ISSN 2689-8268 | Volume 8

American Journal of Surgery and Clinical Case Reports

Case Report Open Access

Operative Management of Gastrosplenic Fistula in A Patient with Diffuse Large B-Cell Lymphoma

Hernandez D[1*], Lajevardi A[2], Horton C[1], Michet CJ[1], Shaw J[1] and Zaragoza B[1]

[1]Broward Health Coral Springs

[2]Nova Southeastern University, Dr. Kiran C. Patel College of Osteopathic Medicine

[*]Corresponding author:

Hernandez Damian,
Broward Health Coral Springs, USA

Received: 26 July 2024
Accepted: 29 Aug 2024
Published: 02 Sep 2024
J Short Name: AJSCCR

Citation:

Hernandez D. Operative Management of Gastrosplenic Fistula in A Patient with Diffuse Large B-Cell Lymphoma. Ame J Surg Clin Case Rep. 2024; 8(2): 1-4

1. Abstract

Gastrosplenic Fistula (GSF) is an extremely rare manifestation of splenic and gastric malignancy, with fewer than 30 cases reported. Lymphoma of the spleen is the most common etiology, although benign conditions have also been associated with GSF formation. Without urgent surgical management, complications including massive splenic hemorrhage are detrimental. Given its rarity, exact guidelines for diagnostic workup and management remain unclear. Hence, it is critical to maintain a healthy suspicion for this diagnosis in patients with a history of splenic or gastric lymphoma presenting with left upper quadrant pain or gastrointestinal bleeding. We present a case of a 78-year-old woman undergoing chemotherapy for diffuse large B-cell lymphoma, presenting with palpitations, and found to have splenomegaly and incidental GSF on abdominal imaging. While the patient remained hemodynamically stable, due to concern for impending splenic hemorrhage, we proceeded with splenic artery embolization followed by splenectomy and en bloc gastric wedge resection. (149 words)

2. Introduction

Gastrosplenic Fistula (GSF) is a rare complication of various benign or malignant diseases of the stomach or spleen.[1] The first case of GSF was reported by de Scoville et al in 1962.[2] They described radiographic findings of an enlarged spleen filled with air, termed “aerosplenomegaly,” suggestive of an abnormal fistulization between the stomach and spleen.[2] Primary splenic lymphoma, specifically, Diffuse Large B-Cell Lymphoma (DLBCL) is reported the most common cause of GSF.3 GSF has also been associated with lymphoma or adenocarcinoma of the stomach, Crohn’s disease, peptic ulcer disease, metastatic colorectal adenocarcinoma, T-cell lymphomas, and as a complication of sleeve gastrectomy.[1,4,11-8] The pathophysiology of GSF is proposed to be due to rapid, extensive necrosis of lymphoma tissue.4 This pathologic process potentiates injury to the splenic capsule resulting in invasion into the gastric wall, and thus, formation of a fistula.[4] This can be further exacerbated by chemotherapy, which can elicit tumor lysis syndrome and tissue necrosis, which increases the risk of fistulization.[5-10] Clinical presentation of GSF typically includes left upper quadrant pain due to splenomegaly, and fever, weight loss, and hematemesis may also be present.[21,22] Delayed diagnosis increases risk of fatal upper gastrointestinal bleeding. Due to its rarity, a GSF is often overlooked as a differential diagnosis until found incidentally on abdominal imaging. To date, fewer than 30 cases of GSF have been reported in the literature. Here we present a patient with DLBCL who underwent successful surgical treatment for GSF.

3.Case presentation

A 78-year-old woman with past medical history of diffuse Large B-Cell Lymphoma (DLBCL) with splenomegaly, was admitted to the medical service with complaints of palpitations and left-sided abdominal pain. These symptoms began eight days after her first chemotherapy session. Due to concern for acute coronary syndrome, the patient underwent cardiac catheterization which was negative for coronary vessel disease. One month prior, the patient had presented to the hospital with similar left upper quadrant abdominal pain. Computed Tomography (CT) scan of the abdomen with intravenous (IV) contrast showed splenomegaly with an 8.5 cm mass with perisplenic and retroperitoneal lymphadenopathy, as can be seen in Figure 1A and 1B. She underwent surgical biopsy of a peri-splenic lymph node as well as port placement for chemotherapy. The biopsy confirmed the diagnosis of high-grade DLB-

CL, and there were no complications. On current admission, CT of the abdomen without contrast revealed a gastrosplenic fistula, as demonstrated in Figure 1C and 1D with splenomegaly, demonstrated in Figure 2. General surgery was consulted for further recommendations. Initially, the general surgery team decided to pursue nonoperative management given her recent chemotherapy and lack of clinical symptoms. The patient was placed on bowel rest and Total Parenteral Nutrition (TPN), and gastroenterology, infectious disease, and oncology teams were consulted. After multidisciplinary discussion with specialists and the patient, the surgical team decided to proceed with splenectomy and en bloc gastric wedge resection due to the risk of impending bleeding. The patient underwent further workup with an upper GI series, as shown in Figure 3, which did not show any contrast extravasation into the spleen to suggest a fistula.

The day of surgery, the patient was taken to the interventional radiology suite first for embolization of the splenic artery with water-soluble embolic agent. She was then transferred to the operating room while under general anesthesia. A long midline incision was made and carried down to the subcutaneous tissue to enter the abdomen. The splenic flexure of the colon was taken down and the short gastric vessels were divided using electrocautery for mobilization of the spleen. The splenic artery was ligated using absorbable, braided ties, then the remainder of the splenic hilum was transected using a surgical stapler. Special care was taken during this approach to avoid damage to the pancreatic tail. The splenorenal and splenophrenic ligaments were dissected to visualize the fistulous tract. The tract involved the upper portion of the fundus and did not involve the gastroesophageal junction. A stapler was used to transect this portion of the fundus, and the staple line was inverted using a running synthetic, absorbable suture. The staple line was further reinforced with omentum. The abdomen was then thoroughly irrigated and a round drain was positioned in the splenic fossa. The fascia was then closed with running synthetic, absorbable suture and the skin was closed with staples.

The patient was monitored in the intensive care unit for 48 hours. She continued to recover with return of bowel function on postoperative day four. She was weaned off TPN and her diet was advanced accordingly. She was kept on multiple antibiotics and antifungals for empiric coverage. Her hospital course was complicated by atrial fibrillation with rapid ventricular response, postoperative anemia requiring blood transfusion, and a left pleural effusion requiring thoracentesis. She was discharged on postoperative day 10 to a skilled nursing facility and was advised to follow up with her medical oncologist to continue systemic therapy.

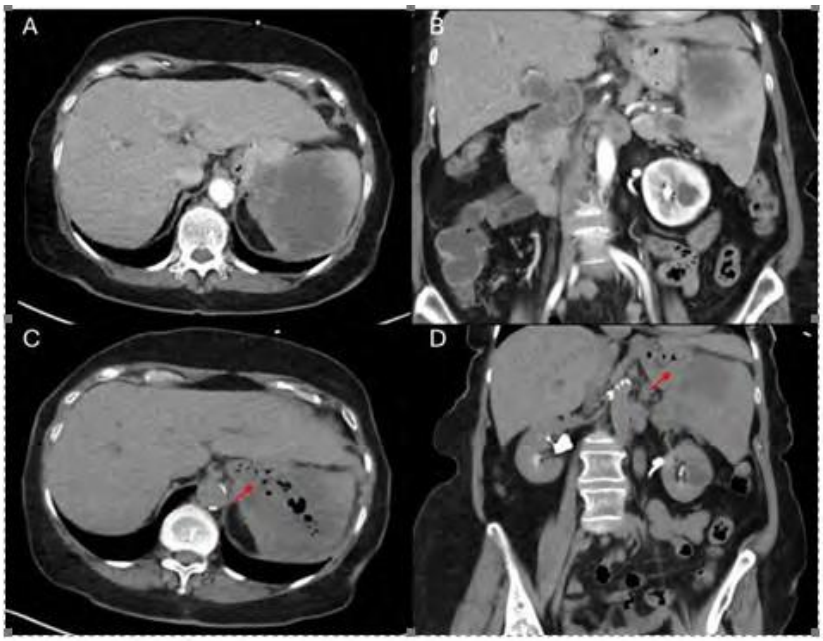

Figure 1: Abdominal CT scans. (A) Axial view and (B) coronal view of initial abdominal CT with intravenous contrast 1 month prior to admission without any sign of fistula. (C) Axial view and (D) coronal view of fistula (indicated by red arrow) in CT without contrast performed on admission.

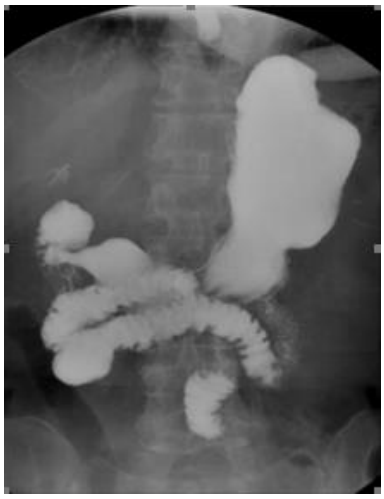

Figure 2: Upper gastrointestinal series with water-soluble contrast shows no evidence of contrast extravasation into the spleen.

4. Discussion

Our patient likely developed a GSF from DLBCL in a splenic mass that eroded into the stomach secondary to a local inflammatory reaction. Frenkel et al determined that 75% of GSF cases were associated with lymphoma, with 57% from DLBCL.[23] Interestingly, of the 28 cases reported since 2016, GSF was identified 48% of patients following administration of chemotherapy, such as occurred in our patient. [23-25]

Diagnosis of GSF may include utilization of imaging studies such as CT, MRI, or UGI series, or direct visualization with upper endoscopy or diagnostic laparoscopy. However, it may be difficult to ascertain the presence of a GSF with these conventional imaging methods, as our patient clearly had signs of a fistula on CT imaging (Figure 1C and 1D) but no evidence of contrast extravasation on UGI series (Figure 3). It is possible the fistula tract in this patient was small and intermittently closed, which would explain lack of contrast to the spleen at that given point in time, only for the surgical team to subsequently identify a fistula in surgery.

While a very rare entity, it is important for surgeons to have a high level of suspicion for this condition, particularly in patients with splenic or gastric lymphomas presenting with UGI bleeding, as GSFs can rapidly progress and become fatal without urgent surgical intervention.[26] A rapidly fatal complication includes splenic rupture with massive hemorrhage upon splenic expansion with involvement of the splenic artery or vein. This unfortunate event occurred in four cases documented in the literature. [23,25,27]

Given its infrequency, appropriate treatment of GSF is unclear. Most patients underwent splenectomy with or without partial gastrectomy with closure of the fistula track. Some patients underwent distal pancreatectomy depending on involvement of the pancreas. In patients with splenic artery involvement, management with embolization prior to surgical intervention is likely beneficial to prevent this complication, as was performed with our patient. Our team intentionally conducted embolization with a soft, pliable, embolic agent rather than metallic coils to optimize success in stapling across the area. However, embolization may be foregone in the setting of hemodynamic instability in which case emergent laparotomy and exploration is warranted. [19,23] Sousa et al conducted post-surgical splenic artery embolization to prevent future chemotherapy cycles from causing tumor necrosis and splenic artery hemorrhage. [24]

In conclusion, GSF is a rare complication frequently associated with DLBCL. There have been scant cases documented in the literature, which is why optimal diagnosis and treatment are unclear. Given the potential risks for morbid complications such as splenic artery rupture, we recommend swift surgical intervention with splenectomy with closure of the fistula tract via partial gastrectomy. This may be conducted via a minimally-invasive or open approach depending on surgeon comfort level and stability of the patient. Further management may involve consideration of IR for preoperative splenic vessel embolization or hepatobiliary specialist expertise if distal pancreatectomy if warranted. This case is unique as the patient did not have symptoms of internal bleeding, rather, her case allowed for non-urgent, multidisciplinary planning, with splenic embolization and without need for pancreatectomy.

References:

1. Kerem M, Sakrak O, Yilmaz TU, Gultekin FA, Dursun A, Bedirli A. Spontaneous Gastrosplenic Fistula in Primary Gastric Lymphoma: Surgical Management. Asian J. Surg. 2006; 29(4):287-90.
2. Scoville AD, Bovy P, Demeester P. [Radiologic "aerosplenomegaly" caused by necrotizing splenic lymphosarcoma with double fistulization into the digestive tract]. Acta Gastroenterol Belg.1967 ;30(12):840-6.
3. Borgharia S, Juneja P, Hazrah P, Lal R, Kapur N, Chaudhary P. Gastrosplenic Fistula: a Systematic Review. Indian J Surg Oncol. 2022;13(3):652-660.
4. Kang DH, Huh J, Lee JH, Jeong YK, Cha HJ. Gastrosplenic fistula occurring in lymphoma patients: Systematic review with a new case of extranodal NK/T-cell lymphoma. World J Gastroenterol. 2017;23(35):6491-6499.
5. Choi JE, Chung HJ, Lee HG. Spontaneous gastrosplenic fistula: a rare complication of splenic diffuse large cell lymphoma. Abdom Imaging. 2002;27(6):728-730.
6. Blanchi A, Bour B, Alami O. Spontaneous gastrosplenic fistula revealing high-grade centroblastic lymphoma: endoscopic findings. Gastrointest Endosc. 1995;42(6):587-589.
7. Aribaş BK, Başkan E, Altinyollar H, Üngül Ü, Cengiz A, Erdil HF. Gastrosplenic fistula due to splenic large cell lymphoma diagnosed by percutaneous drainage before surgical treatment. Turkish J Gastroenterol. 2008;19(1):69-70.
8. Carolin KA, Prakash SH, Silva YJ. Gastrosplenic fistulas: a case report and review of the literature. Am Surg. 1997;63(11):1007-10.

9. Bubenik O, Lopez MJ, Greco AO, Kraybill WG, Cherwitz DL. Gastrosplenic fistula following successful chemotherapy for disseminated histiocytic lymphoma. Cancer. 1983;52(6):994-6.
10. Moghazy KM. Gastrosplenic fistula following chemotherapy for lymphoma. Gulf J Oncolog.2008;(3):64-7.
11. Moran M, Bilgiç I, Dizen H, Dilektaşli E, Köseoğlu T, Özmen MM. Spontaneous Gastrosplenic Fistula Resulting from Primary Gastric Lymphoma: Case Report and Review of the Literature. Balkan Med J. 2011;2011(2):205-208.
12. Krause R, Larsen CR, Scholz FJ. Gastrosplenic fistula: complication of adenocarcinoma of stomach. Comput Med Imaging Graph. 1990;14(4):273-76.
13. Martinez JD, Moya L, Hernandez G, Viola L. Rev Gastroenterol Peru. 2015; 35:165-167.
14. Pizzirusso F, Gillet JP, Fobe D. Isolated spleen metastatic involvement from a colorectal adenocarcinoma complicated with a gastrosplenic fistula. A case report and literature review. Acta Chir Belg. 2004;104(2):214-6.
15. Cary ER, Tremaine WJ, Banks PM, Nagorney DM. Isolated Crohn's disease of the stomach. Mayo Clin Proc.1989;64(7):776-9.
16. Stoica T, Dragomir T, Gabrovescu D, Laky D, Marinescu M. Ulcer of the fornix perforated into the spleen, simulating a gastric diverticulum. Chirurgia (Bucur) 1973;22(509):12.
17. Joffe N, Antonioli DA. Penetration into spleen by benign gastric ulcer. Clin Radiol.1981; 32(2):177-181.
18. Glick SN, Levine MS, Teplick SK, Gasparaitis A. Splenic penetration by benign gastric ulcer: Preoperative recognition with C T. Radiology. 1987;163(3):637-9.
19. Montana L, Genser L, Cortes A, Poupardin E, Barrat C, Carandina S. Gastrosplenic Fistula with Gastrointestinal Bleeding: a Rare and Potentially Fatal Complication After Sleeve Gastrectomy. Obes Surg. 2018;28(7):2135-39.
20. Toman D, Foltýs A, Ostruszka P, Tulinský L, Martínek L. Successful treatment of gastrosplenic fistula after sleeve gastrectomy. Rozhl Chir. 2024;103(1):26-30.
21. Rothermel LD, Chadwick CL, Pillai TT. Gastrosplenic fistula: etiologies, diagnostic studies, and surgical management. Int. Surg. 2010;95(3):270–2.
22. Harris NL, Aisenberg AC, Meyer JE, Ellman L, Elman A. Diffuse large cell (histiocytic) lymphoma of the spleen. Clinical and pathologic characteristics of ten cases. Cancer. 1984;54(11):2460-7.
23. Frenkel A, Bichovsky Y, Perry ZH, Peiser J, Shapira AR, Brotfain E, et al. Management of gastrosplenic fistula in the emergency setting - A case report and review of the literature. Ann. Med. Surg (lond). 2018; 29:26-2.
24. Rizzo JF, Tomassetti EL, Esparragón JC, Cabrera LS, Hernandez JRH. Massive upper gastrointestinal bleeding secondary to gastrosplenic fistula. Rev. Española Enfermedades Dig. 2013;105(9):570-571.
25. Sousa M, Gomes A, Pignatelli N, Nunes V. Massive gastrointestinal bleeding after chemotherapy for gastric lymphoma. Int. J. Surg. Case Rep. 2016; 21:41-3.
26. Dellaportas D, Vezakis A, Fragulidis G, Tasoulis M, Karamitopoulou E, Polydorou A. Gastrosplenic fistula secondary to lymphoma, manifesting as upper gastrointestinal bleeding. Endoscopy. 2011;43(SUPPL. 2).
27. Gulati D, Tama M, Mutchnick M. Unusual Cause of Upper GI Bleed in a Patient with Lung Cancer. Gastroenterology 2014;147(2):287-8.