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Single Case

Obscure Gastrointestinal Bleeding Due to a Small Intestinal Gastrointestinal Stromal Tumor in a Young Adult

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Keywords

Gastrointestinal stromal tumors · Young adult · Small bowel bleeding · Small bowel tumor

Abstract

The source of most cases of gastrointestinal bleeding is the upper gastrointestinal tract. Since bleeding from the small intestine is very rare and difficult to diagnose, time is required to identify the source. Among small intestine bleeds, vascular abnormalities account for 70–80%, followed by small intestine tumors that account for 5–10%. The reported peak age of the onset of small intestinal tumors is about 50 years. Furthermore, rare small bowel tumors account for only 1–2% of all gastrointestinal tumors. We describe a 29-year-old man who presented with obscure anemia due to gastrointestinal bleeding and underwent laparotomy. Surgical findings revealed a well-circumscribed lesion measuring 45 × 40 mm in the jejunum that initially appeared similar to diverticulosis with an abscess. However, the postoperative pathological diagnosis was a gastrointestinal stromal tumor with extramural growth.

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Introduction

Gastrointestinal (GI) bleeding is a common emergency [1] and bleeding from the upper GI tract and small intestine account for 75% and 2–10% [1] of all GI bleeds [2], respectively. We describe a 29-year-old man who presented with obscure anemia due to small intestine bleeding. Vascular abnormalities account for 70–80%, followed by small intestinal bleeds that account for 5–10%, respectively [3].

Adenocarcinoma is the most common primary malignant tumor of the small bowel (PMTSB) [4, 5]. However, our patient had a gastrointestinal stromal tumor (GIST), which accounts for <1% of all GI neoplasms [6]. The peak age for developing GIST is 50–60 years [6, 7], and thus GIST is rarely the source of small intestinal bleeds. This report describes obscure GI bleeding due to GIST of the jejunum of a young adult.

Case Report

A 29-year-old man presented to our hospital with a 2-week history of gross hematochezia and anemia. He was free of nausea, vomiting and localized abdominal pain. Laboratory findings were as follows (values are shown with range in parentheses): aspartate aminotransferase, 11 (normal, 5–35) U/L; alanine aminotransferase, 11 (5–30) U/L; alkaline phosphatase, 185 (115–359) U/L; γ -glutamyl transferase, 49 (0–50) U/mL; lactate dehydrogenase, 96 (106–211) U/L; albumin, 3.0 (3.7–5.5) g/dL; total bilirubin, 0.26 (0.2–1.0) g/dL; and C-reactive protein, 1.2 (0–0.3) mg/dL. Other than C-reactive protein, almost all blood biochemistry findings were essentially within normal ranges. Hematological findings were as follows (values are shown with range in parentheses): white blood cells, 11,600 (5,000–8,000)/ μ L; red blood cells, 165 (400–530) $\times 10^4$ / μ L; and hemoglobin, 4.5 (14–18) g/dL.

These findings indicated inflammation and anemia. Tumor markers were as follows (values are shown with range in parentheses): CEA <0 (0–5) ng/mL; CA19-9, 3 (0–37) U/mL; and IL-2, 280 (145–519) U/mL. All tumor markers were within normal ranges.

Upper GI endoscopy and total colonoscopy findings were normal. Abdominal computed tomography revealed a well-delineated 45 \times 40-mm mass in the jejunum (Fig. 1). These findings indicated infected diverticulitis such as Meckel's diverticulitis with abscess [8].

The lesion in the jejunum was resected and removal of a possible tumor was assured by lymphadenectomy (Fig. 2). Thereafter, a jejuno-jejunostomy proceeded. The postoperative course was uneventful and the patient was discharged on postoperative day 10.

The resected specimen initially appeared similar to diverticulosis with abscess, so we diagnosed infected diverticulosis (Fig. 3). However, the pathological findings revealed GIST with extramural growth. Histopathological assessment revealed a submucosal tumor comprising proliferative atypical spindle or epithelioid cells arranged in fascicle whorls with a dense acute inflammatory infiltrate, abscess and necrosis. Immunohistochemical findings showed that the atypical cells were positive for c-kit, and some were faintly positive for CD34 and S-100 (Fig. 4). The MIB-1 labeling index was <1%. These features were compatible with those of low-risk GIST [8–10].

Discussion

GI bleeding is a common medical emergency [1, 2]. Accurate, prompt diagnosis of a bleeding source is crucial because such bleeding can result in death. The source of 3–5% of GI bleeds cannot be identified by esophagogastroduodenoscopy and/or colonoscopy, but bleeding from the upper and lower GI tracts accounts for 75 and 15%, respectively, of all GI bleeds [1, 7]. The reported rate of bleeding from the small intestine is 2–10% [1]. Vascular abnormalities and small intestinal tumors account for 70–80% and 5–10%, respectively, of small intestinal bleeds [3] and PMTSB accounts for only 1–2% of all primary GI tumors [11, 12].

Most patients with PMTSB have nonspecific clinical symptoms and signs. The most frequent symptoms are abdominal pain (67.4%), abdominal mass (31.2%) and bowel obstruction (24.1%), followed by hematochezia (21.3%), jaundice (16.3%), and fever (14.2%) [4]. Our patient had only hematochezia.

Adenocarcinoma is the most common type of PMTSB [4, 5]. Leiomyoma and leiomyosarcoma in the small intestine are associated with massive bleeding, whereas lymphoma, GIST and carcinoids are associated with relatively slow bleeding [4, 7]. Our patient had a 2-week history of gross hematochezia and his condition roughly corresponded to GIST.

Mazur et al. [13] originally described GIST in 1983. Less than 1% of all GI neoplasms are GIST [6], and they are defined as specific, generally Kit (CD117)-positive and/or platelet-derived growth factor receptor- α mutation-driven tumors [9, 10, 13]. Among mesenchymal tumors of the GI tract, GIST are the most common, and primary GIST can occur anywhere along the GI tract between the esophagus and the anus. Between 50 and 70% of clinically manifested tumors arise in the stomach, 20–30% are found in the small bowel, 5–15% are located in the large bowel, and <5% are located in the esophagus and elsewhere [14]. The reported peak age of GIST is 50–60 years [6, 7]. Therefore, to initially consider GIST in a differential diagnosis of GI bleeding in a younger patient is unlikely.

Meckel's diverticulum and Crohn's disease are common when considering only GI bleeds in younger patients. Meckel's diverticulum is the most common congenital anomaly of the GI tract, with an incidence of 2–4% in the general population. Meckel's diverticulum occurs twice as often in men than in women, and acute diverticulitis can occur at any age, but the incidence peaks in children [15, 16]. Meckel's diverticulum is always located within 1 m of the oral side of the ileocecal valve and straying ectopic stomach tissue is the key pathological feature in 27% of patients with this disease [16]. The specimen resected from our patient macroscopically resembled diverticulitis, but it was not obtained from a location within 1 m from the oral side of the ileocecal valve. Small intestinal diverticula other than the Meckel type can be differentially diagnosed, but the incidence ranges from only 0.06 to 1.3% [17]. Finally, microscopic assessment ruled out these types of diverticulosis.

Crohn's disease is also common among younger patients. Crohn et al. [18] first described this condition in 1932 as a chronic inflammatory disease of unknown origin that can occur not only in the terminal ileum but anywhere in the GI tract. It commonly affects men and women aged in the early 20s and late teens, respectively [18–20]. Preoperative upper and lower GI endoscopy and pathological findings excluded Crohn's disease as the bleeding source in our patient.

Conclusion

We described hemorrhage due to GIST of the small intestine in a young adult male. The possibility of neoplastic disease must be considered even among young adults who present with GI bleeding.

Statement of Ethics

Written informed consent was obtained from the patient for publication of this paper and any accompanying images. All authors read and approved the final manuscript.

Disclosure Statement

The authors have no conflicts of interest to disclose.

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Fig. 1. Computed tomography findings. White arrow, well-delineated 45 × 40-mm mass in the jejunum.

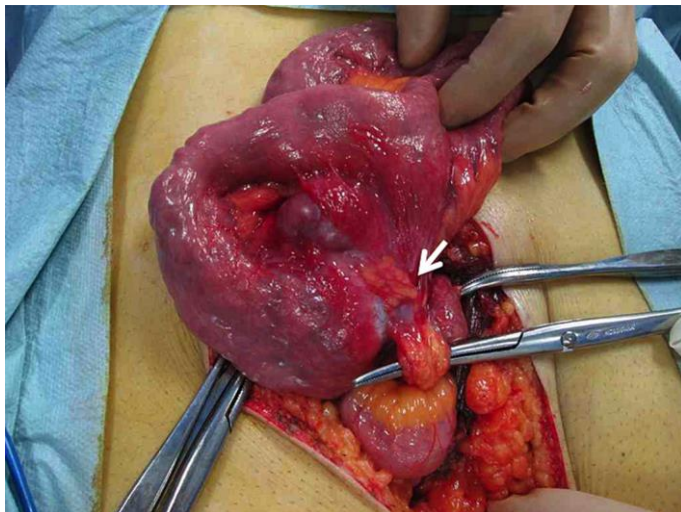


Fig. 2. Intraoperative view. White arrow, well-circumscribed 45 × 40-mm lesion with inflammatory adhesion.



Fig. 3. Resected specimen. **a** Dissected jejunum shows swollen diverticulosis. **b** Cross-sectional slice shows infected diverticulosis with abscess. Pathological findings revealed GIST with extramural growth.

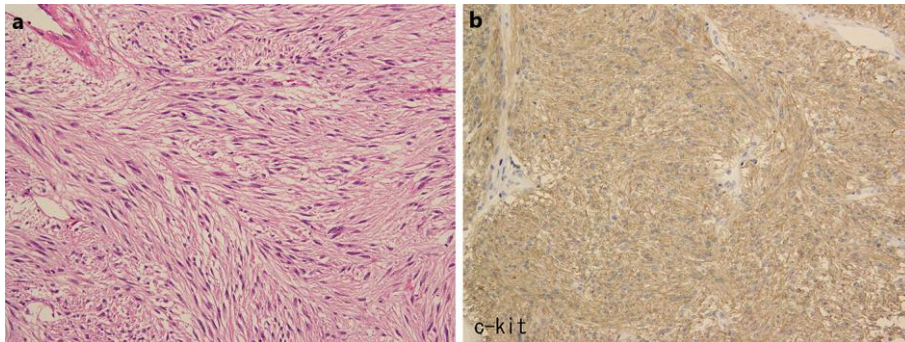


Fig. 4. Pathological examination of the tumor. **a** The tumors consisted of spindle cells (hematoxylin and eosin staining, $\times 100$). **b** Immunohistochemically, the atypical cells positively reacted to c-kit ($\times 100$). MIB-1 labeling index was $< 1\%$.