Abdominal Basidiobolomycosis a Diagnostic Challenge

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INTRODUCTION

Basidiobolomycosis is a disease ranging from simple subcutaneous infection to deep ones that affect the bowel and may lead to death if not treated properly; sometimes it mimics cancer¹. *Basidiobolusranarum* is a fungus found in dead plants, soil, reptile excrement, and fish that feed on insects. The disease is mainly confined to the tropics (Africa, Asia, and South America)².

THE CASE

A 47-year-old woman (from Qunfudhah governorate, Makkah region, West Saudi Arabia) had presented with abdominal pain and weight loss for 4 months. She neither had diarrhea or constipation nor rectal bleeding. She had no family history of colorectal cancer, but she complained of having vague abdominal discomfort and was being treated for irritable bowel syndrome (IBS). The pain was dull and colicky in nature. A surgeon examined her physically, and she was found to have a fixed mass in the right lower quadrant of her abdomen. Then an ultrasound, a CT scan and colonoscopy were requested.

LABORATORY DATA

Guaiac test (stool occult blood): Positive (+ve)

CBC: WBC result: $1300 \times 10^{9}/L$

Films showed repeatedly marked eosinophilia.

Serology: HIV and other viruses were negative.

Microbiology: Blood culture was negative.

CT scan: It showed a diffuse circumferential wall thickening and partially obstructing the cecum and terminal ileum-suggestive of malignant tumors. The presence of multiple lymph nodes was found, which appeared reactive.

Colonoscopy: The report showed partially obstructed lesion affecting mainly the ascending colon, which was biopsied.

Biopsy: The result showed nonspecific, chronic mucosal inflammation with no evidence of malignant cells or features of inflammatory bowel disease.

The professional diagnosis of inflammatory mass was made.

Unfortunately, the patient did not follow (show) up, and, 3 weeks later, she presented with intestinal obstruction. Then she underwent exploratory laparotomy. The intraoperative finding was a large mass that included a part of terminal ileum, cecum, ascending colon, and the appendix.

SPECIMEN SUBMITTED

Right hemicolectomy was performed on terminal ileum, cecum, ascending colon, part of the transverse colon, and appendix, with mesentery and related lymph nodes.

GROSS DESCRIPTION

The report of right hemicolectomy was received, and it showed the following findings: terminal ilium (3 cm), cecum (6 cm), appendix (6 \times 1.5 cm), ascending colon, and part of transverse colon (18 cm).

(Figure 1) The colonic mucosa was red and edematous, with ulcerations. Shows swollen, thickened (3 cm thick), and partially obstructed colonic wall with firm-to-soft lesion, prominently affecting the ascending colon. Nine lymph nodes (ranging from 3 to 5 cm) were isolated from pericolic fat. Around 46 representing cassettes were taken from the proximal surgical margin, distal margin, and representative sections from the appendix.

MICROSCOPIC EXAMINATION

Multiple sections from the colon showed dense and marked transmural infiltration by eosinophils, neutrophils and Chronic granulomatous inflammation with central abscesses, multinucleated giant cells (foreign body type) (Figure 1), plasma cells, and fibrosis. Fungal hyphae are noted invading the bowel wall, with characteristic Splendore-Hoeppli phenomenon/bodies-demonstrating that the fungus was surrounded by eosinophilic material (Figure 2). The fungus was also surrounded by inflammatory cells, with some fungi engulfed by multinucleated giant cells.

(Figure 3) GMS stains showed fungus with thin wall, pauciseptate hyphae, and irregular outlines.

Sections from appendix, multiple pericolic lymph nodes, and surgical excision margins show the same inflammatory response and reactive follicular lymphoid hyperplasia with germinal centers, with fungal elements. No evidence of malignancy was found.

DIAGNOSIS

Right hemicolectomy specimen: Colonic/intestinal fungal infection, typical for basidiobolomycos.

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Figure 1: Section from the colon showed dense and marked transmural infiltration by eosinophils and granuloma with central abscesses, multinucleated giant cells (foreign body type)



Figure 2: Fungal hyphae invading the bowel wall (arrows head), with characteristic Splendore-Hoeppli phenomenon /bodies (arrows)



Figure 3: GMS stains showed fungus with thin wall, pauciseptate hyphae and irregular outlines

DISCUSSION

The causative agent is a saprophytic B. ranarum that belongs to Zygomycetes family³. Gastrointestinal basidiobolomycosis is considered as a serious disease. To prevent complications and eradicate the disease completely, it must be surgically intervened as soon as possible, and it needs a close follow-up with the treating doctors⁴. Gastrointestinal basidiobolomycosis is a rare disease, and it affects more men, but sometimes it can affect children and women as well. The most important presenting symptom is abdominal pain. The diagnosis of this disease is difficult, because the symptoms are not specific. Some patients may undergo surgery, suspecting it as malignancy⁵. It causes intestinal obstruction, so early diagnosis and prompt treatment are one of the most important ways of preventing complication and death. This disease is rare in people with normal immunity, making early diagnosis difficult and requirement of more research necessary to explore the causes and risks of this disease in people with normal immunity⁶. Any person with normal immunity presenting with a tumor in the abdomen, associated with the presence of eosinophil leukocytes in the peripheral blood, should be alerted to this disease. The presence of necrosis, leukocytes in the tissues, and granules may be a strong indication of the presence of this disease. All attempts must be made to determine if it is caused by a fungus. It is important for the pathologist and the treating doctor to be familiar with this disease7. The leading presenting symptom of basidiobolomycosis in most reported cases was abdominal pain. The other symptoms, in order from most to common least common, are as follows: abdominal mass, constipation, abdominal distension, fever, weight loss, diarrhea, vomiting, and lower gastrointestinal bleeding⁸. Since the most commonly involved parts of gastrointestinal tract are colon and rectum, followed by small bowel, liver, and gall bladder and stomach, around 50% of cases were provisionally diagnosed as intraabdominal malignancy or inflammatory bowel disease, and diverticulitis came second9. The commonest finding for the presence of this disease is peripheral blood eosinophilia. Characteristic histopathology features is a common finding in surgically removed bowel, but may be difficult to obtain in endoscopic biopsies, especially if the sample was taken from a necrotic area¹⁰.

CONCLUSION

Basidiobolomycosis is a rare but serious disease, and hence should not be overlooked by both the surgeon and pathologist. In this area of Saudi Arabia, clinicians should keep this disease in mind during their differential diagnosis, especially when biopsy shows chronic, nonspecific inflammation that is not explained by inflammatory bowel disease.

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REFERENCES

- 1. Humber RA. Entomophthoromycota: A new phylum and reclassification for entomophthoroid fungi. Mycotaxon 2012;120(1):477-92.
- Geramizadeh B, Heidari M, Shekarkhar G. Gastrointestinal basidiobolomycosis, a rare and under-diagnosed fungal infection in immunocompetent hosts: A review article. Iran. J. Med. Sci 2015;40(2):90-97.
- Vikram HR, Smilack JD, Leighton JA, et al. Emergence of Gastrointestinal Basidiobolomycosis in the United States, With a Review of Worldwide Cases. Clin. Infect. Dis 2012;54(12):1685-91.
- 4. Al-Shanafey S. AlRobean F, Hussain IB. Surgical management of gastrointestinal basidiobolomycosis in pediatric patients. J. Pediatr. Surg 2012;47(5):949-51.
- Mohammed SA, Abdelsatir AA, Abdellatif M, et al. Challenging Presentations of Seven Cases of Gastrointestinal Basidiobolomycosis in Sudan: Clinical Features, Histology, Imaging,

and Recommendations. J Lab Physicians 2020;12(4): 281-84.

- Aljohani AE, Alshemesi B, Alshubaisheri A, et al. A rare case of colon obstruction due to gastrointestinal basidiobolomycosis in a 36-year-old woman. Int J Surg Case Rep 2020;77:762-65.
- 7. Alahmadi R, Sayadi H, Badreddine S, et al. Gastrointestinal basidiobolomycosis, the experience of a tertiary care hospital in the western region of Saudi Arabia and a report of four new cases. Life Sci J 2014;11(1):344.
- Alabdan L, Amer SM, Alnabi Z, et al. Gastrointestinal Basidiobolomycosis in a 45-Year- Old Woman. Cureus 2020;12(12):e12109.
- 9. Khan ZU, Prakash B, Kapoor MM, et al. Basidiobolomycosis of the rectum masquerading as Crohn's disease: Case report and review. Clin Infect Dis 1998;26(2): 521-23.
- Kurteva E, Bamford A, Cross K, et al. Colonic Basidiobolomycosis-An Unusual Presentation of Eosinophilic Intestinal Inflammation. Front Pediatr 2020;8:142.