BASIDI BOLO MYCOSIS
A CASE REPORT

INTRODUCTION:
Basidiobolomycosis is a rare disease caused by the fungus *Basidiobolus ranarum*. Patients with *Basidiobolus ranarum* infection may present with subcutaneous, gastrointestinal, or systemic lesions. Recently, its etiologic role in gastrointestinal infections has been increasingly recognized. We report this case of gastrointestinal and retroperitoneal basidiobolomycosis because of its rarity and also to emphasize the need to consider gastrointestinal and retroperitoneal basidiobolomycosis in the differential diagnosis of inflammatory bowel diseases, tuberculosis and malignancy.

Key words: Basidiobolomycosis, gastrointestinal, retroperitoneal, Splendore-Hoeppli phenomena

CASE REPORT:
A previously healthy two and a half year old, male, Sudanese patient residing in Jeddah presented to the emergency room three weeks ago with fever, abdominal pain, vomiting, watery diarrhea and history of weight loss. The patient had been residing in poor neighborhood, however gave no history of any prior skin infection in him or other members of the family. There was history of having ingested a small lizard when he was eight months old. On examination the patient was febrile but the rest of the vitals were stable. Abdominal examination revealed a hard, tender, globular, smooth, non-pulsatile mass in the right iliac fossa. Hematological investigations revealed marked increase in Erythrocyte Sedementation Rate and ‘C’ reactive protein, leukocytosis (41.9K/uL) with an absolute neutrophil count of 33.1 k/ul, and absolute eosinophil count of 1.6 k/ul. Liver function tests revealed high liver enzymes and bilirubin. The values were as follows; Alkaline phosphatase 208 U/L, Aspartatate amino transferase 66U/L, Gammagutaryl transferase 227U/L, Total bilirubin 109\mu mol/l, Direct bilirubin 56\mu mol/l. Bacterial cultures showed no organisms. Serum creatinine was slightly reduced. Ultrasound showed a huge right iliac mass extending from the terminal ileum and involving the right colon up to the hepatic flexure with minimal ascitis and large necrotic mesenteric lymph nodes. There was also diffuse

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thickening of the entire right colonic wall with non homogenous enhancement and considerable narrowing of lumen.

Based on the clinical findings the clinical differential diagnosis included: gastrointestinal tuberculosis, Crohn’s disease and lymphoma. However Polymerase Chain Reaction (PCR) done on sputum and Purified Protein Derivative (PPD) skin test were negative and Acid Fast Bacilli (AFB) culture on gastric aspirate revealed no organisms. Endoscopy was performed revealing marked diffuse circumferential thickening, ulceration and pseudopolyp formation of the wall of the ilium, caecum and ascending colon with thick exudate. (Fig.1). Exploratory laparotomy was performed revealing a huge retroperitoneal mass at the terminal ilium and right colon with a high clinical suspicion of malignancy. Biopsy was taken from the mass with a tru –cut specimen from the liver. Resected material from the retroperitoneal mass received for histopathologic investigations consisted of a 3-4cm piece of soft tissue. Histopathological examination of the biopsy from the mass revealed fibrocollagenous tissue heavily infiltrated by eosinohils, plasma cells, and foamy histiocytes with fungal elements having broad pleomorphic sparsely septate hyphae surrounded by a thick eosinophilic cuff (Splendor-Hoeplli phenomenon) (Fig.2). Spindle cells with an epithelioid appearance exhibiting mitotic figures including abnormal forms were also seen.(Fig.3) The fungal hyphae were demonstrated on Periodic acid-Schiff stain (PAS) and Gömöri methenamine silver stain (GMS). (Fig.4 & 5) Histopathological examination of the tru-cut liver biopsy was unremarkable. Based on the clinical presentation of intestinal obstruction, ultrasound and endoscopic findings, and the histopathological examination of the biopsy from the mass, a diagnosis of Zygomycetes infection consistent with gastrointestinal basidiobolomycosis was made. Tissue culture for fungal colonies was still in progress at the time of reporting this case. Antifungal treatment with amphotericin B was started with promising response.

Fig.1: Endoscopic view of the right colon showing multiple pseudopolyps in the lumen
Fig. 2: 40 X showing broad sparsely septate transparent hyphae (arrows) amid eosiophils on Hematoxylin & Eosin (H&E) stain.

Fig. 3: 40 X showing plump spindle cells with epitheliod appearance with occasional mitotic figures.
**Fig.4:** 40X showing the fungal hyphae (arrow) on PAS stain.

**Fig.5:** 40X showing fungal hyphe and spores (arrows) on GMS stain.
DISCUSSION:

Basidiobolus ranarum was first isolated in 1955 from decaying plants in the United States and subsequently has been found in soil and vegetations throughout the world.[3] It is sometimes present as a commensal in the intestinal tracts of frogs, toads, turtles, chameleons, horses and dogs.[4] This probably explains the mode of infection in our case as being related to the history of ingestion of a lizard. Subcutaneous mycosis was recognized as the first human case in Indonesia in 1956.[5] The subcutaneous form of the disease has been reported mainly from tropical Africa and sporadically from Asia, Australia, and South America.[6] Disseminated forms of subcutaneous type have been reported from South India.[7] Endemic and occasional submucosal forms have been reported from South East India.[8] The Entomophthorales are true pathogens, infecting primarily immunocompetent hosts. Most patients are in apparent good health before acquiring infection. Basidiobolus ranarum organisms generally do not invade blood vessels and rarely disseminate. However, Guido EL van den Berk et al[1] have presented a case of obstructing colon tumor with associated liver mass and Bigliazzi et al[7] have presented a case of disseminated basidiobolomycosis in an immunocompetent woman, with lung involvement as the first clinical manifestation. The mode of acquisition of the disease remains poorly understood. Ingestion of contaminated food, soil, animal feces or use of contaminated “toilet leaves” for cleaning of the skin after defecation has been considered the likely possibility.[2,9,10] Some authors have also suggested rectal inoculation as a likely route of infection[11].

There are only 21 case reports of gastrointestinal basidiobolomycosis in the literature: 8 from the United States, 6 from Saudi Arabia, 4 from Brazil, 2 from Nigeria, and 1 from Kuwait. All patients had abdominal pain and fever as their main symptoms[12], as in our case, with no response to conventional therapy. Only a few cases of retroperitoneal[13] and pulmonary[14] basidiobolomycosis have been reported.

A case-control study was conducted to identify potential risk factors for gastrointestinal basidiobolomycosis.[6] Owing to sample size limitations, few associations reached statistical significance, but many were suggestive of possible modes of exposure. Ranitidine may contribute to the pathogenesis of gastrointestinal basidiobolomycosis by decreasing stomach acidity and allowing the organism to survive gastric passage, which was identified as a risk factor for bacterial gastroenteritis. A longer duration of smoking also was identified as a potential risk factor. Smoking was proposed to adversely affect mucosal white blood cell (WBC) function, possibly facilitating infection with B. ranarum.[6]

The prognosis of gastrointestinal basidiobolomycosis is usually favorable with two fatal cases reported so far.[1,7] On the basis of the limited information available from a review of the literature, it appears that optimal treatment of gastrointestinal basidiobolomycosis is combined surgical and prolonged medical treatment. Nemenqani et al[10] reported gastrointestinal basidiobolomycosis mimicking colon cancer. Zavasky et al[15] have reported a case of gastrointestinal basidiobolomycosis that was initially treated as inflammatory bowel disease. The main differential diagnosis of gastrointestinal basidiobolomycosis with granuloma are inflammatory bowel disease (IBD) (Crohn’s disease), intestinal tuberculosis, sarcoidosis and amebiasis.[9,15] Perivisceral inflammation, fistulization, perforation, and abscess formation, may be present and mimic Crohn’s disease.[16,17]

Gastrointestinal basidiobolomycosis may masquerade as another clinical entity, delaying definitive diagnosis and treatment with more morbidity especially because of the nonspecific signs and symptoms.[6] It may be emerging as a result of various environmental and demographic factors, such as migration of susceptible persons.
to areas that were not previously populated or contamination of food etc. Increased awareness of this clinical entity will lead to earlier diagnosis and probably less morbidity. As more cases become reported, larger studies may improve our understanding of the risk factors for this disease and ways to prevent it.

In conclusion, gastrointestinal basidiobolomycosis is an emerging infection that leads to diagnostic confusion, morbidity and mortality. Diagnosis of this disease requires a high index of suspicion and awareness.

REFERENCES: