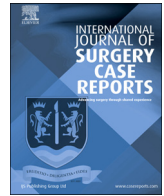




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A rare cause of ischemic gut: A case report

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ABSTRACT

INTRODUCTION: Mucormycosis is a fungal infection that is a rare cause of gastrointestinal ischemia, usually found in the immunocompromised or patients with extended surgical intensive care unit stay. This case report brings to light the rare presentation of a total gastric infarction secondary to a rare fungal infection where the overall outcome of the patient depends on timely diagnosis and management.

CASE PRESENTATION: An elderly Malay female patient in our institution developed severe sepsis. Radiological investigations revealed an intra abdominal source of sepsis likely secondary to total gastric ischemia. Both the abdominal X-ray and computed tomography scan showed evidence of gastric pneumatosis. She was diagnosed with gastric ischemia and underwent a total gastrectomy. However post surgically she continued to deteriorate and passed away 5 days later.

CONCLUSION: Recognition of mucormycosis infection is pertinent to commence anti fungal treatment early with timely implementation of subsequent surgical management. Early access to surgery is necessary to improve cure rates in patients with mucormycosis as antifungal treatment alone is usually not adequate for cure.

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1. Introduction

Anatomical or functional changes in the local vasculature that compromise blood flow can lead to ischemia. Common causes of occlusive obstruction to the blood flow include vascular emboli or thrombosis and strangulation of the venous blood supply. Primary splanchnic vasoconstriction is the most common cause for non-occlusive obstruction.

In order for a total infarction, all vasculature feeding the organ must be compromised. The stomach is a highly uncommon area for total ischemia due to its rich vasculature. In this case report, we report a rare cause of vascular compromise to the stomach, which presented as a diagnostic challenge.

2. Case presentation

A 60 year old Malay lady with a significant past medical history of poorly controlled type 2 diabetes mellitus and bone marrow transplant for acute myeloid leukemia was admitted for the presenting complain of right otalgia associated with purulent otorrhea. She was diagnosed with right malignant otitis externa and started

on intravenous Augmentin. It was however found to be refractory to antibiotic treatment. A follow up computed tomography scan of the right temporal bone performed 5 days after the commencement of antibiotic therapy showed bony destruction of external auditory canal, mastoid and right mandibular fossa, associated with osteomyelitis of the right temporal bone.

Infectious disease consult was obtained to assist in the management of her sepsis, and the patient was started on a planned 6-week intravenous antibiotic therapy of linezolid and piperacillin/tazobactam. Management included a full septic work up with blood, urine and pus cultures. However the patient developed severe sepsis 8 days after the commencement of antibiotic therapy. It was complicated by metabolic acidosis and acute renal failure. She was subsequently intubated and managed in the surgical intensive care unit.

Cultures performed on pus obtained from the right ear grew multi-resistant staphylococcus aureus and pseudomonas aeruginosa. Her urine culture grew *Candida albicans*. Following the positive culture results, the antibiotic regime was changed to intravenous meropenem and vancomycin. Intravenous caspofungin was also started for the positive *Candida* culture results.

During her stay in the surgical intensive care unit, she became anuric and was started on dialysis, initially on sustained low efficiency dialysis for 2 days before being converted to continuous veno-venous hemodialysis. Patient developed abdominal distension and an abdominal X-ray was performed as part of investigations. It showed mottled lucencies over the gastric lumen,

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Fig. 1. Presence of mottled gas lucencies overlying the gastric lumen is highly suspicious for intramural gas secondary to mucormycosis infection.

suspicious of gastric pneumatosis. A computed tomography scan of the abdomen and pelvis performed confirmed the findings of interval development of extensive gastric intramural gas.

She underwent an exploratory laparotomy and intra-operative findings revealed an infarcted stomach from the cardia to the pylorus. A total gastrectomy was performed. It was noted that the vasculature of the coeliac axis and the superior mesenteric artery had good palpable pulsations. However post operatively, the patient continued to deteriorate, developing persistent hypotension and bradycardia despite maximal inotropic support. She eventually passed away 5 days after the surgery.

The final histopathology report of the patient's total gastrectomy specimen reported a total gastric infarction associated with the presence of widely disseminated fungal organisms. Microscopically, an infarction of the entire gastric wall was seen with a “ghost outline” preservation of architectural features of the main mural components. Stains applied to the specimen showed numerous fungal organisms disseminated through the entire gastric wall that was also present within the vascular channels. The fungus consisted of spores and hyphae of variable irregular sizes, showing branching at right angles.

3. Discussion

Total gastric infarction is a highly unusual presentation as the stomach is a richly vascular organ. This is due to the multiple vascular supplies originating from the coeliac axis. Therefore in order to have total gastric infarction, most if not all of the vessels supplying the stomach must be involved. A fungal infection by mucormycosis is one such rare cause that can involve multiple vessels resulting in this catastrophic presentation (Fig. 1).

Mucormycosis is a life threatening opportunistic infection caused by fungi of the subphylum Mucoromycotina, order Mucorales. *Rhizopus oryzae* is the most common organism isolated from infected patients [1]. Other genera causing mucormycoses include *Mucor*, *Lichtheimia*, *Cunninghamella*, *Rhizomucor*, *Saksena* and *Apophysomyces* [2].

Risk factors for the development of invasive mycormycosis include immunocompromised states such as in diabetes, Acquired Immunodeficiency Syndrome (AIDS), malnutrition, defects in host phagocytes, corticosteroid use, and organ or stem cell



Fig. 2. Transverse Computed Tomography Scan Cuts showing extensive intramural gas affecting the stomach secondary to mucormycosis infection.

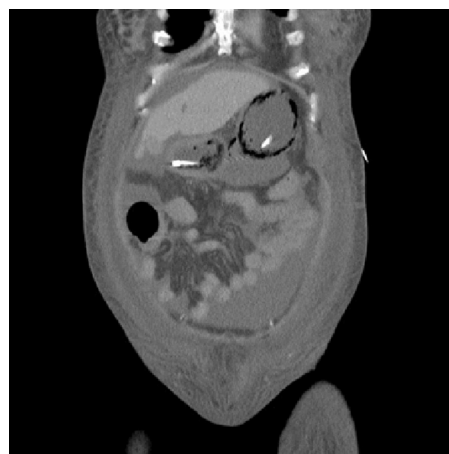


Fig. 3. Coronal cuts showing extensive intramural gas affecting most of the stomach secondary to mucormycosis infection.

transplantation. However there are also reports of mucormycosis in patients without any of the above-mentioned risk factors [3].

The fungus produces either superficial or deep mycotic infections, involving predominantly the skin, nails and external ear. Gastrointestinal mucormycosis is generally rare, especially in developed nations. It is seldom diagnosed in living individuals. While gastrointestinal infections comprises of only 7% of the reported cases of mucormycosis, the reported mortality is around 85%. The stomach is the most common site of gastrointestinal mucormycosis, followed by the colon, ileum, duodenum and jejunum [4–6]. This begets the question of how mucormycosis infection of the gastrointestinal tract occurs. It is commonly believed that infection of the alimentary tract is due to ingestion of the spores. The stomach is the first major alimentary organ that comes into contact with the ingested spores, possibly resulting in it being most commonly infected. In severe cases, infarction of the infected organ may occur, as described in our patient. The fungus has a predilection for the walls of the vessels, both arterial and venous. The invasion of different types of blood vessels by fungi hyphae leads to different complications. In arterial invasion, arterial thrombosis can occur and lead to tissue infarction and eventual necrosis. Venous invasion usually causes hemorrhage [4] (Fig. 2).

Eventually the diagnosis of mucormycosis can be confirmed on either the growth of mold on culture or the histopathological demonstration on biopsy of aseptate, wide, ribbon-like hyphae that branches at right angles. However due to the difficulty of growing

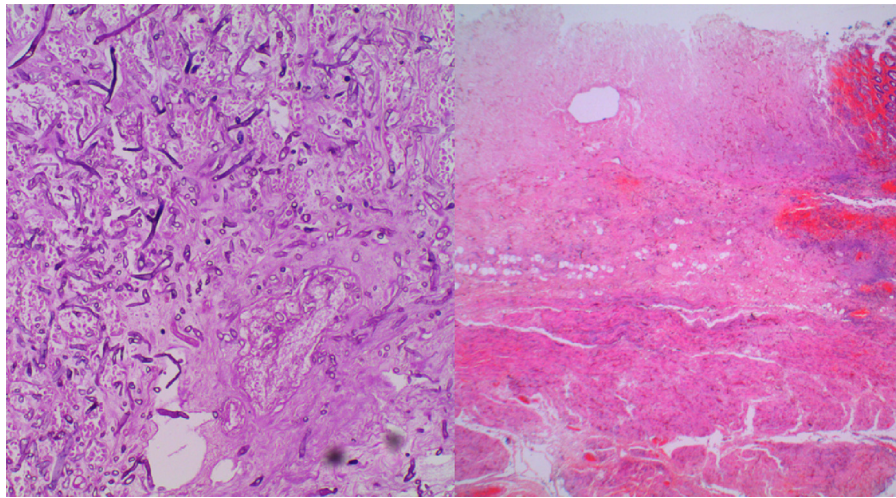


Fig. 4. Histopathologic examination revealed total gastric infarction associated with the presence of abundant, widely disseminated fungal hyphae with morphologic features of mucormycosis within the vascular channels as demonstrated.

the fungal organism from tissue culture, biopsy with histological identification is usually the preferred mode of diagnosis [7]. The advantage of culture is the identification and susceptibility testing of the organism. While identification and susceptibility testing allows for a more directed anti-fungal treatment, there is no strong evidence proving that identification is important to guide treatment. It will however allow subsequent epidemiological studies for this rare fungal infection [8,9].

In event of mucormycosis infection, anti-fungal treatment is usually inadequate to control mucormycosis. Surgery to debulk the fungal infection with resection of all the infected necrotic tissue is often required for cure [8]. The inadequacy of anti-fungal treatment is due to various factors including resistance to the anti-fungal agents, angioinvasion, thrombosis of vessels resulting in less optimal penetration of the medications to the site of interest (Fig. 3).

The European Society for Clinical Microbiology and Infectious Disease and the European Confederation of Medical Mycology has released a joint clinical guideline in the diagnosis and management of mucormycosis. It is recommended that in addition to first line anti fungal treatment with liposomal or lipid complex amphotericin B, surgical debridement be instituted [10] (Fig. 4).

Surgery was found to be an independent variable for favorable outcomes in patients with mucormycosis [7]. It was found that patients who did not undergo surgical intervention had a higher mortality rate than those who did [11–13]. Reversal of the predisposing condition is also recommended such as controlling hyperglycaemia in diabetic patients, and limiting the use of glucocorticosteroids or reducing the dosage in immunosuppressed patients [10].

4. Conclusion

Mucormycosis is a rare cause of gastrointestinal ischemia. Its presentation may be insidious and an underlying fungal infection is usually not known until specific investigations for the presenting signs and symptoms of the gastrointestinal ischemia are made. Even so, by the time the diagnosis of mucormycosis is made, it may be too late for favorable outcomes. Therefore when suspecting ischemic gut in patients, especially in those who are immunocompromised, or one who has had extended surgical intensive care unit stay, mucormycosis/fungal infections of the gut should be considered. If mucormycosis is suspected, early consult by the infectious disease specialty should be made. This allows appropriate anti-fungal treatment to be commenced quickly and the timely

implementation of subsequent surgical management. With better recognition of mucormycosis as an insidious cause of gastrointestinal ischemia, prompt management may be instilled to result in better outcomes.

Conflict of interest

The authors declare that they have no conflicts of interests.

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The authors declare that they have no sources of funding for the research.

Ethical approval

This is a observation case report. No institutional review board approval is required.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Authors' contributions

WWK is the senior consultant in charge of the patient and was involved in the primary care of the patient. CSL was the consultant involved in the care of the patient and played a major role in guiding the direction of the manuscript. ZJ analyzed the interpreted the patient's data and was a major contributor in writing the manuscript. All authors read and approved the final manuscript.

Guarantor

Wai Keong Wong, Claramae Chia, Zhen Jin Lee.

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