Case Report

We report a case of 19 year old girl, who presented with complaints of fever, loose stools (blood stained), vomiting and pain abdomen for 4 days. Before she came to our hospital, she was evaluated at nearby clinic and found to have pancytopenia, acute kidney injury, coagulopathy and hepatic derangement and treated conservatively. She gave history of two episodes of generalized tonic clonic seizure on day before admission. She denied any previous co-morbidities. On admission she was tachypneic, hypotensive and altered sensorium. On examination she had pancytopenia, acute kidney injury, hematuria and proteinuria with preceding acute gastrointestinal bleeding persisted, despite conservative management. So endoscopy was repeated which revealed healed esophageal ulcer and extensively bleeding gastric ulcer (Figure 1). CT abdomen was conducted with oral contrast which revealed leakage from gastric ulcer. Then surgical gastroenterology opinion was taken and exploratory laparotomy was performed. Intra-operatively posteriorly perforated gastric ulcer was found and partial gastrectomy was done and excised tissue was sent for histopathological examination. During postoperative course her general condition gradually improved. Subsequently the biopsy report showed extensive ulcerative gastric mucosa filled with necrotic granulation tissue with polymorphonuclear infiltrate, suggestive of ischemic ulcer. Fungal stain of ulcer tissue showed presence of granulomatous necrotic areas positive for mucormycosis. Then she was managed with amphotericin-B, posaconazole with which she improved.

Abstract

Mucormycosis, is an emerging fungal infection in immunocompromised and diabetic individuals, usually affects rhino-orbito-cerebral, cutaneous and pulmonary regions. But mucormycosis in immunocompetent environment is rare and occurrence of gastric mucormycosis is unusual. We report a case of 19 year old female, with no pre-existing co-morbidities, presented with fever, dysentery, vomiting, and melena for 4 days. On evaluation she was found to have pancytopenia, acute kidney injury, hemolytic anemia, coagulopathy and hepatic derangement and treated with hemodialysis, plasmapheresis along with antibiotics and packed cell RBC transfusion. Upper gastrointestinal endoscopy revealed presence of extensive esophageal and gastric ulcer. In view of persistent bleeding despite endoscopic sclerotherapy, repetition of upper gastrointestinal endoscopy and CT abdomen with oral contrast was done, which revealed perforated gastric ulcer. Exploratory laparotomy and excision of ulcer was done. The biopsy of gastric ulcer had shown the presence of granulomatous necrotic areas positive for mucormycosis. Then she was managed with amphotericin-B, posaconazole with which she improved.

Introduction

Invasive life-threatening fungal infections like mucormycosis usually occur in immunocompromised and diabetic individuals but the same in immunocompetent conditions is very rare. Mucormycosis is caused by fungi of order Rhizopus (Zygomycetes). It most commonly affects rhino-cerebral region followed by pulmonary and cutaneous involvement. But involvement of gastrointestinal system is very rare and mortality rate in gastric mucormycosis is high (98%). Along with immunocompromised states, malnourishment also predisposes to gastric mucormycosis. Early suspicion, diagnosis and treatment is the key for survival. Surgical resection of involved tissue (in possible cases) and antifungals (amphotericin-B and posaconazole) are the treatment modalities. We report a case of young malnourished girl who presented with manifestations of hemolytic uremic syndrome and on evaluation she was found to have gastric mucormycosis and treated successfully with antifungals and gastric ulcer excision.

Gastric Mucormycosis with Hemolytic Uremic Syndrome

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Absidia, Rhizopus, Rhizomucor, the four genera include those caused by members of infections caused by Zygomycetes distribution of fungi. The most common takes place in immunocompromised material. Disease manifestation usually which often present in soil and decaying material. Ingestion of spores and common in gastrointestinal endoscopy was repeated which showed healed gastric ulcer. During the hospital course she got secondary bacterial sepsis which treated conservatively. Finally she was discharged in a stable condition and on follow-up after 3 months she is doing well.

Discussion

Zygomycetes are ubiquitous fungi, which often present in soil and decaying material. Disease manifestation usually takes place in immunocompromised patients, despite widespread distribution of fungi. The most common infections caused by Zygomycetes include those caused by members of the four genera Rhizopus, Rhizomucor, Absidia, and Mucor, with R. oryzae being the species most frequently isolated from patients. Gastrointestinal mucormycosis is thought to arise from ingestion of spores and common in malnourished patients. Traditional risk factors for the development of invasive mucormycosis include diabetes, defects in host phagocytes, corticosteroid use, organ or stem cell transplantation, and increased levels of available serum iron as a result of acidosis or administration of desferoxamine. Initial manifestations may be abdominal pain and distension, fever and hematochezia. Gastric mucormycosis can be categorized into three forms as colonization, infiltration and vascular invasion. The most frequent presentation is perforation, bleeding or epigastric distention. To diagnose mucormycosis, direct microscopy preferably using optical brighteners, histopathology and culture are strongly recommended. Pathogen identification to species level by molecular methods and susceptibility testing are strongly recommended to establish epidemiological knowledge. Surgical resection of involved tissue always gives good prognosis in mucormycosis. Among antifungal agents, Amphotericin B is the licensed agent. Liposomal amphotericin (LAmB) and lipid formulation Amphotericin B (LFAB) are safer and better agents than conventional amphotericin B. But in poor patients, conventional amphotericin is better economically. Posaconazole does contribute beneficiary effect as a part of combination therapy, but it cannot recommended as primary therapy for mucormycosis on the basis of available data. Regarding duration of treatment no guidelines are available, it should be individualized for each patient. In general, antifungal therapy for mucormycosis should be continued until all of the following objectives are attained: (a) there is resolution of clinical signs and symptoms of infection, (b) there is resolution or stabilization of residual radiographic signs of disease on serial imaging, and (c) there is resolution of underlying immunosuppression.

Hemolytic uremic syndrome (HUS) and thrombotic thrombocytopenic purpura (TTP) are the entities which usually preceded by acute gastroenteritis/dysentery or drug toxicity. According to center for disease and control (CDC), presence of anemia with microangiopathic changes, thrombocytopenia and acute kidney injury within 3 week after acute or bloody diarrhea is enough to diagnose HUS/TTP. Plasma exchange and hemodialysis are the principle modalities of hemolytic uremic syndrome along with treatment of cause. Even though acute gastroenteritis can present as hemolytic uremic syndrome (HUS), in literature, no studies existed to explain the association of HUS with mucormycosis. But in our case as the patient clinically improved after treated for mucormycosis, retrospectively gastric mucormycosis was considered as a cause of hemolytic uremic syndrome. As of our knowledge this would be a rare case of “gastric mucormycosis presented as hemolytic uremic syndrome”.

References