was inserted into the jejunum. Intra-abdominal fluid was sent for culture and Gram stain.

The patient was once again brought to the ICU and eventually transferred to a step-down unit. Preliminary cultures from the abdominal fluid were positive for *Pseudomonas aeruginosa* for which the patient was already appropriately treated with piperacillin-tazobactam. After five days of incubation, the fluid culture was positive for fungus and the patient was started on voriconazole for suspected aspergillus.

The patient's clinical status deteriorated however and she developed sepsis, respiratory failure, acute kidney injury and increasing diffuse non-peritonitic abdominal pain. One of the JP tubes began draining fluid resembling enteric feeding formula. Her enteral feeds were stopped and a CT of the abdomen was done which demonstrated a perforated small bowel and probable anastomotic leak. Final fungal cultures came back positive for *Mucor* sp. and voriconazole was replaced by liposomal amphotericin B.

In the context of a worsening clinical picture and the poor prognosis of mucormycosis, the patient and her family decided to refrain from any aggressive therapy. Comfort measures were started and the patient died a few hours later.

3. Discussion

This case exemplifies the aggressive nature of mucormycosis and its often-late diagnosis. As previously mentioned, mucormycosis (sometimes referred to as zygomycosis) is a rare infectious disease caused by the *Mucorales* species of fungus. The most common pathogens are of the *Rhizopus*, *Lichtheimia* or *Mucor* genus [1].

Although they are ubiquitous in nature, these fungi are rarely pathogenic as they are opportunistic and commonly infect patients with certain recognized risk factors. Mucormycosis is independently associated with diabetes mellitus, hematological malignancies, chemotherapy, stem cell or solid organ transplants and trauma [1,3]. Another possible risk factor is the prolonged use of voriconazole, that may select for those particular pathogens, presumably because *Mucorales* species are resistant to anti-fungal

therapy [4]. Recently, cases of nosocomial mucormycosis associated with surgical trauma or contaminated medical equipment have been described in patients with no recognized risk factors. However, unlike the present case, these patients typically present with the cutaneous variant of the disease [1].

Mucorales are vasotropic in nature and cause thrombosis and necrosis of the involved tissue [5,6]. The most recognized clinical syndromes of mucormycosis (in decreasing order of occurrence) are rhinocerebral, pulmonary, cutaneous, gastrointestinal, disseminated and other uncommon presentations [1].

Gastrointestinal mucormycosis presents clinically as gastrointestinal bleeding, obstruction or bowel perforation with subsequent peritonitis. Diagnosis is often difficult to make as there are for more common clinical entities that have similar manifestations. A high degree of suspicion is warranted in presence of recognized risk factors for the disease. Unfortunately, cultures on pathologic specimens are often inconclusive and there are no specific imaging findings compatible with gastrointestinal mucormycosis [6]. Nonetheless, liberal use of abdominal imaging and endoscopy is recommended when working up for possible mucormycosis. The most common organs involved are the stomach (57.5%), colon (32.3%) and ileum (6.9%) [6]. Infrequently, the liver, spleen, pancreas and kidneys can be affected as well [1].

If a prompt diagnosis is made, treatment generally consists of source control by debriding all infected and necrotic tissue followed by systemic antifungal therapy with liposomal amphotericin B. Extensive bowel resection is often required as was the case with the aforementioned patient [5,7].

Even in cases treated aggressively with surgery and antifungals, mortality remains high. One study reported mortality of 85% in patients with gastrointestinal mucormycosis when diagnosis was delayed and found that an ante-mortem diagnosis was only made in 25% of cases [1,8].

4. Conclusion

Mucormycosis is an aggressive and opportunistic fungal infection that typically affects patients with a number of recognized risk factors. Recently, however, there are increasing numbers of cases of nosocomial mucormycosis affecting patients not considered to have the typical risk factors. *Mucorales* is a vasotropic fungus that causes thrombosis and necrosis of involved tissue. One variant of the disease is gastrointestinal mucormycosis. It carries a particularly high mortality rate and is often not diagnosed in time for prompt treatment. Management consists of surgery to remove all infected and necrotic tissue along with systemic antifungal treatment with liposomal amphotericin B.

Conflicts of interest

None.

Funding

None.

Ethical approval

There was no ethics approval required for this case report.

Consent

Informed consent was obtained from the patient's next of kin for publication of this case report. A copy of the written consent

is available for review by the Editor-in-Chief of this journal on request.

Author contribution

A. Di Palma: Reviewing patient data, writing the manuscript.

F. Schwenter: Surgeon involved in patient's care, revising manuscript.

Guarantor

Frank Schwenter.

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