



## Dramatic Response of Gastric Outlet Obstruction with Ileocecal Involvement of Basidiobolomycosis to Amphotericin B: A Case Report

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### ABSTRACT

Gastrointestinal Basidiobolomycosis (GIB) is a fungal infection in the stomach, small intestine, colon, and liver that can play a role in malignancy. It is a rare disease and only about 73 cases of GIB exist in the literature. Abdominal pain, weight loss, fever and abdominal mass are the most common symptoms which are clues of definitive diagnosis by microbial culture from surgical specimens. In present article, we have reported a case of GIB with gastric outlet obstruction that dramatically has responded to treatment of Amphotericin B. A 44 y/o male was admitted by chief complaint of early post meal vomiting and dyspepsia. He also had a noticeable weight loss in the last 6 months. There were not any other sign or symptoms as fever or etc. However, two masses were observed by abdominopelvic contrasted CT scan; one in second portions of duodenum and the other one in caecum. Therefore, he was planned for surgery and a jejunostomy tube was placed for the elimination and operation was terminated because the lack of definite diagnosis. Final results of pathology confirmed the fungal infection compatible with basidiobolomycosis. After the 3<sup>rd</sup> day of laparotomy diet and Intravenous Amphotericin B were started for the patient. After 2 weeks of start of amphotericin B patient scheduled for second laparotomy for permanent gastrojejunostomy. Finally, in the next follow up in 3 months patient was completely relieved of all sign and symptoms. A Contrasted abdomen and pelvic CT scan was performed with any signs of previous masses. In conclusion, surgery could be avoidable by early diagnosis and treatment of GIB by administration of Intravenous Amphotericin B.

**Keywords:** Gastric Outlet Obstruction, Basidiobolomycosis, Amphotericin

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### INTRODUCTION

Gastrointestinal Basidiobolomycosis (GIB) is an unusual, rare but emerging fungal infection in the stomach, small intestine colon, and liver it has been rarely reported in the English literature and most of the reported cases have been from US, Saudi Arabia, Kuwait, and Iran [2]. Infections

caused by fungi in the subphylum Entomophthoromycotina, called entomophthoromycosis, include both conidiobolomycosis and basidiobolomycosis these are rare infections of the paranasal sinus and subcutaneous tissues principally encountered in the tropics [1]. Gastrointestinal basidiobolomycosis (GIB) can play a role of malignancy. For example GIB can be manifestation of liver and colorectal tumors with chief complain of abdominal pain and rectal pain and abdominal mass [2]. Basidiobolomycosis is a rare fungal disease that caused by basidiobolomycosis.

member of Zygomycete class [6]. Visceral involvement of basidiobolomycosis is rare, a 6 years old boy was the first case of gastrointestinal basidiobolomycosis in 1964 [3]. Approximately 73 cases of GIB exist in the articles [6]. The most common presenting symptom is abdominal pain, weight loss, fever and abdominal mass. A definitive diagnosis of basidiobolomycosis depends on microbial culture from surgical specimens or fresh aspiration [7]. Over the last two decades most articles shows increasing gastrointestinal basidiobolomycosis in immunocompromised persons, especially in the Middle East and United states [2]. Treatment of GIB often depends on Non-surgical intervention, such as anti-fungal therapy. More articles have treatment basidiobolomycosis with Itraconazole or Variconazole, here we have reported a case of GIB with gastric outlet obstruction that dramatically has responded to treatment of Amphotericin B. Nearly all patients had abdominal pain and fever as their initial symptoms; however, some cases additionally had constipation, diarrhea, and gastrointestinal bleeding.

#### CASE PRESENTATION

A 44 y/o male admitted in our hospital because of early post meal vomiting and dyspepsia. Vomits were not Projectile, bloody or bilious, in about 15 minutes after eating. There were not any other sign or symptoms as fever or etc. He manifest with weight loss in 6 months. Plain radiography did not show something particular. In contrasted abdomen and pelvic CT scan there were 2 masses. One in second portions of duodenum and the other one in ceccum. Upper GI endoscopy showed bulb deformity and D1, D2 narrowing. Biopsy was taken but there were no pathologic changes and no definite diagnosis. With operation planning, Exploration was performed through midline laparotomy. In macroscopic appearance there were three masses. The first in pyloric region in 7\*8 Cm, creamy yellowish, elastic and necrotic. The second one was in distal ileum and ileocecal part of intestine. (premedication pictures) Three metastatic lesions were in liver. Biopsy was taken from all of the lesions. A jejunostomy tube was placed for the elimination and operation was terminated because the lack of definite diagnosis. Final results of pathology confirmed the fungal infection compatible with basidiobolomycosis (pathologic pictures).

After the 3<sup>rd</sup> day of laparotomy diet started for the patient. Intravenous Amphotericin B was started for the patient. In day eight after the first laparotomy clinical symptoms of patient were tolerated and the patient feels better.

After 2 weeks of start of amphotericin B patient scheduled for second laparotomy for permanent gastrojejunostomy. At the second operation all the three masses were shrunk about 75% (postmedication pictures). The pyloric was open and so jejunostomy tube site was closed. Diet started again orally after 5 days of second laparotomy. The patient tolerated the regimen and gained weight and discharged.

In the next follow up in 3 months patient was completely relieved of all sign and symptoms. We performed a Contrasted abdomen and pelvic CT scan that did not show any sign of previous masses.

#### DISCUSSION AND CONCLUSION

Gastrointestinal basidiobolomycosis is an unusual fungal infection of the gastrointestinal tract caused by *B. ranarum*. It is rarely reported in the medical literature although sporadic case has been reported worldwide [7]. Unintentional ingestion of the fungus from contaminated soil or fruits or vegetables is touted as the route of entry preceding GIB. Although living in tropical and subtropical areas are well established as risk factors for basidiobolomycosis, no risk factors are identifiable for GI basidiobolomycosis per se. Incidentally, basidiobolomycosis affects men more likely due to their indulgence in outdoor activity compared to women in the tropics [7-9].

Literature review by Vikram *et al.* [8] showed 44 cases, Most of these patients were diagnosed with abdominal mass on CT scan and endoscopy after having been evaluated for indolent nonspecific signs and symptoms. Patients were normally immunocompetent men with no underlying serious medical problems. Basidiobolomycosis which is resulted of a rare fungal infection, Basidiobolus ranarum, is belonging to the Zygomycetes [14]. Despite to the other order of the Zygomycetes, Mucorales that involve the immunocompromised patient, Entomophthorales which is other order of the Zygomycetes, include *Basidiobolus* genera, causes infection in immunocompetent patient [15].

Basidiobolus ranarum there is in the nature as an environmental saprophyte found in soil and decaying vegetable materials. It has been occasionally seen as a commensal in the gastrointestinal tracts of amphibians, reptiles, fish, dogs, frogs, and bats. *B. Ranarum* has hyphae and zygospores. Branching and septation with thin wall show in hyphae [16].

In the histopathologic specimen stained with hematoxylin and eosin stain in gastrointestinal Basidiobolomycosis, sections show infection seems not to involve the mucosal layers of the gastrointestinal tract; and specimen were characterized by marked mural thickening with fibrosis.

Microscopic examination reveals areas of necrotizing inflammation and thin wall and broad hyphae surrounded by an eosinophilic and amorphous hyalinized proteinaceous material. Which called Splendore-Hoeppli phenomenon accompanied by marked infiltration of eosinophils, mixed infiltration of PMN leukocytes and granulomatous inflammation included foreign body type multinucleated giant cells.

Splendore-Hoeppli phenomenon refers to radiating or annular amorphous eosinophilic deposits of host-derived materials and possibly of parasite antigens. It usually forms around fungi, helminths or their ova, or bacterial colonies and, on rare occasions, suture material in tissues. It is usually surrounded by inflammatory cells including eosinophils, neutrophils, histiocytes, lymphocytes, and multinucleated giant cells [17]. Periodic Acid-Schiff (PAS) and Gomori Methenamine Silver (GMS) can intensify the fungal wall staining.

Case series by Saud Al Shanafey reported 9 pediatric cases of GIB at King Faisal Specialist Hospital and research center between 2001 and 2010 [10]. Distribution of lesions included left colon involvement in 11%, right colonic involvement in 33%, liver involvement in 78% and diffuse abdominal disease in 22%. Patients with colonic involvement in GIB had right or left hemicolectomy. Liver lesions were managed with partial hepatectomy [10, 11].

Susceptibility of Basidiobolomycosis to antifungal agents are known to be highly variable and isolate

dependent [12,13]. Empiric treatment could be started with Itraconazole or Voriconazole, but susceptibility testing is recommended to ensure effective treatment. Taghipour *et al.* reported mortality with GIB from septic shock and pulmonary insufficiency despite successful diagnosis and treatment with Itraconazole and amphotericin B. [13].

In our case the disease dramatically cured with Amphotericin B and surveillance of treatment response could be performed with MRI or CT scans. We propose the surveillance imaging could be performed with CT scan or MRI scans depending on the local availability and expertise. In summary, GIB is a rare invasive fungal infection involving the gastrointestinal tract and liver. If identified early and treated appropriately in this case Amphotericin B, it is curable and surgery can be avoided. Surgery is unavoidable if GIB is complicated by bowel obstruction. Awareness and knowledge is important for early diagnosis and treatment of GIB and prevent morbidity and mortality from delay in diagnosis.

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