

BASIDIOLOMYCOSIS OF COLON: CASE REPORT

DOI: [10.33762/bsurg.2023.137789.1038](https://doi.org/10.33762/bsurg.2023.137789.1038)

Bahzad Hamad

Lecturer and General Surgery Specialist, College of Nursing, University of Raparin, Rania, Sulaimany, Iraq

Email: bahzad.hamad@uor.edu.krd

Receive Date: 14 January 2023

Revise Date: 27 January 2023

Accept Date: 18 February 2023

First Publish Date: 18 February 2023

Abstract :

Basidiobolomycosis of the colon is a very rare fungal infection disease. We report here primary colonic Basidiobolomycosis as the first female case in Iraq. A 48 years old Insulin dependent diabetic patient presented with abdominal pain , right side abdominal mass and weight loss for few months duration. CT scan revealed caecal wall thickening without intestinal obstruction. Colonoscopy revealed large fungative ulcerated mass in caecum but the mucosal biopsy was negative for malignancy and in favour of chronic inflammation .Laparotomy with right hemicolectomy and ilio-transverse anastomosis done. Histopathological examination of the excised specimen prove Basidiobolomycosis of colon for which she received 6 months course of oral Itraconazole . She recovered well from the disease with no sequela or recurrence .

Key Words: Basidiobolomycosis, Gastrointestinal mycoses, Iraq.

Introduction:

Gastrointestinal Basidiobolomycosis (GIB) is extremely rare fungal infection with only 122 cases had been reported worldwide till 2019, as the first reported case was reported in Nigeria in 1964^{1, 2}. Most cases of Basidiobolomycosis have been reported from tropical and subtropical regions of Africa, South America, and Asia³.

GIB caused by the fungus *Basidiobolus ranarum* (*B. ranarum*), belonging to the family Basidiobolaceae, order of Entomophthorales, class Zygomycetes⁴.

In current paper, to our best knowledge, GIB is being reported for the first time in female gender in Iraq although, It had been reported previously in 6 males⁵.

B. ranarum was first described in 1886 and isolated from frogs. It has been found in decaying vegetation, foodstuffs, fruits, and soil⁶. It's occasionally found as commensal

in the gastrointestinal tract of amphibians, reptiles, fish, dogs, frogs and bats⁷.

Basidiobolomycosis can result in subcutaneous infections causing subcutaneous zygomycosis, however, the disease can involve visceral organs such as the gut causing gastrointestinal Basidiobolomycosis⁸. The most likely route of infection in GIB is through ingestion of soil, animal feces, and food contaminated by either, as well as rectal inoculation³. This route of infection explains the higher incidence in children age group specially males when they play outdoor and lying on the grounds and playing with decaying plants^{2, 9}. Uncontrolled diabetes, prolong neutropenia, steroids use, hematological malignancies and transplant recipients are risk factors for the development of the disease¹⁰.

GIB usually has non specific clinical manifestations and its diagnosis is challenging . The disease commonly manifests with fever, nausea, vomiting, abdominal pain, diarrhoea and/or an abdominal mass ¹¹. In most reported cases the initial diagnosis was malignant neoplasm, tuberculosis or inflammatory bowel disease ¹¹, but definitive diagnosis proved after laparotomy and excision of the affected organ.

Informed consent had been taken by the author from the patient for reporting this case and publishing it in future.

Case presentation:

In December 2021 , 48 years old Insulin dependent diabetic lady presented to the

clinic with abdominal pain and right sided lower abdominal mass for 3 months duration. She had also anorexia ,weight loss but no fever and normal bowel motion . On physical examination a non tender mass palpated in right lower abdomen . Ultrasound examination revealed right side hypoechoic mass 73*66*89 mm , pictures of bowel mass. CT of the abdomen and pelvis with oral and intravenous contrast revealed thickening of the wall of ileum as well as of caecum with multiple enlarged (10mm short axis diameter) mesenteric and para-aortic lymph nodes but no CT finding of intestinal obstruction found Figure 1.

Figure 1: Radiological features of colonic Basidiobolomycosis .Abdominal CT scan with oral and intravenous contrast revealed thickening of the wall of the caecum (white arrow) and ileum.



Colonoscopic examination revealed large fungate ulcerated mass in the caecum . Multiple mucosal biopsies taken for histopathological examination which revealed only colonic mucosa with mild increase in chronic inflammatory cells ,mainly lymphocytes and plasma cells ,and necrotic fragments of tissue . Complete blood count revealed mild anaemia (HB 10gm/dl) and leucocytosis (12.3×10^9 cells/L) but no Eosinophilia. Preoperative diagnosis was presumed to be malignant lesion of caecum and decision for surgical intervention was taken.

Mechanical and antibacterial bowel preparation of the colon done the night before operation.

Midline laparotomy done , huge caecal mass extending to ascending colon found, the mass was adherent to right ovary and sigmoid colon and all these were wrapped with the greater omentum but the terminal ileum was normal. Regional lymph nodes were enlarged. My first impression on the intraoperative findings was in favour of neoplastic lesion that is why decision of En-bloc resection of terminal ileum , appendix caecum and ascending colon done with ileo-transverse end to end anastomosis , and segmental (10cm) excision of sigmoid colon with end to end anastomosis . A summary of resected parts is shown in figure 2.

Figure 2: Excised specimen showed a mass of the caecum, ascending colon , terminal ileum and segment of sigmoid colon with omentum.



Postoperative period was uneventful and the patient discharged on fifth postoperative day.

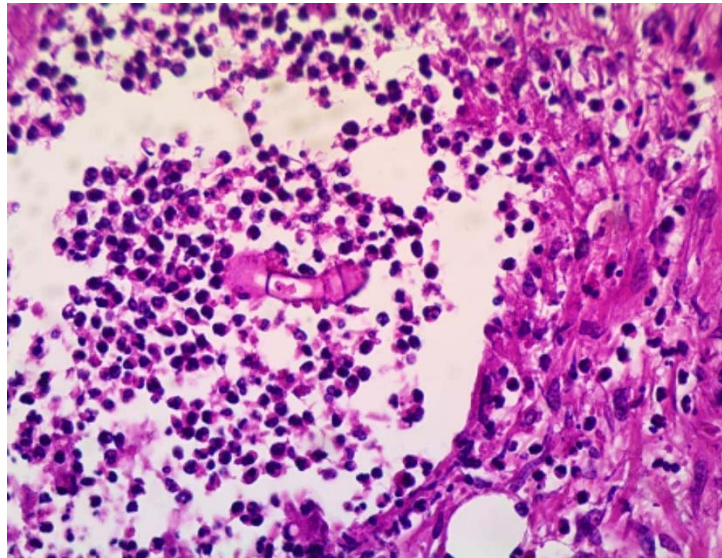
Histopathological examination of the sections showed involvement of caecum,

sigmoid colon ,and omental fat by necrotizing granulomatous inflammation associated with extensive dense mixed inflammation composed of mixed inflammatory cells including large number of eosinophils with abundant broad hyphae with septation with zygo spores ,the inflammatory process involving the submucosa up to the serosal fat. All excised lymph nodes show

reactive hyperplasia. The picture was consistent with Basidiobolomycosis. A summary of the histological findings is shown in Figure 3.

She received postoperatively Itraconazole tablet 200mg twice daily for 6 months with good response and no evidence of disease recurrence till now.

Figure 3: Histopathologic examination of the colon revealed a broad ,thin walled ,septated Hyphae of *Basidiobolus ranarum* (black arrow), surrounded intensely by a cuff of eosinophilic (Splendore-Hoepli phenomenon), and numerous eosinophils (white arrow) and other mixed inflammatory cells.



Discussion :

This is the first reported adult female case of gastrointestinal Basidiobolomycosis in Iraq and thirteenth in the world ¹. GIB is extremely rare disease caused by the fungus *Basidiobolus ranarum* .The fungi of *B. ranarum* are commonly found in the soil and therefore gardeners, farmers, and landscapers are more exposed to GIB ¹². GIB is acquired in most cases by ingestion of contaminated water, or food including fruits, and

vegetables contaminated by animal fecal matters¹². Recent taxonomic classifications dictate *B.ranarum* is pathogenic fungus causing GIB for 122 reported cases worldwide to 2018 ⁹. There is only 12 cases had been reported world wide in adult females till 2018¹.

Most of the cases has been reported in Saudi Arabia 62 cases , next Iran 24 cases , then USA 22cases ,Iraq 6 cases, Kuwait 2 cases, France 2 cases and lastly sporadic cases has

been reported in Brazil, Netherland, Qatar, Oman and Thailand¹³. The six cases which has been reported in Iraq , all of them were male , four adult and two children, and all of them were in Sulaimanyah /north of Iraq in 2012⁹. The infection is more common in hot ,arid climate , that is why we expect much cases are present in south of Iraq but has not been reported yet. The occupation of our case, is house wife , living in a rural area raising cows and dealing with dried grass daily as food for the livestock so the source of infection in our case most probably was from the plants and grass .

In a review of 102 cases, the commonest clinical manifestations of the GIB was abdominal pain in 86.6%, fever in 40.2% , weight loss 33.3% , and abdominal mass in 30.4%¹. Current patient has all these symptoms except fever . The disease rarely diagnosed preoperatively because of the similarity of the condition with the neoplastic lesions and other chronic inflammatory lesions.

GIB most commonly affects colon and rectum , then small bowel and gall bladder and liver, but stomach has least chance of infection with *B.ranarum* probably due to low PH of the stomach, that's why those patients received Ranitidine had been infected more¹⁴.

The risk for Basidiobolomycosis may be higher in individuals with uncontrolled diabetes mellitus and probably this the risk factor in our patient, as she was in uncontrolled state .Regarding laboratory test results , she has no Eosinophilia although 85% of GIB has eosinophilia.

Diagnosis of colonic Basidiobolomycosis can be confirmed through multiple colonoscopic mucosal biopsy¹⁴, although the biopsy in current case was not diagnostic due to inadequacy of the sample or the presence of the fungal hyphae in the submucosal or muscular layer .

In previous reported cases ,only few cases had been diagnosed preoperatively after colonoscopy biopsy , because the hyphae usually is not living in the mucosa , but its detected in the submucosa and muscular layer that is why most colonoscopic biopsy reveal only necrotic tissue and negative for the microorganism⁶ .

The confirmatory diagnosis is done by culturing the sample, but this is excluded due to the use of formalin when taking the surgical sample, so the histological examination was relied upon. Typical morphologic features in the histopathological sample include hyphae that are irregularly branched, thin walled, occasionally septated, and surrounded by a thick eosinophilic cuff (Splendore-Hoeppli phenomenon); the presence of a few associated spore like spherules, although not entirely specific, are characteristic histologic features³. The gold standard for definite diagnosis of GIB is microbial culture of *B. ranarum* from fresh aspiration or surgical specimens and requires a high level of clinical suspicion by the clinician and the mycologist¹³.

Treatment require both surgical intervention and prolong antifungal medical treatment. It has been reported in few cases that only medical treatment started on the result of the colonoscopy biopsy with good outcome and full recovery⁸. Its diagnosis is challenging due to non specific symptoms and rarity of the disease , that is why it need high degree of suspicion and multidisciplinary team of gastroenterologist, general surgeon, radiologist and pathologist .

Conclusions:

GIB clinically is mimicking malignancy or inflammatory bowel disease, with non specific radiological findings may mimics cancer.

1. Adequate tissue for histological examination is necessary to identify *B.ranarum* and definitive diagnosis need culture of the organism .

2. Colonoscopy biopsy usually give false negative result because the fungus living in the submucosa and muscular layer .
3. Rarity of the GIB with its variable symptoms and resemblances to other more common clinical conditions like cancer and inflammatory bowel disease , make early diagnosis difficult.
4. Surgical removal of the affected part in GIB along with treatment with an antifungal for several months is necessary to cure the disease.

References :

1. Pezzani MD, Di Cristo V, Parravicini C, Sonzogni A, Tonello C, Franzetti M, et al. Gastrointestinal basidiobolomycosis: An emerging mycosis difficult to diagnose but curable. Case report and review of the literature. *Travel Med Infect Dis*. 2019;31:101378. doi: [10.1016/j.tmaid.2019.01.013](https://doi.org/10.1016/j.tmaid.2019.01.013)
2. Balkhair A, Al Wahaibi A, Al-Qadhi H, Al-Harthy A, Lakhtakia R, Rasool W, et al. Gastrointestinal basidiobolomycosis: Beware of the great masquerade a case report. *IDCases*. 2019;18:e00614. doi: [10.1016/j.idcr.2019.e00614](https://doi.org/10.1016/j.idcr.2019.e00614)
3. Nemenqani D, Yaqoob N, Khoja H, Al Saif O, Amra NK, Amr SS. Gastrointestinal basidiobolomycosis: an unusual fungal infection mimicking colon cancer. *Arch Pathol Lab Med*. 2009;133(12):1938-42. doi: [10.5858/133.12.1938](https://doi.org/10.5858/133.12.1938)
4. Vikram HR, Smilack JD, Leighton JA, Crowell MD, De Petris G. Emergence of gastrointestinal basidiobolomycosis in the United States, with a review of worldwide cases. *Clin Infect Dis*. 2012;54(12):1685-91. doi: [10.1093/cid/cis250](https://doi.org/10.1093/cid/cis250)
5. Hemmin A Hassan RAM, Nawshirwan G Rashid, Bryar E Nuradeen, Qalandar H Abdulkarim, Tahir A Hawramy, Rekawt M Rashid, Alton B Farris, Jeannette Guarner and Michael D Hughson. Eosinophilic granulomatous gastrointestinal and hepatic abscesses attributable to basidiobolomycosis and fasciolias: a simultaneous emergence in Iraqi Kurdistan. *BMC Infectious Diseases*. 2013;13(91)
6. Abd El Maksoud WM, Bawahab MA, Ashraf TH, Al Shehri DM, Mirza NI. Surgical management of colonic basidiobolomycosis among adolescent and adult patients: presentation and outcome. *Colorectal Dis*. 2018;20(4):296-303. doi: [10.1111/codi.13939](https://doi.org/10.1111/codi.13939)
7. Bitar Geramizadeh H, M, ; Golsa Shekarkhar2, . Gastrointestinal Basidiobolomycosis, a Rare and Under-diagnosed Fungal Infection in Immunocompetent Hosts: A Review Article. *IJMS*. 2015;40(20). doi: [10.30476/ijms.2023.48937](https://doi.org/10.30476/ijms.2023.48937)
8. Al Jarie A, Al Azraki T, Al Mohsen I, Al Jumaah S, Almutawa A, Mohd Fahim Y, et al. Basidiobolomycosis: Case series. *J Mycol Med*. 2011;21(1):37-45. doi: [10.1016/j.mycmed.2010.11.002](https://doi.org/10.1016/j.mycmed.2010.11.002)
9. Mohammadi R, Ansari Chaharsoghi M, Khorvash F, Kaleidari B, Sanei MH, Ahangarkani F, et al. An unusual case of gastrointestinal basidiobolomycosis mimicking colon cancer; literature and review. *J Mycol Med*. 2019;29(1):75-9. doi: [10.1016/j.mycmed.2018.11.004](https://doi.org/10.1016/j.mycmed.2018.11.004)
10. Omar Takrouni A, Heitham Schammout M, Al-Otaibi M, Al-Mulla M, Privitera A. Disseminated intestinal basidiobolomycosis with mycotic aneurysm mimicking obstructing colon cancer. *BMJ Case Rep*. 2019;12(1). doi: [10.1136/bcr-2018-225054](https://doi.org/10.1136/bcr-2018-225054)
11. Amal S. Al-Maani GP, 1 Amina Jardani, 2 Madhavan Nayar, 3 Fatma Al-Lawati, 4 Sheikha Al-Baluishi, 5 Ibrahim B. Hussain. Gastrointestinal Basidiobolomycosis First case report from Oman and literature review. *Sultan Qaboos University Med J*. 2014;14(2)

- 12.Hussein MRA, Alqahtani AS, Alhagawi Y, Al Saglan NS, Albishi AM, Ahmed FO, et al. Primary Basidiobolomycosis of the Cecum in Immunocompetent Patients: Two Case Reports. *Gastroenterology Res.* 2021;14(2):116-24. doi: [10.14740/gr1368](https://doi.org/10.14740/gr1368)
- 13.Parisio¹ EM, Mattei¹ R, GC, Maurizio Sanguinetti², MN, Elena De Carolis² ,. Gastrointestinal basidiobolomycosis in a patient suffering from duodenal ulcer with perforation: First case report from Italy. *New Microbiologica.* 2019;42(2)
- 14.Aljohani AE, Alshemesi B, Alshubaisheri A, Alkraidis A, Alzahrani A, Sairafi R. A rare case of colon obstruction due to gastrointestinal basidiobolomycosis in a 36-year-old woman. *Int J Surg Case Rep.* 2020;77:762-5. doi [10.1016/j.ijscr.2020.11.054](https://doi.org/10.1016/j.ijscr.2020.11.054)

Acknowledgement: None

Funding: None

Conflict of interest : Authors declare no conflict of interest

Availability of Data and Material:

The corresponding author is prompt to supply datasets generated during and/or analyzed during the current study on wise request.

This is an open access article under the CC BY 4.0 license: <http://creativecommons.org/licenses/by/4.0/>

Cite this article: Hamad, B. Basidiobolomycosis of colon: Case report". *Basrah Journal of Surgery*, 2023;29 (1): 84-90. doi: [10.33762/bsurg.2023.137789.1038](https://doi.org/10.33762/bsurg.2023.137789.1038)
