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Case report

Spontaneous hemoperitoneum as a rare presentation of gastric lesions: Two case reports

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ABSTRACT

Introduction: Spontaneous hemoperitoneum is a rare but life-threatening condition. Clinical presentation is usually nonspecific. The aim of this report is to document a rare clinical presentation of two different intramural stomach tumors.

Clinical cases: A 40-year-old patient with 24 h epigastric pain was admitted to the emergency, pale, with signs of peritoneal irritation. Computed tomography showed an hemoperitoneum with active bleeding in the posterior wall of the stomach. A wedge resection was performed. Histological report revealed a plexiform fibromyxoma (PF). The second case presents a 79-year-old patient with 24 h abdominal pain, fullness and dizziness, pale and with signs of peritoneal irritation. A voluminous exophytic lesion on gastric wall with active bleeding was diagnosed. Wedge resection was performed and histological report demonstrated a gastrointestinal stromal tumor (GIST).

Discussion: Imaging plays a role in the diagnosis of spontaneous hemoperitoneum, in elucidating a cause and detecting active hemorrhage. Tumor hemorrhage may be the first presentation of an underlying mass. The presence of a bleeding gastric mass of uncertain nature may result in a challenging situation for the surgeon, who is forced to perform a gastric resection without knowing the exact nature of the tumor and hence the extent of gastric resection required. To our knowledge, our case is the first of PF presenting as hemoperitoneum. Hemoperitoneum is rare as first presentation of GIST, with few cases reported in literature.

Conclusion: We report two extremely rare cases of spontaneous hemoperitoneum as first presentation of gastric tumor. For the diagnosis a high level of suspicion is required.

1. Introduction

Spontaneous hemoperitoneum (SH) is a rare but life-threatening condition that is defined as blood within the peritoneal cavity of non-traumatic and non-iatrogenic etiology. Common sources of spontaneous abdominal hemorrhage are visceral, gynecologic, coagulopathy-related, and vascular. The clinical presentation is usually nonspecific; and frequently the diagnosis is made on the basis of radiologic findings [1].

Intramural stomach tumors can be asymptomatic or can present with an abdominal mass or pain. The overlying mucosa can be focally ulcerated due to pressure necrosis from the intramural mass, resulting in signs of gastrointestinal bleeding, such as hematemesis, melena, and iron-deficiency anemia. Larger masses may result in bowel obstruction or intussusception [2]. Hemoperitoneum is a rare complication of gastric lesions. Due to its rarity, the diagnosis of this lesions as the cause of hemoperitoneum may be challenging [3].

The aim of this report is to document a rare clinical presentation of spontaneous hemoperitoneum due to different intramural gastric tumors. This case reports have been elaborated in accordance with the SCARE criteria [4].

2. Clinical cases

The first case is a 40-year-old male patient, admitted to the

Abbreviations: PF, plexiform fibromyxoma; GIST, gastrointestinal stromal tumor; SH, spontaneous hemoperitoneum; CT, computed tomography; MRI, magnetic resonance imaging; EUS-FNA, endoscopic ultrasound-guided fine-needle aspiration biopsy; PDGFR, platelet-derived growth factor receptor.

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emergency with a 24 h of evolution epigastric pain. The patient had no prior medical history. In the objective examination the patient was pale, dehydrated, hemodynamically stable and with signs of peritoneal irritation. Hemoglobin of 15,5 g/dl. The ultrasound detected a moderate intra-abdominal fluid, an abdominal computed tomography (CT) showed an hemoperitoneum with active bleeding in the posterior wall of the stomach (Fig. 1). After clinical case evaluation and management options discussion with the patient, informed consent was obtained for an exploratory laparotomy. A hemoperitoneum originated in a lesion of the posterior gastric wall was found. An intra-operative upper endoscopy was performed, showing an intact mucosa. A wedge resection was performed (Fig. 2). The histology showed a PF with R1 resection. In the early post-operative period, the patient had clinical symptoms of outlet obstruction: nausea and vomiting after meals. The endoscopy showed torsion of the distal stomach with stenosis that could be franked by the endoscope. An antrectomy with a roux-y-anastomosis was performed, with complete resolution of obstructive symptoms. The histology showed no lesions on the surgical piece.

The second case is a 79-year-old male patient who was admitted to the emergency with abdominal pain, fullness and dizziness, with 24 h evolution. The patient had a medical history of type 2 diabetes, dyslipidemia, hypertension, atrial fibrillation hypocoagulated with edoxaban, and was submitted to elective laparoscopic cholecystectomy. Patient was pale, hemodynamically stable and with signs of peritoneal irritation. Arterial blood gas sample showed lactates of 2.8 and hemoglobin of 7,6 g/dl. CT showed voluminous exophytic lesion on the posterior surface of the stomach body with active bleeding and hemoperitoneum (Fig. 3). After hypocoagulation reversal an atypical gastrectomy was performed (lesion of 10,1 × 9,1 × 8,9 cm with active bleeding who was attached to the posterior wall of the stomach by a pedicle) (Fig. 4). The histology demonstrated an GIST, Fletcher intermediate risk, Miettinnen low risk, R0. In the post-operative period, the patient needed a blood transfusion, although there was no evidence of blood loss. The patient was discharged on the eighth post-operative day.

Both patients denied any abdominal trauma in the days before.

3. Discussion

SH is a rare condition, so diagnosis is almost always unsuspected until the time of imaging, which is undertaken in patients who present with acute abdominal pain and/or distention and anemia [5].

Imaging plays a role, not only in the diagnosis of spontaneous hemoperitoneum, but also in elucidating a cause. It may be idiopathic or related to spontaneous rupture of either a known or an unknown pathology [6]. CT is the most commonly used modality in patients with acute abdominal pain. CT can point to a specific organ as the source of the bleeding, detect active hemorrhage and provide information on how long ago the hemorrhagic episode took place [5].

Like the cases presented, tumor hemorrhage may be the first presentation of an underlying mass. The mechanism of spontaneous tumor hemorrhage is not entirely known and is likely multifactorial. Rapid tumor growth with necrosis may make masses prone to spontaneous hemorrhage. In the cases of an underlying malignancy, hemorrhage occurs in part due to associated coagulopathy, neovascularization from released growth factors, metastatic marrow invasion, and thrombotic microangiopathy [7].

The presence of a bleeding, ruptured gastric mass of uncertain nature may result in a challenging situation for the surgeon, who is forced to perform a gastric resection without knowing the exact nature of the tumor and hence the entity of gastric resection required, or whether a lymphadenectomy is needed [3].

The case reports presented above are of gastric lesions, with different histologic findings. We will discuss the two entities found in the pathological anatomy.

PF is a very rare mesenchymal neoplasm of the stomach that was first described in 2007 and was officially recognized as a subtype of gastric mesenchymal neoplasm by World Health Organization in 2010. To date, a total of 130 PF cases have been reported in the English literature [8].

The most common site for PF is the gastric antrum [9]. The clinical presentation is usually nonspecific. Most common symptoms are abdominal pain/discomfort, fullness, nausea and vomiting, followed by symptoms of blood loss (bleeding, syncope, anemia). To our knowledge, it is the first case of PF presenting as hemoperitoneum [8].

Endoscopic and radiologic findings are nonspecific and most cases were misdiagnosed as GISTs [10]. Radiological and endoscopic findings reveal a submucosal tumor-like elevated lesion that is often ulcerated and endoscopic ultrasonography demonstrate a multinodular mass. CT images demonstrate areas of low attenuation owing to presence of mixoid tissue, interspersed with foci of vascularity [9].

The treatment of choice is surgical resection, although there have been reports of endoscopic resection. The surgical technique depends on the size and location of the tumor. Most studies in the literature report treatment by distal gastrectomy [10]. To date, no malignant progression, local recurrence, or metastasis was reported [8].

PF demonstrates a multinodular, myxoid or gelatinous appearance with or without hemorrhage. Histologically, PF shows a multinodular, plexiform growth pattern with a proliferation of ovoid to spindle cells within myxoid stroma, and an increased vascularity. Immunohistochemically, the tumor cells are positive for smooth muscle actin and vimentin, and can show focal or partial staining for CD10, desmin and caldesmon [8].

Despite their rarity, GISTs are the most common gastrointestinal

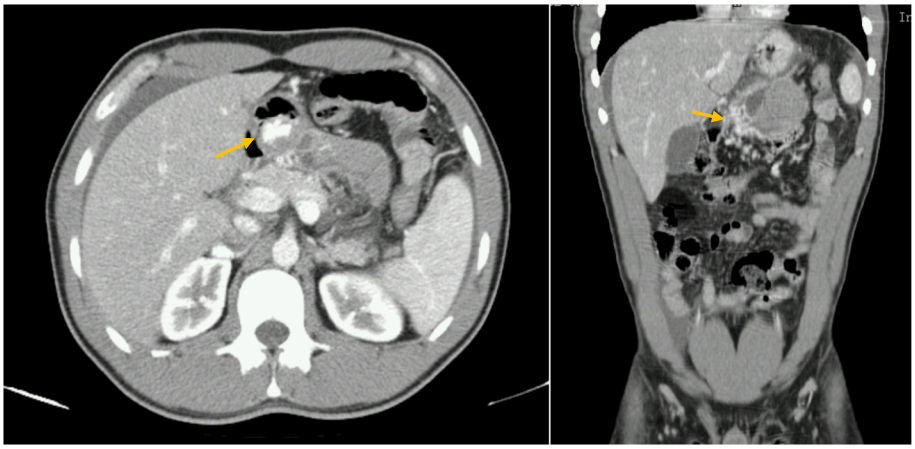

Fig. 1. Computed tomography image on admission: hemoperitoneum with active bleeding (arrows) in the posterior wall of the duodenum/stomach.

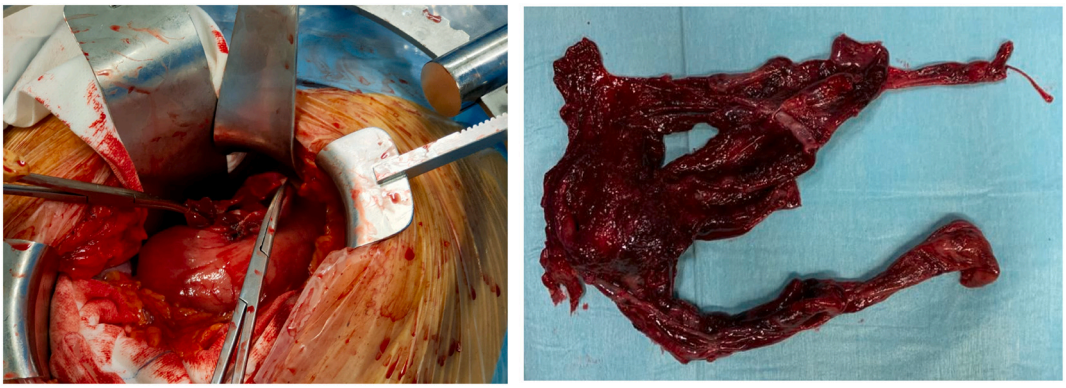

Fig. 2. Exploratory laparotomy: a hemoperitoneum originated in a lesion Ruptured cystic lesion of the posterior wall of the stomach. An atypical gastric resection was performed.

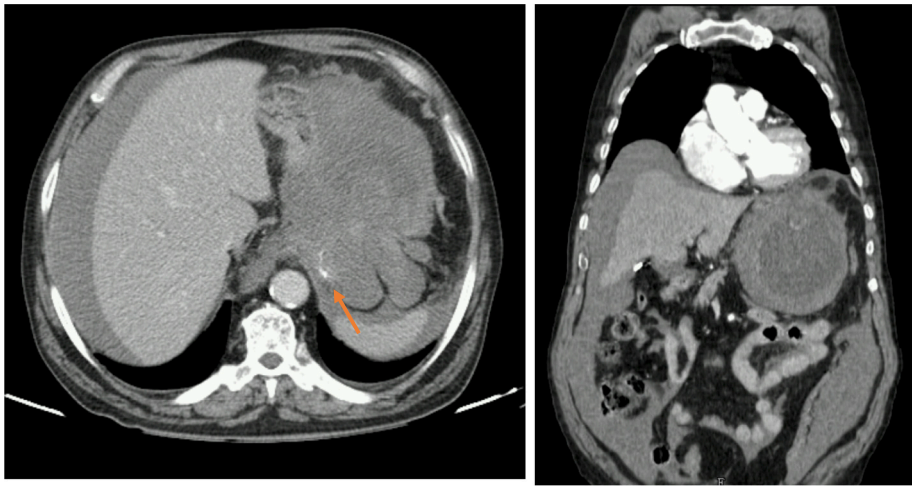

Fig. 3. Computed tomography image on admission: voluminous exophytic lesion on the posterior surface of the stomach body with active bleeding (arrow) and hemoperitoneum.

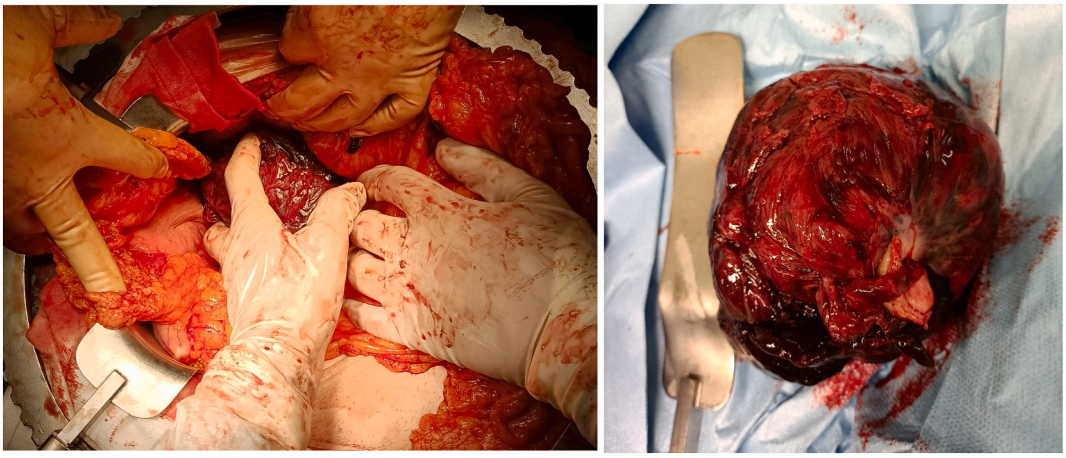

Fig. 4. Exploratory laparotomy: lesion of 10,1 × 9,1 × 8,9 cm with active bleeding was attached to the posterior wall of the stomach by a pedicle. Atypical gastrectomy was performed.

mesenchymal tumors (90%). The most common location is the stomach (70%), representing 2%–3% of all gastric malignancies. In the stomach, the body is the most common site [2].

Clinical manifestations of GISTs depend on its size and location. Ulcerated lesions can manifest with symptoms of gastrointestinal bleeding. Patients with larger tumors can present with abdominal pain and early satiety. However, because of the exophytic growth pattern of these tumors, some patients remain asymptomatic until the tumor has

become quite large [2]. Hemoperitoneum is rare, with few cases reported in literature [3].

The diagnostic evaluation in patients with GISTs often requires attention to specific clinical findings as well as a high level of suspicion. Both imaging and endoscopic procedures are necessary to the initial diagnostic evaluation of GISTs [11].

CT is the imaging method of choice to characterize an abdominal mass suspicious. The main endoscopic finding of GISTs is a nonspecific smooth bulge covered with normal mucosa [12]. The addition of ultrasound to endoscopy (EUS) can distinguish intramural from extramural tumors by identifying the layer of origin. On EUS, GISTs are typically hypoechoic, homogeneous lesions with well-defined margins. Most GISTs originate from within the muscularis propria [13].

EUS imaging characteristics alone provide insufficient accuracy in the diagnosis of GISTs, tissue sampling for immunohistochemical analysis using EUS-FNA or biopsy is required for a definite diagnosis [12].

The diagnosis of GIST is establish using histopathology, immunohistochemistry, and identification of disease-specific molecular alterations: approximately 95% of GISTs are positive for expression of the receptor tyrosine kinase KIT (CD117). A subset of GISTs lacking *KIT* mutations have activating mutations in the tyrosine kinase platelet-derived growth factor receptor alpha (*PDGFRA*) gene [15].

The standard treatment of a GIST without metastasis is surgical resection (R0). Administration of tyrosine kinase inhibitors such as imatinib is the primary approach for unresectable, metastatic or recurrent GISTs. The introduction of imatinib has dramatically improved the management of GISTs, prolonging recurrence-free survival after surgery and extending overall survival in metastatic or unresectable cases. Nearly half of patients present with metastatic disease, most commonly to the liver and peritoneum. Lymph node metastasis is infrequent, so lymph node dissection is not recommended except when lymph node metastasis is clinically suspected [12].

Risk factors that influence prognosis in patients with GIST are mainly based on the characteristics of the primary tumor. The most clinically relevant risk factors are mitotic rate, size and location [14].

4. Conclusion

We report two extremely rare cases of patients with a spontaneous hemoperitoneum as first presentation of gastric tumors. There are few reports in literature for gastric GIST, but none for PF. Imaging plays a critical role in the diagnosis of hemoperitoneum and can detect and characterize the underlying pathology.

When a patient presents with unspecific abdominal pain and spontaneous hemoperitoneum, the surgeon should consider a potential bleeding from gastric tumor that may require gastric resection.

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Ethical approval

Not applicable.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Registration of research studies

This study is not ‘First in Man’ study.

Guarantor

Yoshito Wada, MD, PhD.

Provenance and peer review

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CRediT authorship contribution statement

Y Wada, S Taniwaki and H Soh performed the operations. Y Wada, S and H Terabe managed the postoperative intensive care. All authors conceived the study and participated in its design and coordination. T Imamura supervised the study and drafted the manuscript. Y Morimitsu contributed to the pathological diagnosis. All authors declare that the paper is being submitted for consideration for publication in Journal of Medical Case Reports, that the content has not been published or submitted for publication elsewhere. All authors read and approved the final manuscript.

Declaration of competing interest

None.

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