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Abdominal imaging findings in gastrointestinal basidiobolomycosis

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Abstract

Purpose: To describe the abdominal imaging findings of patients with gastrointestinal *Basidiobolus ranarum* infection.

Methods: A literature search was performed to compile the abdominal imaging findings of all reported worldwide cases of gastrointestinal basidiobolomycosis (GIB). In addition, a retrospective review at our institution was performed to identify GIB cases that had imaging findings. A radiologist aware of the diagnosis reviewed the imaging findings in detail. Additional information was obtained from the medical records.

Results: A total of 73 GIB cases have been published in the medical literature. The most common abdominal imaging findings were masses in the colon, the liver, or multiple sites and bowel wall thickening. Initially, many patients were considered to have either a neoplasm or Crohn disease. We identified 7 proven cases of GIB at our institution, of which 4 had imaging studies (4 computed tomography [CT] examinations, 4 abdominal radiographs, and an upper gastrointestinal study). Imaging studies showed abnormalities in all 4 cases. Three-fourths of our study patients had an abdominal mass at CT. Two of 3 masses involved the kidneys and included urinary obstruction. All masses showed an inflammatory component with adjacent soft tissue stranding, with or without abscess formation.

Conclusions: Radiologists should consider GIB when a patient from an arid climate presents with abdominal pain, weight loss, and an inflammatory abdominal mass on CT. Abdominal masses of the colon or liver, bowel wall thickening, and abscesses are the most common imaging findings.

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Abbreviations

CT	Computed tomography
GIB	Gastrointestinal basidiobolomycosis

Basidiobolus ranarum is an environmental saprophytic fungus found worldwide in soil and in the dung of amphibians, fish, bats, and insectivores [1–3], with transmission to humans by insect bite or by local inoculation into open wounds [2, 3]. Basidiobolomycosis is characterized by chronic subcutaneous swelling and induration involving the limbs, trunk, or buttocks and is most commonly seen in tropical areas of South America, Africa, and Asia. Gastrointestinal basidiobolomycosis (GIB) is an uncommon manifestation of this fungal infection, and sporadic cases have been reported from arid regions worldwide. A recent publication described the emergence of GIB in the arid Southwestern United States [3]. In patients with GIB, the fungus is believed to be transmitted through contaminated food or the ingestion of soil or animal feces [2, 3]. In this same study [3], 34% of patients had a chronic underlying illness. For example, 18% of patients had diabetes mellitus. No patients were immunocompromised. A second study, by Geramizadeh et al. [4], also reported 14 cases of GIB in immunocompetent patients. In addition, GIB has been reported in healthy children [1, 5].

Histopathologic findings in tissue infected by *B. ranarum* are distinctive. They consist of necrotizing granulomatous inflammation and severe tissue eosinophilia and of the organism itself, with its floppy hyphae

and rare septae [3]. Fungal hyphae are characteristically surrounded by bright, intensely eosinophilic, granular material (called the *Splendore-Hoeppli phenomenon*) [3].

A comprehensive review of GIB cases by Vikram et al. [3] reported that gastrointestinal imaging most often found a mass in various abdominal organs. The most common clinical symptom was abdominal pain, followed in frequency by abdominal distension, constipation, fever, weight loss, and a palpable abdominal mass [2, 3, 6, 7]. Patients were believed to have an intra-abdominal malignancy, inflammatory bowel disease, diverticulitis, or other diagnosis before GIB was confirmed [2, 3, 6, 7].

Except for a single case report [8], no published studies in the radiology literature detail the imaging findings of GIB. Herein, we summarize abdominal imaging findings of reported GIB cases in the medical literature and describe in greater detail such findings in 4 cases from our institution.

Methods

A comprehensive review of the English-language literature was performed to identify all cases of GIB through search and inclusion criteria, as previously described [3]. Abdominal imaging findings were recorded and categorized (when possible) in accordance with the involved organ, imaging modality, and the findings.

Cases of GIB at our institution were identified through our microbiology and pathology databases, with the inclusion criteria described elsewhere [3]. Details of the abdominal imaging findings were collected. All imaging studies were reassessed by a radiologist (C. D. J.) who is an expert in abdominal imaging. This study was approved by the Mayo Clinic Institutional Review Board. The initial radiologic report was recorded, as well as an unblinded review by the gastrointestinal radiologist (C. D. J.). The combined review of the initial report and the unblinded read was used to generate a final composite report for each case for this study.

Results

Literature review

Vikram et al. [3] conducted a literature review through March 2010 of all cases of GIB worldwide (including cases from our institution) and studied the imaging findings of these cases. In their study, 30 patients with GIB had computed tomography (CT) of the abdomen and pelvis, among which 1 patient had a barium swallow and 1 patient had abdominal ultrasonography. A colorectal mass was the most common finding (21 patients [48%]), followed by a hepatic mass (9 [20%]) or a mass in the small bowel (5 [11%]). Bowel wall thickening, perforation, or abscess was found in 11 patients (25%), 2 patients, and 2 patients, respectively. Masses were infrequently found in the gallbladder or retroperitoneum (e.g., pancreas and kidney). A duodenal-biliary fistula, hydronephrosis, and ascites have also been reported.

From March 2010 through February 2014, a total of 29 additional cases of GIB were reported [1, 4, 5, 7, 9–15]. All patients had CT of the abdomen and pelvis. The most common imaging finding was a mass, seen in 22 of the 29 patients (76%). The locations of the masses were the liver ($n = 3$), the colon ($n = 1$), and 2 or more organs ($n = 9$; 2 with masses in the small bowel and colon, 5 with masses in the colon and liver, and 2 with extensive, nonresectable multiorgan abdominal involvement). The location was not described in 9 cases [9]. For the 7 patients who presented without an abdominal mass, 6 (21% of the 29) had colonic wall thickening and 1 (3%) had an abscess in the colorectum.

Cases from our institution

Seven patients with pathologically proven GIB seen at our institution comprise the cohort of cases reported in the present study. All 7 cases were included in a prior study of GIB from our institution [3]. Three patients were excluded; 1 patient had no imaging study available and 2 patients had CT imaging performed 10 and 15 years after the diagnosis and treatment of GIB. The 4 patients included in the study were men (mean [range] age, 60 [45–74] years).

All 4 patients had CT examinations of the abdomen and pelvis between 1997 and 2009. Three patients had CT examinations with oral and intravenous contrast material; 1 patient had oral contrast material only. Three patients had chest radiographs and abdominal plain films and 1 patient had an upper gastrointestinal study.

Among these 4 patients, 3 presented with abdominal pain, weight loss, and a palpable abdominal mass. The fourth patient presented with diarrhea, likely due to a prior pancreatectomy. On abdominal CT, 3 patients had an abdominal mass, 2 of whom had hydronephrosis. The fourth patient did not have a visible mass on CT but did have gastric and proximal duodenal wall thickening. We describe the details of these 4 cases.

Case 1

A 66-year-old man presented with a 3-month history of abdominal pain radiating to his back and to the right side greater than the left side. An outside colonoscopy reported Crohn disease with ulcerations in the ileum. On abdominal CT at the outside institution, a 4.9 × 4.2-cm mass was identified adjacent to the terminal ileum, with surrounding soft tissue stranding and mildly enlarged lymph nodes. The patient started therapy for inflammatory bowel disease. Repeat CT examination at 1 month later showed enlargement of the ileal mass to 10 × 6 cm. The superior mesenteric vein and artery were encased in the mass. In addition, the mesenteric mass extended into the retroperitoneum, to obstruct the right ureter (Fig. 1).

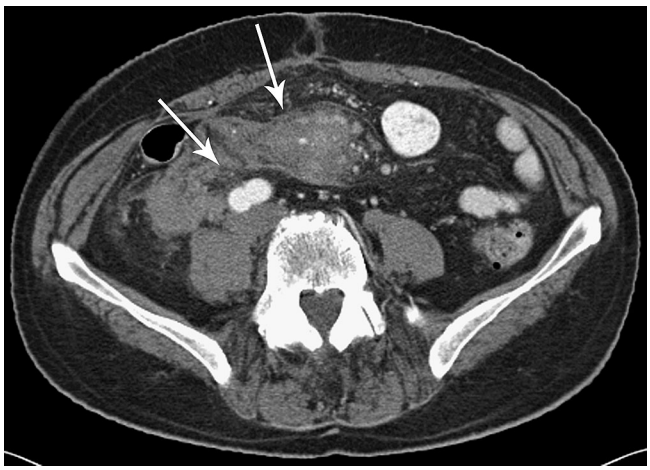

Fig. 1. Gastrointestinal basidiobolomycosis in a 66-year-old man. Computed tomography of the abdomen shows a large mesenteric mass (*arrows*) that obstructs the right ureter (not shown).

The patient underwent a partial ileal and colonic resection, with incomplete removal of the mass. The patient was then transferred to our institution for care. Fine-needle aspirate and core needle biopsies of the right lower quadrant mass showed granulomatous inflammation, as well as eosinophilia and fungal organisms consistent with *B. ranarum*. Treatment with an appropriate antifungal medication was started, and the patient had not had recurrence of symptoms at 16 months later.

Case 2

A 74-year-old man with a past medical history of diverticulitis requiring a bowel resection presented with a 4- to 5-week history of anorexia, a 40-pound weight loss, left-sided abdominal pain, and diarrhea. Two palpable masses were found in the abdomen. At an outside hospital, a left upper quadrant abscess had been identified, and the patient started treatment with metronidazole and had no symptomatic improvement. At colonoscopy, a high-grade stricture was found at 40 cm.

After evaluation at an outside facility 2–3 weeks earlier, the patient was transferred to our institution. CT of the abdomen and pelvis showed a 10-cm mixed attenuation mass that engulfed the upper descending colon with luminal expansion (i.e., aneurysmal dilation). The mass abutted the lateral abdominal wall and left anterior renal capsule (Fig. 2A). An abdominal radiograph showed an abnormal collection of gas present in the region of the descending colon (Fig. 2B).

The patient underwent colonic resection and a diverting colostomy. Histopathologic evaluation was diagnostic of *B. ranarum* infection and cultures were positive. The patient was treated with antifungal medication for 3 months and improved dramatically.

Case 3

A 56-year-old diabetic man presented to an external hospital with a 6-month history of upper abdominal pain. He was treated for a peptic ulcer without improvement. Two months later, he had a palpable mass in his upper abdomen. He underwent a subtotal gastrectomy and partial colon resection. Histopathologic findings showed granulomatous disease of the colon, and the patient received a diagnosis of Crohn disease. He continued to have symptoms of diarrhea

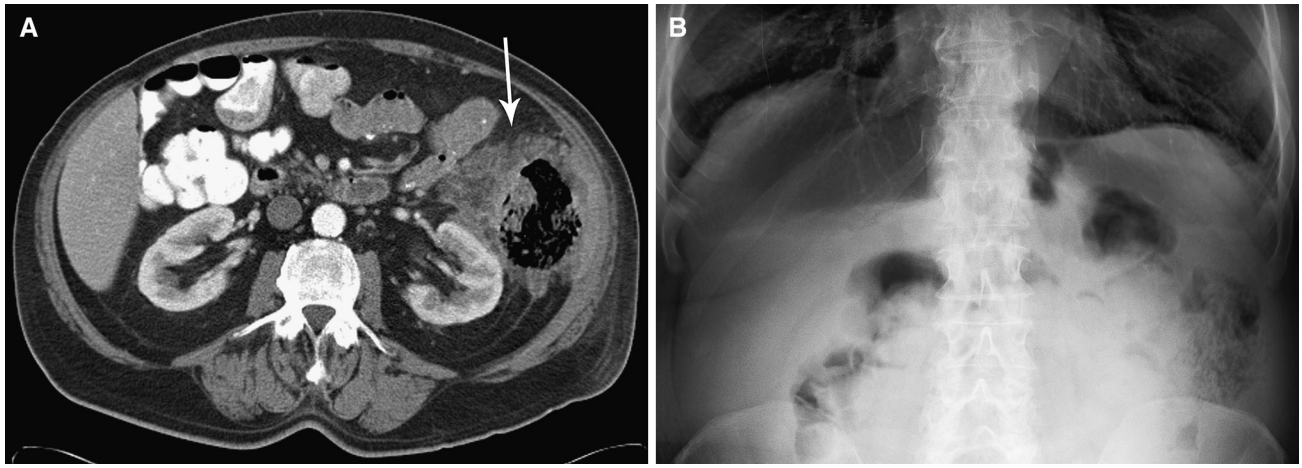

Fig. 2. Gastrointestinal basidiobolomycosis in a 74-year-old man. **A** Computed tomography of the abdomen shows a 10-cm mixed mass with a markedly thickened wall of the upper descending colon and luminal expansion (aneurysmal dilation) (*arrow*). The mass abuts the lateral abdominal wall and left anterior renal capsule. **B** An abdominal radiograph shows an abnormal gas collection in the region of the descending colon. The gas collection corresponds to the abnormal loop of colon seen on computed tomography (Fig. 2A).

and fever. A CT examination at an external institution showed left hydronephrosis, perinephric phlegmon, and rectal wall thickening. The patient underwent subsequent left ureteral stenting and right colectomy with removal of a 6.5 × 4.0 × 2.7-cm mass in the ascending colon. The histopathologic examination confirmed *B. ranarum* infection. The patient was treated with antifungal medications, but anorexia and fever developed. He was then transferred to our institution.

A repeat CT examination showed extensive perinephric inflammatory changes and abscess around the left kidney (Fig. 3A) and rectosigmoid regions (Fig. 3B). Left hydronephrosis was also present. Pathologic review was consistent with *B. ranarum*. Cultures were also positive. The patient underwent rectosigmoid resection. He restarted taking itraconazole and continued the medication for more than a year.

Case 4

A 45-year-old man with an extensive medical history, including alcohol-induced necrotizing pancreatitis requiring pancreatectomy, diabetes mellitus, hepatitis C, and chronic *Clostridium difficile* colitis, presented with persistent diarrhea (despite long-term use of oral vancomycin). Small-bowel aspirates and stool culture were positive for *B. ranarum*. The patient received voriconazole and the diarrhea resolved [3].

Retrospective review of the patient's CT abdomen and pelvis examination showed extensive postoperative changes and thickened folds within the fundus of the stomach (Fig. 4A). An upper gastrointestinal study showed thickened folds in the fundus and body of the stomach and within the proximal duodenum (Fig. 4B). Abdominal plain films were negative for disease.

Fig. 3. Gastrointestinal basidiobolomycosis in a 57-year-old man. **A** Computed tomography of the abdomen shows extensive perinephric inflammatory changes and abscess (*arrows*) adjacent the left kidney. **B** Computed tomography of the pelvis shows an extensive inflammatory mass containing fluid and air (*arrows*) within the perirectal space.

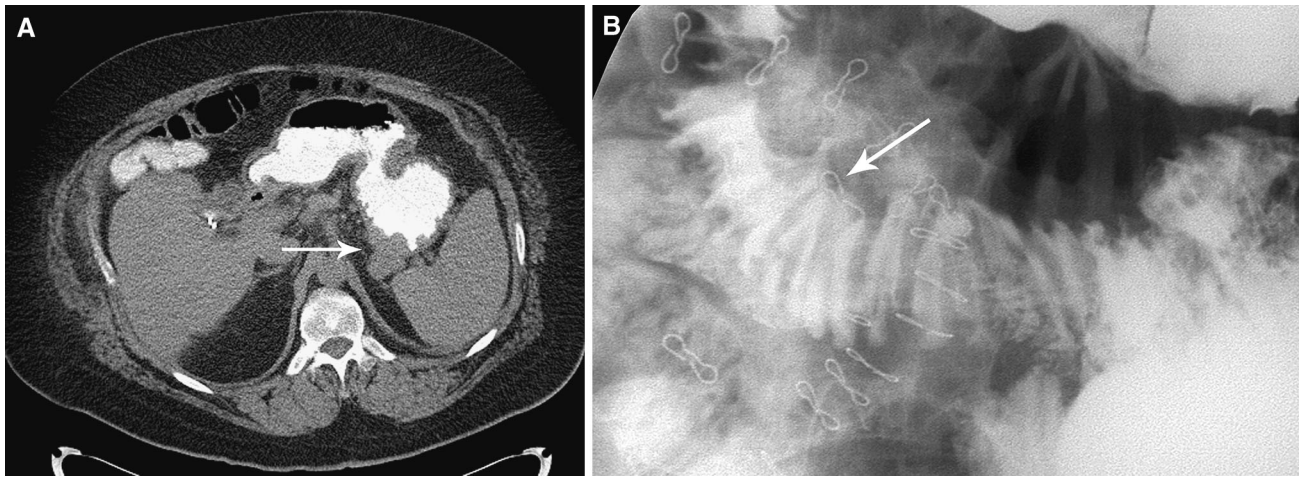

Fig. 4. Gastrointestinal basidiobolomycosis in a 45-year-old man. **A** Computed tomography of the abdomen shows thickened gastric folds (*arrow*) in the fundus. **B** An upper gastrointestinal study shows thickened folds in the duodenum (*arrow*).

Discussion

Basidiobolomycosis is an uncommon subcutaneous fungal infection restricted to arid climates around the world and rarely involves the gastrointestinal tract. GIB usually presents with nonspecific clinical symptoms, including diarrhea, abdominal pain, weight loss, and, occasionally, an abdominal mass [3, 8, 10]. A fast-growing abdominal mass is one of the strongest clues that a patient may have GIB; another is the failure of nonantifungal medical therapies.

Worldwide, the most commonly reported abdominal imaging findings in GIB are abdominal masses in the colon or liver or in 2 or more locations. Focal bowel wall thickening is often reported. These findings are most likely mistaken for a neoplasm or inflammatory bowel disease rather than identified as GIB. The finding of an abscess might lead clinicians and radiologists to consider an infectious cause earlier in the course of the disease process than when GIB is typically diagnosed.

The 4 GIB cases from our institution had similar findings to those reported elsewhere because 3 of the 4 patients (75%) had an abdominal mass detected on CT. A majority of these masses involved the mesentery or colon, with 2 of 3 masses involving the kidney masses (either abutting the kidney or causing urinary obstruction). All the abdominal masses showed an inflammatory component with adjacent soft tissue stranding, with or without abscess formation. Two of our 4 patients initially received a diagnosis of Crohn disease at other institutions [2, 5, 6].

Radiologists should consider GIB when a patient from an arid climate presents with abdominal pain, weight loss, and an inflammatory mass on abdominal CT. Tissue diagnosis is usually required for confirmation.

The present study has several limitations. First, many of the case reports in the English literature had inadequate details regarding the complete imaging findings of GIB. Second, the 4 patients in the present study, evaluated at our institution, had their initial presentation and work-up at an outside hospital. This external evaluation might have resulted in missed data and missed access to outside imaging studies, as well as further progression of GIB, before the patient's evaluation at our institution, when the diagnosis of GIB was established.

In conclusion, GIB is an uncommon fungal infection that in the United States appears to be restricted to the arid Southwest. Abdominal masses in the colon or other organs, bowel wall thickening, and abscess are the dominant imaging findings. These imaging findings are nonspecific and most commonly represent either neoplasm or inflammatory bowel disease. However, the diagnosis of *B. ranarum* infection should be considered, especially when patients are not responding to the prescribed therapy or have unusual findings, such as multiple abdominal inflammatory masses or abscesses. Consideration of GIB in such cases and the biopsy of tissue (either by an interventional radiologist or a surgeon) for histopathologic analysis and culture will facilitate prompt diagnosis and timely initiation of antifungal therapy.

Conflict of interest None.

Ethical standards This study complies with the ethical standards and laws of the United States of America.

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