Chronic mesenteric artery insufficiency produces hyperplasia of the intestine – case report

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Chronic ischemia produces corresponding tissue hypoplasia. However, the present paper reports a distinctive hypertrophy of the intestine due to chronic mesenteric artery insufficiency. Two patients with chronic mesenteric artery insufficiency confirmed by angiography are presented. The first patient demonstrated marked hypertrophy of the intestine, while the second patient was remarkable for pronounced hypoplasia. During the surgery of the first patient, an ileum approximately 60 cm long with a sausage-like consistency and cyanosis was identified proximal to the cecum; wall-attached thromboembolism

material and the hypertrophic segment of ileum were removed. In the other patient, the intestine wall was paper thin; after the aortomesenteric bypass, the intestine wall grew thicker. Postoperative recovery of both patients was uneventful. The commonly observed situation after a longstanding hypoxic insult in a setting of chronic mesenteric ischemia is that the target tissue will, at least, develop slight hypoplasia. However, the cases presented here had either pronounced hyperplasia or severe hypoplasia. The authors find this observation so interesting that they report it in the hope that an explanation for this conundrum may come forth.

Key Words: Embolectomy; Hypotrophy; Hypoxia; Intestinal ischemia; Mesenteric ischemia

Acute mesenteric ischemia is a disease associated with high mortality. Without timely and proper intervention many patients die. Those who survive often develop chronic symptoms, usually with a triad consisting of postprandial pain, small meals and weight loss. Timely revascularization of the superior mesenteric artery (SMA) is likely to correct the fundamental pathology of most cases of acute ischemia, but is rarely effective in chronic ischemia. In the chronic state, the common intestinal presentation is atrophy. However, we have observed another completely different pathological picture – distinctive hypertrophy of the intestine. We find this observation so interesting that we report it in the hope that an explanation for this conundrum may come forth.

CASE PRESENTATION

Case 1

A 65-year-old man was admitted to the Xuan Wu Hospital (Beijing, China) due to severe acute abdominal pain and nausea without vomiting for three days. On examination, the abdomen was soft and bowel sounds were present. The white blood cell count was $11,200 \times 10^9/L$, lactate dehydrogenase 433 IU/L and a plain abdomen x-ray revealed intestinal distension in the right one-half of the abdomen. Stool was positive for occult blood. He did not have a history of intermittent abdominal pain, diarrhea, constipation, weight loss or diabetes. He had a history of atrial fibrillation for eight years. Selective superior mesenteric arteriography showed a filling defect at the root of the SMA with poor blood flow and an occlusion at the distal ileocolic artery. Small collaterals between the common hepatic artery and the SMA could be identified (Figure 1). A diagnosis of acute mesenteric embolism was made before the angiography was performed by the first author; however, before surgical SMA embolectomy, a laparoscopy was performed by the surgeon in charge that showed no bowel necrosis. Thus, it was decided to manage the patient conservatively with

Figure 1) Superior mesenteric arteriography showing a filling defect at the root of the superior mesenteric artery (SMA) and an occlusion of the distal ileocolic artery (arrows). Collaterals between the hepatic artery and SMA can be seen

250,000 IU of urokinase perfusion through the angiographic catheter that had been inserted into the SMA, after which the catheter was withdrawn. He was discharged on the 10th day after admission.

Six days later, he was readmitted having suffered intolerable abdominal pain for nearly 16 h. He was febrile ($38.1^{\circ}C$) and an x-ray of the abdomen showed multiple air fluid levels. Intestinal

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Figure 2) Intestinal wall hypertrophy is distinctive, with involvement of the adjacent mesentery; the lumen of the intestine is remarkably narrowed

obstruction was diagnosed and conservative treatment was accordingly performed. The symptoms were then relieved, but soon recurred, presenting a wax and wane pattern. The patient suffered postmeal pains, could only eat small meals, had repeated intestinal obstructions and was therefore kept in the hospital for a prolonged period. After the patient lost 13 kg, he was again referred to the first author, and this time a diagnosis of chronic mesenteric insufficiency was immediately made. On May 27, 2004, more than three months after the first occurrence of his disorder, the patient underwent surgery. During surgery, an embolectomy of the SMA was performed; the thromboembolism material with partial fibrosis attached to the wall of the SMA was removed. A section of the ileum approximately 50 cm to 60 cm long, with smooth serosa, a sausage-like consistency and cyanosis, was found approximately 50 cm proximal to the cecum, and was removed with end-to-end anastomosis. The proximal intestine was slightly dilated, but no other intestinal, mesenteric or peritoneal lesions were found. The removed ileum was markedly thickened, even somewhat solid, remarkably hypertrophied without irregular appearance but with a much narrowed lumen (Figure 2), which explains why repeated intestinal obstructions occurred.

After the histopathological examination of the resected bowel sample, the pathological report stated that the submucosa and serosa layers were unusually thick, with fibrous proliferation and some cell infiltration (mainly lymphocytes and plasma cells) (Figure 3).

The patient had an uneventful recovery and was discharged after 10 days. The patient has been followed for almost three years, and has been completely free of symptoms. He regained 13 kg within three months following the surgery, and has not reported any further discomfort.

Case 2

In another previously reported case (2), a 69-year-old woman repeatedly suffered from abdominal pain and lower gastrointestinal bleeding for 1.5 years. She was anemic (hemoglobin 40 g/L) and had lost 8 kg. Angiography showed an occlusion of the SMA and formation of a huge Riolo's arch. During surgery, the intestinal wall was found to be paper thin in some areas; at these thin sections of the intestinal wall the light from the colonoscopic illumination of the lumen could be seen clearly. After an aorto-SMA bypass, the SMA pulsation could be seen and felt immediately. Postoperative recovery was uneventful. She became asymptomatic, gained 5 kg and her hemoglobin rose by 10 g/L four weeks after the surgery.

Figure 3) Marked widening of the submucosa layer (left) and the adventitia layer (right) is obvious, with fibrous tissue proliferation and some infiltration of mainly lymphocytes and plasma cells ($\times 10$; Pathology No 156108 of Xuan Wu Hospital, Capital University of Medical Science, Beijing, China)

DISCUSSION

The clinical presentation of acute abdominal pain and non-specific findings may be confusing, and a diagnosis of intestinal ischemia depends heavily on the ability of the clinician to recognize the spectrum of symptoms. However, when common clinical conditions are excluded, the possibility of mesenteric vascular disease should be aggressively pursued. Traditionally, conventional angiography has been regarded as the gold standard among imaging methods. Approximately 90% of patients with acute mesenteric ischemia who underwent angiography before the onset of peritoneal signs survived, which demonstrates the value of angiography and early diagnosis. One advantage of angiography is the option of concomitant endovascular treatment. Multiple therapeutic approaches are available for intestinal ischemia, depending on the acuity and extent of disease. Embolectomy without removal of intestine would be an early treatment. Embolectomy and resection of an infarcted bowel can also be accomplished during surgery for late-stage cases, although this may result in short bowel syndrome.

The various types of vessels involved, the location of the affected bowel and different levels of acuity of illness all result in multiple possible presentations. The detection of this serious condition can therefore be a diagnostic and therapeutic dilemma (1).

The completely different presentation seen in the two cases reported here, in which one patient had intestinal hypoplasia and the other had hyperplasia, is rather perplexing. Could this be related to the rapidity of onset, duration and degree of ischemia? Or could it be that different layers or parts of layers of the intestine behave differently in response to ischemic insult?

Most emboli originate in the heart and are potentiated by cardiac arrhythmias or depressed systolic function due to ischemic heart disease. In our cases, unnecessary delay increased the morbidity of the first patient, and both patients went through unnecessary suffering, although they were fortunate to have uneventful recoveries after obviously delayed surgeries.

Mesenteric ischemia most often results from embolization or thrombosis of the SMA, and less commonly, venous occlusion or nonocclusive process (2). Embolization of the SMA accounts for nearly 50% of cases, while thrombosis is responsible for another 25% of cases (3,4). In our cases, atrial emboli may have been the underlying pathology.

Conservative treatment with thrombolytic therapy did not help our first patient because urokinase was used selectively via the SMA. In fact, this unnecessary delay could have jeopardized the patient's chances of survival. Generally, as in our two cases, surgical exploration is not emergently performed to restore intestinal arterial flow or to resect irreparably damaged bowels. In fact, if surgery is performed at the acute stage, only embolectomy can be applied. The very thin ischemic hypotrophied bowel in the second case contrasted with the first case, in which the ischemic bowel segment was remarkably thickened or hypertrophied with smooth and normal serosa, and other lesions were completely absent from the abdomen.

We do not understand why the same etiology in these two cases of mesenteric ischemia should lead to extremely different pathological processes. We speculate that a mechanism exists

that leads to a different response to longstanding hypoxia in different types of tissue – that is, a fading or degeneration of functional or essential tissue such as smooth muscle, as seen in the second case, in contrast with a remarkable thickening of nonessential tissue, such as fibroblast or connective tissue, as seen in the first case.

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