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## A Rare Case of a Ruptured Metastatic Hepatic Lesion from a Jejunal Gastrointestinal Stromal Tumor (GIST) Treated by Arterial Embolization

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Data Collection B  
Statistical Analysis C  
Data Interpretation D  
Manuscript Preparation E  
Literature Search F  
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**Conflict of interest:** None declared

**Patient:** Male, 68  
**Final Diagnosis:** Ruptured metastatic hepatic lesion from a jejunal GIST  
**Symptoms:** Abdominal discomfort • hypotension  
**Medication:** —  
**Clinical Procedure:** —  
**Specialty:** Gastroenterology and Hepatology

**Objective:** Rare disease

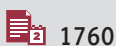
**Background:** Gastrointestinal stromal tumors (GISTs) are rare gastrointestinal neoplasms. The spontaneous rupture of a jejunal GIST is very rare and spontaneous rupture of liver metastasis from an intestinal GIST is even rarer with only a few cases reported in the literature.

**Case Report:** In this article, we reported a case of spontaneous rupture of a liver metastasis from a malignant jejunal GIST that presented with active tumoral bleeding, hypovolemic shock, and hemoperitoneum. The patient was successfully treated with arterial embolization of the tumor.

**Conclusions:** In appropriately selected patients, arterial embolization appears to be an effective safe treatment for a GIST metastasis rupture.

**MeSH Keywords:** Chemoembolization, Therapeutic • Endoscopy, Gastrointestinal • Gastrointestinal Hemorrhage

**Full-text PDF:** <https://www.amjcaserep.com/abstract/index/idArt/910061>



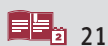
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## Background

Gastrointestinal stromal tumors (GISTs) are the most common mesenchymal tumors of the GI tract and can occur anywhere in the GI tract but are most commonly found in the stomach and the small intestine (39–70% and 20–32%, respectively) [1–3]. GISTs have variable malignant potential as 40% of initially localized lesions give rise to metastasis whereas 10–20% present initially with distant metastasis [4]. The liver is the most common site of metastasis (65%) followed by the peritoneum (21%), whereas the lymph nodes, lungs, and bones are rarely affected [5]. Kin et al. showed that to 50% of GIST patients will have recurrence or metastases after resection of the primary lesion, and the most common affected sites will be the liver and peritoneum [6]. Spontaneous rupture of jejunal GISTs are rare with less than 15 cases reported as of 2013 [7]. Unlike a primary hepatocellular carcinoma, it is very unusual for a hepatic metastasis to rupture, and it is even rarer in the case of a jejunal GIST metastasis [8].

We report the case of a 68-year-old male with a jejunal GIST that was surgically removed but recurred with liver metastasis, and who presented to our academic center with spontaneous rupture of his liver metastasis.

## Case Report

This is the case of a 68-year-old male patient known to have a jejunal GIST that was surgically treated by small bowel resection and primary anastomosis 2 years ago and was maintained on imatinib mesylate 100 mg 4 tablets daily since then. One year following the surgery, the patient developed liver metastasis as a follow-up computed tomography (CT) scan revealed a single 2 cm nodule in segment VIII of the liver. Other past surgical and medical history are unremarkable.

Recently, the patient had an episode of syncope and falling down, for which he was admitted to a primary care center. Upon presentation, the patient was hypotensive with a systolic blood pressure of 70 mmHg, and his laboratory tests showed low hemoglobin and hematocrit levels (5.5 g/dL and 16%, respectively). On physical examination, the patient had mild abdominal distention with diffuse nonspecific tenderness. He denied any melena, hematochezia, or hematemesis.

Urgent stabilization and resuscitation were done followed by an urgent CT scan of the abdomen and pelvis with intravenous (IV) contrast administration revealing multiple liver lesions in segments VI, VII, and VIII, associated with a large subcapsular hepatic hematoma and minimal amount of intra-abdominal fluids.

**Table 1.** Vital signs upon admission to our institution.

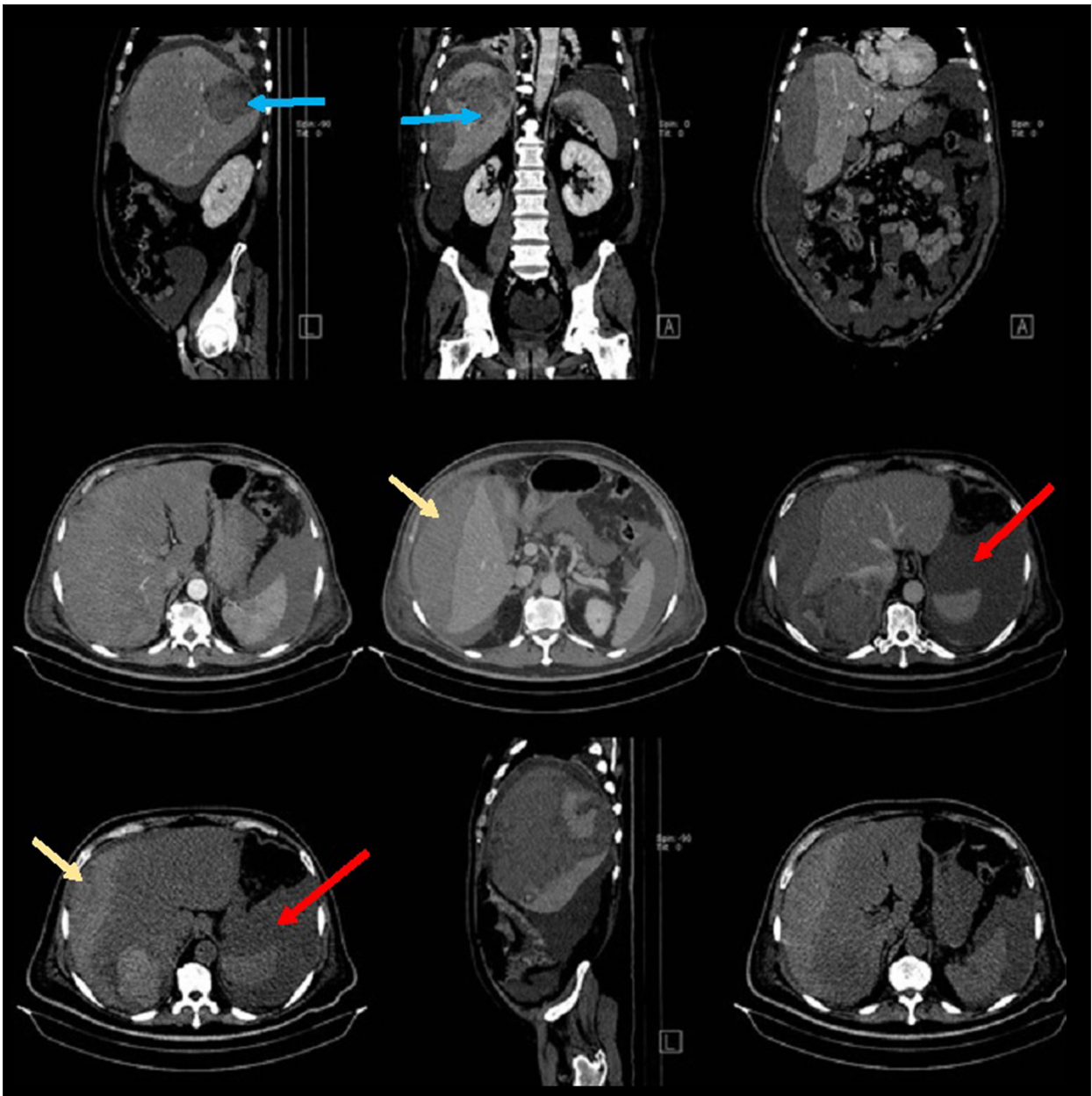
Blood pressure (SBP/DBP) in mmHg	110/70
Heart rate (beats per minute)	106
Respiratory rate (breaths per minute)	16
Oxygen saturation (%)	94
Temperature (°C)	37

**Table 2.** Laboratory test values upon admission to our institution.

	Patient	Normal range
Platelet count (10 <sup>3</sup> /μL)	114	186–353
International normalized ratio (INR)	1.17	0.8–1.1
Partial thromboplastin time (seconds)	36.7	30.5
Serum creatinine (mg/dL)	0.52	0.44–1.00
Total bilirubin (mg/dL)	1.63	0.3–1.2
Direct bilirubin (mg/dL)	0.33	0.1–0.5
Total protein (g/dL)	6.6	6.0–8.0
Serum albumin (g/dL)	3.2	3.5–5
SGOT (U/L)	29	15–41
SGPT (U/L)	79	14–54
Gamma-glutamyltransferase (U/L)	30	7–50
Serum lipase (U/L)	27	73–393
C-reactive protein (mg/dL)	156	<7.48

The patient received 5 units of packed red blood cells and 2 units of fresh frozen plasma over a period of 48 hours. Hemoglobin level increased to 7.7 g/dL; however, the patient's abdominal distention was increasing in intensity progressively. A second CT scan was done and showed increase in the subcapsular hematoma size as well as in the amount of ascites. Once stabilized, the patient was transferred by an ambulance to our institute, a tertiary care center, for further investigations and treatment.

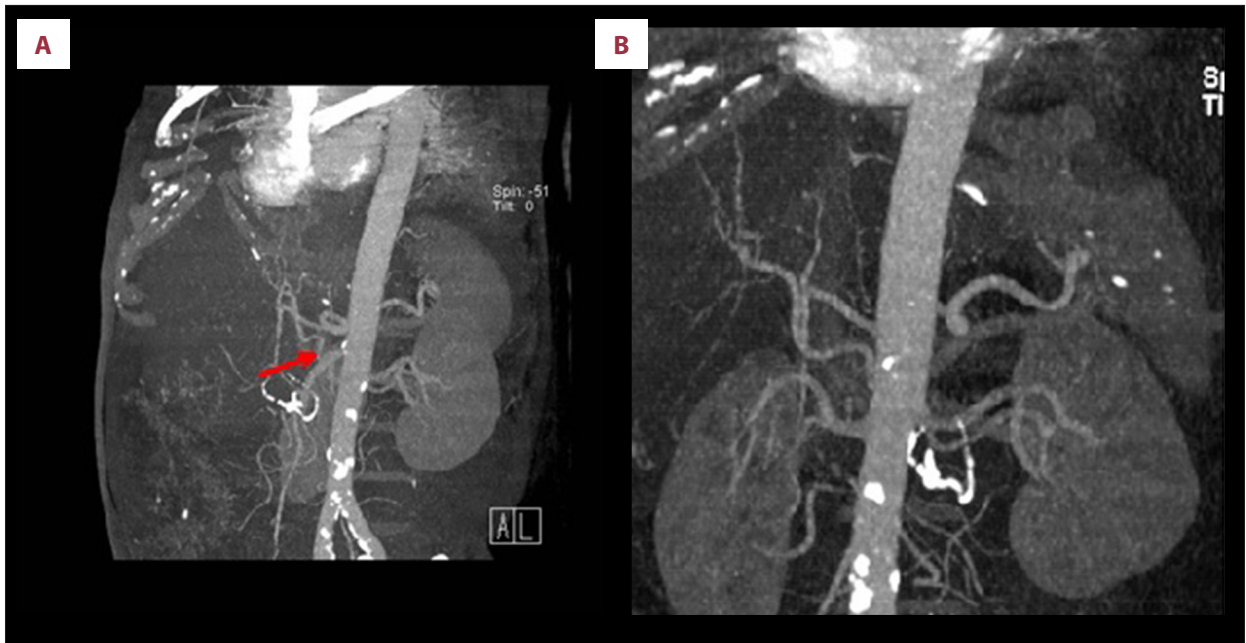
Upon arrival, the patient was hemodynamically stable with normal vital signs (Table 1) Blood pressure was 110/70 mmHg, heart rate was 106 beats per minute, respiratory rate was 16 breaths per minute, SaO<sub>2</sub> was 94%, temperature was 37°C. Hemoglobin was 8.7 g/dL and hematocrit was 25.1%. Other laboratory values are shown in Table 2.



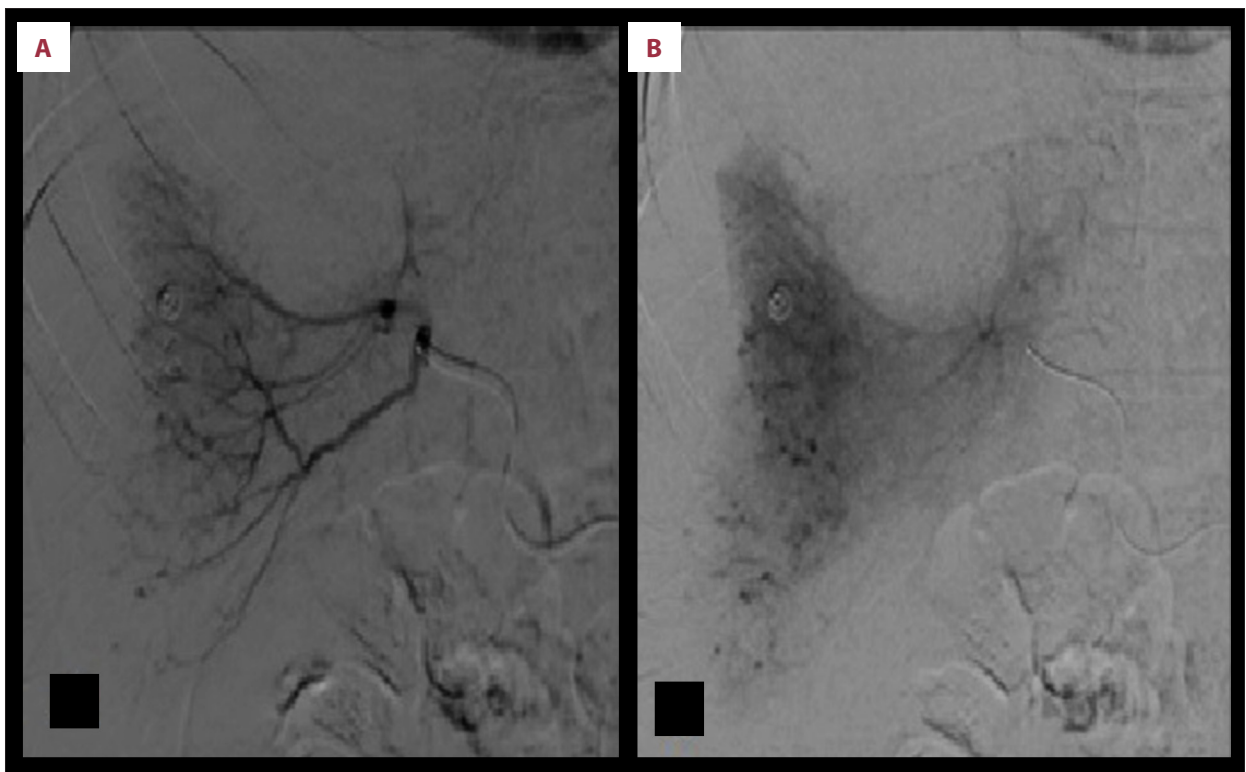
**Figure 1.** Computed tomography scan of the abdomen. A hyperdense liver mass is seen in liver segments VI/VII (blue arrows), along with a subcapsular hepatic hematoma (yellow arrows) and a massive high density (>15 Hounsfield units) ascites (red arrows) consistent with a hemoperitoneum.

A CT angiography of the abdomen and pelvis was performed and showed the presence of a 10.5×8 cm hyperdense liver mass in liver segments VI/VII prior to contrast injection, communicating with a large subcapsular hematoma measuring up to 20 cm; a faint arterial blush was also seen, indicating the presence of active bleeding. These findings were associated with massive ascites of high density (>15 Hounsfield Units) in favor of a hemoperitoneum (Figure 1).

The patient was then transferred to the intensive care unit (ICU) for close monitoring. In the ICU, an abdominal paracentesis was done confirming the bloody nature of the ascitic fluid, consistent with the diagnosis of a hemoperitoneum. A decision was made to perform an embolization of the artery supplying the posterior segment (VI/VII) of the right hepatic lobe in an attempt to intent stop the bleeding. Of note, the CT angiography showed that the right hepatic artery originated from the superior mesenteric artery (SMA) instead of the celiac trunk (Figure 2).



**Figure 2.** CT angiography Scan of the abdomen. The right hepatic artery (red arrow) is arising from the superior mesenteric artery.



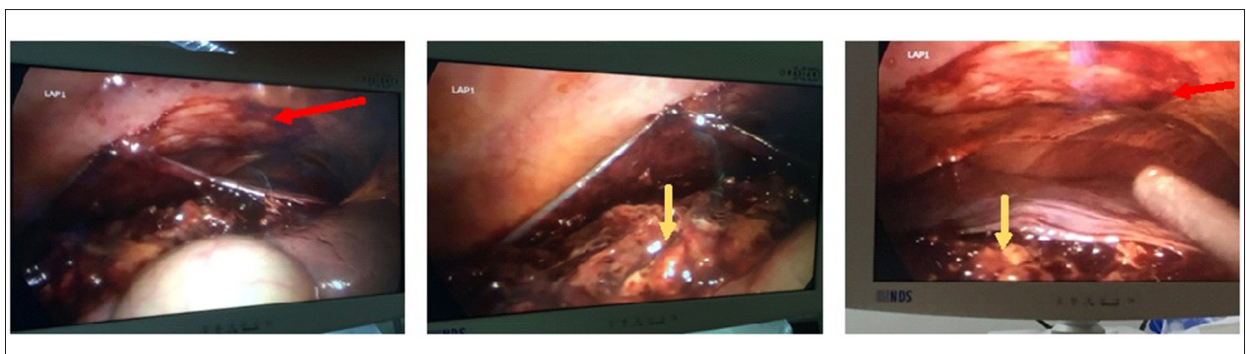
**Figure 3.** Hepatic angiography prior to embolization. A selective right posterior segment angiography was performed (A) and showed tumoral arterial blush (B).

The procedure, performed by an expert interventional radiologist, was done using a right 5Fr femoral access sheath: a 5Fr COBRA catheter was introduced and selective catheterization of the SMA then the right hepatic artery using a microcatheter

and finally the posterior segments branches using a HI-FLO microcatheter (Boston Scientific) (Figure 3).



**Figure 4.** Hepatic angiography post-embolization. There is absence of tumoral arterial blush following embolization (A) and coil release (B).



**Figure 5.** Laparoscopic exploration of the abdomen. The hepatic capsule is ruptured (red arrows) and there is evidence of peritoneal nodules (yellow arrows).

Introduction of microparticles (Boston Scientific) were done selectively, followed by release of two pushable coils (Complex Helical 18/Vortex Diamond 18-Boston Scientific).

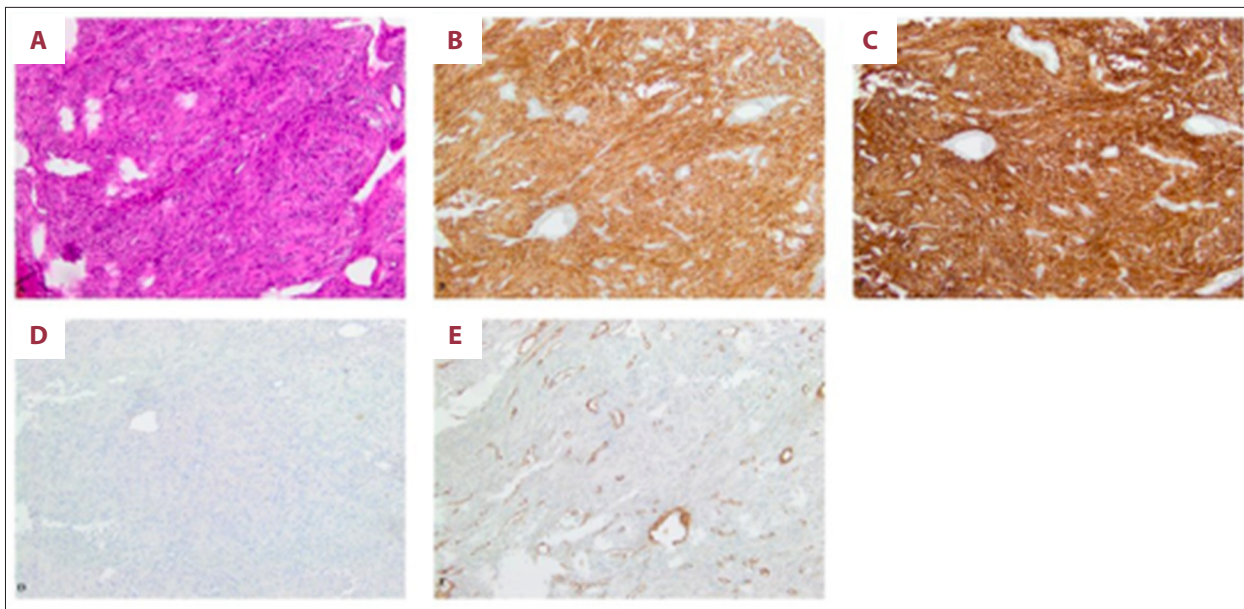
Angiography post-procedure showed absence of any tumoral contrast blush (Figure 4).

After the procedure, the patient became stable as his hemoglobin stabilized around 8.8 g/dL

On day 1 post-embolization, the patient experienced worsening dyspnea and was oxygen dependent. A CT pulmonary

angiogram was performed and revealed to no evidence of pulmonary embolism.

We suspected an increased diaphragmatic pressure due to the abdominal blood collection as the cause of dyspnea, and thus we performed a laparoscopic lavage and drainage of the hemoperitoneum. This procedure revealed a rupture of the liver capsule as well as the presence of peritoneal nodules. Approximately 3.4 liters of blood were extracted from abdominal cavity and biopsy taken from the peritoneal nodules (Figure 5).



**Figure 6.** Histological examination of the peritoneal nodule (magnification 100×). The hematoxylin and eosin staining show a well-demarcated nodule made up proliferation of smooth muscle cells. No atypia and necrosis. Mitotic figures are rare (A). The immunohistochemistry study reveals high expression of CD117 (B) and DOG1 (C) and negative expression of S-100 marker (D) and smooth muscle actin (E). All these findings are consistent with a GIST metastasis.

The patient's condition was improving following the lavage and he was transferred to the regular floor on day 1 post-lavage and discharged home day 2. The patient was followed for 6 months and remained free of complications.

The peritoneal nodules immunohistochemistry was highly positive for DOG1 and CD117 markers and negative for S100 and smooth muscle actin (Figure 6). This was consistent with the diagnosis of secondary peritoneal carcinomatosis as the DOG1 and CD117 have been classically linked to GIST cells [9].

## Discussion

GIST is a rare tumor representing 0.1–3% of all GI neoplasms. Nevertheless, GISTs are the most common parenchymal tumors of the GI tract [1,2] and represents a wide clinical spectrum of tumors with different clinical presentations, locations, histology, and prognosis.

Most GISTs remain silent until reaching a large size. Symptoms vary according to location and size. Patients generally present with non-specific symptoms including abdominal pain, fatigue, nausea, anorexia, weight loss, fever, obstruction, GI bleed, and dyspepsia, among others [4]. Metastasis occurs in patients with aggressive GISTs and the liver is the most common site of metastasis (up to 65%) [10].

Diagnosis is initially made by endoscopy or imaging studies such as MRI and CT scan. Tissue pathology with molecular testing after biopsy or surgical resection is the most accurate and definitive diagnostic tool for GISTs.

Localized tumors are treated with surgical approach especially if it is locally or marginally resectable. Patients with advanced GISTs and those with local unresectable tumors are managed with imatinib mesylate, an oral, selective, small-molecule tyrosine kinase inhibitor [11] that directs the signaling pathways involved in the pathogenesis of the disease. However, there is a high risk of relapse (40%) after complete resection [12].

The treatment of liver metastasis of GISTs depends on multiple factors. After achieving complete resection of the primary site, assessment of metastasis resectability (number and localization of lesions), the presence of other distant metastasis, the liver function reserve, and the predicted remnant volume will determine the proper management [13].

Following resection of the liver metastasis, the patient should be maintained on imatinib mesylate therapy. If recurrence occurs, one should consider upgrading the treatment dose or shifting to other molecules (eg. sunitinib malate, nilotinib hydrochloride monohydrate, or sorafenib) [13].

Hemoperitoneum is a rare presentation of hepatic metastasis of intestinal GIST. Aggressive resuscitation and urgent intervention are the key to therapy. Ruptured liver subcapsular

hematoma might cause massive bleeding and hemorrhagic hypotensive shock, and therefore an eventual deterioration of clinical status of the patient. Few cases of ruptured GIST liver metastasis with hemoperitoneum have been reported in the literature. In 2003, Suzuki et al. reported a case of a 65-year-old male who presented with massive hemoperitoneum secondary to a ruptured liver GIST metastasis that resulted in subsequent death of the patient [14].

Yoon reported another case of a 65-year-old male with a spontaneous rupture of a hepatic metastasis from a gastric GIST that presented as hypovolemic shock. Resuscitation and emergent selective arterial embolization of the tumor stopped the hemorrhage, but recurrence occurred 3 days after embolization imposing surgical intervention [15].

There have also been cases of spontaneous subcapsular liver hematoma described in patients with metastatic GISTs receiving imatinib, raising concerns of bleeding as a potential complication [16].

Liver metastasis of GISTs are highly vascularized as a result of neo-angiogenesis, receiving 90% of their blood supply from the arterial circulation via the hepatic artery, compared to only 25% for normal liver parenchyma [17]. Thus, intra-tumoral hepatic bleeding can be managed by selective arterial embolization. A trans-arterial embolization is supposed to induce necrosis of the tumor by causing severe ischemia, while many portal veins that supply normal parenchyma only get mild, non-lethal ischemia [18].

Assessment of liver function is crucial before undergoing embolization, as well as assessing whether the portal vein is invaded or occluded, for this carries a high risk of hepatic insufficiency/infarction post embolization [19]. Nonetheless, it has

been shown that the procedure can be done if adequate collateral hepatoportal flow is present [20].

Seldinger wire technique is used to secure the arterial access; a diagnostic visceral arteriography is then done to assess the arterial anatomy. Major complications rarely occur with a prevalence of less than 5%, unless there are predisposing factors such as portal vein occlusion, compromised liver function, biliary obstruction, and in cases of non-selective embolization [19].

In cases of interventional arterial embolization failure, surgical treatment should be considered and is considered lifesaving. Surgical resection of the affected hepatic parenchyma is challenging, difficult to perform, and life-threatening. Other options include hepatic artery branch ligation, suture ligation, and packing with hemostatic agents [21]. Recurrence of hemorrhage may occur and carries a poor prognosis.

In our case, the resection of the right hepatic lobe would have been difficult and dangerous because of active hemorrhage and invasion of the right hepatic vein, and would have led to ligation of the right hepatic artery.

## Conclusions

In conclusion, we report a rare case of jejunal GIST with liver and peritoneal recurrence following primary resection that presented with hemoperitoneum secondary to spontaneous rupture of the patient's hepatic metastasis. In appropriately selected patients, arterial embolization appears to be an effective and safe treatment for the rupture of a GIST metastasis.

## Conflicts of interest

None.

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