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Case Report

Gastrointestinal Basidiobolomycosis: An Emerging Disease Mimicking Obstructing Colon Cancer, Case Report

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Abstract

Gastrointestinal obstruction is a common scenario that is seen on daily basis at hospitals having a general surgery service we are discussing a rare case of gastrointestinal Basidiobolomycosis that mimicking a presentation of obstructing colon cancer.

Keywords: ***intestinal obstruction, basidiobolomycosis, colon cancer.***

Background

Gastrointestinal basidiobolomycosis (GIB) is considered to be a rare type of mycosis that is characteristically reported in immunocompetent [1]. Among all reported GIB cases, the colon is the most common organ involved and may reach up to 82%. Moreover, surgical intervention is required in most of these cases [2]. The majority of the reported cases have been from the desert region of the USA (mainly Arizona) and from Middle Eastern countries [1].

A review by Vikramet and his colleagues reported that the worldwide occurrence of GIB cases between 1964 and 2010 was 44 cases, with 19 from the USA [3]. The other large pool of cases (18 cases) was reported in a multicentric study from Saudi Arabia by Shreef et al., in 2018 [2]. Many authors observed that most of GIB cases were from the southern region of Saudi Arabia, a region that has warm and humid climate which might enhance the growth of the fungus with subsequent environmental contamination.

No specific risk factors for GIB have been identified; however, prior ranitidine use and prolonged residence in endemic areas may contribute to the risk [5,6].

The organism *B ranarum* may be found all over the world but basidiobolomycosis is most reported in tropical and subtropical areas of the world [6]. It typically causes a subcutaneous disease of arms, trunks and buttocks. The mode of transmission is assumed to be minor skin trauma and insect bites [7].

Clinically, gastrointestinal basidiobolomycosis may mimic malignancy, inflammatory bowel disease or chronic infections like tuberculosis [8,9]. Due to the rarity of the disease and its wide spectrum of presentations, a high index of suspicion is required for accurate diagnosis [1]. Histopathological findings can help in diagnosing the condition; however, the most definitive diagnostic test is fungal culture.

Case report

A 70-year-old diabetic and hypertensive male was admitted to the hospital after complaining of left lower quaderant pain science about ten years back as an on/off attacks. Three weeks before the presentation the pain became progressive but more aggressive for the last week, associated with alternative constipation, mucus diarrhea, anorexia, loss of appetite and weight loss.

He lived in Aseer, in the southern part of Saudi Arabia. Examination was essentially normal apart from tenderness in the lower abdomen. No palpable masses could be felt. The rectum was empty on rectal examination. Investigation revealed a white cell count of 10.000 /ml, hemoglobin of 16 g/dL, and platelets of 277,000/ml. The eosinophil count was normal 7.5 (NR 0.0-0.6! 10^9/l). Carcinoembryonic antigen (CEA) and cancer antigen 125 (CA-125) not ordered. The C-reactive protein (CRP) was normal.

An abdominal computerized tomography (CT) scan (Fig. 1A & B) confirmed a localized segment of diffuse irregular circumferential mass (cm length X cm transverse diameter) involving the sigmoid colon with surrounding soft tissue stranding, multiple gas foci and small fluid pockets are seen within the thickened wall. Furthermore, no definitive lymphadenopathy, solid organ liver spleen, pancreas and adrenal glands are grossly unremarkable. A left colon mass extending from descending colon to lower sigmoid and no nodule seen in the liver during emergency exploratory laparotomy, so a Hartmann's procedure, involving resection of the rectosigmoid colon with closure of the anorectal stump and formation of an end colostomy, was performed. The postoperative course was uneventful.

A tow site of perforation with serosal hemorrhage was noted on the gross histopathology. Microscopically, Acute diverticulitis with abscess formation and there is chronic mural necrotizing granulomas and abscesses with fungal hyphae. The hyphae have eosinophilic cuff and numerous eosinophils noted with characteristic Splendore-Hoeppli phenomenon.

This is an intense amorphous esoinophilic substance with star-like appearance surrounding the micro-organism. Periodic acid-Schiff (PAS) and Gomori methenamine silver (GMS) stains were positive in the fungal walls. These features were highly suggestive of colonic basidiobolomycosis (Fig.1). Since the disease was not suspected preoperatively, the tissue was not sent for fungal culture.

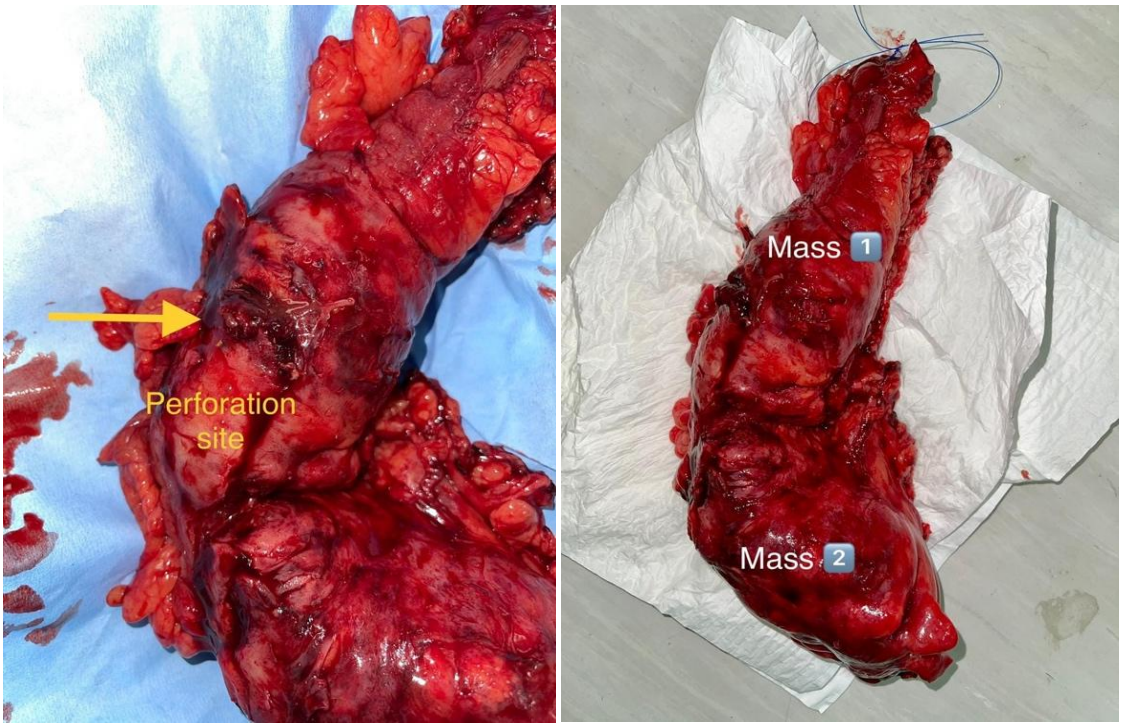

Figure 1: Rectosigmoid colon

Discussion

Basidiobolomycosis is an emerging fungal infection that manifests in the skin and rarely involves other systems. Visceral involvement by basidiobolomycosis is uncommon.

Within the past 3 decades, sporadic cases of gastrointestinal involvement with B. ranarum have been reported worldwide, and it appears to be an emerging infection in the southwestern United States [1]. (89%) of the 19 US patients with GIB were from Arizona [10]. Within the Middle Eastern countries, most cases of basidiobolomycosis have been reported from Saudi Arabia and the largest number of reported cases came from the southern region of the Saudi Arabia [11-13]. This finding may be explained by the nature of the environment in southern Saudi Arabia, which is warm and humid and considered optimal for fungal growth. In addition, some species of wall lizard that are found in this region may be one of the reasons for the increased prevalence of the disease due to transmission via their excreta [14]. However, the real cause for the relatively high number of cases in this region cannot entirely be explained and should be an area for further research. B. ranarum can be found in soil; decaying vegetable matter gastrointestinal tracts of amphibians (eg, frogs and toads), reptiles (eg, garden lizards and geckos) [15].

The clinical presentation is non-specific Abdominal pain, anorexia, and loss of weight were found in all patients with colonic basidiobolomycosis, and less common symptoms include diarrhea (9.0%) and lower gastrointestinal bleeding (13.6%) [1]. Our case presentation was suspicious for obstructive colon cancer in view of the patient's age. The presence of bowel perforation. Although thickening of the bowel wall was seen in 25% of cases, bowel perforation, as seen in our patient, is very uncommon and occurred in only two patients [1].

Remarkably, our patient's eosinophil count remained normal before and following surgery. Peripheral eosinophilia is observed in 76-94.0% of the cases [1,13,14]. Our patient Specimens are sent for histopathology and did not have a tissue culture and he is from Aseer area, which is endemic for GIB, and had a classical pathological picture. Although Splendore-Hoeppli phenomenon can occur in a number of infectious and eosinophilic non-infectious conditions, its presence in a male patient from an endemic area should raise suspicion of GIB [16]. On the other hand, the tissue should be sent to the laboratory for a culture. Lack of awareness of these points and putting the specimen in formalin or even saline, may be the cause of high negative culture rates [3,7].

Optimal treatment of GIB requires combined early surgical intervention and prolonged use of antifungals. A combination of surgery plus antifungal treatment was used in the majority (77.5%) of patients [17].

Azoles are the usual effective antifungals used. There is well-established experience with prolonged itraconazole treatment. Recently, voriconazole has been used with good success, while posaconazole is described in a number of case reports. Despite aggressive treatment, the mortality remains high at 20% within 2 years [1].

High index of suspicion, early intervention, and close follow-up are essential to improving the prognosis.

Conclusion

GIB is a potentially lethal fungal infection and considered to be a surgical condition, which affects immunocompetent individuals in hot arid regions of the world, such as Arizona in the United States and southern region of Saudi Arabia. High index of suspicion, early intervention, and close follow-up are essential to improving the prognosis.

Due to a limitation of the cases and non-specific presentation, GIB should be considered in the differential diagnoses specifically in those who sharing the same environmental and geographical distribution as The Southwestern region of Saudi Arabia. The definitive diagnosis was based on characteristic histopathologic findings after a surgical intervention, tissue culture should be considered in such a case. However early diagnosis of GIB with prolonged treatment with Antifungal treatment offering the best chance for curing this disease are remain challenging in order to avoid major surgeries. We recommend to a targeted screening in the endemic area.

List of abbreviations

GIB: Gastrointestinal basidiobolomycosis

CEA: Carcinoembryonic antigen

CA-125: Cancer antigen 125

CRP: C-reactive protein

CT: Computerized tomography

PAS: Periodic acid-Schiff stain

GMS: Gomori methenamine silver stain

Ethics approval and consent to participate

Taken

Data Availability

Available on the corresponding author upon a responsible request.

Conflicts of Interest

There is no conflict of interest

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Authors' contributions

All authors equal contribution

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