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CASE REPORT

Gastrointestinal Basidiobolomycosis (GIB) in Jeddah, Saudi Arabia

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INTRODUCTION

Basidiobolomycosis is a well known, but rare and under diagnosed condition. It is adeep-seated fungal infection caused by the organism Basidiobolusranarum a worldwide environmental saprophyte belonging to the Entomophthorales order of the class zygomycetes[1,2] and is found in soil, decaying plants,and gastrointestinal tracts of amphibians, reptiles, horses, dogs and bats. It has been found to be of higher incidence in males (a male to female ratio of 11:1) and is seen in different age groups, the mean age being 37 years[2]. Eidam first isolated Basidiobolusranarum in 1886[3,9]. Unlike other fungi, Basidiobolus Ranarum, can cause significant disease in the immune-competent hosts[4].

It is usually known to affect the skin and soft tissues and is uncommonly seen to involve the extracutaneous systems. Systemic dissemination is extremely uncommon. It has caused diagnostic difficulties as the clinical manifestations are various and have not been well defined[3]. The first reported case of GIB was in a 6-year-old Nigerian boy in 1964[3]. Basidiobolus Ranarum requires a warm and humid environment for its growth and so GIB is most commonly reported in the tropical and subtropical areas.

The majority of the reported cases have been from the desert region of the USA (mainly Arizona) and from Middle Eastern countries[5].

Gastrointestinal basidiomycosis has been increasingly recognized in Saudi Arabia with the largest number of the cases being detected in Tohama, in the southern area[5].

The clinical features of gastrointestinal basidiobolomycosis may vary but include abdominal pain, nausea, vomiting, diarrhea, or an abdominal mass[3,6].

Diagnosis is made based on characteristic histopathology findings of fungal hyphae and the gold standard for diagnosis is culture[3]. Treatment includes long-term antifungal treatment (6 months to 2 years) and possible surgical resection. Whether surgery is performed or not, prolonged antifungal therapy is always required[7].

CASE REPORT

Our case is a 71-year-old gentleman who presented to our hospital for the first time on March 2021. He's known to have type 2 diabetes, hypertension and presented to our hospital with right hypochondriac abdominal pain with an intermittent fever, fatigue and loss of appetite for a period of 2 weeks.

The pain was continuous, moderate to severe, radiating to the back and was not related to food intake or relieved by analgesics. It was associated with an unintentional 12 kg weight loss over 2 months. The patient also noted a history of yellow sclera, fatigue and muscle pain.

He had an exploratory laparotomy done 4 years prior to presentation for a gunshot wound and a history of cataract surgery but no recent surgeries. There was no history of a subjective fever before this episode; there was no abdominal distention, nausea, vomiting, diarrhea or constipation. There was also no history of bleeding from any orifices. No urinary or respiratory symptoms were noted. No contact with sick individuals. He is a non-smoker and does not have any known allergies.

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Upon examination the patient wasvitally stable and initially afebrile.He was conscious and oriented,fatigued, with dry mucous membranes and muscle wasting. On abdominal examination he had an enlarged liver with right hypochondrial discomfort on deep palpation. His chest was clear and he had a normal cardiovascular examination. The rest of his examination was unremarkable.

Laboratory investigation showed the presence of leukocytosis, WBC count 16.8 (monocytes 1.07, eosinophil 0.567), Hemoglobin 8.3 (which was normocytic, normochromic with a high ferritin level, low serum iron and TIBC), Platelet count461. High inflammatory markers (CRP 356), ESR?, Procalcitonin level of 9.1

He had an acute kidney injury with an uncontrolled blood sugar and pseudo-hyponatremia (no previous high renal function or history of CKD, and he had a good urine output). His renal function improved with good hydration and treatment of the infection with adjusted antibiotic doses.

Hepatitis serology: negative HCV PCR, negative HB sAg, low surface Ab 5.55 (non immune), positive hepatitis B core Ab Liver function tests were normal apart from a rising ALP to 690. Tumor markers: CEA 1.7, Ca 19.9 (upper limit 37), AFP normal (<2), Ca125?

Covid negative, Sputum for AFB stain was negative. Blood culture: enterococcus faecium (vancomycin and ampicillin resistant)

Hospital course: He was admitted in the hospital with a liver lesion found on ultrasound (well defined 7.5 cm hyperechoic lesion at the right hepatic lobe) and cultures were sent. He was also started on tazocin for a possible infection. Further imaging with a CT Chest Abdomen and pelvis with IV and oral contrast was carried out.

CT Chest showed multiple mild mediastinal and moderate right anterior cardiophrenic lymphadenopathy, a mild right pleural effusion with no obvious pulmonary masses or nodules.

CT Abdomen and pelvis showed segmental transverse colon and focal splenic flexure marked mural thickening versus a mass lesion with adjacent severe inflammatory fat stranding and a mesenteric soft tissue mass. A right hepatic mass (measuring 7 x 9 x 7.9cm) was also seen extending beyond the liver capsule to involve the posterior abdominal wall muscles. There was also evidence of a well-defined hypodense soft tissue mass seen just posterior to the transverse colon measuring 3.2x3.1cm suggesting a pathological lymph node versus a mesenteric deposit. Other mild para-aortic, aortocaval and mesenteric lymph nodes were seen with bilateral cardiophrenic lymphadenopathy. Both kidneys showed lobulated outline with perphrenic fat stranding and minimal fluid.

An MRI of the Liver showed a right hepatic lobe, well-defined round shaped focal liver lesion at segment VI with irregular micro lobulated shaggy outlines measuring 8 x 7 x 7with the differential including a liver abscess, but a possibility of an infiltrative mass could not be ruled out.There was re-demonstration of the previously described colonic mass and inflammatory fat stranding with multiple lymphadenpopathy.

There were 3 spikes of fever despite his antibiotic coverage (metronidazole 9 days and tazocin) and without a good clinical and laboratory response to antibioticstazocin was escalated to meropenem.

A liver biopsy was arranged and two core biopsies weretaken from the liver by interventional radiology through U/S guidance. The result revealed two cores of necrotic tissue heavily

infiltrated by neutrophils and it contained numerous broad septateth in walled fungal hyphae with Splendore-Hoeppli phenomenon; no evidence of malignancy was seen on these biopsies.

Culture of the Liver tissue came out for enterococcus faecium (vancomycin resistant) in addition to basidiobolus species.
The gastroenterology team was involved in patient's care as the MRI showed colonic thickening and a **colonoscopy** was done. It showed the presence of a large circumferential ulcerated, partially obstructing mass at the distal transverse colon with friable mucosa that is easy to bleed and multiple biopsies were taken.

The biopsies showed mild mixed inflammatory cells infiltrating the lamina propria with the presence of eosinophilia.An ulcer slough contained neutrophils and fibrin material. No evidence of microorganisms or a granuloma was identified and there was no evidence of dysplasia or malignancy.

The infectious disease team was consulted and voriconazole was started at 6mg/kg for 2 days and a maintenance dose of 4mg/kg BID.

The patient markedly improved on voriconazole, clinically with a decrease in abdominal pain in addition to lowering of inflammatory markers and WBC levels on the antifungal and adjusted antibiotics.

Possible drainage of the abscess was considered but upon review, the lesion was found to be mainly of necrotic tissue and so no drainage was required.

Preliminary blood cultures showed the presence of gram-positive cocci so vancomycin was added. Final cultures then came out forVRE (enterococcus faecalis) and vancomycin was switched to Daptomycin.

Two weeks after admission the patient developed tachypnea with a slight drop in saturation and a few scattered wheezeswhere heard on auscultation with decreased air entry in the right base.A CXR was done which showed no local infiltration but had increased vascular markings with the presence of a fissure edema. He developed pulmonary edema with the IV fluids so an echo was done and reported an ejection fraction of 40%, started on a low dose of diuretics and cardiology were contacted for which they recommended starting an ACEI and provided a follow up appointment to reassess the patient for possible cardiac catheterization.

He also had a persistently low hemoglobin level requiring 3 to 4 units of blood. Iron study revealed serum iron level of 3. There was no melena or other forms of active bleeding.He was scheduled for an upper endoscopy

He received a 13-day course of antifungal treatment and 10 days of daptomycinin hospital and was then discharged on voriconazole 200mg BID for minimal 6 months with clinical and laboratory investigations, radiologic follow up of liver lesions, in addition to linezolid for 1 week for his VRE bacteremia. Meropenem was discontinued upon discharge.

DISCUSSION

Extracutaneous involvement is extremely rare but in the past decade, many cases of extracutaneous basidiobolomycosis have been reported. GI basidiobolomycosisis an emerging fungal infection that causes a serious, and occasionally fatal, disease. Most cases were misdiagnosed as other chronic granulomatous diseases or malignancies[7]. The differential diagnosis of GIB with granuloma includes inflammatory bowel disease, intestinal tuberculosis, sarcoidosis, amebiasis and malignancy. (6)
In this case, the patient was admitted for over a month with extensive workup.

It was thought to be a colonic tumor with liver metastasis; a biopsy was taken and was negative for malignancy. Diagnosis was made by the characteristic fungal hyphae on histopathology with Splendore-Hoeppli phenomenon and confirmed by microbiologic methods through culture and sensitivity.

History and radiologic findings were consistent with an underlying malignancy and it is very unlikely to find an infection with the same signs and symptoms but eventually it turned out to be a very rare fungal infection which can clinically mimic a malignancy but this is a treatable condition and if left untreated can be very fatal and carries a high mortality rate.

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