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July 2018

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Recommended Citation

Khan, M., Khan, I., Humza, A., Hadi, B., Haider, Z. (2018). Enteroenteric fistulae in acute bowel ischemia. *Journal of the College of Physicians and Surgeons Pakistan*, 28(7), 568-571.

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Enteroenteric Fistulae in Acute Bowel Ischemia

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ABSTRACT

Gastrointestinal fistulae are classified as enteroenteric or enterocutaneous. Most gastrointestinal fistulae are formed after surgical procedures for inflammatory bowel disease or malignancy. For spontaneous enteroenteric fistulae, ischemia has been reported as a possible etiology. We report two cases of spontaneous enteroenteric fistulae arising after bowel ischemia; a 38-year male with a 10-day history of severe abdominal pain with fever and vomiting, and a 22-year female with a one-week history of abdominal pain and diarrhea. Arterial and venous thrombi in association with enteroenteric fistulae were identified on computed tomography. These cases point towards acute mesenteric ischemia as a rare cause of spontaneous enteroenteric fistulae. Surgical management of these fistulae can be effective in resolving this complication.

Key Words: *Infarction. Intestinal fistula. Mesenteric ischemia. Acute abdomen. Enteroenteric fistula. Abdominal radiography. Computed tomography.*

INTRODUCTION

Fistulae are abnormal connections between two organs. Enteric fistulae are classified as internal or external, the former draining to an abnormal site in the gastrointestinal lumen or any other internal organ; and the latter draining to a perforation in the skin.¹ Abdominal surgery is the most commonly reported cause of such fistulae, followed by Crohn's disease which accounts for about 20-30% of them.² Ischemia has been hypothesised as a possible etiology for spontaneous enteroenteric fistulae,^{3,4} but as yet no cases of enteroenteric fistulae definitively arising from mesenteric ischemia have been reported. Given the rarity of enteroenteric fistulae secondary to mesenteric ischemia, there is no guideline regarding their ideal course of management.

We report two cases of spontaneous enteroenteric fistula formation in association with mesenteric ischemia resulting from arterial or venous thrombi that were surgically managed and resulted in complete resolution of symptoms.

CASE REPORT

Case 1: A 38-year male presented to the emergency room with a 10-day history of severe abdominal pain associated with fever and vomiting. Past medical and surgical history was unremarkable as was drug history. Family history was positive for hypertension in the patient's father. He was an alcoholic with a 10-year

history of alcohol consumption. Review of systems was unremarkable. Physical examination revealed a vitally stable patient with generalised abdominal tenderness and no other positive findings.

Computed tomography (CT) scan of the abdomen showed an eccentric plaque-like partial thrombus at the origin of superior mesenteric artery (SMA). Based on clinical presentation and CT scan findings, a diagnosis of bowel ischemia secondary to SMA thrombosis was made. He was kept on *nil per oral* (NPO) regimen and a nasogastric tube was passed. Conservative management with total parenteral nutrition, intravenous (IV) antibiotics and warfarin infusion was started. Subsequently, his condition improved and he was discharged. A follow-up visit after one week was planned.

The patient presented again 28 days later with complaints of abdominal pain and vomiting. On examination, the only positive finding was tachycardia, with a heart rate of 123 beats per minute. CT scan of the abdomen revealed progression of mesenteric ischemia with an increase in SMA thrombus size, dilated thick-walled jejunal, and ileal loops and developing pneumatosis intestinalis (Figure 1). Again, he was conservatively managed and discharged on the fifth day of admission upon resolution of symptoms.

Four months later, he presented with abdominal pain and 23 kgs weight loss. Physical examination did not yield any positive findings. CT scan of the abdomen showed ischemic strictures in jejunal loops with multiple loops coalescing to form an enteroenteric fistula. These findings were confirmed on CT scan and barium follow through (Figures 2a and 2b). Based on these findings, a diagnosis of chronic SMA thrombosis with ischemic jejunal stricture and enteroenteric fistula was made.

The patient underwent small bowel resection anastomosis with stricturoplasty. Intraoperatively, the small bowel was found to be dilated with loops adherent to omentum and

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Received: May 17, 2017; Accepted: October 29, 2017.

colon, forming a closed loop obstructed region. The presence of the enteroenteric fistula was confirmed intraoperatively. Three to four matted loops with multiple strictures were also noted. Postoperative recovery was unremarkable. At a one year follow-up period, the patient did not report any symptoms.

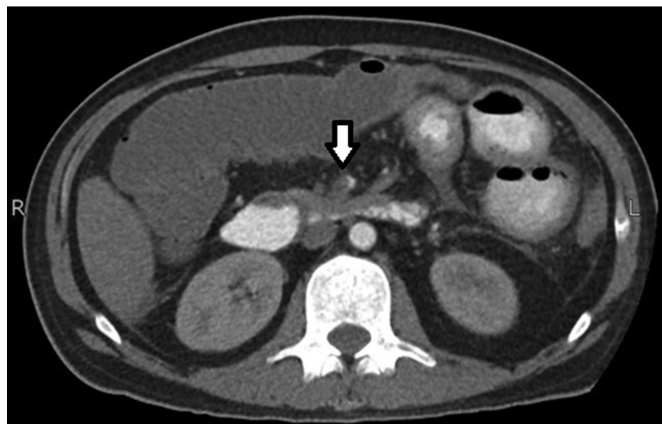


Figure 1: Axial post-contrast arterial phase CT image shows dilated thick-walled small bowel loops in left mid abdomen showing fat stranding in the adjacent mesentery, suggestive of ischemic loops. Note the partial thrombus in SMA (arrow).

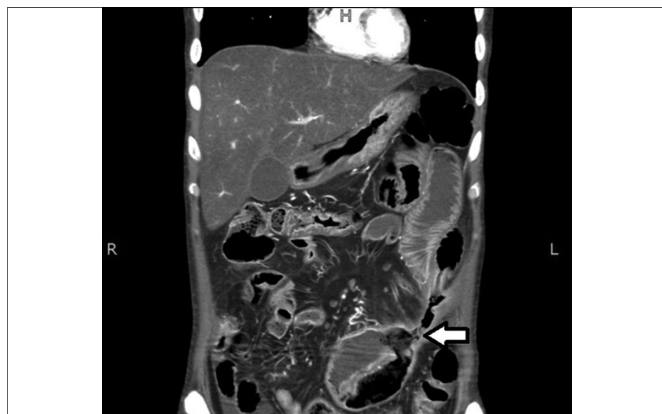


Figure 2a: Same patient 5 months later coronal post-contrast CT scan demonstrating development of ischemic strictures in left-sided jejunal loops with multiple loops coalescing and forming enteroenteric fistula (arrow). There is resolution of bowel thickening and enhancement of bowel walls with dilated lumen proximal to stricture. Note development of fatty infiltration of liver.



Figure 2b: Axial post-contrast CT image of the same patient after 5 months of initial ischemic ictus, demonstrating coalescing small bowel loops showing enteroenteric fistula formation in left abdomen with stricture and enhancement.

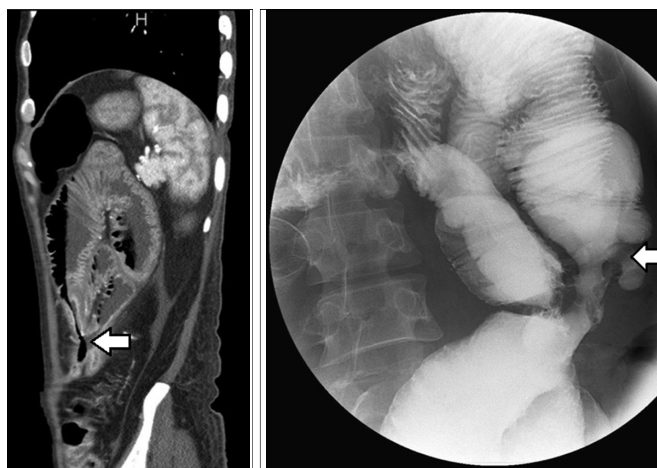


Figure 3a: Sagittal-post contrast CT image of the same patient after 5 months of initial ischemic ictus, demonstrating coalescing small bowel loops showing enteroenteric fistula formation in left abdomen with stricture and enhancement. Note after coalescing, there is tiny outpouching simulating a diverticulum (arrow).

Figure 3b: Same patient after the CT, barium follow through. Image after 5 months of initial ischemic ictus, demonstrating clustered and strictured small bowel loops showing enteroenteric fistula with superimposed strictured and a few dilated small bowel loops in left abdomen showing tiny outpouching simulating a diverticulum (arrow).

Case 2: A 22-year female presented to the general surgery service with a one week history of abdominal pain and diarrhoea. The pain was sudden in onset and was aggravated by oral intake. The patient also complained of bilious vomiting and nausea. She had no significant comorbid condition and the review of systems was unremarkable. On examination, she was vitally stable. Abdominal exam revealed generalised tenderness, while a ballooned rectum with smooth walls was found on digital rectal examination. Other systemic examinations were unremarkable.

Abdominal CT scan showed thrombosis in the mesenteric and portal veins with ischemia of the duodenum and proximal jejunal loops. She was kept on NPO regimen and a nasogastric tube was passed. Conservative management was planned and the patient was started on IV fluids, antibiotics and anticoagulants, which resulted in clinical improvement and complete resolution of symptoms by the fifth day of admission. The nasogastric tube was removed on the seventh day of admission and the patient was discharged two days later with the plan for a follow-up visit after one week.

The patient presented to the emergency room five days later with complaints of abdominal pain and vomiting for the past two days. Physical examination revealed a vitally stable patient with a mildly tender abdomen that was dull to percussion. A CT scan was performed which showed a stricture at the duodenojejunal junction with marked gastric and duodenal dilatation proximal to the stricture. An enteroenteric fistula was found between the proximal jejunal loops (Figures 3a and 3b). Exploratory laparotomy and gastrojejunostomy was performed. She was discharged on the 14th day of admission after an uneventful recovery. The patient was stable and symptom-free at a nine-month follow-up period.

DISCUSSION

Gastrointestinal fistulae are uncommonly encountered in surgical practice. The etiological categories of gastrointestinal fistulae include postoperative fistulae, spontaneous fistulae, and trauma-induced fistulae. About 75-85% of gastrointestinal fistulae form following surgical procedures for Inflammatory Bowel Disease (IBD), pancreatitis or malignancy, while 15-25% form spontaneously, without any preceding surgical intervention.⁴

The most common cause of spontaneous fistula formation is Crohn's disease with around half of all patients with Crohn's disease developing a fistula during the disease course.⁵ Abscess formation, appendicitis, diverticulitis, perforation and obstruction of bowel loops resulting from inflammatory conditions, such as pancreatitis and IBD, can also cause spontaneous fistula formation.⁴⁻⁶ Pancreatic and gynaecologic malignancies and radiotherapy for malignancies are other causes of spontaneous fistula formation.⁴

The patients presented in this report developed spontaneous fistulae in the absence of the above-mentioned conditions. Both developed spontaneous enteroenteric fistulae against a backdrop of ischemic bowel documented on radiological imaging. The first patient had SMA thrombosis, while the second had mesenteric vein and portal venous thrombosis. In both patients, there was an obvious progression from bowel ischemia to enteroenteric fistulae in the ischemic region, as documented on radiology (Figures 2a and 3a). Although bowel ischemia has been mentioned as a possible cause of spontaneous enteroenteric fistula formation in literature,^{4,9} an extensive literature review failed to yield any case report or other record of spontaneous enteroenteric fistula formation as a result of ischemic bowel disease. Thus, this is the first report of spontaneous enteroenteric fistulae resulting from bowel ischemia. Notably, although post-contrast CT scan was effective in diagnosing enteroenteric fistulae in the reported cases, the sensitivity of this modality, or the most sensitive modality, for diagnosis of this complication remains undetermined.

Mesenteric ischemia can be categorised into three categories based upon the underlying pathophysiology: acute mesenteric arterial embolism, acute mesenteric arterial thrombosis and acute venous thrombosis.⁷ In contrast to embolic vascular occlusion which causes a sudden onset of abdominal pain, thrombotic insults usually result in a relatively insidious onset of abdominal pain and progressive weight loss.⁷

A recent meta-analysis concluded that acute abdominal pain (sensitivity range 60-100%), nausea and vomiting (sensitivity range 39-93%) and pain out of proportion to physical examination findings (sensitivity range 45-54%) were the most common presenting complaints in

patients with acute mesenteric ischemia (AMI) while diffuse abdominal tenderness and peritoneal signs (sensitivity ranges 54-90% and 13-65%, respectively) were the most common physical examination findings.⁸ The World Society of Emergency Surgery (WSES) 2017 guidelines recommend that severe abdominal pain out of proportion to physical examination findings should be assumed to be AMI until disproven.⁷

Laboratory results can help in diagnosis of AMI; around 90% of patients with AMI have an abnormally elevated leukocyte count while 88% have metabolic acidosis with elevated lactate level.⁷ WSES recommends an early CT angiogram in patients with lactic acidosis in combination with abdominal pain that do not otherwise appear clinically ill.⁷ The multi-detector CT (MDCT) angiogram - which has a sensitivity of 93% and a specificity of 100% - has replaced formal angiography as the diagnostic study of choice for AMI.⁷ MDCT findings in AMI can include intestinal dilatation and thickness, portal venous gas, reduced or absent visceral enhancement, pneumatosis intestinalis and pneumoperitoneum.⁷ Mesenteric ischemia can lead to mucosal ulceration, gangrene and bowel perforation resulting in peritonitis, stricture formation or haemorrhage.⁹ Both the patients presented here developed ischemic strictures after AMI (Figures 2a, 2b and 3a). It is noteworthy that, based on this report, enteroenteric fistula formation is a new addition to the list of possible complications of mesenteric ischemia.

In conclusion, this article serves to highlight enteroenteric fistulae as a rare complication of AMI, which can significantly alter the disease course and choice of treatment. A high level of suspicion and appropriate radiological investigations (e.g. CT scan, as in this case) can help in diagnosis of this condition. There is no definite guideline for the management of enteroenteric fistulae secondary to bowel ischemia. In our experience, surgical management of these fistulae can be effective in completely resolving this complication and its associated symptoms.

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