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CASE REPORT ARTICLE

GASTROINTESTINALBASIDIOBOLOMYCOSIS.

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Abstract

Basidiobolomycosisis a rare infection caused by the fungus *Basidiobolusranarum*, which is a known cause of subcutaneous zygomycosisbut rarely gastrointestinal manifestations have been described; the latest review reported 44 patients most were from the United States (19 patients [43%], of whom 17 [89%] were from Arizona) or Saudi Arabia (11 [25%]). Gastrointestinal basidiobolomycosis poses diagnostic difficulties; its clinical presentation is nonspecific, there are no identifiable risk factors, and all age groups are susceptible. This report presents yet another case of this disease in a Saudi male involving the gastrointestinal tract, he received hemicolectomy followed by oral Voriconazole, with successful outcome.

Case presentation:-

A 19-year-old male originally from the south, Najran, Saudi Arabia, was admitted to King Fahad hospital of university in May 2011 with a history of abdominal pain of 2 months duration associated with diarrhea and subjective weight loss. He denied any history of nausea, vomiting, hematemesis nor melena. No history of fever, night sweat, mouth ulcers, joint pain nor skin rash. No contact with febrile or TB patient.

On examination, he was afebrile, and his abdomen was soft but particularly tender at the right ileac fossa where a palpable, firm mass (5 x 5cm) was located.

His laboratory works showed leukocytosis with marked eosinophilia (13.8×103/µL, reference range 4–11 × 103 with 40% eosinophils) and apart from an elevated C-reactive protein 2.2 mg/dl (reference range 0.0 – 0.3 mg/dl) his liver functions; renalvalues, ESR and electrolytes were all within normal. Tuberculin skin test was negative. Computerized tomography scan of abdomen showed irregular circumferential wall thickness and mucosal enhancement of cecum and ascending colon till the hepatic flexure with multiple enlarged necrotic pericolic lymph nodes. (Figure1A)

Barium enema showed an apple core like circumferential focal stricture noted in right colonic flexure measuring 5 cm. (Figure 1B)

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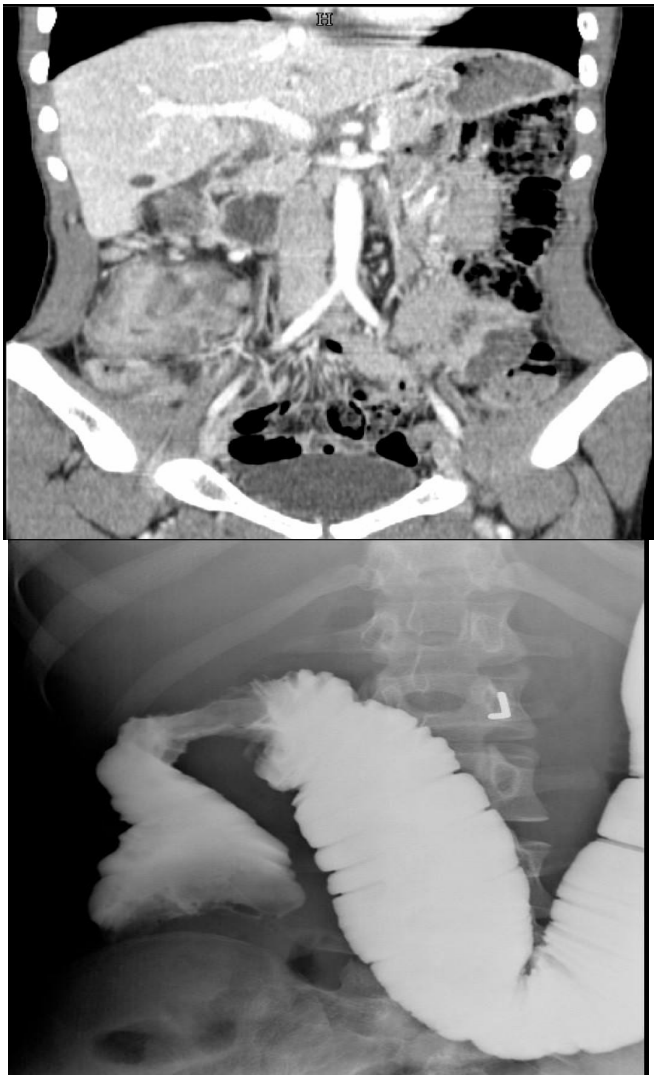

Figure 1:- (A) CT-scan abdomen showing thickened wall of cecum, ascending and transverse colon with multiple enlarged necrotic LN that suggests inflammatory process. (B) Barium enema showing destruction of haustral and mucosal pattern of right colonic flexure and ascending colon, an **APPLE-CORE** like circumferential focal stricture noted in right colonic flexure measuring 5 cm.

Colonoscopy revealed two masses, semicircular cecal mass with necrotic ulcer and a polypoid ascending colon mass (figure 2A). Biopsies were taken and histopathology revealed active eosinophilic colitis and negative AFB culture. (Figure 2B)

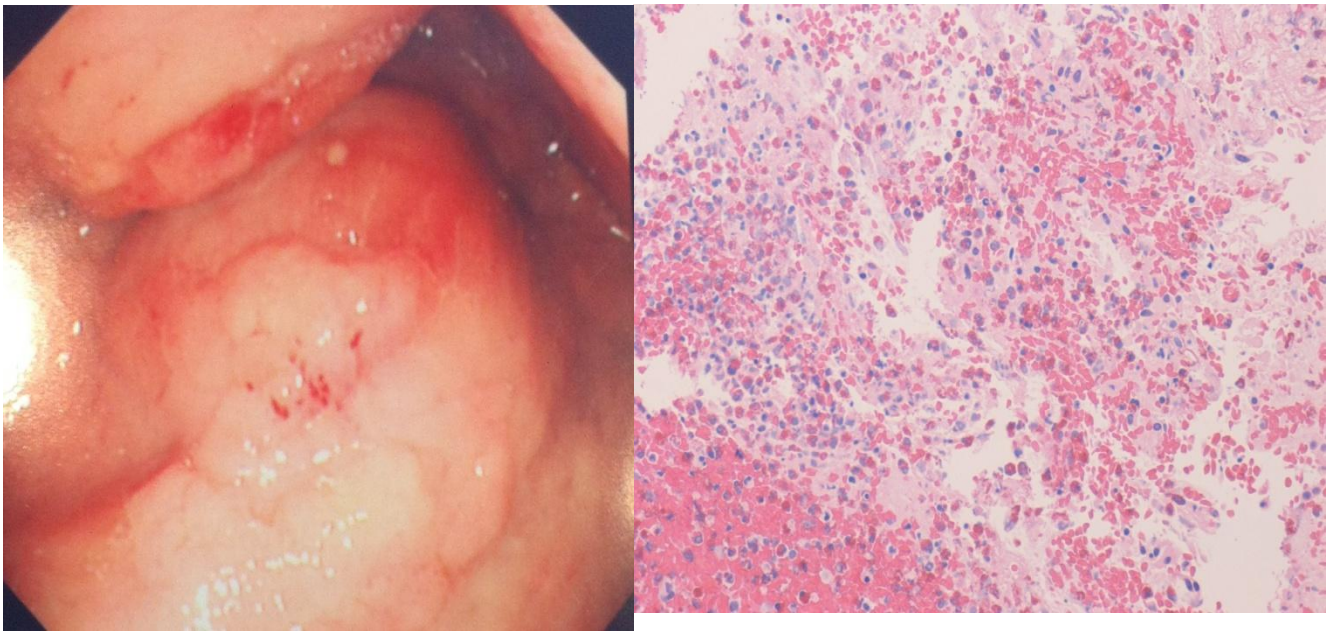

Figure 2:- (A) colonoscopic picture of ascending colon polypoid mass. (B) Histopathology of colonoscopic biopsies revealed active chronic colitis with tissue eosinophilia.

Based on pathological findings patient was started on prednisolone, had period of remission and discharged from hospital, after which he lost follow up for nearly 12 months when he was readmitted again with exacerbation of his previous symptoms that were intractable to even higher doses of corticosteroid. This time his laboratory investigation showed even higher leukocytosis with marked eosinophilia (17x103/µL) and elevated ESR reaching 52 mm/hr.

A repeated CT-scan (Figure 3-A) showed progressive involvement of ileum, the right colon with significant intraluminal narrowing (there was a rim enhancement Structure with pocket of air at ileocecal region most likely perforated appendicitis incorporated in the inflammatory process), Also left colon was involved with submucosal edema which signifies an acute inflammatory process.

As the radiological picture was suggestive of obstructive lesion with impending perforation, the patient was subjected to laparotomy where a right hemicolectomy and ileocolic anastomosis was performed.

Histopathology of resected specimen showed suppurative granulomata with many multinucleated giant cells, engulfing septated irregularly branched wide fungal hyphae surrounded by radiating annular amorphous eosinophlic deposit (splendore-Hoeppil phenomenon), which suggested the diagnosis of basidiobolomycosis. (Figure 3-C) Oral voriconazole, in a daily dose of 200 mg, was started. In his follow-up outpatient visits, the patient remained well, his investigations, including liver and renal function tests, were normal.

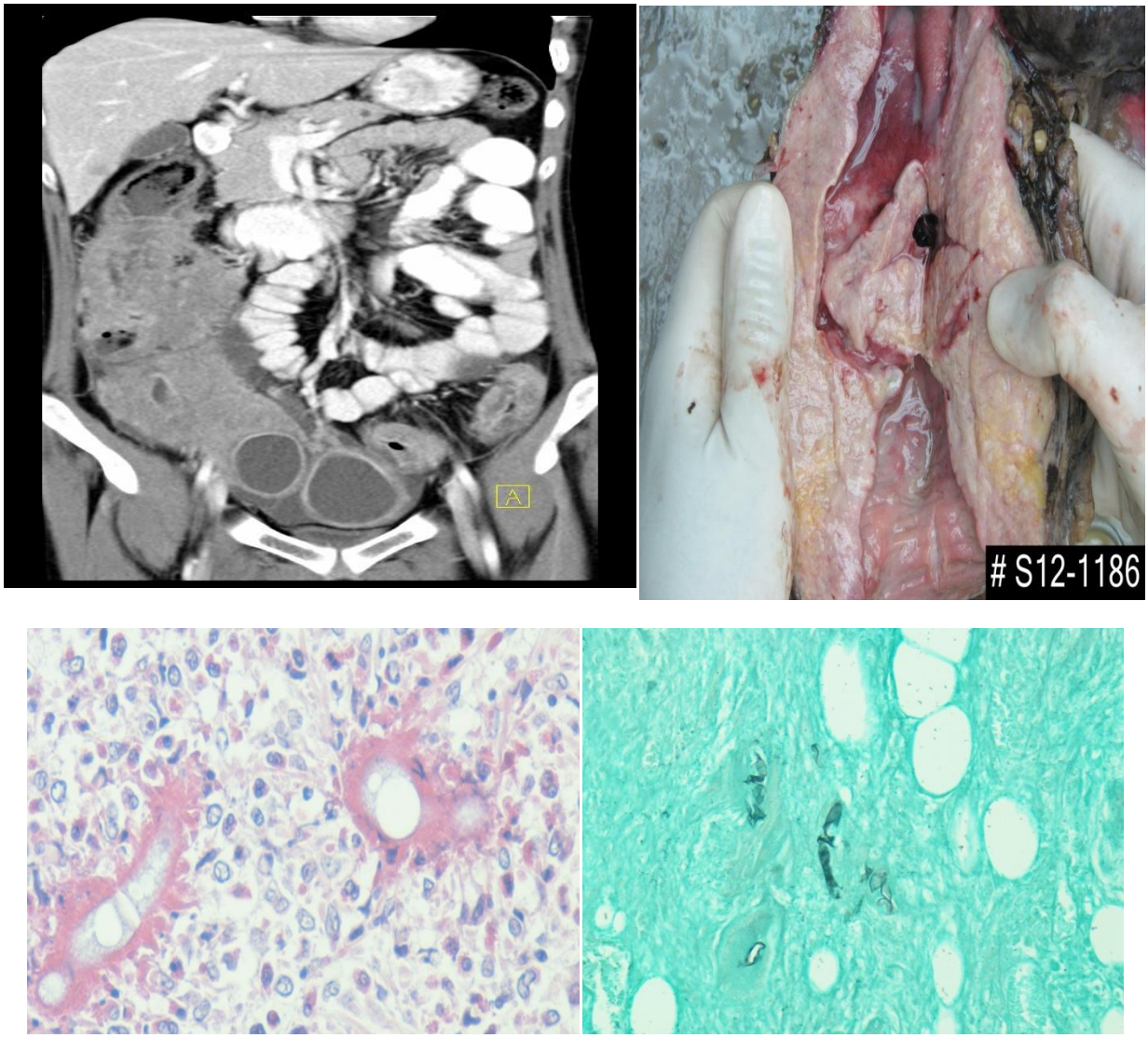

Figure 3:- (A) CT-scan of abdomen showing progressive involvement of ileum which is dilated with wall enhancement, right colon with significant intraluminal narrowing increased wall thickness and enhancement, rim enhanced structure with pocket of air at ileocecal region most likely perforated appendix incorporated in the inflammatory process. (B) Gross picture of the resected colon showing diffusely thickened colonic wall with intraluminal narrowing. (C) Broad, thin-walled, occasionally septated fungal hyphae, surrounded by dense eosinophilic reaction (Splendore-Hoeppli phenomenon). (D) Fungal hyphae stained black by Grocott staining.

Discussion:-

Basidiobolomycosis is a rare infection caused by the fungus *Basidiobolusranarum*, of the order Entomophthorales, of the class Zygomycetes [1]. The most recent review of worldwide cases published by Mayo researchers in March 2012 studied 44 patients (mean age, 37 years [range, 2–81 years]) with gastrointestinal basidiobolomycosis, most were from the United States (19 patients [43%], of whom 17 [89%] were from Arizona) or Saudi Arabia (11 [25%]).

Basidiobolomycosis, which is characterized by chronic subcutaneous induration affecting limbs, trunk, and buttocks, originates mostly in tropical areas of South America, Africa, and Asia [2–4]. Minor trauma, local inoculation, and insect bites appear to be the predominant modes of acquisition [2-4].

Other rare sites include the gastrointestinal tract [1, 5,6], which proved fatal in exceptional occasions [7] (8 out of 44 patient died) and the nasal sinuses [8]. It is unclear how the fungus is introduced into the host's gastrointestinal tract, but this probably occurs through ingestion of contaminated soil, animal feces or food. [6]

The disease, which has been described relatively recently, has radiological features suggestive of inflammatory bowel disease or malignancy. However, contrary to these pathological entities, leukocytosis and eosinophilia are usually present [6]. In the casepresented here, our patient had leukocytosis with eosinophilia, and as previously reported [6], leukocytosis resolved after surgical resection.

In gastrointestinal basidiobolomycosis the colon is the most frequently involved part of the gastrointestinal tract, and patients usually present with mild abdominal pain with a subacute onset, eosinophilia, and on histopathology examination inflammatory changes with many eosinophils [9]. The difficulty in diagnosing the condition is multifactorial. Firstly, the non-specific clinical presentation. Secondly, there are no identifiable risk factors [6]. Thirdly, as the causative agent lies deep beneath the mucosa, colonoscopic biopsies may be no representative. The fourth factor and contrary to expectations, victims of this infection are usually healthy immunocompetent subjects [10].

In regard to the diagnosis,the organism can be isolated from surgically resected tissues. It should be inoculated immediately because it does not survive at 4°C [11]. Sabouraud agar is an adequate medium, and visible growth is usually present 2 to 3 days after incubation at 25 to 30°C. On microscopic examination of the culture material, B. ranarum, can be identified by its characteristic beaked zygospores. The fungal elements appear as broad, pleomorphic, sparsely septated hyphae, which stain faintly with fungal stains (Grocott, Gomorimethenamine silver and periodic acid Schiff, PAS). Definitive diagnosis requires culture of the organism. Serodiagnosis with immuno-diffusion can be employed as an adjunctive diagnostic method. This test is very specic for B. ranarum with no cross-reactivity with other species of the order Entomophthorales[11,12,13,14].

Optimal treatment of GIB combines surgical and medical methods. Patients should undergo resection of all affected bowel segments and debridement of involved tissue that is followed by prolonged treatment with Azoles antifungal for up to two years is usually needed [6, 10, 17]. The best choice of antifungal agent is not clear, but Itraconazoleseams to be reasonable [16].

In conclusion, gastrointestinal basidiobolomycosis is a recently recognized disease, which leads to diagnostic confusion, morbidity and mortality. Diagnosis of this disease requires high index of suspicion, awareness and consideration of its possibility in the differential diagnosis of patients with abdominal masses and eosinophilia.

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