



## Case report

# Management of gastrosplenic fistula in the emergency setting - A case report and review of the literature



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## ABSTRACT

**Introduction:** A gastrosplenic fistula (GSF) is a very rare complication that arises mainly from a splenic or gastric large cell lymphoma. The proximity of the gastric fundus to the enlarged fragile spleen may facilitate the fistulisation. This complication can lead to massive bleeding, which, though uncommon, may be lethal. We present a patient with massive upper gastrointestinal bleeding secondary to a GSF.

**Case presentation:** We present a 48-year-old man with a refractory diffuse large B-cell lymphoma who was admitted to our hospital due to hematemesis. On arrival, he was in hemorrhagic shock, and was taken directly to the intensive care unit. The source of bleeding could not be identified on gastroscopy, the patient remained hemodynamically unstable and a laparotomy was performed.

A fistula between a branch of the splenic artery and the stomach was identified. The stomach appeared to be involved in the malignant process. After subtotal gastrectomy and splenectomy, the bleeding was controlled. After stabilization, the patient was admitted to the intensive care unit, and 24 hours later was discharged in stable condition.

**Discussion:** We describe a fistula between a branch of the splenic artery and the stomach, which was accompanied by massive bleeding. An emergency laparotomy saved the patient's life.

**Conclusion:** The purpose of this report is to alert physicians that surgical intervention can be lifesaving in this rare malignant condition. A literature review focusing on the presenting symptoms and the epidemiology of GSF is presented.

## 1. Introduction

We present a patient who reported to the emergency department of a university-affiliated hospital with massive upper gastrointestinal (GI) bleeding secondary to a gastrosplenic fistula (GSF). GSF is very rare (28 cases have been described during the last 27 years) and a potentially fatal complication of various diseases, including lymphoma, gastric adenocarcinoma, Crohn's disease, splenic abscess, and trauma [1]. Of these diseases, the majority have occurred in patients with diffuse, large, B-cell lymphoma (DLBCL). Other complications of DLBCL that involve the GI tract are: perforation, obstruction and intractable bleeding.

In the described case, the fistula was a complication of DLBCL that

involved the spleen and the stomach. The case is unusual because, of the 28 cases of GSF reported in the English literature, only four presented with massive upper GI bleeding [1–4].

As detailed below, we attribute the successful management of this case to early aggressive surgical treatment of the bleeding site. We believe that surgical treatment may rescue patients and offer a chance for long term survival even when the malignancy is not localized.

This paper describes the management of the index case and reviews prior publications, in line with the SCARE criteria [5].

## 2. Case presentation

A 48-year old man with a history of refractory DLBCL was admitted

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**Table 1**  
Characteristics of reported GSF cases.

Age	Gender	Etiology	Presenting symptom	Chemotherapy ?	Survived the event ?	year of publication
66	male	large B cell lymphoma	weakness	yes	yes	2016
79	female	diffuse B cell	LUQ abdominal pain	none	yes	2016
52	male	gastric B cell lymphoma	GI bleeding	yes	yes	2016
70	male	gastric adenocarcinoma	?	none	no	2015
22	male	bariatric surgery	abdominal pain	none	yes	2015
55	male	large B cell lymphoma	abdominal pain, GI bleeding	none	yes	2013
57	male	large B cell lymphoma	fever, cough	yes	?	2014
62	male	B cell lymphoma	fever, abdominal pain	none	yes	2012
68	male	large B cell lymphoma	hematemesis	none	yes	2011
55	male	large B cell lymphoma	weakness, melena	none	yes	2011
43	female	large B cell lymphoma	weakness, melena	none	yes	2010
49	male	B cell lymphoma	weakness, melena	yes	yes	2009
76	female	large B cell lymphoma	GI bleeding	none	no	2009
76	male	?	weakness	none	yes	2009
25	male	large B cell lymphoma	leukocytosis	none	no	2008
50	female	lymphoma	?	yes	yes	2008
56	male	B cell lymphoma	fever	yes	yes	2008
57	male	B cell lymphoma	abdominal pain	none	yes	2006
70	male	post traumatic	abdominal pain, weight loss	none	yes	2005
66	male	metastatic colon adenocarcinoma	malaise	yes	yes	2004
24	male	large B cell lymphoma	routine CT followup	yes	yes	2002
21	male	large B cell lymphoma	abdominal mass	none	yes	2002
62	male	large B cell lymphoma	left abdominal pain, fever	none	yes	1995
45	male	large B cell lymphoma	epigastric pain	none	yes	1995
46	female	splenic abscess	left flank pain	none	yes	1991
36	male	centroblastic lymphoma	GI bleeding	yes	yes	1991
36	female	adenocarcinoma of stomach	abdominal pain	yes	no	1990
52	female	crohn's	nausea, vomiting	none	yes	1989

to our hospital due to hematemesis. Four months before the current event, a B cell lymphoma was diagnosed, and the patient was treated by chemotherapy: rituximab plus cyclophosphamide, vincristine, doxorubicin, and prednisone (CHOP) for 3 cycles, followed by rituximab plus cisplatin, cytarabine, and dexamethasone (DHAP) for 2 cycles. He had a remission but quickly relapsed, and gemcitabine - oxaliplatin (GEMOX) was given. A PET-CT scan performed 3 weeks before the described event demonstrated return of the disease: lymphatic hyperplasia with hypermetabolic disease above and below the diaphragm, with nodular and extranodal involvement and involvement of the spleen. No fistula between the spleen and the stomach was identified on the scan.

Upon arrival to the emergency room, the patient described two vomiting episodes of “fresh bright content in large amount” 30 minutes before his arrival. On physical examination he was alert, oriented, and diaphoretic; blood pressure (BP) was 100/55, heart rate (HR) 110, respiratory rate 24, and saturated oxygen (SaO<sub>2</sub>) 96% in room air. His hemoglobin level was 7.6 g/dL. A nasogastric tube was inserted and 150 cc of fresh bright blood was identified. Focused assessment with sonography for trauma (FAST) was not performed due to the lack of a trauma history, and a decision was made to transfer the patient immediately to the intensive care unit (ICU) to perform blood product resuscitation and an urgent gastroscopy. Two liters of Hartman's solution were given through two peripheral 16-gauge intravenous catheters.

On arrival to the ICU, the patient was lethargic, BP 90/40, HR 120, respiratory rate 26 and SaO<sub>2</sub> 96% with an oxygen mask (FiO<sub>2</sub> = 1.0). A digital rectal examination revealed normal sphincter tone, no masses, and brown stool. The hemoglobin level dropped to 6 g/dL. The patient was sedated and intubated due to the hemorrhagic shock. Considering the patient's history, differential diagnosis included a gastric and/or duodenal ulcer, severe or erosive gastritis/duodenitis, mass lesions and an aortoenteric fistula.

Large volumes of blood products were rapidly transfused, according to our local massive transfusion protocol: 8 units of packed cells, 10 units of thrombocytes and 8 units of fresh frozen plasma. Coagulation laboratory results were not available at the time. Blood pressure dropped and reached a minimum of 60/30, and noradrenalin was started simultaneously. When blood pressure reached 90/60,

gastroscopy demonstrated traces of blood in the esophagus, and large quantities of fresh blood in the stomach and in the first part of the duodenum. However, the source of the bleeding could not be identified because of ongoing active bleeding in the gastric cavity.

The patient was immediately taken to the operating room and an urgent laparotomy was performed. Angiography with embolization was not a possibility due to hemodynamic instability. In the course of the operation, the stomach was found to be filled with blood, and a bleeding artery from a gastric ulcer located in the large curve penetrating the spleen gate was identified. The stomach was separated from the spleen, and total splenectomy with subtotal gastrectomy and gastrojejunostomy were performed. After control of the hemorrhage, the patient's condition stabilized and he was readmitted to the ICU. Several hours later, he regained full consciousness and underwent extubation. Twenty-four hours later the patient was transferred to a hospital unit in stable condition.

Examination of the gastrectomy specimen demonstrated transmural infiltration of the gastric wall by medium to large atypical cells with vesicular nuclei and, in part, with clear cytoplasm. Areas of necrosis, involving whole thickness of the muscularis propria were seen (Fig. 4). The tumor cells stained positive for LCA, CD79a, PAX5, CD43, CD10 and Bcl6 (Fig. 5) and negative for CD20, MUM1, Bcl2, TcT, C-myc and CD99. The Ki67 proliferation index was as high as 99%. In addition, foci of intestinal metaplasia were found in the gastric mucosa.

The proper examination of the spleen has been precluded by severe autolytic changes. However, the widespread infiltration by medium to large cells with the same immunophenotype was identified in the splenic tissue and in surrounding fat.

### 3. Review of the literature

A PubMed search (1989–2016) identified 28 case reports of GSF (Table 1). Twenty-one (75%) were associated with lymphoma (Fig. 1); of them, 12 (57%) were of the DLBCL type. Ten (48%) patients received chemotherapy before the episode [1,2]. Median age was 55 years, range 21–79; the mean age was 52.7 years (Fig. 2). Twenty-two patients (78%) were men.

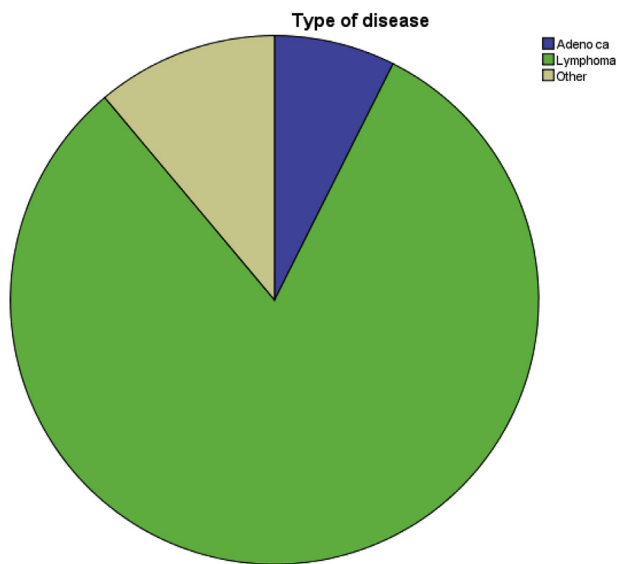


Fig. 1. Background diseases. This graph represents the distribution of background diseases that presented in 28 patients described in the literature.

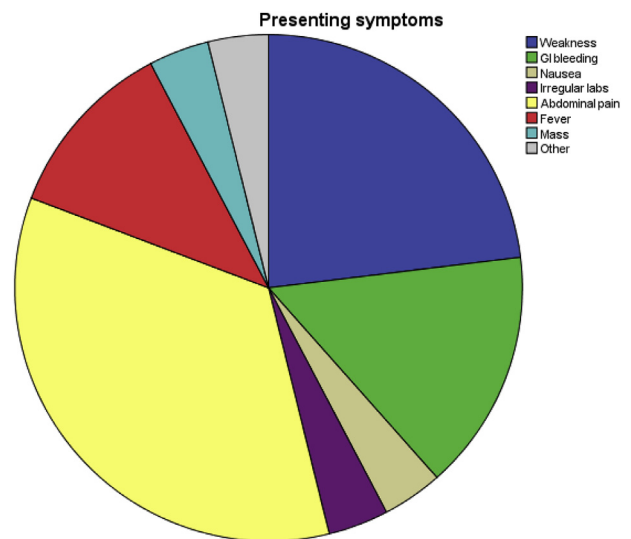


Fig. 3. Presenting symptoms. This graph represents the distribution of presenting symptoms of the 28 cases described in the literature.

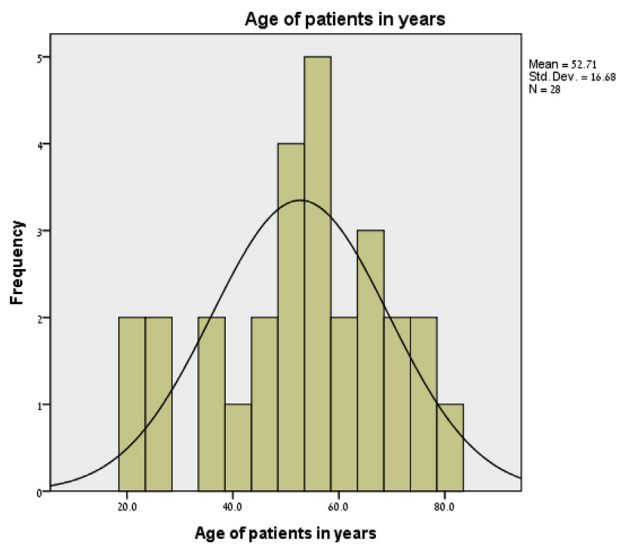


Fig. 2. Patient age. This graph represents the age distribution and the mean age of the patients described in the literature.

Nine patients (32%) presented with abdominal pain, six (21%) with weakness, and the remaining (13 patients) with other symptoms. Only four patients (14%) presented with upper GI bleeding (Fig. 3); of them, three (75%) survived the event. In addition, GI bleeding was reported in four other patients (14%) as a secondary symptom (Table 1). Twenty-three patients (82%) survived the event, four died (14%), and the outcome of the remaining patient was not reported.

#### 4. Discussion

GSF is a very rare condition that arises mainly from splenic or gastric large cell lymphoma. Gastric and splenic lymphomas can fistulate with other organs, including the bronchus and colon [6]. Based on the literature review presented above, GSF does not usually present as massive bleeding. As shown in Table 1, when the presentation is not massive bleeding, the prognosis, at least in the short term, is good (about 82% survival). In the current case, we believe that the GSF was a direct complication of DLBCL, since the histological examination showed malignant cells in the spleen, as well as in the stomach.

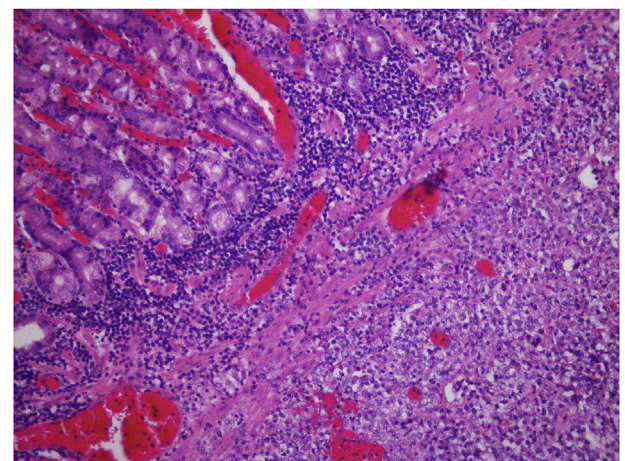


Fig. 4. Gastrectomy specimen. Transmural infiltration of the gastric wall by medium to large atypical cells with vesicular nuclei and, in part, with clear cytoplasm. Areas of necrosis, involving whole thickness of the muscularis propria were seen.

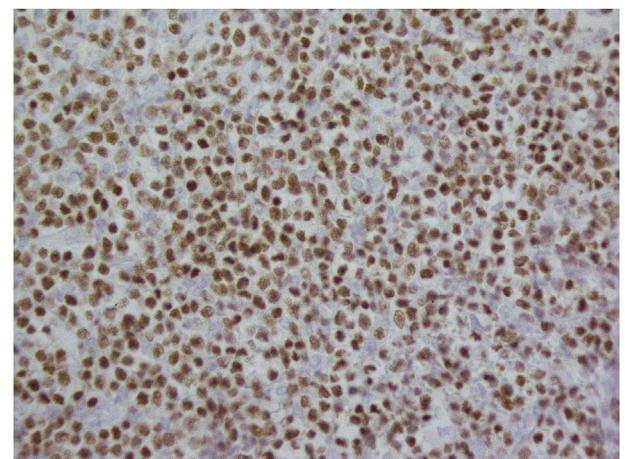


Fig. 5. PAX5 positive. Gastric wall cells stained positive for PAX5.

Early diagnosis of GSF may be life-saving, but can be challenging, and should be considered in every DLBCL patient undergoing routine abdominal imaging. Abdominal CT seems superior to other radiological tests for diagnosing GSF ([10]). The presence of air in the spleen should alert physicians to the possibility of GSF. Upper gastrointestinal endoscopy can confirm the presence of GSF by direct visualization of the fistulous opening.

When GSF is diagnosed, with or without active bleeding, radical surgical resection with splenectomy and gastrectomy is the most common treatment option. However, a few cases treated by distal pancreatectomies have been reported ([7]). Although open procedures are more commonly described, one laparoscopic case was reported ([8]). When active bleeding is the presenting symptom, and the patient is hemodynamically stable, interventional radiology in the form of splenic artery embolization can serve as the definitive treatment [9]. Massive bleeding is associated with significant mortality. In patients with refractory lymphoma, some surgeons may be inclined to avoid an aggressive approach [4]. In the current case, due to the lack of response to conventional chemotherapy, long term prognosis was expected not to be good. Nevertheless, the postoperative course was uneventful, and recovery was rapid.

## 5. Conclusion

We strongly recommend that GSF should be considered in regard to every DLBCL patient who undergoes routine abdominal imaging, and if found – should be treated surgically to avoid the complication of fatal bleeding. In cases in which active bleeding is a presenting symptom of GSF – embolization vs laparotomy should be considered, depending on the patient's hemodynamic condition. Aggressive surgical treatment may be warranted in selected cases of hemorrhage from gastrosplenic fistulae, despite the presence of refractory lymphoma.

## Ethical approval

This is a review of the current literature, no ethical approval needed.

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## Authors' contributions

A.F and ZP drafted the manuscript. Y.B, E.B, A.R.S and L.K scanned

and brought citations from the relevant literature, and M.K and J.P supervised and brought the manuscript to its final version. All authors read and approved the final manuscript.

## Conflicts of interest

The authors declare that they have no competing interests.

## Guarantor

Amit Frenkel, MD.

## Consent

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

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