



Ischemic Colitis From Idiopathic Myointimal Hyperplasia of the Mesenteric Veins in a Post–Liver Transplant Patient

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ABSTRACT

Idiopathic myointimal hyperplasia of the mesenteric veins (IMHMV) is a rare cause of nonthrombotic, noninflammatory ischemic colitis. IMHMV classically presents in men with abdominal pain and bloody diarrhea and is frequently misdiagnosed as inflammatory bowel disease. However, IMHMV causes a more protracted, relapsing course of abdominal pain that does not respond to medical therapy. The diagnosis can be secured by colonoscopic biopsy. Surgical resection is curative and should be considered even in high-risk patients. Here, we describe a case of IMHMV diagnosed preoperatively in a post–liver transplant patient with residual portal hypertension who ultimately underwent successful surgical resection.

INTRODUCTION

Idiopathic myointimal hyperplasia of the mesenteric veins (IMHMV) is a rare cause of ischemic colitis. It causes venous congestion and ischemia of the sigmoid colon and rectum most commonly and may clinically mimic inflammatory bowel disease (IBD). Colonoscopic biopsies can be helpful to distinguish between ischemia and inflammation. However, successful nonsurgical management of IMHMV has rarely been reported. We present here a case of IMHMV in a post–liver transplant patient and highlight the need for early surgical planning because surgical resection appears to be curative and unavoidable.

CASE REPORT

A 72-year-old man with cirrhosis because of nonalcoholic steatohepatitis complicated by hepatocellular carcinoma status-post recent liver transplant 5 months before presentation (on immunosuppression with tacrolimus and mycophenolate mofetil), with post-transplant course complicated by *Cytomegalovirus* viremia, was admitted to an outside hospital for 1 day of left lower-quadrant abdominal pain and nonbloody diarrhea. Initial abdominal computed tomography scan demonstrated left-sided colitis from the splenic flexure to the sigmoid colon. Infectious stool studies were negative. Initial empiric antibiotic therapy improved his symptoms. However, he was readmitted 3 days later with recurrent abdominal pain and new-onset bloody diarrhea. Repeat computed tomography revealed persistent left-sided colitis (Figure 1). He was also noted to have splenic and gastroesophageal varices that extended into the rectosigmoid region. Laboratory test results were unremarkable, including negative serum *Cytomegalovirus* viral load and repeat negative stool studies.

Subsequent colonoscopy was notable for edematous left colon with friability and intermittent ulceration, with difficulty advancing the colonoscope into the proximal sigmoid colon (Figure 2). Colonoscopic biopsies were interpreted as consistent with ischemic colitis, based on the damage to the surface epithelium, small withering crypts, and hyalinization of the lamina propria. Arterialization of the lamina propria capillaries was also noted, concerning for IMHMV. Given concerns regarding increased surgical risk in the setting of intra-abdominal varices and immunosuppression, he was initiated on medical therapy with budesonide, resulting in initial improvement of his abdominal pain. However, he soon developed acutely worsening abdominal pain with peritoneal signs and required urgent

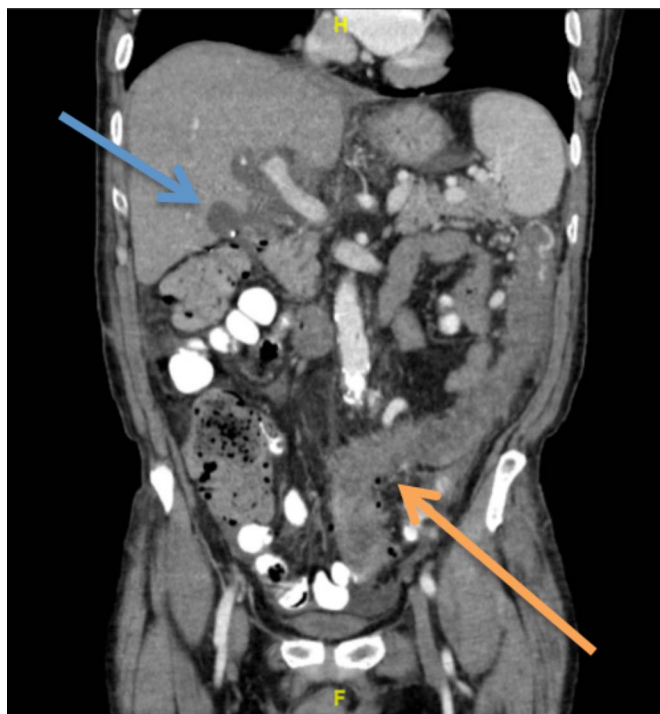


Figure 1. Left-sided colitis (orange arrow) extending from splenic flexure to the sigmoid colon on computed tomography. Post-liver transplant infrahepatic seromas are stable compared with previous imaging (blue arrow).

surgical resection with end colostomy. Careful resection was performed, given his known splenic and rectosigmoid varices. Final surgical pathology showed submucosal wall thickening, consistent with IMHMV (Figure 3). His post-operative course was uncomplicated, and his symptoms remain in remission.

DISCUSSION

To the best of our knowledge, this case is the first case of IMHMV to be reported in a post-liver transplant patient. IMHMV is an uncommon cause of chronic ischemic colitis, more often seen in middle-aged men. The pathogenesis is unknown, but the pathology findings suggest vascular remodeling in the setting of venous hypertension and localized trauma from sigmoid hypermobility and intermittent volvulus.¹ Although no portal vein thrombus or anastomotic stricture was observed in this patient, he did have evidence of venous congestion and prominent varices that extended to the rectosigmoid area. We suspect that this residual sinistral portal hypertension after liver transplant likely increased his risk of developing IMHMV. Often clinically misdiagnosed as IBD, IMHMV is important to consider in patients with protracted courses of relapsing abdominal pain and findings of left-sided colitis that do not improve with medical therapy. Although IMHMV typically affects the descending, sigmoid colon, and rectum, there have been few case reports of small bowel involvement.²⁻⁴ Only 1 other case report has been described in an

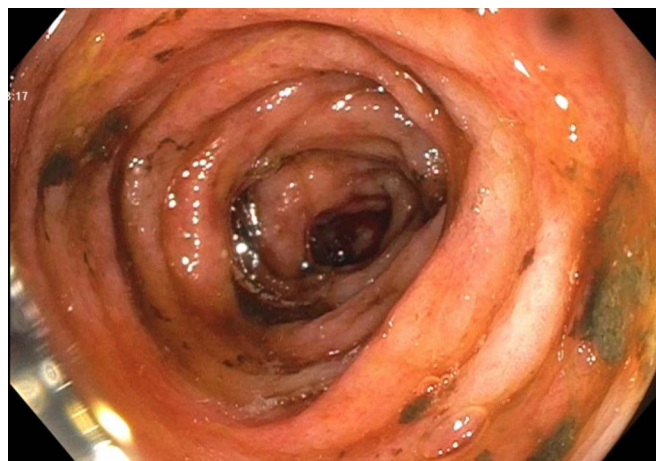


Figure 2. Edematous descending colon with friability and intermittent ulcerations on colonoscopy.

immunosuppressed transplant patient, although the patient had undergone kidney transplantation as opposed to liver.²

Most cases are diagnosed postoperatively from surgical resection specimen. However, this case is a rare example of colonoscopic biopsies that were diagnostic of IMHMV preoperatively.⁵ Although it can be difficult to differentiate ischemic colitis from IBD clinically, endoscopic evaluation and biopsies can help distinguish between ischemic and inflammatory colitis.⁶ Diffusely edematous and congested colonic mucosa without deep ulceration particularly in the left-sided colon, in combination with evidence of venous congestion or venous outflow tract problems in the patient, can increase one's suspicion for IMHMV. If suspicion for IMHMV is high, it is important to obtain biopsies deep enough to sample the submucosal layer to allow for optimal pathological evaluation.⁷ To minimize the risk of perforation from biopsy, performing a lower endoscopic ultrasound-guided biopsy would be of higher yield and safety. Diagnosis based on colonoscopy biopsy can then guide patient-shared decision-making regarding timing of surgical intervention because surgical resection can be curative.⁸ In the literature, although the average time to surgery is about 6 months and can be delayed up to 2 years depending on disease severity, sustained remission of symptoms cannot be achieved with medical therapy alone.^{9,10} Surgery was ultimately required in all cases to avoid complications such as bowel necrosis and perforation.^{9,11,12} Therefore, despite the higher risk of surgery in these liver transplant patients with intra-abdominal varices, immunosuppression, and adhesions from previous abdominal surgery, surgery for IMHMV should still be pursued early to avoid even higher surgical risk when performed emergently after complications develop.^{13,14}

DISCLOSURES

Author contributions: R. Wong wrote the manuscript and reviewed the literature. D. Westerveld provided the images and edited the manuscript. H. Yeo edited the manuscript. J. Jessurun

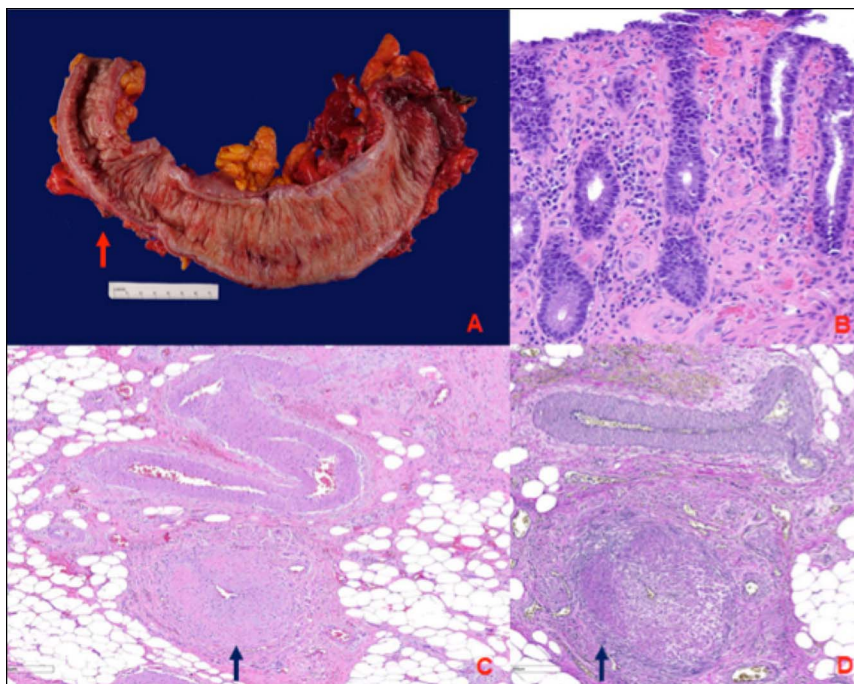


Figure 3. (A) Resection specimen showing narrowing of the distal colon by a thickened wall (arrow). (B) Section of the mucosa showing arterIALIZATION of lamina propria capillaries on hematoxylin and eosin (H&E) stain. These changes were present in the preoperative mucosal biopsy specimen. (C) Mesenteric vein obliteration by myointimal hyperplasia with obliteration of the lumen (arrow) on H&E stain. (D) An elastic tissue stain highlights a normal artery (top) and an obliterated vein (bottom, arrow).

provided the pathological images. A. Jesudian revised the manuscript for intellectual content and is the article guarantor.

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