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GASTROINTESTINAL BASIDIOBOLOMYCOSIS A FUNGAL INFECTION THAT MAY BE POTENTIALLY LETHAL

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ABSTRACT

Background: Basidiobolus ranarum belongs to the Entomophthorales order and the Zygomycetes class. This fungus is an environmental saprophyte that can be found in soil and rotting vegetables. Primarily restricted to tropical regions including Asia, Africa, and South America. It might cause chronic inflammatory diseases, mostly affecting subcutaneous tissue. Systemic infections involving the gastrointestinal tract are extremely rare.

Objective: studying the possibility of infection by basidopolomycosis in emergency surgical cases and its management and follow up.

Case presentation:Herein, we presented 5 patients from the emergency department with acute abdominal pain with or without Abdominal mass. Leukocytosis with esinophilia was found in all patients. CT abdomen and pelvis with oral and Intravenous (IV) contrast were done. Colonoscopy was done for 2 patients.

Results: In those five patients, the diagnosis was established after extensive colonic surgery in three patients, and after colonoscopy with biopsy for the other two patients. After establishing the diagnosis, all patients received antifungal therapy. In those who had undergone extensive colonic surgery, two patients died and one recovered. In those who were diagnosed early by colonoscopy, both patients recovered without surgery.

Conclusion: Fungal infection should be among the differential diagnoses for patients presented with an abdominal pain or masses in endemic regions of the world.

Keywords: Gastrointestinal basidiobolomycosis, Fungal infection, Abdominal mass, Basidiobolus ranarum.

INTRODUCTION

Basidiobolomycosis is a rare disease caused by the fungus Basidiobolus ranarum (B. ranarum), an environmental saprophyte found worldwide. It can saprophytically live in the intestines of mainly cold-blooded vertebrates and on decaying fruits and soil*(Pezzani et al., 2019).*It has been isolated from decaying vegetation, foodstuffs, fruits, and soil and from the gastrointestinal tracts of reptiles, amphibians, fish, and insectivorous bats (*Mendoza et al.*, 2015).Most cases of basidiobolomycosis have been reported from tropical and subtropical regions of Africa, South America, and Asia *(Ezzedien et al.,2019).* Patients with B.ranarum infection may present with subcutaneous, gastrointestinal, or systemic lesions. Recently, theetiologic role of B. ranarum in gastrointestinal infectionshas been increasingly

recognized (*Dalal et al., 2009)*. Ingestion of soil, animal feces, or contaminated food is the most likely route of infection reported with GIB, most often involving the colon. Most of these cases have been reported from Saudi Arabia, and the majority came from the southern region of the country**(***Mohammadiet al., 2019).* Symptoms include fever, abdominal pain, diarrhea, constipation, weight loss, and rarely, chills and rigors. Gastrointestinal basidiobolomycosis (GIB) poses diagnostic difficulties, as its clinical presentation is nonspecific, with no identifiable risk factors. All age groups are susceptible, and the condition was reported in children and adults (*Abdollahi A, and Sadeghpour A, 2018).* It was first reported in 1964, and there have been 174 cases reported till July 2021,colonic involvement was reported in 111 cases **(***Maisa et al., 2022).*

the present work aimed to study the possibility of infection by Basidiobolomycosis in patients presented by acute abdominalpain, abdominal masses or intestinal obstructionespecially in hot areas of the world.

PATIENTS AND METHODS

This retrospective study was carried out in the general surgery department, of MuhayilGeneral Hospital, (a hot temperate area in the southwestern part of Saudi Arabia) from January 2019 toDecember 2020. A total of 5 patients included in this study presented to the emergency department with acute abdominal pain with or without Abdominal mass.All patientswere subjected to a full detailedclinical history, with meticulous general and local abdominalexaminations. Laboratory tests in the form ofdifferentiated complete blood pictures, liver, renal function tests, and blood glucoselevels,with coagulation profiles, were done.Radiological examination in the form of abdominopelvic ultrasonography, chestx-ray, andCT abdomen and pelvis with oral and intravenous (IV) contrast were done.Colonoscopy was done for2patients.

RESULTS

Five patients were either born in Muhayilor lived for a long time there. Three Saudi, one Yamani, and one Palestinianpatient were included in the present study. Three of them were males (60%) and two were females (40%). A variety of ages were founded including 8,29,30,40 and 65 years. One patient was diabetic, but the others were immunocompetent. Laboratory investigations revealed leukocytosis with esinophilia. The diagnosis was established after extensive colonic surgery in three patients and after colonoscopy with biopsy for the other two patients. After establishing the diagnosis, all patients received antifungal therapy. In those who had undergone extensive colonic surgery, two patients died and one recovered. In those who were diagnosed early by colonoscopy, both patients recovered without surgery (Table 1).

Representative cases (Table 1)

FIRST CASE

A Male patient 65 years old has lived for about 30 years in the southwesternarea of Saudi Arabia (Muhayil) which is a hot temperate area. He presented to the emergency room witha picture suggesting acute intestinal obstruction in the form of colicky abdominal pain,distension, and absolute constipation. Urgent resuscitation by intravenous fluids with antibiotics followed by insertion of nasogastric tube and urinary catheter.X-ray abdomen in

erectand supine positions was done that showed multiple air fluid levels.CT scan abdomen and pelvis with oral and intravenous contrastwerealso done that showed a left colonic mass that rose the suspicion of malignancy.left hemicolectomy with colostomy was done. The specimen was sent for histopathological assessment with a surprising result ofbasidiobolomycosis. The patient was consulted by a team from the tropical medicine department, and started an IV antifungal itraconazole 200 mg iv /12 h during admission, then started oral itraconazole tab (100) mg was given orally and planned to continue for 6 to 18 months.The patient was discharged from the hospital in a stable condition. Three weeks later, thepatient returned to the emergency room with generalized edema and jaundice. The patient was shifted to the ICU and underwent a pan CT scan that showed multipleliver abscesseswith brain edema. The condition deteriorated into a coma in spite of massive therapy and the patient died after 1 month.

SECOND CASE

Male patient 40 years old who was born and lived in Muhayil presented to the ER (emergency room)with acute abdominal pain,anda palpable mass in the right iliac fossa. Blood tests showed leukocytosis(30000) with esinophilia. CT scan abdomen and pelvis with oral and intravenous contrast showed an ileocecal mass and abdominal collection. He had undergoneurgent laparotomy and right hemicolectomy with ileo-transverse anastomosis. The specimen was sent for histopathological examination that revealedbasidiobolomycosis.The patient was referred to the infectious disease team and started antifungal treatment.He was discharged in a stable condition. Three months later, he developed severe hypoalbuminemia with generalized edema.Hehas admittedin the ICU, resuscitation started but the condition deteriorated, and the patient died from heart failure.

THIRD CASE

A male patient 29 years old who born and lived in Muhayil presented to the ER with a picture suggesting acute appendicitis with leukocytosis and esinophilia.US revealed acute appendicitis witha pelvic collection. Appendectomywas done followed by discharge after 2 days in a stable condition. Two weeks later,the patient returned to the ER with acute abdominal pain,CT scan abdomen and pelvis with contrastdone, and revealeda mass in the ileocecal region (Fig.3). The patient underwent right hemicolectomy, histopathological examination revealedbasidiobolomycosis. He started oralantifungal therapy and followed up for2years with a team of infectious diseases with no symptoms or signs of recurrence.

FOURTH CASE

A female patient 30 years old presented to the ER with acute abdominal pain, watery diarrhea, and repeated vomiting.Bloodtestsshowedleukocytosis with esinophilia. Basidiobolomycosis was suspected, so colonoscopy was done that showed superficial ulceration and congestion of the rightcolon with a biopsy revealed basidiobolomycosis.(Fig.5,6). Thepatient had consulted by the infectious disease team with oral antifungaltherapy started with follow-upfor2years without signs or symptoms of recurrence.

FIFTH CASE

A female child 8 years old presented to the ER with right iliac fossa pain,nausea,vomiting, and low-grade fever. The blood tests revealed leukocytosiswith esinophilia.The US showed no signs suggesting appendicitis,colonoscopy was orderedwhichrevealedileocecal congestion and edema. A biopsy was taken and basidiobolomycosis was confirmed. Oral antifungal therapywas started by the infectious diseaseteam for 1 year without signs of recurrence.

Table (1): Site of the disease, presentation, professional diagnosis, management, and outcome:

NO of cases	PRESENTATION	DIAGNOSTIC WORKUP	MANAGEMENT	OUTCOME
2	Watery diarrhea, vomiting, abdominal pain.	Leukocytosis with esinophilia Thickened terminal ileum and cecum Colonoscopy and biopsy	Antifungal therapy and follow up	Cured
1	Right iliac fossa pain, tenderness,with rebound tenderness	-Leukocytosis with esinophilia -Diagnosed as appendicitis laboratory and radiological. Histopathology after right hemicolectomy	Appendectomy followed by right hemicolectomy. Then antifungal therapy	cured
1	Right iliac fossa mass, loss of weight	Leukocytosis with esinophilia histopathology afterright hemicolectomy	Rt hemicolectomy followed by antifungal	Died
1	Left colon mass with large bowel obstruction	Leukocytosis with esinophilia Histopathology after left colon resection Eosinophilia	Left hemicolectomy with colostomy followed by antifungal	Died

Operative data

Under general anesthesia, laparotomy with right hemicolectomy (Fig. 4) was done in two patients, a left hemicolectomy was done in onepatient, appendectomy was done in onepatient. The fungal mass was soft mass, large in size, easily dissectible from the surrounding structure, with large soft multiple mesenteric lymph node enlargements.

Post-operative outcome:

1- One patient developed a fungal liver abscess (Fig. 3) followed by death.

2-Recurrent mass in the cecum after appendectomy (Fig. 3) in one patient.

3-One patient died from severe hypoalbuminemia and generalized edema that cannot be corrected by albumin supply then died from heart failure.

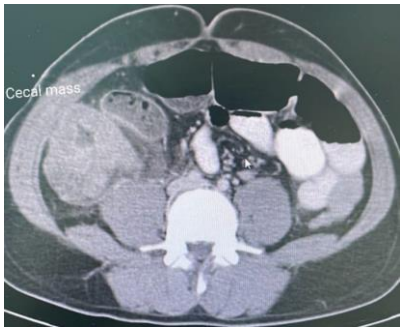


Figure (1) CT showing fungal ileocecal

Figure (2): multiple fungal liver abscess in CT

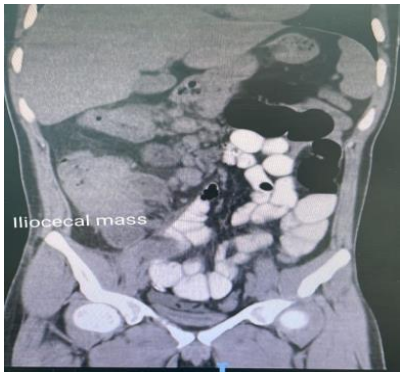
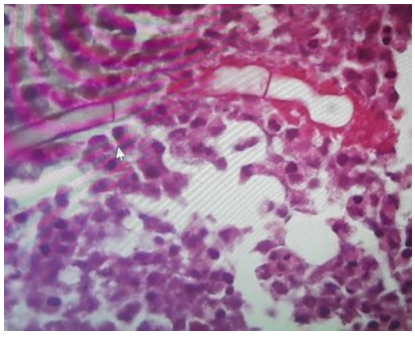


Figure (3): CT showing ileocecal fungal

Figure (4) Right hemicolectomy for resected fungal mass



Figure(5):slide show basidopolomycosis fungus

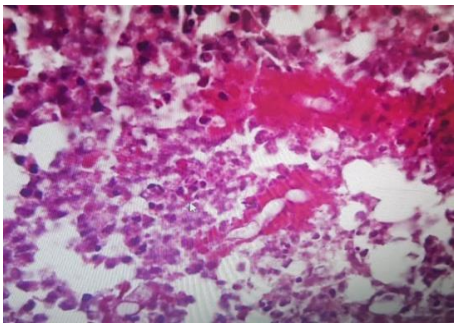


Figure (6): slide show dense infiltration around the fungus by eosinophil

DISCUSSION:

Gastrointestinal basidiobolomycosis is a known rare infection caused by the fungus Basidiobolus ranarum which belongs to the Entomophthorales order and the Zygomycetes classthat affects immunocompetent patients. This fungus is an environmental saprophyte that can be found in soil and rotting vegetables**(***Takrouni et al., 2019).*Primarily, it is restricted to tropical regions including Asia, Africa, and South America. It might cause chronic inflammatory diseases, mostly affecting subcutaneous tissue. Systemic infections involving the gastrointestinal tract are extremely rare*(Shreef, et al. 2018)*. In all cases in this study the fungus affected the gastrointestinal system.

Colonic with liver involvement was reported in six cases (*Bander et al.,2014).*Most of the previous series involve one or two cases. one case of left colon affection with liver abscess and colonic perforation was reported in two cases**(***El-Shabrawi, M.H.; and Kamal, N.M 2011)*. In this study,5 cases of basidiobolomycosis colon, appendix, and terminal ileum were diagnosed by histopathological examination of resected colon, appendix, or by colonoscopic biopsy, one case complicated by liver abscess, no cases presented with GIT perforation.

In this study, there were one pediatricand one adult female whowasdiagnosedearly.So, they didn't require surgery, and theywere treated by antifungaltherapy only.The newly discovered cases increased refer to the development in diagnostic modality and thinking in this fungal diseaseas one of the differential diagnoses (you cannot come across rarities unless you are aware of it).

The rarity of this disease added to the unfamiliarity of the concerned physicians with it, and its clinical endoscopic, and radiologic similarity to the more common abdominal malignancy, inflammatory bowel disease, or tuberculosis**(***Van den Berket al., 2009).*

In this study, three cases were presented by abdominal masses like tumors, two on the right colonone in the left colon, and the fourth one with appendicitis.

On histopathology, thin‑walled irregularly branching hyphae with occasional septae may be seen surrounded by dense eosinophilic reaction (Splendore–Hoeppli phenomenon).**(***Zekavat, et al., 2015),*these morphologic characters were seen in all histopathological examinations of our cases.

Prolonged antifungal treatment was followed in all cases either who had undergone surgery or not. (*Almoosa, et al.,2017).* In this series, two patients received antifungal therapy without surgery for 18 months which is the longest period of therapy. One patient with right colon and liver affection had received antifungal therapy for one and a halfmonthsbut died from liver cell failure. The second patient had received antifungal therapy for three months but died also from heart failure. In this regard, the choice and duration of the antifungal therapy were at the hands of the physicians in conjunction with the clinical pharmacist, and as the disease is rare with no previous local experience exists, different drugs were chosen without a clear indication of the superiority of one over the other. At

anyhow, a regular short-term follow‑up at the start of antifungal therapy with periodic liver and renal functions is mandatory (Saeed *et al.,2014)*.In this study, IV antifungal itraconazole 200 mg iv /12 h was given during admission then oral itraconazole tab 100 mg was given orally for 6 to 18 months.

Awareness of the disease amongst physicians and pathologists would contribute to the successful treatment of more recent cases and would result in a significant reduction in the disease's morbidity and mortality rates. Further research on the risk factors that lead to GIB diagnosisis highly recommended.

Conclusion: The diagnosis of GIB requires a high index of suspicion, and must be included in the differential diagnosis of abdominalpain, masses associated with fever, weight loss, and eosinophilia, especially in tropic and subtropics regions. Early diagnosis saves the patient from extensive surgery and givesa better prognosis.

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عدوي الجهاز الهضمي بفطر باسيدوبولاس مرض نادر ولكن خطير

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خلفية البحث: الالتهاب الفطري باسيدوبولاس من الالتهابات الفطرية النادرة التي تصيب الانسان حيث تم اكتشاف 174 حاله في جميع انحاء العالم حتى عام 2021 وتنتقل العدوي عن طريق اللمس او الطعام الملوث بالفطر الذي ينتقل له عن طريق البرص والحشرات ذات الدم البارد، وايضا يوجد في الخضروات والفواكه المتعفنة. ويعيش هذا الفطر في الاماكن الحارة الرطبة من العالم، ويتسبب بآلام بالبطن تشبه اعراض النزلة المعوية أو أعراض تشبه أعراض التهاب الزائدة الدودية.

أو كتله نسيجيه بالبطن قد تحدث انسدادا معويا أو تتشابه مع اورام الجهاز الهضمي ولا يشترط أن تكون مناعة المريض ضعيفة حتى يصاب بهذا الفطر بعكس ما هو شائع في الإصابة بالفطريات.

المرضي وطريقة البحث: في هذه الدراسة تم تسجيل المسار الاكلينيكي لخمسة حالات تم اكتشافهم في الجنوب الغربي للمملكة العربية السعودية. وهذه الحالات إما ولدوا وعاشوا في هذه المنطقة أو عاشوا زمنا طويلا بها وقد تم دراسة طرق العلاج ونتيجته.

نتائج البحث: كانت أعمار الحالات تتراوح من 8 سنوات الي 65 سنه، وقد اصاب المرض الناحية اليمني من القولون في أربع حالات حيث تم تشخيص حالتين منهم بمنظار القولون وأعطوا علاج مضاد للفطريات لمدة سنة ونصف وتمت متابعتهم لمدة سنتين بدون علامات تدل علي عودة الإصابة، وتم استئصال القولون الايمن لحالتين منهم، واصاب الناحية اليسرى في حالة واحدة وكانت عبارة عن كتلة بالقولون الأيسر، وتم استئصاله وتوفي المريض بعد ثلاثة أشهر.

الاستنتاج: يوضع في الاعتبار احتمال الاصابة بهذا الفطر في حالات الالام الحادة للبطن، أو حالات الكتل البطنية خصوصا في الأماكن الشائع بها هذا المرض حيث ان التشخيص المبكر يقي المريض من الجراحه ، ونتائج العلاج افضل.

الكلمات الدالة: عدوي الجهاز الهضمي بفطر باسيدوبولاس، الاتهاب الفطري ، كتل البطن ، باسيدوبولاس.