Inferior Mesenteric Vein Thrombosis Resulting in Pylephlebitis in a Patient With Inflammatory Bowel Disease

Erica C. Becker, MD, MPH1, Dheera Grover, MBBS1, Sanket Patel, DO2, Purabi Dhakras, MD3, and Lisa Rossi, MD4

1Department of Internal Medicine, University of Connecticut Health Center, Farmington, CT
2Karsh Division of Gastroenterology and Hepatology, Cedars-Sinai Medical Center, Los Angeles, CA
3Division of Pathology, St. Francis Hospital and Medical Center, Hartford, CT
4Division of Gastroenterology, St. Francis Hospital and Medical Center, Hartford, CT

ABSTRACT

Although uncommon, pylephlebitis is a life-threatening complication of mesenteric vein thrombosis. It is reported to affect adult men more commonly than women, with a median age of 57 years. Furthermore, few cases have been reported of pylephlebitis in patients with inflammatory bowel disease. Owing to low prevalence, there is a paucity of data regarding the diagnosis and management of pylephlebitis. Treatment approach ranges from conservative management (antibiotics and/or systemic anticoagulation) to surgical management. We report a case of a 62-year-old man diagnosed with inflammatory bowel disease with a course complicated by pylephlebitis of the inferior mesenteric vein managed successfully with antibiotics and anticoagulation.

INTRODUCTION

Pylephlebitis is an uncommon complication of mesenteric vein thrombosis (MVT) that has been associated with inflammatory bowel disease (IBD). Mesenteric pylephlebitis in Crohn’s disease (CD) is rare, and there are few published cases with pylephlebitis occurring in patients with ulcerative colitis (UC). It is more likely to affect adult men with a median range of 57 years. The incidence is unclear, but it has been estimated to be 2.7 per 100,000 person-years with a mortality rate between 11% and 32%.1,2 Studies suggest that only 30% of patients with pylephlebitis have localizing symptoms (ie, abdominal pain, nausea, and vomiting). There is often a delay in diagnosis because clinical symptoms may be nonspecific, which can lead to complication rates as high as 20%.1 Furthermore, there are no guidelines for the management of these patients, and often they are treated at the physician’s discretion. Thrombosis typically occurs in the superior mesenteric vein (SMV), and current literature reports a 5% prevalence of thrombosis within the inferior mesenteric vein (IMV). We present a case of pylephlebitis occurring within the IMV in a patient during an IBD flare.

CASE REPORT

A 62-year-old non-Hispanic White man diagnosed with ulcerative pancolitis s/p induction of infliximab 2 weeks earlier presented with a 1-day onset of chills, diaphoresis, and generalized nonradiating abdominal pain. On presentation, the patient was afebrile, normotensive with a regular heart rate, and on room air. Physical examination was notable for an ill-appearing man with suprapubic and left lower quadrant tenderness without any peritoneal signs. Laboratory work revealed a normal white blood cell count, anemia with hemoglobin 10.8 g/dL (reference range 12–16 g/dL), and a normal platelet count. A comprehensive metabolic panel and thyroid profile were within normal limits. Lactic acid was 3.4 mmol/L (reference range 0.5–2.2 mmol/L). C-reactive protein was elevated to 13.6 mg/dL (reference range < 0.9 mg/dL). Blood cultures were positive for group C Streptococcus and Staphylococcus epidermidis. Fecal calprotectin was 1,610 μg/g (reference range < 50 μg/g). Clostridium difficile was negative. Stool culture for Salmonella, Shigella, and Campylobacter were negative. Stool ova and parasites were negative for Giardia and Cryptosporidium. Viral hepatitis
serologies were within normal limits. Abdominal and pelvic computed tomography (CT) revealed inferior MVT, involvement of its branches, and foci of air within the splenic vein along with significant colitis of the left colon.

Colonoscopy during this hospitalization revealed erythema with mild friability, and histology was negative for *Cytomegalovirus* (Figure 2). The Mayo score was 8 points. The patient was promptly started on antibiotics with cefepime and vancomycin. He was also started on intravenous heparin. Transthoracic echocardiogram did not reveal any vegetations. Other infectious workup, including chest x-ray, urine analysis, and spinal magnetic resonance imaging (MRI), was unrevealing. Cutaneous infections, lines, and medical hardware were ruled out as an infectious source as well. Before discharge, he had resolution of abdominal pain. On discharge, he was transitioned to Lovenox for a duration of at least 6 months and his antibiotics were adjusted to cefpodoxime and metronidazole based on culture and sensitivities. His colitis flare was managed with a prednisone taper on discharge. Per patient’s preference and because of previous mild nonanaphylactic reaction during induction therapy, the outpatient treatment was switched from infliximab to vedolizumab.

Nine months later, the patient had a repeat abdominal and pelvic CT for left lower quadrant and suprapubic abdominal pain associated with hematuria. Imaging now showed extensive proctocolitis with the development of colovesical fistula and a perineal abscess. The thrombus noted on previous imaging was now resolved. He underwent surgery with abscess drainage, debridement, sigmoid colectomy, and diverting loop ileostomy. Pathology was notable for diverticulosis complicated by acute diverticulitis with rupture and peridiverticular abscess formation with associated serositis and serosal fibrous adhesions within the sigmoid colon and proximal rectum (Figure 3). Pathology was negative for dysplasia and malignancy. It was believed that vedolizumab did not have adequate time to work, and after initiation, the disease manifested itself over ensuing months to CD with fistulizing and perianal complications. After surgery, vedolizumab was continued to prevent postoperative recurrence as the patient was high-risk. The ileostomy was reversed after disease improvement with vedolizumab.

The first case of pylephlebitis was reported in 1846 during an autopsy; however, the first case in a patient with IBD was not reported until 100 years later. Pylephlebitis is commonly associated with diverticulitis, pancreatitis, cholangitis, peptic ulcer disease, liver abscess, and gastroenteritis. Based on our

**DISCUSSION**

The first case of pylephlebitis was reported in 1846 during an autopsy; however, the first case in a patient with IBD was not reported until 100 years later. Pylephlebitis is commonly associated with diverticulitis, pancreatitis, cholangitis, peptic ulcer disease, liver abscess, and gastroenteritis. Based on our
literature review, less than 10 published cases have described pylephlebitis occurring in CD and only 5 cases are published in patients with UC (Table 1). These cases demonstrate a male predominance. Abdominal pain, fever, and weight loss were the most commonly reported symptoms while bacteremia was reported in less than half of the cases. Treatments included antibiotics with or without systemic anticoagulation, and roughly half required surgical management needing bowel resection (Table 1).

Pylephlebitis is most commonly reported to involve the right portal vein, main portal vein, and SMV. The SMV drains the second portion of the duodenum to the right two-thirds of the transverse colon, whereas the IMV drains the remaining one-third of the transverse colon, descending and sigmoid colon, and rectum. The SMV is more commonly thrombosed, and studies suggest that MVTs are found within the same territory in which IBD is located. Our case follows a similar presentation. The prevalence of MVT remains unknown within the IBD population; however, the risk seems to be increased and is speculated to be 8 times higher during a flare. Bowel wall inflammation and localized venous engorgement from MVT could be correlating factors. Pylephlebitis may ensue after bacteria translocate from the site of bowel damage or infection into small veins, draining this area and then become thrombosed. This process extends to involve the portal vein and/or mesenteric veins.

Current diagnostic criteria and management have not been standardized. Diagnosis is difficult because symptoms are often

Figure 3. (A) 2×: Diffuse chronic colitis with severe activity and transmural inflammation. (B) 2×: Inflammatory pseudopolyps. (C) 4×: Diffuse chronic colitis with activity in the form of acute cryptitis and crypt abscess formation. (D) 10×: Diffuse chronic colitis with activity in the form of acute cryptitis and crypt abscess formation.
nonspecific and requires a high clinical index of suspicion. Laboratory parameters may help but are neither sensitive nor specific. Leukocytosis and bacteremia may not be present. D-dimer testing is nonspecific and has not been well studied in MVT or pylephlebitis. Imaging must be correlated with clinical presentation. Duplex sonography, CT, and MRI are the available options. A diagnosis of thrombosis is demonstrated by a flow defect and dilation or the absence of compressibility on sonography. CT and MRI may be diagnostic with central hypodense contrast compared with peripheral hyperdense contrast media. CT and MRI are more frequently used because they help rule out other intra-abdominal pathology while being more sensitive at detecting thrombi within the splenic and mesenteric veins and also detect its sequelae (ie, abscesses or ischemia). Owing to limited data, standardized treatment options have not yet been established because of lack of robust randomized clinical trials. Combination treatments have been reported in the literature for pylephlebitis with varying efficacies. These include using antibiotics, anticoagulation, and surgical intervention. The decision to use an anticoagulant remains controversial. Published case reports have demonstrated good outcomes with and without the use of systemic anticoagulation for at least 6 months. In a review of 96 patients by Choudhry et al, 54 patients received antibiotics alone, 46 patients received anticoagulation (usually with warfarin) alone, and 64 patients received both antibiotics and anticoagulation. Only 1 patient required surgical intervention. However, in patients with concurrent IBD, successful treatment varies from conservative, nonsurgical management to surgical intervention. Conservative management, including intravenous fluids, antibiotics, and anticoagulation, showed improved outcomes when patients have concurrent IBD by allowing for the time to optimize health before undergoing a procedure, thus allowing for bowel-sparing surgery and minimizing complications.

Septic MVT is a medical emergency with the potential to have poor prognosis. Symptoms may be absent, nonspecific, or masked by an IBD flare or other abdominal disease; thus, clinicians should consider pylephlebitis as a differential diagnosis. Poor outcomes such as septic shock, acute respiratory distress syndrome, abscess, infarction, perforation, or death may be avoided with prompt diagnosis and treatment with antibiotics and systemic anticoagulation. Unfortunately, bowel resection is more common when patients have pylephlebitis and IBD compared with pylephlebitis alone. Further studies in this area may help illustrate why this is and help improve outcomes.

### DISCLOSURES

Author contributions: E. Becker and D. Grover wrote the manuscript. S. Patel and L. Rossi revised the manuscript for intellectual content. E. Becker, D. Grover, S. Patel, and L. Rossi approved the final manuscript. All authors have participated

| Reference | Type | Sex/age | Symptoms | Image findings | Bacteremia | Treatment |
|-----------|------|---------|----------|----------------|------------|-----------|
| Aguas et al | CD | M/25 | Abdominal pain and fever | Mesenteric partial thrombosis and multiple liver abscess | No | Antibiotics, anticoagulants, and percutaneous liver abscess drainage |
| Ajzen et al | CD | M/64 | Abdominal pain, anorexia, and weight loss | Portal vein gas and septic ascending portal thrombophlebitis | No | Antibiotics and terminal ileum resection |
| Baddley et al | CD | M/41 | Jaundice, fever, vomiting, and weight loss | Portal-MVT, ileal abscess and perforation secondary to CD | No | Antibiotics and terminal ileum and right colon resection |
| El-Matary et al | CD | F/14 | Anorexia, weight loss, and jaundice | Portal-MVT and gas | No | Antibiotics, anticoagulant, and terminal ileum and right colon resection |
| Ng et al | CD | M/19 | Abdominal pain and fever | Portal vein gas and thrombosis peritonitis and pneumopertitoneum | No | Antibiotics and total colectomy |
| Scaringi et al | CD | M/47 | Abdominal pain and fever | Portal-MVT and gas | Yes | Antibiotics and ileocolic resection |
| Shin et al | CD | M/16 | Abdominal pain and fever | Portal-MVT | Yes | Antibiotics and anticoagulants |
| Tung et al | CD | F/18 | Abdominal pain, fever, diarrhea, and weight loss | Portal-MVT, liver abscess | No | Antibiotics, anticoagulants, and CT-guided liver abscess needle aspiration |
| Felsen and Wolarsky | UC | F/62 | Abdominal pain, fever, and diarrhea | Diagnosed on autopsy | Yes | Antibiotics |
| Taylor | UC | F/37 | Abdominal pain, fever, and diarrhea | Diagnosed on autopsy | Yes | Antibiotics |

CD, Crohn’s disease; CT, computed tomography; MVT, mesenteric vein thrombosis; UC, ulcerative colitis.
significantly in writing of this manuscript and approve of its content. The final manuscript has been seen and approved by all authors. L. Rossi is the article guarantor.

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REFERENCES

1. Choudhry AJ, Baghdadi YMK, Amr MA, Alzghari MJ, Jenkins DH, Zielinski MD. Pylephlebitis: A review of 95 cases. J Gastrointest Surg. 2016;20(3):656–61.
2. Hamra L, Abraham S, Jordan J. Pylephlebitis as a rare complication of ulcerative colitis: A case report. Cureus. 2019;11(5):e4792.
3. Rutgeerts PR, Sandborn WJ, Feagan BG, et al. Infliximab for induction and maintenance therapy for ulcerative colitis. N Engl J Med. 2005;353:2462–76.
4. Clinical Review Report: Adalimumab (Humira). Canadian Agency for Drugs and Technologies in Health: Ottawa, ON, Canada, 2016.
5. Aguas M, Bastida G, Nos P, Beltran B, Grueso JL, Grueso J. Septic thrombophlebitis of the superior mesenteric vein and multiple liver abscesses in a patient with Crohn’s disease at onset. BMC Gastroenterol. 2007;7(1):22.
6. Scaringi S, Giudici F, Gabbani G, et al. Pylephlebitis and Crohn’s disease: A rare case of septic shock. Int J Surg Case Rep. 2017;39:106–9.
7. Hartpence J, Woolf A. Pylephlebitis. In: StatPearls. StatPearls Publishing: Treasure Island, FL, 2021.
8. Felsen J, Wolarsky W. Suppurative pylephlebitis with bacteremia in chronic ulcerative colitis. Ann Intern Med. 1995;33(1):211–6.
9. Shin AR, Lee CK, Kim HJ, et al. Septic pylephlebitis as a rare complication of Crohn’s disease. Korean J Gastroenterol. 2013;61(4):219–24.
10. Violi NV, Schoepfer AM, Fournier N, Guix B, Bize P, Denys A. Prevalence and clinical importance of mesenteric venous thrombosis in the Swiss inflammatory bowel disease cohort. Am J Roentgenol. 2014;203(1):62–9.
11. Russell CE, Wadhera RK, Piazza G. Mesenteric venous thrombosis. Circulation. 2015;131(18):1599–603.
12. Ajzen SA, Gibney RG, Cooperberg PL, Scudamore CH, Miller RR. Enterovenous fistula: Unusual complication of Crohn disease. Radiology. 1988;166(3):745–6.
13. Baddley JW, Singh D, Correa P, Persich NJ. Crohn’s disease presenting as septic thrombophlebitis of the portal vein (pylephlebitis): Case report and review of the literature. Am J Gastroenterol. 1999;94(3):847–9.
14. El-Matary W, Jaffray B, Scott J, Hodges S. Portal pyaemia as a presenting feature of paediatric Crohn disease. J Pediatr Gastroenterol Nutr. 2006;43(2):260–2.
15. Ng SSM, Yiu RYC, Lee JFY, Li JCM, Leung KL. Portal venous gas and thrombosis in a Chinese patient with fulminating Crohn’s colitis: A case report with literature review. World J Gastroenterol. 2006;12(34):5582–6.
16. Tung JY, Johnson JL, Llaucaras CA. Portal-mesenteric pylephlebitis with hepatic abscesses in a patient with Crohn’s disease treated successfully with anticoagulation and antibiotics. J Pediatr Gastroenterol Nutr. 1996;23(4):474–8.
17. Taylor FW. Regional enteritis complicated by pylephlebitis and multiple liver abscesses. Am J Med. 1949;76(6):838–40.

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