



Spontaneous Hemoperitoneum Because of a Ruptured GIST without Abdominal Pain

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Abstract

We experienced a rare case of spontaneous hemoperitoneum because of a ruptured malignant gastrointestinal stromal tumor (GIST), with only abdominal bloating and no abdominal pain. GIST is a common mesenchymal tumor of the gastrointestinal tract. Because of the high vascularity of the tumor, GISTs are frequently associated with gastrointestinal bleeding. Most patients present with abdominal pain at the onset of rupture, and the absence of abdominal pain complicated the management of this patient. To confirm or exclude a diagnosis of hemoperitoneum combined with a gastric submucosal tumor and intra-abdominal fluid, careful observation is required.

Keywords: Gastrointestinal stromal tumor; Rupture; Hemoperitoneum; Emergency laparotomy

Background

A gastrointestinal stromal tumor (GIST) is a common mesenchymal tumor of the gastrointestinal tract [1-3]. GISTs are characterized by the over expression of the tyrosine kinase receptor through mutations of the c-kit or PDGFRA genes [4]. Because of its high vascularity, GISTs are frequently associated with gastrointestinal bleeding; however, spontaneous hemoperitoneum because of a ruptured GIST is relatively rare [5,6]. Previous studies reported that most patients with GISTs present with abdominal pain at the onset of rupture [5-7].

Case Presentation

An 83-year-old Asian male presented to our clinic with increasing abdominal bloating without pain. The patient was in his usual good health until 5 days ago when he began to experience unexplained abdominal bloating. He had no past medical history of a serious illness, surgery, or hospitalization.

The patient was alert, with a temperature of 36.3°C, blood pressure of 112/60 mmHg, regular pulse of 84 beats/min, and a normal respiration rate. A physical examination revealed pale palpebral conjunctiva, symmetrical abdominal distention, and a palpable mass in the epigastrium; however, there was no abdominal tenderness. There was no other specific finding on physical examination.

Routine hematological analysis revealed normocytic normochromic anemia with a hemoglobin level of 8.9 g/dL, white blood cell count of 6000 cells/ μ L, and blood platelet count of 185,000/ μ L.

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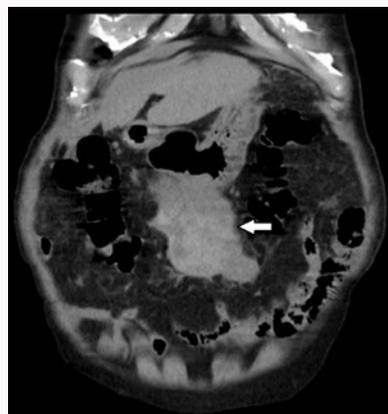


Figure 1: Plain abdominal CT scan revealed a large heterogeneous gastric submucosal mass measuring approximately 70 × 80 mm (white arrow).



Figure 2: Plain abdominal CT revealed a massive accumulation of intra-abdominal fluid in Douglas' pouch.

Blood chemistry tests revealed a mild renal dysfunction with a urea nitrogen level of 40 mg/dL and creatinine level of 1.3 mg/dL. Other blood chemistry parameters were all within normal limits.

Abdominal plain computed tomography (CT) was performed, with no contrast dye because of the renal dysfunction, to evaluate the palpable abdominal mass and bloating, which revealed a large heterogeneous gastric submucosal mass, measuring approximately 70 × 80 mm (Figure 1), with a massive amount of intra-abdominal fluid (Figure 2). Based on the tumor size and imaging pattern, this mass was diagnosed as a malignant GIST. However, it was difficult to identify the causative underlying disease because of the massive amount of intra-abdominal fluid. We suspected that the acute intra-abdominal bleeding was because of a ruptured GIST, although the absence of acute abdominal pain was contradictory to this diagnosis. In addition, we considered a differential diagnosis of cancerous ascites due to a malignant GIST because the patient had not undergone hematological or imaging testing for quite some time, and it was unclear whether the intra-abdominal fluid and anemia were acute or chronic.

At this point, we decided to admit the patient to our clinic for careful observation until the possibility of acute intra-abdominal bleeding was completely excluded. Three hours after the initial examination, a second hematological analysis revealed that the hemoglobin level decreased to 7.1 mg/dL, suggesting acute intra-abdominal bleeding.

Therefore, we decided to perform emergency laparotomy. Intraoperative findings showed a total of 1700 mL of blood in the



Figure 3: The resected tumor was a solid mass with a punctured capsule.

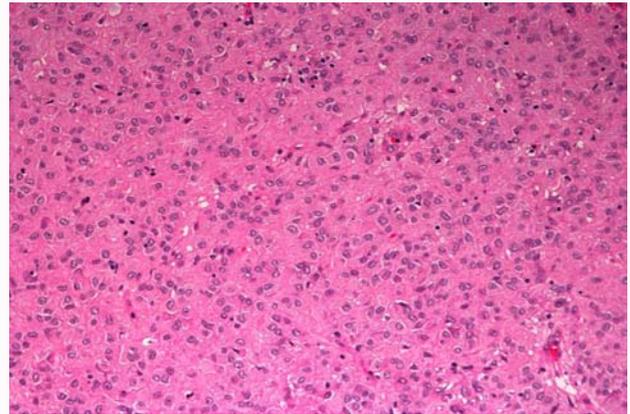


Figure 4: Histological examination showed spindle cells (HE, x200).

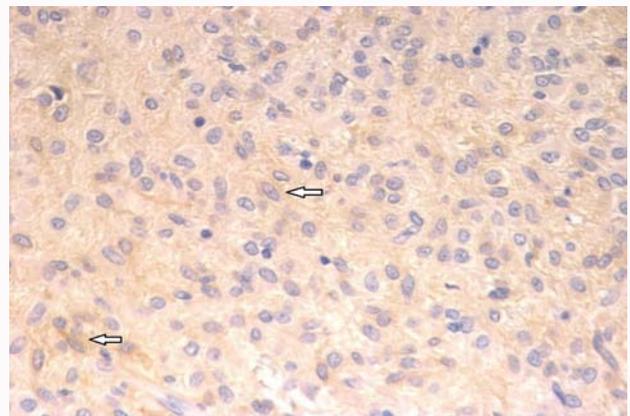


Figure 5: Immunohistological finding was positive for c-kit protein (White arrows) (x400).

abdominal cavity and a 70 × 80-mm, extraluminal, pedunculated, solid tumor that arose from the greater curvature of the gastric body, with no signs of peritoneal dissemination. The peritoneal cavity was lavaged with 7000 mL of saline, and the tumor was completely dissected from the stomach. The capsule of the resected tumor had a puncture (Figure 3) that was sealed with massive blood clots.

Histological examination revealed spindle cells (Figure 4), and immunohistological findings were negative for α -smooth muscle actin, desmin, and S-100 protein but positive for c-kit protein (Figure 5) and CD34.

Based on these findings, the definitive diagnosis was spontaneous hemoperitoneum because of the rupture of a malignant GIST. The patient had an uncomplicated postoperative course and was scheduled for 3 years of adjuvant chemotherapy with imatinib. He has been followed up for 4 months with no evidence of recurrence.

Discussion

We experienced a case of spontaneous non-traumatic hemoperitoneum without abdominal pain because of a ruptured GIST. GIST is a common mesenchymal tumor of the gastrointestinal tract and is most often located in the stomach [1-3]. GISTs are characterized by the over expression of the tyrosine kinase receptor through mutations of c-kit or PDGFRA genes and the expression of the c-kit protein, which is a highly specific marker for the differentiation of GIST from other mesenchymal tumors [4].

Because of the high vascularity of the tumor, GISTs are frequently associated with gastrointestinal bleeding. Although several reports have described spontaneous hemoperitoneum because of the rupture of GIST, it is a relatively rare condition [5,6]. Most patients present with abdominal pain at the onset of rupture [5-7]. In contrast, our patient only presented with abdominal bloating without pain. To the best of our knowledge, this is a very rare case. However, the reason why the bleeding was not persistent but rather intermittent remains unclear. Intraoperative findings revealed that the capsule of the resected tumor had a puncture that was sealed with a massive hematoma, suggesting that the intermittent bleeding through the punctured capsule was the cause of the hemoperitoneum, while hemostatic properties allowed the clotting blood to seal the puncture.

The absence of abdominal pain made it difficult to arrive at a differential diagnosis. CT revealed a large gastric mass and a massive amount of intra-abdominal fluid. The combination of imaging and clinical findings can mislead physicians to an incorrect diagnosis such as an advanced gastric tumor with cancerous peritonitis. The treatment for an advanced gastric tumor with cancerous peritonitis is commonly chemotherapy or/and palliative care, while emergency laparotomy is rarely performed [3,8].

Several steps are considered to arrive at a differential diagnosis. First, the existence of anemia can be helpful in the differential diagnosis. If a previous hematological study reveals a normal hemoglobin level that drastically decreases over time, acute intra-abdominal bleeding is strongly suggested [9]. However, one-time laboratory findings of anemia are insufficient to indicate acute blood loss because an advanced gastric tumor is often concomitant with chronic anemia due to gastrointestinal hemorrhage [3,10]. Laboratory tests were not performed for quite some time in our patient; therefore, it was unclear whether the anemia was acute or chronic. Second, previous imaging findings can aid in a differential diagnosis. If recent imaging findings show a gastric tumor without hemoperitoneum, the presence of intra-abdominal fluid indicates acute accumulation of the intra-abdominal fluids such as blood. Imaging exams had never been performed for this patient; thus, it could not be determined whether the accumulating intra-abdominal fluid was acute or chronic. Third, an abdominal tap can be useful. Bloody ascites detected using an abdominal tap is suggestive of intra-abdominal bleeding. However, a definitive diagnosis of hemoperitoneum should not solely be based on using an abdominal tap because cancerous ascites is frequently bloody [11]. Furthermore, some specific findings of contrast-enhanced CT, such as extravasation and sentinel clot sign, suggest a diagnosis of tumor-associated hemorrhage [12]. However, such findings cannot be detected when bleeding is minimal, intermittent, or has been terminated [12]. In this case, contrast dye could not be used because of a renal dysfunction; thus, it was not possible to arrive at a definitive diagnosis of hemoperitoneum based on CT findings.

Although we suspected that the gastric mass was a malignant gastric submucosal tumor based on CT findings, we could not determine whether the intra-abdominal fluid was because of chronic cancerous ascites or acute intra-abdominal bleeding. We carefully observed the patient and rechecked the hemoglobin level 3 h after admission, which revealed a decrease in the hemoglobin level, indicating acute blood loss because of hemoperitoneum. Finally, emergency laparotomy was performed, and a definitive diagnosis of spontaneous hemoperitoneum because of a ruptured GIST was made.

The choice for operative or nonoperative management of

hemoperitoneum is dependent on the causative disease or general condition of the patient [13]. Angiographic embolization is a typical nonoperative management modality and is often selected for hepatic or splenic injury [13]. In contrast, the management of hemoperitoneum because of a ruptured gastric tumor is usually emergency laparotomy to control bleeding by the resection of the tumor and lavage of the peritoneal cavity to decrease the dissemination of cancer cells from the ruptured tumor and to arrive at a definitive pathological diagnosis [5,7].

Conclusion

We experienced a case of spontaneous hemoperitoneum without abdominal pain because of a ruptured malignant GIST. To confirm or exclude the diagnosis of hemoperitoneum combined with a gastric submucosal tumor and accumulation of intra-abdominal fluid, careful observation is required, and blood tests and imaging examinations should be rechecked to confirm the diagnosis.

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