ADITUM
Enrich your Research

Journal of Clinical Case Reports and Clinical Study

Case Series

Gastrointestinal Basidiobolomycosis: A Case Series and Literature Review

Mohaned M Mohammed[1*], Ahmed Albishri[2], Ali Alabbas[3], Sami Elsamani Ibrahim[4], Badriah G Alasmari[5], Jameelah Ali Alqahtani[6], Samah E Mohammed[7], Ali Hawan[8] , Mahmoud Rezk Abdelwahed Hussein[9] , Muhammad Saeed[10] , Yassin Hamid[11] and Eman Jaber Ghazwani[12]

[1]Certificate of Saudi board of pediatric consultant general pediatric (AFHSR).
[2]Pediatric Infectious Diseases, Armed Forces Hospital Southern Region
[3]Pediatric, Armed Forces Hospital Southern Region (AFHSR).
[4]Pediatric, Armed Forces Hospital Southern Region (AFHSR).
[5]Pediatric hematology Oncology, Armed Forces Hospital Southern Region (AFHSR).
[6]Pediatric, Armed Forces Hospital Southern Region (AFHSR).
[7]Pediatric, Armed Forces Hospital Southern Region (AFHSR).
[8]Consultant Microbiologist, Chief of Pathology and Laboratory Medicine, Armed Forces Hospital Southern Region (AFHSR).
[9]European Board of Pathology, Diploma in Dermatopathology, Professor of Pathology/Consultant Histopathologist, Department of Pathology, Armed Forces Hospitals Southern Region (AFHSR).
[10]Pediatric Neurology, Armed Forces Hospital Southern Region (AFHSR).
[11]Pediatric Gastroentrology, Armed Forces Hospital Southern Region (AFHSR).
[12]Pediatric Infectious disease, Armed Forces Hospital Southern Region (AFHSR).

Article Info

Received: February 20, 2024
Accepted: June 28, 2024
Published: August 01, 2024

***Corresponding author:** Mohaned M Mohammed, Certificate of Saudi board of pediatric consultant general pediatric (AFHSR).

Citation: Mohaned M Mohammed, Ahmed Albishri, Ali Alabbas, Sami Elsamani Ibrahim and Badriah G Alasmari.et.al... (2024) "Gastrointestinal Basidiobolomycosis: A Case Series and Literature Review.", Clinical Case Reports and Clinical Study, 11(1); DOI: 10.61148/2766-8614/JCCRCS/168

Abstract:

Basidiobolomycosis is a chronic granulomatous infection of skin and subcutaneous tissue which is caused by Basidiobolus ranarum.

Here we are reporting three cases of Gastrointestinal Basidiobolomycosis from the southern region of Saudi Arabia. The three patients were diagnosed based on histopathology result and CT abdomen. They had different pathological sites with multiple gastrointestinal tract (GIT) involvement, all of them were treated successfully with antifungal monotherapy (Itraconazole) without surgical intervention, and all of them responded well without any relapses.

Keywords: basidiobolomycosis, basidiobolus ranarum, GIT basidiobolomycosis, itraconazole, southern region saudi arabia.

Introduction:

Basidiobolomycosis is an enigmatic fungal infection clinically and radiologically. Basidiobolomycosis is uncommon fungal infection caused by Basidiobolus ranarum that occurs in immunocompetent individuals [1]. Saudi Arabia has the second highest overall reported Gastrointestinal Basidiobolomycosis patients [2].

Basidiobolus ranarum is an environmental Saprophyte it is a member of the order Entomophthorales of the class zygomycete [3]. Found in soil decaying vegetable materials and the amphibians GIT, reptiles, dogs, frogs and bats, it is usually infecting subcutaneous tissues, it can affect visceral organs causing significant disease. The diagnosis of Gastrointestinal Basidiobolomycosis required high suspicion of index because the clinical presentation resemble other disease like lymphoma, inflammatory bowel disease, other GIT infection.

We are reporting three cases of gastrointestinal basidiobolomycosis with

multiple site GIT involvement treated successfully with single antifungal agent (itraconazole) without surgical intervention also discussion of relative literature.

Case I

5 years old Saudi male, from Jizan (southern region of Saudi Arabia) presented to our hospital complain of abdominal pain for 7 months associated with weight loss, fever with night sweating for one month, anorexia, and decrease appetite. On examination, the abdomen was soft, lax, with right upper quadrant tenderness, no organomegaly, with unremarkable other systemic examination. Laboratory tests revealed microcytic hypochromic anaemia (HB 9.5 g/dl), leucocytosis (WBC 21.4 10^g/l), eosinophilia (eosinophil 21,6%) with absolute eosinophilia (4.54 10^g / l), platelets (678 10^g/l), CRP (192,7 MG/L), ESR in the first hour (120 mm/hr.),the patient was admitted for further investigation to exclude malignancy, abdominal tuberculosis, ultrasound abdomen showed liver(span- 101mm), hyperechoic lesion with hypoechoic rim noted in the right lobe of the liver measuring (47X33 mm). CT scan of the abdomen with iv contrast (Figure-1A).

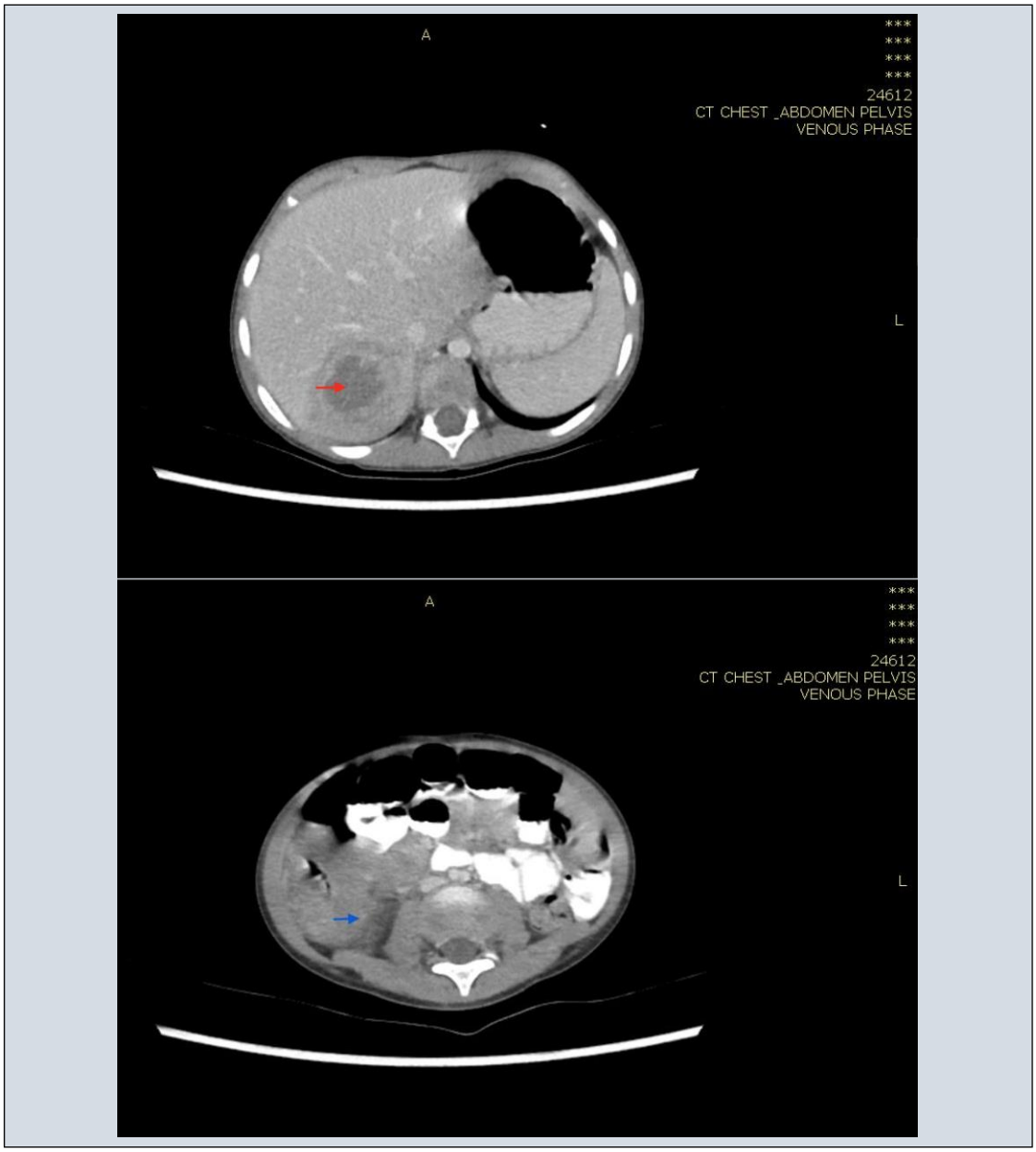


Figure 1A: The liver shows fairly well define peripherally enhancing, centrally hypoattenuating lesion around 45 Hu seeing in the 6th to 7th liver segments it has thick irregular wall measuring (3.8 cm X 4cm X2.8 cm) associated with surrounding peripheral edema (red arrow) otherwise, the liver has normal size and homogenous enhancement with no obvious underlying the parenchymal disease. Significant circumferential wall thickening is noted involving ascending colon (blue arrow) and hepatic flexure and mesentery lymphadenopathy with impression of Basidiobolomycosis fungal infection with right sided bowel wall thickening and the hepatic abscess formation.

CT guided biopsy done from colonic mass biopsy revealed fungal elements are noted by special stain PAS (Figure 1B), was diagnostic for Gastrointestinal Basidiobolomycosis liver and bowel wall.

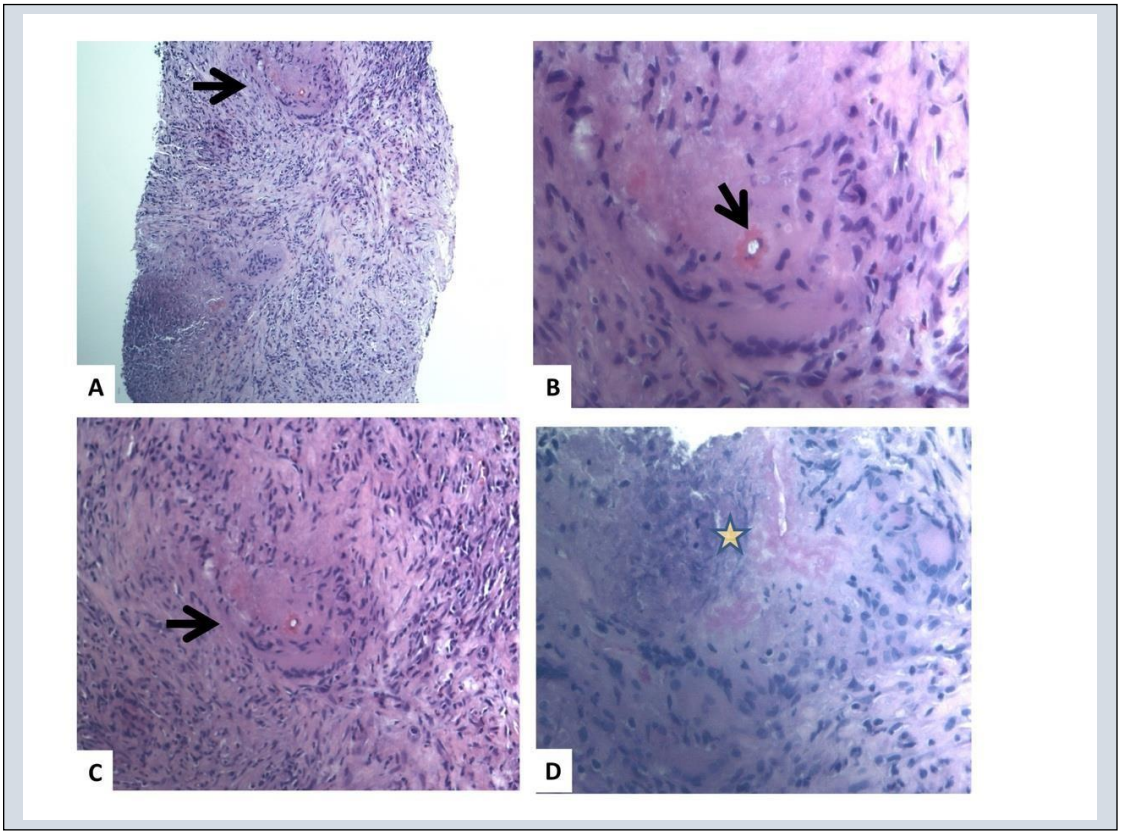
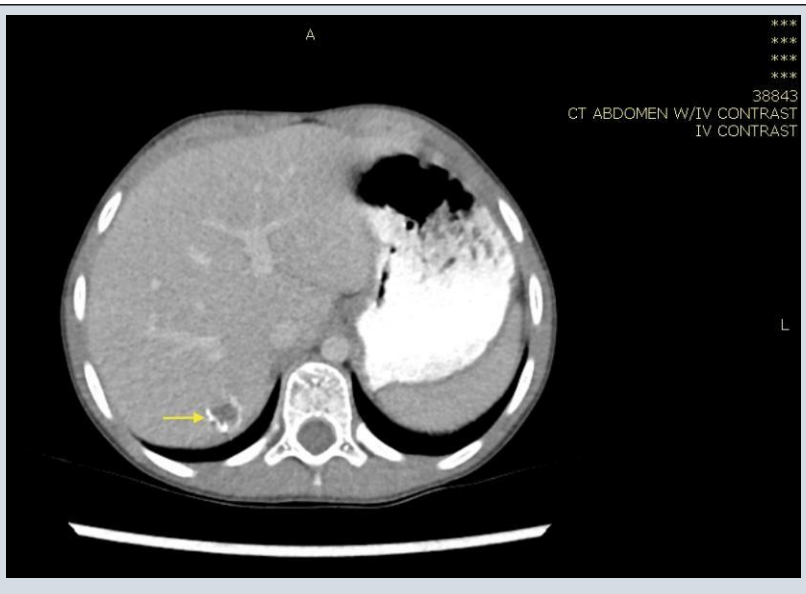


Figure 1B: Basidioblomycosis of the liver, The routinely stained histological sections show liver and fibroconnective tissues with non-necrotizing eosinophil-rich granulomatous inflammation. The constituent cells include mixed granulocytes (lymphocytes, histiocytes and giant cells), and epithelioid cells with giant cells (granulomatous inflammation). Few fungal elements with thin walls (Periodic Acid Schiff stains. A-B-C: Arrows) intensifies the appearance of the fungal walls. Splendore–Hoeppli phenomenon (D:yellow star) is evident by the presence of brightly eosinophilic materials surrounding the hyphae in starburst pattern. Magnifications: A: x200, B:x400, C: x200, D: x400.

Patient was started on iv voriconazole for two weeks the patient showed significant clinical improvement fever and abdominal pain subsided with normalization of laboratory parameters then shifted to full therapeutic dose of oral itraconazole for one year with regular follow up in our clinic, follow up CT scan of the abdomen with iv contrast (Figure1C).



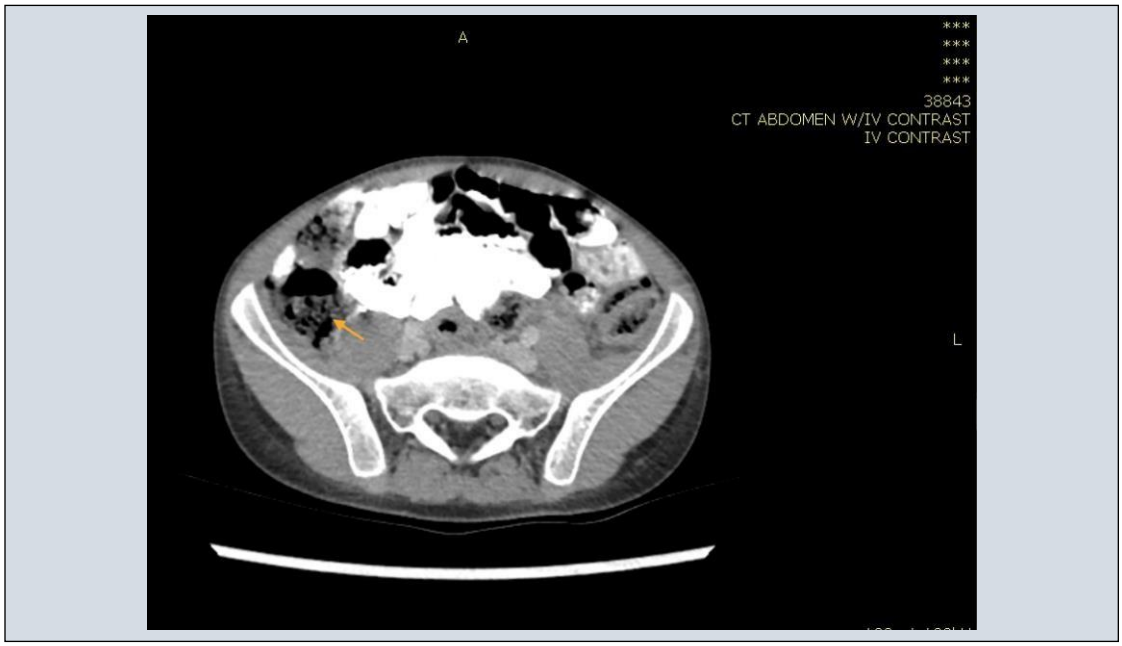
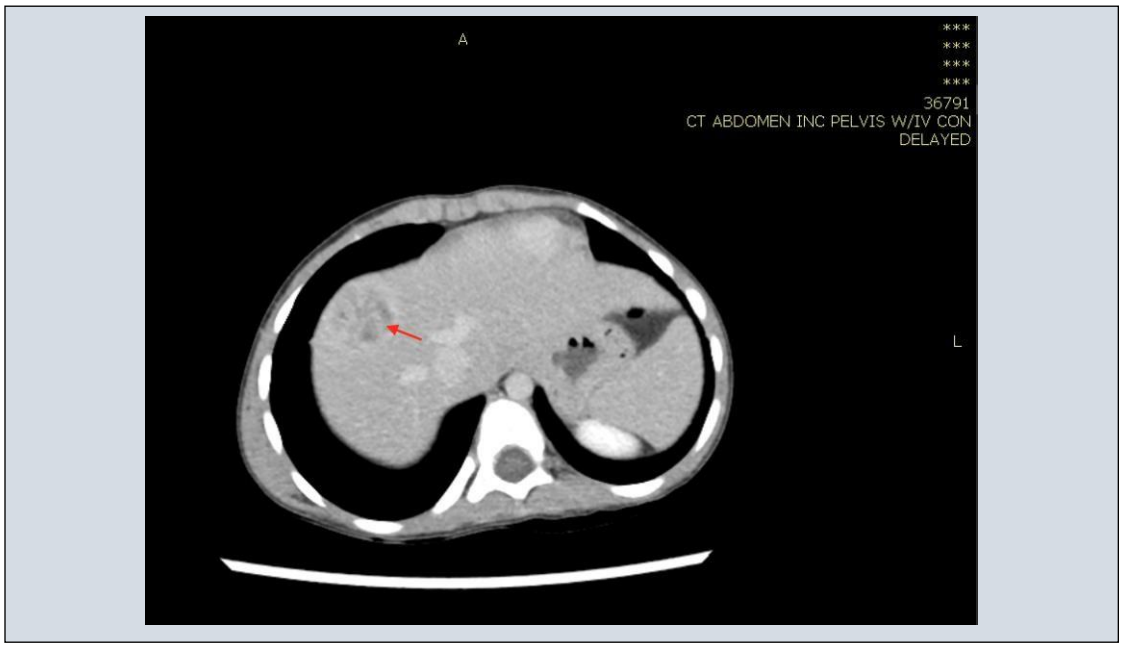


Figure 1C: CT scan of the abdomen with iv contrast revealed no evident bowel wall thickening (orange arrow). The liver is average in size, showing well defined marginally calcified lesion about 1.8 cm in the posterior segment of the right lobe, consistent with calcified granuloma, impressive of healing of the previously noted hepatic abscesses (yellow arrow). No further hepatic lesions. Patient was put on regular follow up both radiological and clinical every 3 months with no relapsing feature

Case II:

A5-years old Saudi male from Jizan (southern region of Saudi Arabia) previously healthy brought by his parent's complaint of fever for one month spike on daily basis, generalized abdominal pain, vomiting, decrease appetite weight loss 4 kg in one month, medical and surgical history unremarkable. On examination weight 13.8 kg at third centile. Abdominal examination revealed distended abdomen, right hypochondrial tenderness, hepatomegaly 4 cm below costal margin, no splenomegaly or lymphadenopathy with unremarkable other systemic examination. Haematological investigations show microcytic hypochromic anaemia (HB 9.5 g/dl), leukocytosis (WBC 31.32 10^g/l), eosinophilia (12.3%) with absolute eosinophilic count (3.85 10^g/l) platelets count (819^g/l). CRP 335.4 mg/l, erythrocyte sedimentation rate in the first hour 115mm/hr. The patient was admitted to the paediatric word as case of pyrexia of unknown origin for further workup, done for him CT scan of the abdomen with iv contrast (Figure 2A).



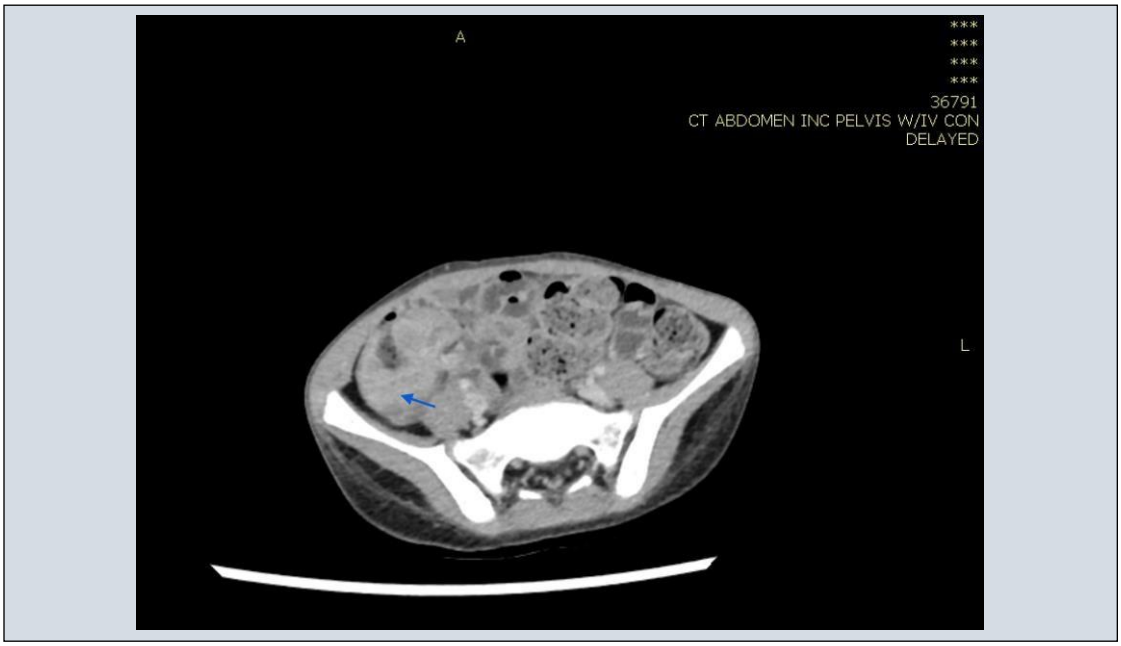


Figure 2A: CT scan of the abdomen with iv contrast showed the liver is enlarged, showing large ill defined mass lesion of the medial segment of the left hepatic lobe, the lesion is about 6 cm this lesion is mildly hypodense to isodose (red arraw), there is another similar lesion at the right lobe about 3.6 cm, there are associated pathologic significantly enlarged lymph nodes at the porta hepatis and peripancreatic head region about 2.3 cm in short diameter. The caecum and proximal ascending colon are presenting segment of circumferential moderate wall thickening about 5 cm with significant submucosal oedema and lumen narrowing (blue arrow), multiple significantly enlarged mesenteric lymph node largest about 1.5cm in short diameter and 2.3cm in long diameter, conclusion large two focal lesions of the right and left hepatic lobes, associated circumferential thickening and oedema of the caecum and proximal ascending colon, pathologic enlarged necrotic lymph node at the porta hepatis and bowel mesentery. The described finding are suggestive of infection with special attention for fungus infection, especially Gastrointestinal Basidiobolomycosis.

US guided true cut biopsy was done from the right hepatic lobe solid lesion was taken, section showed liver parenchyma with eosinophilic-rich mixed inflammatory, necrosis, few fungal elements (hyphae) with variable thickness (Periodic Acid-Schiff/D stain) surrounded by histiocytic reaction with giant cell formation, stains for fungal elements were positive (Periodic Acid-Schiff stain/ D stain). The result was diagnostic for (GIB) Gastrointestinal Basidiobolomycosis (Figure 2B).

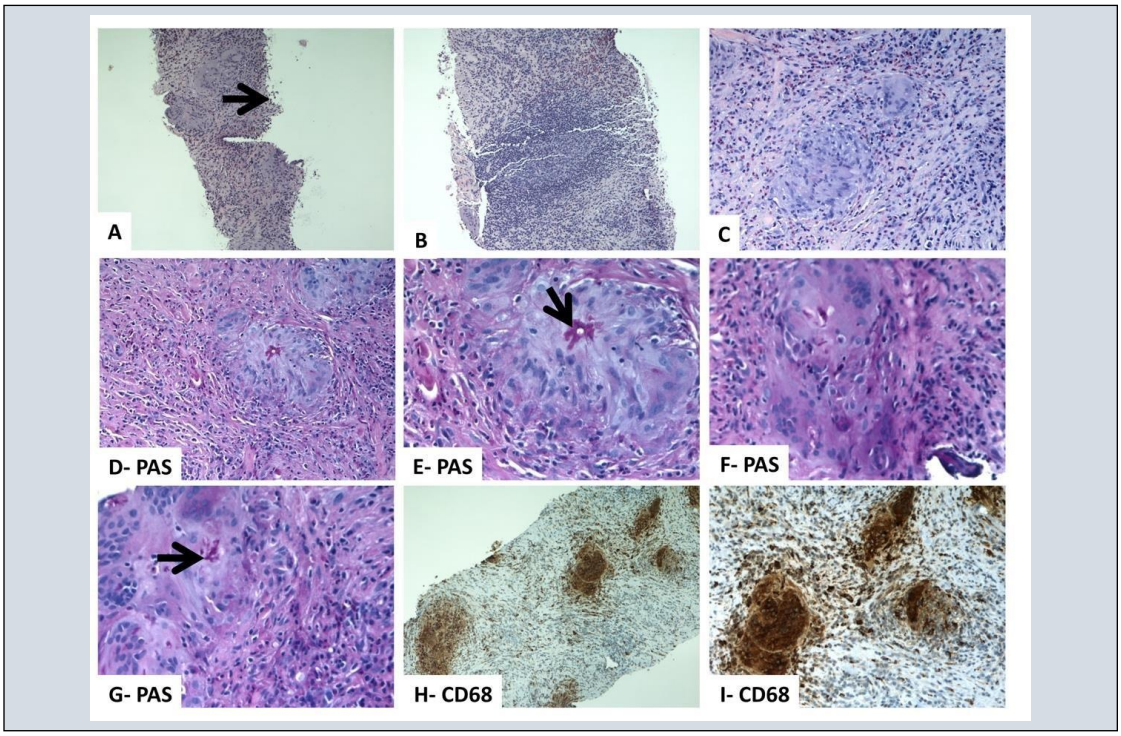


Figure 2B: Basidioblomycosis of the retroperitoneal tissues.

The Hematoxylin and Eosin-stained histological sections reveal cores of densely inflamed fibroconnective tissues. There is lymphohistiocytic and eosinophil-rich mixed inflammatory cell infiltrate with formation of epithelioid cell granulomas with multinucleated giant cells. Rare broad, thin-walled hyphae walls (Periodic Acid Schiff stains, D-E-F-G) intensify the appearance of the fungal walls. Splendore–Hoeppli phenomenon (Arrows: D-E) is indicated by the presence of deeply eosinophilic material, typically surrounding the hyphae. Scan fragments of the hyphae are seen inside the giant cells. The polymorphic nature of the inflammatory cells is indicated by the presence of admixture of CD20 (B lymphocytes), CD3 (T lymphocytes), and CD68 (histiocytes) positive inflammatory cells. Molecular testing for Mycobacterial DNA (PCR amplification of the ITS region of the 16sRNA gene cluster and the IS6110 sequence of the mycobacterium tuberculosis complex). Magnifications: A: x40, B:x100, C: x200, D: x200, E:x400, F:x200, G:x400, H:x100 and I:x200.

Patient started initially on iv voriconazole for three weeks then discharged on full therapeutic course of oral itraconazole was continued for 11 months, the patient improved clinically become asymptomatic, all labs normalized, had regular follow up as outpatient clinic, follow up CT abdomen with iv contrast showed complete resolution of the previous lesions (Figure 2C). after medication was stopped the patient had regular follow up in our clinic clinically and radiologically every 6th month with no evidence of relapse.

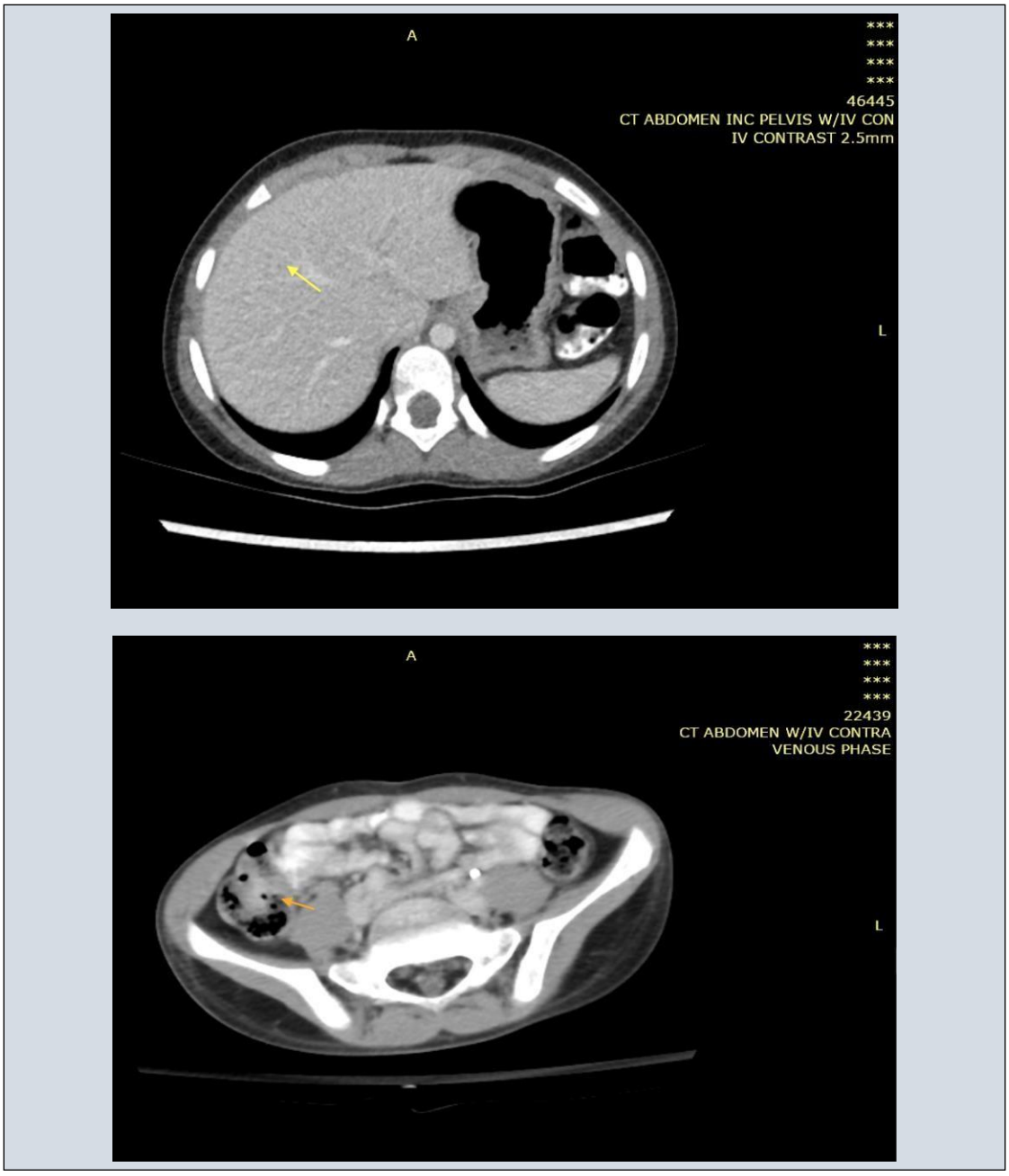
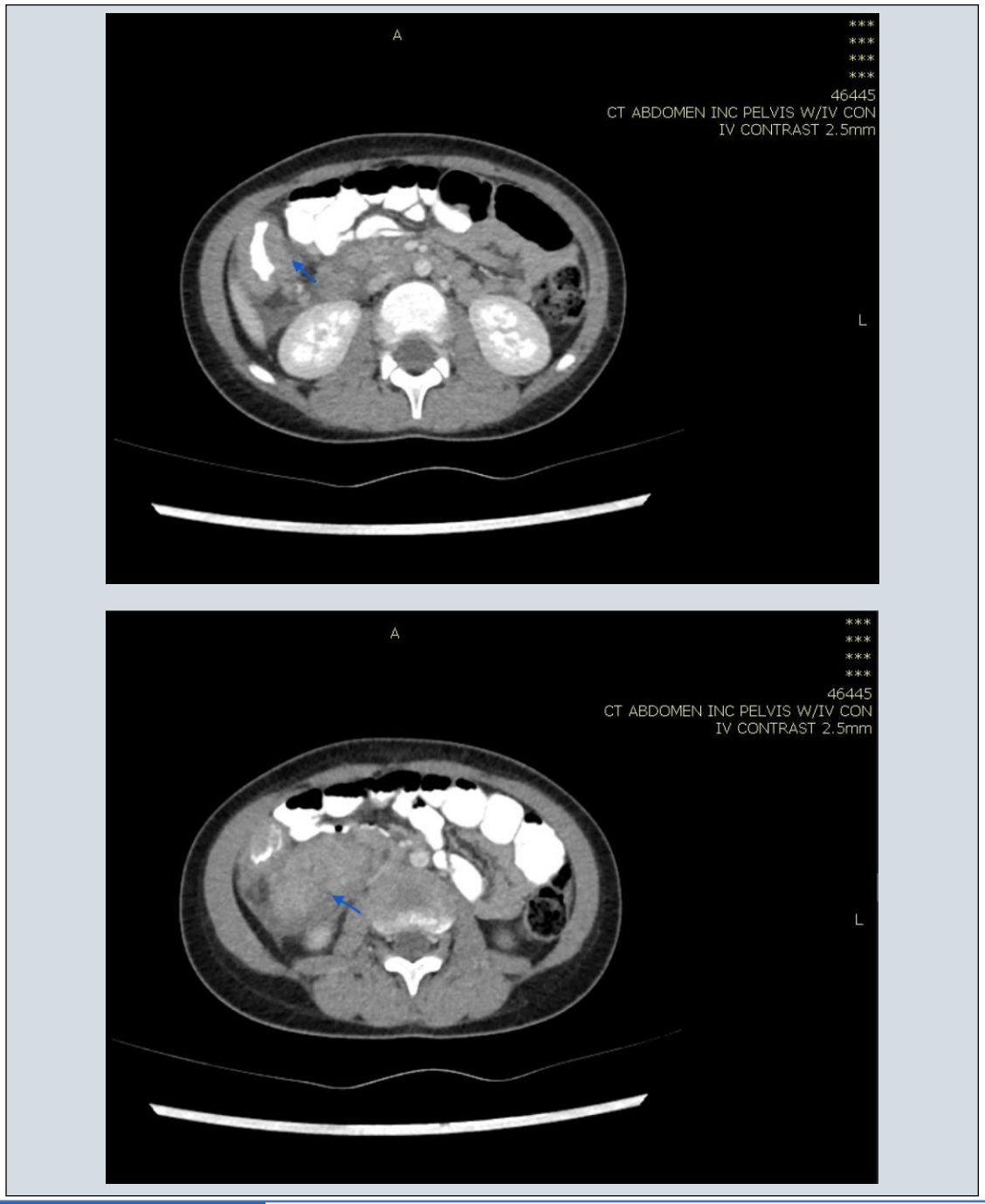


Figure 2C : CT abdomen with iv contrast showed complete resolution of the previously seen two hepatic lesions (yellow arrow) and lymphadenopathy with almost complete resolution of the previously described wall thickening in the caecum and ascending colon region (orange arrow)

Case III:

7 years old female from Jizan presented to our hospital complaint of abdominal pain for one month, associated vomiting and constipation, decrease appetite fever for two weeks. Examination of the abdomen revealed localized tenderness positive rebound tenderness in the right iliac fossa, with un remarkable other systemic examinations. US abdomen done for her showed large soft tissue mass lesion is seen at right iliac fossa measuring about 58X32 mm, suggesting enlarged suspected mesenteric lymph nodes associated with surrounding smaller lymph nodes, as well as para aortic region, patient was admitted under pediatric surgery for further investigations , initial lab works showed microcytic hypochromic anemia (HB 11,9 g/dl) thrombocytosis platelets 580 10^g/l) leukocyte (WBC 10.97 10^g/l) eosinophilia 18.5% with absolute eosinophilic counts (2.02 10^g/l), CRP 24 mg/l ,ESR in the first hour 51 mm/hr. CT abdomen suggestive either lymphoma or inflammatory bowel disease(involving the large and small bowel) (Figure 3A).



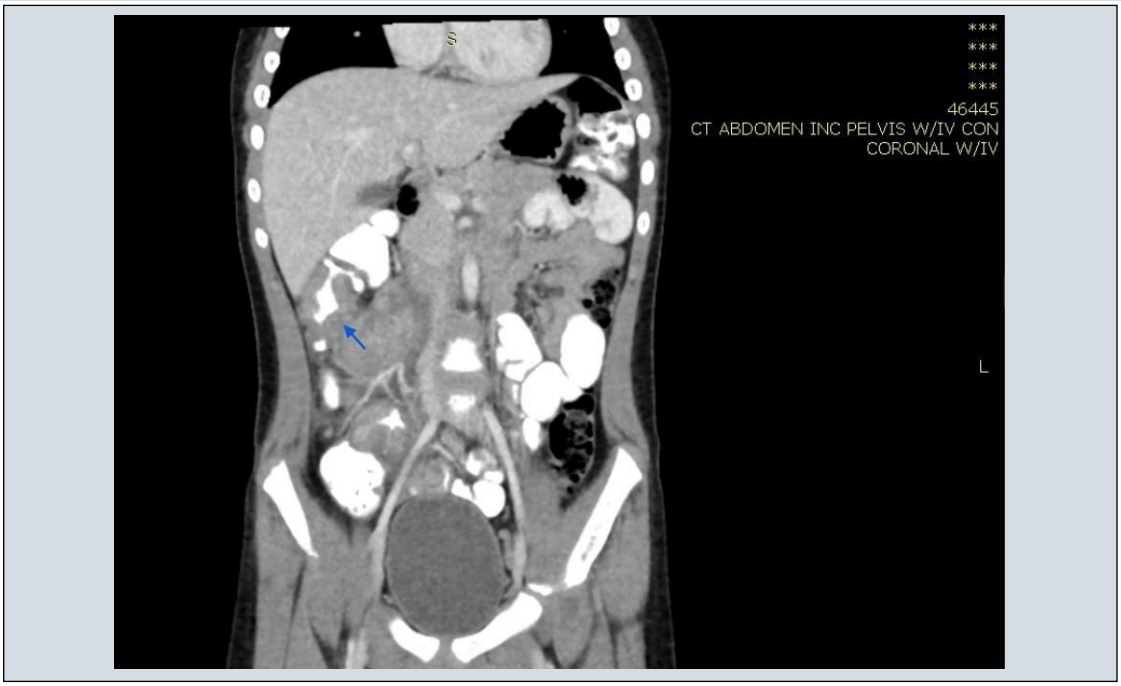
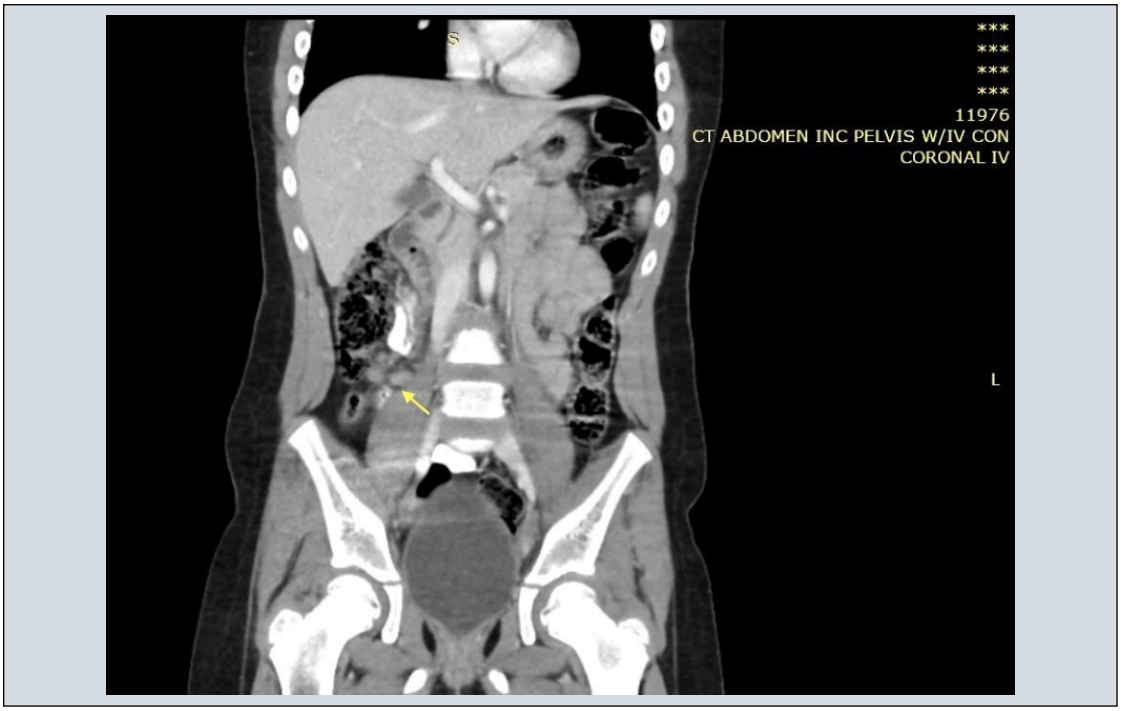


Figure 3A: CT abdomen showed focal area of significant wall thickening of the ascending colon is seen and terminal ileum with luminal narrowing (blue arrow), accompanied by multiple variable sized lesions seen in the mesenteric fat of the right side of the abdomen suggestive of enlarged and pathological lymph nodes (the largest measure about 40 X 23 mm) finding suggestive either lymphoma or inflammatory bowel disease(involving the large and small bowel).

US guided true cut biopsy was done from the retroperitoneal mass lesion. Histopathology result as follow (there is eosinophilic -rich lympho histiocytic, plasmacytic infiltrate and occasional lymphoid follicle, epithelial cell granulomas, some giant cell lomas with PAS/PASD material, suggestive of fungal hyphae in side them confirming the diagnosis of (GIB) Gastrointestinal Basidiobolomycosis. We started iv voriconazole for two weeks patient improved clinically become asymptomatic, all laboratory parameters came back to normal, shifted to therapeutic dose of oral itraconazole, For 10 months, with regular follow up in infectious diseases clinic, repeated CT scan of the abdomen with iv contrast compared with previous study near total disappearance of the previously noted colonic and ileal masses (Figure 3B). all labs investigations was normal medication was stopped, patient given follow up every 3 months in infectious disease clinic



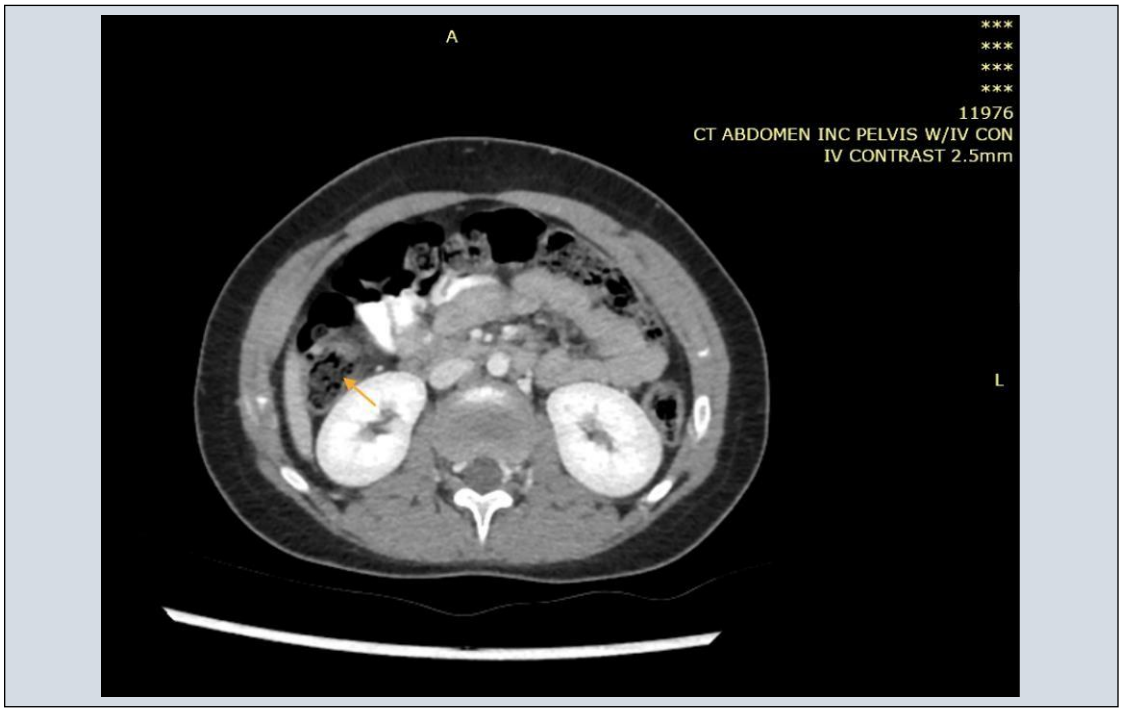


Figure 3B: Repeated CT scan of the abdomen with iv contrast compared with previous study near total disappearance of the previously noted colonic and ileal masses (orange and yellow arrows), now mild residual thickening is seen in the secum and terminal ileum, few residual lymph nodes are noted.

Discussion:

Entomophthoromycosis is caused by B. ranarum which represents saprophytic fungus, Subcutaneous zygomycotic is the commonest clinical form of basidiobolomycosis [3], is a rare fungal infection caused by B. ranarum, an environmental saprophyte [4], it can cause significant disease and occasionally fatal, large number of cases (18cases) from Saudi Arabia multicentric study by shreef et al, in 2018[5]. Basidiobolus Ranarum grow in warm and humid climate which was the climate of the southern region of Saudi Arabia for that most of cases from this area, also environmental contamination plays a role. Mode of transmission remained un known, the portal of entry is believed to be the skin after loss of the integrity by scratch or insect bite [3], also the root of infection could be via contaminated food from soil which may lead to involvement of the gut [5].

We have here three cases with Basidiobolomycosis from same area Jizan (southern region of Saudi Arabia), clear point toward mode of infection in the history for this un usual organism, non-specific presentation, the symptoms and sign may be lead to other diagnosis which resembling Gastrointestinal Basidiobolomycosis, this entities includes intestinal lymphoma, inflammatory bowel disease, abdominal tuberculosis. Sarcoidosis, for that may be misdiagnosed or delayed the diagnosis which may lead to morbidity.

To reach the diagnosis of Basidiobolomycosis needs alert physician aware about the endemic area in Saudi Arabia and high index of suspicion in Such patient presented with prolonged fever, long duration of abdominal pain with changing in bowel habits. Abdominal mass, weight loss, with other features of chronic infection or cancer with anemia thrombocytosis, leukocytosis, high inflammatory markers ESR, CRP, we should put in our mind the Gastrointestinal Basidiobolomycosis as a part of differential diagnosis of these patients.

We are reporting three cases of Gastrointestinal Basidiobolomycosis from the same area Jizan, in our three cases we observed had same presentations inform of abdominal pain, prolong fever, weight loss, abdominal mass, change in bowel habits, all had no clear point how they get infections, laboratory finding microcytic hypochromic anemia, leukocytosis, with obvious eosinophilia, high inflammatory marker, the above mentioned data may help physician to become more alert to reach diagnosis as early as possible to avoid complications that may necessitate surgical intervention, also to keep doctors oriented about this rare condition, it is common in young children as disease of skin or subcutaneous tissue which support the transmission via loss of integrity of the skin[3]. Culture of the organism is the accurate method for diagnosis, also diagnosis can be reached depending on typical finding of histopathological features of Basidiobolus ranarum (granulomatous inflammation rich in eosinophils and the Splendore-Hoeppli phenomenon and broad-based hyphae on Gomori Methenamine Silver) [3].Splendor-Hoeppli phenomenon (also known as a steroid bodies) was first described by Splendore and in bilhazziasis by Hoeppli [3], it was described in mucocutaneous tissue and internal organs, Basidiobolomycosis diagnosis can be reached by immune diffusion test by detecting immune response against agent [6]. In our series the diagnosis of Gastrointestinal Basidiobolomycosis was based on clinical presentation, radiological finding accompanied by unique and characteristic histopathological findings for B. ranarum in human tissue, the three of them were immunocompetent.

Appropriate treatment has not been yet outlined; some centers preferred combination of surgical intervention for removal of pathology and confirming of the diagnosis to prevent recurrence with minimization the duration of antifungal and it is side effect [7], this approach difficult to be applied in our patients they had multiple sites involvement (colon and liver abscess in case I, in case II involving caecum, ascending colon with involvement of right and left hepatic lobes, while in case III involving ascending colon and terminal ileum with multiple lymph nodes involved in all cases) , because of this multiple sites with involvement of lymph nodes we preferred medical therapy initially than to start by surgery because it carries high risk of complications. To confirm their diagnosis, we used US guided true cut biopsy for histopathology which is one of method of diagnosis.

All of them was started on iv voriconazole as a broad-spectrum antifungal agent for 2 weeks in case I and case III, and 3 weeks for case II, as it is potent to reduce fungemia all of them showed response to treatment inform of subside of fever, abdominal pain, appetite improved and general condition as well, inflammatory markers came down, eosinophilia resolved, the three of them changed to therapeutic regiment of itraconazole and discharged on good conditions with regular follow up as outpatient clinic with monitoring them clinically and radiologically, for case I treated for one year, repeated CT abdomen with iv contrast (Fig 1c) resolved lesions medication was stopped, case II treated for 11 months after repeated CT scan showed resolved lesions (Fig2c), case III treated for 10 months, repeated CT scan of the abdomen with IV contrast fig 3c , compared with previous CT scan of the abdomen showed near total disappearance of the previously noted colonic and ileal masses, few residual lymph nodes. As we mentioned before alert physician toward clinical presentation and endemic area lead to early diagnosis carrying better outcome when we started treatment as early as possible.

Conclusion:

Gastrointestinal Basidiobolomycosis can be easily misdiagnosed as malignancy, inflammatory bowel disease, abdominal TB, we reported this case series for all pediatrician to keep in their mind as a part of differential diagnosis in any patient with prolonged fever, abdominal pain, weight loss, with eosinophilia, high inflammatory marker, endemic area, because early diagnosis and starting of management had excellent outcome, we recommended medical therapy, even if multiple lesion and avoid surgical intervention, for diagnosis histopathology had great value, CT scan can be used for diagnosis and follow up to stop medication.

References:

1. Carr EJ, Scott P, Gradon JD. Fatal gastrointestinal mucormycosis that invaded the postoperative abdominal wall wound in an immunocompetent host. Clin infect Dis 1999; 29:956-7.
2. Vikram HR, Smilack JD , Leighton JA, Crowell MD , De Petris G. Emergence of gastrointestinal Basidiobolomycosis in the United States , with a review of world wide cases. Clin infect Dis 2012;541685-91.
3. Hussein MR, Musalam AO, Assiry MH, Eid RA, El Motawa AM, Gamel AM. Histological and ultra structural features of gastro intestinal basidiobolomycosis. Mycol Res 2007;111:926-30.
4. Bennetl JE. Diagnosis and therapy of systemic mycosis in the immunosuppressed host. Transplant proc 1973;5:1255-7.
5. Shreef K, Saleem M, Saeedd MA, Eissa M, Gastro intestinal basidiobolomycosis (GIB) : an emerging confusing disease in children. (multiple.centers experience). European Journal Surgery. Eur J pediatr Surg 2018;2:194-9.
6. Nemenqani D, Yagoob N, Khoja H, Al Saif O, Amra NK, Amr SS. Gastrointestinal basidiobolomycosis: An unusual fungal infection mimicking colon cancer. Arch pathal Lab Med. 2009;133:1938-42.
7. Al-Shanafey S, Alrobean F, Bin Hussain I. Surgical management of gastrointestinal basidiobolomycosis in pediatric patients. J pediatr Surg 2012; 47:949-5.