Case Notes Notes de cas

Ischemic pancolitis: recognizing a rare form of acute ischemic colitis

Noha Al-Saleh, MB; Bret M. Wehrli, MD; David K. Driman, MD; Brian M. Taylor, MD

I schemic colitis is the most common form of gastrointestinal ischemia.^{1,2} The diagnosis, however, is often elusive, as patient signs and symptoms are nonspecific. Colonic involvement is usually segmental; ischemic colitis rarely involves the entire colon, but in such cases it is associated with increased morbidity and mortality.³

Case 1

A 62-year-old woman presented with septic shock and was referred after the results of an initial laparotomy were negative. Afterward, she was persistently hypotensive and required mechanical ventilation. Abdominal computed tomographic imaging revealed free air in her upper abdomen and diffuse thickening of the entire colon. Flexible sigmoidoscopy showed patchy areas of mucosal necrosis in the colon.

At a second laparotomy, the serosa of the rectosigmoid and splenic flexure were noted to be dusky red, with multiple perforations. A total colectomy and ileostomy were performed. Histological examination revealed extensive areas of fullthickness necrosis throughout the entire length of the colon.

Case 2

A 74-year-old woman was referred to our institution 2 weeks after a Hartman's resection for perforated segmental ischemic colitis. She developed abdominal distention and acute hemodynamic instability. Repeat emergency laparotomy revealed a

grey colon proximal to the colostomy and patchy necrosis of the transverse colon. The cecum showed multiple areas of serosal ischemia. The results of histological examination indicated ischemic colitis involving the entire colon (Fig. 1).

Discussion

Ischemic colitis can be categorized as occlusive or nonocclusive.¹ Low-flow nonocclusive ischemia occurs most frequently in response to diminished cardiac output, shock, hypovolemia and the use of medications known to diminish splanchnic blood flow.² Critically ill patients are the most susceptible to ischemic colitis. The precise biological process leading to colonic ischemia remains unknown. The regions of the colon typically affected are shown in Fig. 2.²

Ischemic pancolitis is extremely rare,



FIG. 1. Ischemic colitis. Note the diffuse, continuous necrosis of the colonic epithelium with focal sparing of the base of the crypts (withering crypts), submucosal edema, vascular congestion, hemorrhage and fibrin thrombi.

with only a handful of cases documented.^{3,4} When Longo and colleagues³ reviewed cases of ischemic colitis over a period of 6 years, 12 of 43 patients presented with diffuse peritonitis and were found to have total colonic ischemia. The mortality rate for this group of patients was 75%. The authors concluded that the prognosis for total colonic ischemia is far worse than for segmental colonic ischemia, which, in their study, had an associated mortality of 22%.

Guivarc'h and associates⁴ reviewed 88 patients with ischemic colitis, of whom 18 had total colonic ischemia. The importance of perioperative colonoscopy was highlighted by these authors, as the endoscopic appearance of the mucosa, not the external appearance of the bowel,



FIG. 2. Incidences of colonic ischemia, by site. The data are from a 1999 review by S.V. Alapati and A.A. Mihas, published in volume 105 of *Postgraduate Medicine*, pages 177-80, 183-4 and 187.

London Health Sciences Centre, University of Western Ontario, London, Ont.

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Correspondence to: Dr. Brian M. Taylor, London Health Sciences Centre — University Campus, 339 Windermere Rd., London ON N6A 5A5; fax 519 663-3068; brian.taylor@lhsc.on.ca

best revealed the extent of ischemic damage and in turn determined the extent of resection.

Both of our patients fared well, presumably because the pancolonic nature of the ischemia was recognized at their respective second laparotomy and the entire colon removed.

Conclusions

Ischemic pancolitis should be considered in the differential diagnosis of ischemic colitis, particularly in patients with a hypotensive prodrome. Although indications for surgery do not differ from those of segmental ischemia, vigilant examination of the entire colon at laparotomy as well as cautious pre- or intraoperative colonoscopy are necessary in order to recognize the diffuse nature of the disease and to initiate appropriate management.

Competing interests: None declared.

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Chylous ascites secondary to small-bowel angiosarcoma

Amanda Selk, MD;* Bret Wehrli, MD;† Brian M. Taylor, MD‡

A 57-year-old man arrived at hospital after 10 weeks of progressive abdominal distention and shortness of breath. His medical history included multiple hereditary osteochondromatosis and development 8 years before of a secondary chondrosarcoma of his right hemipelvis, for which he underwent a right internal hemipelvectomy and subsequent postoperative radiotherapy. There was no evidence of recurrent chondrosarcoma.

Upon physical examination, his abdomen was distended and a fluid wave could be elicited. There was no evidence of hepatosplenomegaly or lymphadenopathy.

Paracentesis drained 9.5 L of milky fluid with total protein content 38 g/L, glucose 6.2 mmol/L, cholesterol 2.68 mmol/L and triglycerides 8.45 mmol/L; cytological examination found no cells.

Abdominal CT, repeated paracentesis, hepatitis B and C serology, 1-step tuberculosis test, radionucleotide lymphangiography, abdominal MRI, colonoscopy and upper gastrointestinal endoscopy all failed to yield any etiology for the ascites.

Laparotomy to search for an underlying malignancy found multiple fluctuant, 0.3–1.4 cm cream-coloured cysts studding the surface of the patient's small bowel. They drained chylous fluid when incised. A segment 75–90 cm long of convoluted, indurated small bowel was resected and a small-bowel anastomosis performed. The right-hemipelvis transection line felt clean.

Histopathological analysis revealed multiple foci of high-grade angiosarcoma in the small bowel and mesentery with associated lymphatic obstruction corresponding to the cystic lesions identified grossly. Tumour was identified microscopically at both proximal and distal resection margins.

After surgery the patient refused further treatment. He was managed with palliative care and died 4 months after surgery.

In the Western world, chylous ascites

results most often from malignancy. The proposed pathophysiological mechanism¹ is obstruction of lymphatic flow between the bowel and cisterna chyli by a tumour, with resultant serosal lymphatic dilatation and leakage of lymph into the peritoneal cavity. Radiation therapy to the abdomen can cause chylous ascites from radiationinduced fibrosis and obstruction of lymphatic vessels in the small bowel and mesentery; it occurs, on average, a year after radiation therapy,² and was therefore an unlikely cause in this case. Other causes of chylous ascites, including congenital abnormalities, filiarial parasites, tuberculosis, liver cirrhosis, trauma and nephrotic syndrome, were ruled out by clinical history, biochemical and imaging studies.

After exploratory laparotomy, smallbowel angiosarcoma was deemed to be the likely cause of the ascites. Angiosarcomas, as a group, are uncommon. Primary small-bowel angiosarcomas are even rarer.³ In this case, we suspect that the

From the *Faculty of Medicine, †Department of Pathology and ‡Division of General Surgery, University of Western Ontario, London, Ont.

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Correspondence to: Dr. Brian M. Taylor, Division of General Surgery, 339 Windermere Rd., London ON N6A 5A5; fax 519 663-3068





FIG. 1. Well-differentiated area of angiosarcoma with numerous ecstatic, irregular blood vessels lined by enlarged endothelial cells with prominent nucleoli. The small normal arteriole offers comparison.

FIG. 2. Poorly differentiated area of angiosarcoma composed of solid sheets of spindled cells with enlarged nuclei containing coarse chromatin, prominent nucleoli and mitotic figures. Numerous extravagated red blood cells are present; bloodvessel formation is rudimentary.

angiosarcoma resulted from radiation to the small bowel. Long-term follow-up of patients who have had radiation therapy has shown an overall risk for postirradiation sarcoma development of between 0.03% and 0.8%, of which one-third are angiosarcomas.4 Criteria for diagnosing postirradiation sarcoma proposed in 1948 by Cahan and colleagues5 include no evidence either clinically or microscopically of the antecedent malignant lesion, development of the malignancy in the radiation field, a sufficiently long latency between irradiation and the second malignancy, and histological confirmation of the sarcoma. This patient fulfilled these criteria.

This case presented a challenge in

finding an etiology. It serves as a reminder that malignancy is the most common cause of chylous ascites in the western world and should be suspected even when no radiologic or clinical evidence of an underlying malignancy is obvious. In this situation, exploratory laparotomy can be a useful diagnostic tool.

Competing interests: None declared.

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