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Emergency surgery for hemorrhagic shock caused by a gastrointestinal stromal tumor of the ileum: A case report

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Abstract. In this report, a case of hemorrhagic shock caused by a gastrointestinal stromal tumor (GIST) of the ileum, which was successfully treated by emergency surgery, is presented. A 67-year-old male patient presented to the Ibaraki Medical Center, Tokyo Medical University (Ami, Japan) in July 18, 2014, with dizziness and blood in the stool. Upper endoscopy and colonoscopy failed to reveal the source of the hemorrhage, although abdominal contrast-enhanced computed tomography revealed extravasation of the contrast medium into the small intestine. The patient developed hemorrhagic shock; thus, double-balloon enteroscopy (DBE) was performed, which revealed a Meckel's diverticulum and a submucosal tumor with excessive bleeding at 60 and 100 cm proximal to the ileocecal valve, respectively. Subsequent emergency partial resection of the ileum, including the tumor and the Meckel's diverticulum, was performed in July 20, 2014. Histological examination of the excised tumor revealed proliferation of spindle-shaped cells, and immunohistochemical staining of the tumor was positive for CD34, KIT and α-smooth muscle actin, but negative for S-100 protein. These immunohistological results supported the diagnosis of GIST of the ileum. The patient had an uneventful recovery and has been monitored at our outpatient clinic for 14 months after surgery. This case demonstrated the efficacy of DBE for the diagnosis of small intestinal bleeding, and immediate emergency surgery should be considered for cases of small intestinal GISTs with excessive bleeding.

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Key words: gastrointestinal stromal tumor, emergency surgery, hemorrhagic shock, gastrointestinal bleeding, double-balloon enteroscopy

Introduction

Gastrointestinal stromal tumors (GISTs) are rare tumors that may arise from any site of the GI tract and are generally associated with abdominal pain, GI bleeding, or a palpable mass. However, a small intestinal GIST rarely causes hemorrhagic shock. We herein report a case of hemorrhagic shock with excessive bleeding caused by an ileal GIST that was managed by emergency surgery. The patient provided written informed consent for the publication of this case report.

Case report

A 67-year-old male patient presented to the Department of Gastroenterology of the Ibaraki Medical Center, Tokyo Medical University (Ami, Japan) in July 18, 2014, with dizziness and blood in the stool. The patient's medical history included treatment for hypertension by a local physician. The findings of the subsequent physical examination were unremarkable, except for low blood pressure (97/60 mmHg) and mild pallor of the palpebral conjunctiva. Laboratory data revealed mild anemia (hemoglobin, 10.2 g/dl) and increased blood urea nitrogen (34.1 mg/dl). Upper endoscopy revealed no hemorrhagic lesion of the duodenum, stomach, or esophagus. Colonoscopy revealed fresh blood with clotting discharged from the proximal side of the ileocecal valve; no hemorrhagic lesion of the colon or rectum was identified. Abdominal contrast-enhanced computed tomography (CT) revealed extravasation of the contrast medium into the small intestine (Fig. 1). The intestinal bleeding continued, and the patient eventually developed hemorrhagic shock (blood pressure, 76/42 mmHg; hemoglobin 4.5 g/dl). Hence, 22 units of red blood cells stored in mannitol-adenine-phosphate and 10 units of fresh-frozen plasma were administered. Subsequent double-balloon enteroscopy (DBE) revealed a Meckel's diverticulum and a submucosal tumor with excessive bleeding at 60 and 100 cm proximal to the ileocecal valve, respectively (Fig. 2). Endoscopic hemostasis was not possible, as the enteroscope could not approach the tumor. Following DBE marking near the tumor, emergency partial resection of the ileum, including the tumor and the Meckel's diverticulum, was performed 2 h from developing hemorrhagic shock in July 20, 2014. There were no signs of lymphadenopathy, peritoneal dissemination, or liver

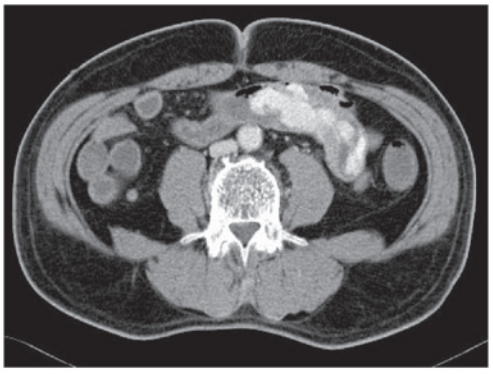

Figure 1. Abdominal contrast-enhanced computed tomography revealed extravasation of contrast medium into the small intestine.

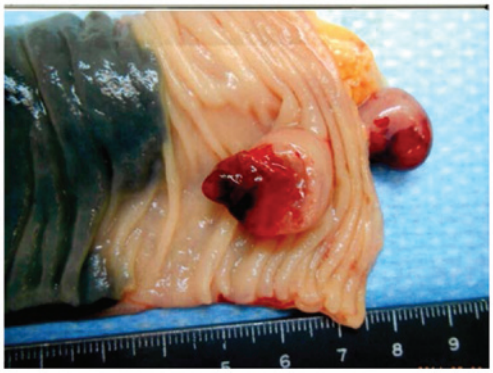

Figure 3. Macroscopic examination of the excised specimen. The excised tumor measured 1.3x0.8 cm and exhibited ulcerative mucosal change.

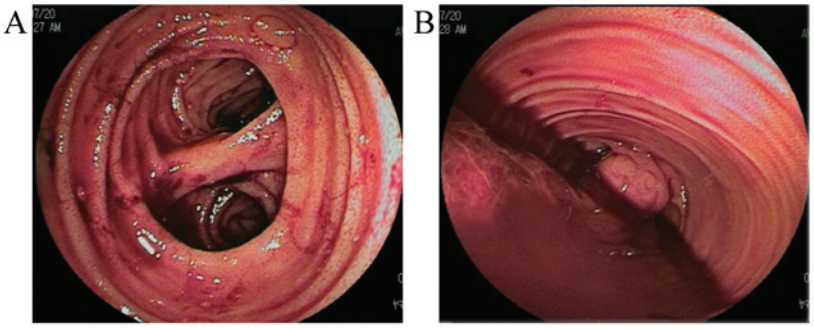

Figure 2. Double-balloon enteroscopy revealed (A) a Meckel's diverticulum and (B) a submucosal tumor with excessive bleeding at 60 and 100 cm proximal to the ileocecal valve, respectively.

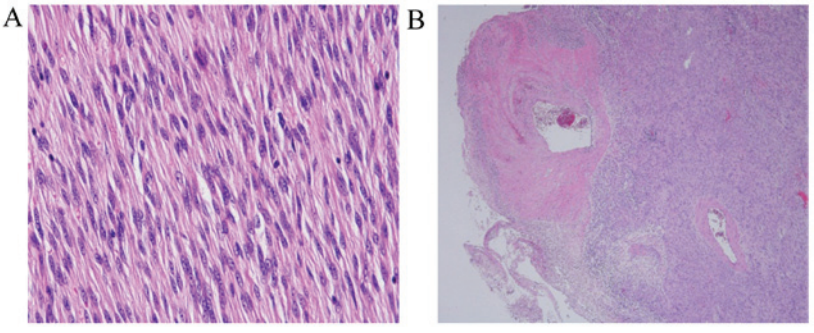

Figure 4. Histological analysis of the excised specimen. (A) Spindle-shaped cell proliferation (hematoxylin and eosin staining; magnification, x400). (B) Ruptured artery on the tumor surface (hematoxylin and eosin staining; magnification, x40).

metastasis. The excised tumor (1.3x0.8 cm) exhibited ulcerative mucosal changes (Fig. 3). Sectioning of the tumor revealed a solid and grayish white tissue. Histological examination of the excised tumor revealed proliferation of spindle-shaped cells in the submucosa to the subserosa of the ileum and a ruptured intratumoral artery at the tumor surface (Fig. 4). Immunohistochemical staining of the tumor was positive for CD34, KIT and α-smooth muscle actin, but negative for S-100 protein (Fig. 5). The MIB-1 labeling index using Ki-67 was 1.0-5.0%. The tumor size and the immunohistological findings supported the diagnosis of a low-risk GIST of the ileum (1). The patient had an uneventful recovery, was discharged on postoperative day 14 and has been monitored at our outpatient clinic for 14 months after the surgery.

Discussion

GISTs constitute ~0.2-0.5% of all GI tract tumors, with ~70% of the cases occurring in the stomach, 20% in the small intestine, and <10% in the colon, rectum, or esophagus (2,3). The clinical presentation of GISTs is variable and the most frequent symptoms are abdominal pain, GI bleeding, or a palpable mass. In GISTs arising in any site, including the stomach, duodenum, small intestine, colon and rectum, Sorour *et al* (4) reported that GI bleeding was the most serious symptom. However, with respect to small intestinal GISTs, abdominal pain (35.5%) is the most frequent symptom, while hemorrhagic shock (6.4%) is relatively rare (5). In the present case, the patient developed hemorrhagic shock; thus,

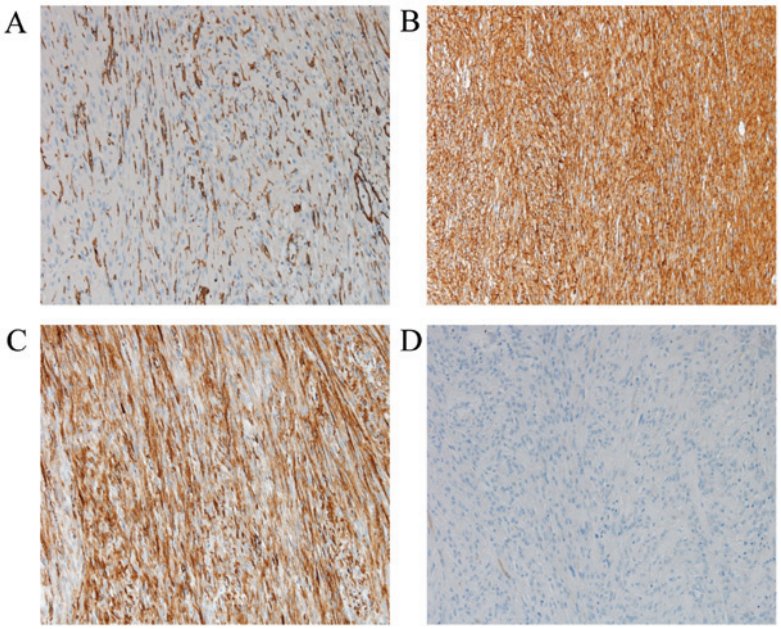

Figure 5. Immunohistochemical staining of tumor spindle cells (magnification, x200) revealed positive staining for (A) CD34, (B) KIT and (C) α-smooth muscle actin and (D) negative staining for S-100 protein.

upper endoscopy, colonoscopy, abdominal contrast-enhanced CT, and DBE were performed to investigate the source of the hemorrhage. Abdominal CT plays an important role in the diagnosis of small intestinal GISTs, particularly contrast-enhanced CT in cases with excessive bleeding (6,7). In the present case, abdominal contrast-enhanced CT revealed that the hemorrhagic source was the small intestine, although no tumor was detected. DBE is an efficacious procedure for the diagnosis and treatment of small intestinal disease. The most common clinical indications of GISTs include obscure bleeding, abdominal pain, anemia, chronic diarrhea and inflammatory bowel disease (8). Robles *et al* (9) reported that the histological detection rate of GISTs by DBE biopsy was 71.4%. The first-line treatment for small intestinal GISTs with excessive bleeding remains debatable (10,11). The endoscopic treatment by DBE is very limited in massive GIST bleeding, but it may be possible, delaying or averting emergency surgery (9). In the present case, DBE was unable to avert emergency surgery, but located the level of the bleeding and guided resection. Interventional digital subtraction angiography has been reported to be effective for GISTs with bleeding (12). Basile *et al* (13) reported that interventional radiological procedures for the detection of bleeding GISTs of the small intestine were superior to other diagnostic approaches. In the present case, we performed emergency partial resection of the ileum, including the GIST, as i) the patient had developed hemorrhagic shock and ii) an interventional radiology specialist was unable to urgently respond in our hospital.

There are certain differences in the follow-up examinations after surgery for GISTs between the guidelines of the National Comprehensive Cancer Network (NCCN) and the Japanese Society of Clinical Oncology (JSCO) (14,15). After surgical resection of all tumors, the NCCN guidelines recommend abdominal and pelvic CT imaging every 3-6 months for 3-5 years. However, the JSCO guidelines recommend CT testing every 6-12 months for GISTs with low or very low risk for recurrence, and every 4-6 months for GISTs with a high or intermediate risk or clinically malignant. Our patient has been monitored by CT scans every 6 months in our outpatient clinic, as recommended by the JSCO guidelines.

In conclusion, DBE is an efficacious procedure for identify the source of bleeding in the small intestine. Moreover, immediate emergency surgery should be considered for cases of small intestinal GISTs with excessive bleeding.

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