Gastrosplenic fistula in Hodgkin's lymphoma treated successfully by laparoscopic surgery and chemotherapy

Hamad I. Al-Ashgar, MD, Mohammed Q. Khan, MD, MRCP (UK), Abduallah M. Ghamdi, MD, Fahad Y. Bamehriz, MD, Irfan Maghfoor, MD.

ABSTRACT

A gastrosplenic fistula is a rare complication of a gastric or splenic lesion. We report a case of Hodgkin's lymphoma (nodular sclerosis) involving the spleen that was complicated by spontaneous gastrosplenic fistula. The fistula was closed laparoscopically, and the patient underwent partial gastrectomy and gastric wall repair, followed by successful chemotherapy. This is also the first reported case in published literature where closure of gastrosplenic fistula and partial gastrectomy was carried out laparoscopically. We recommend that extensive open surgical procedures including total gastrectomy, splenectomy, and pancreatectomy may be avoided in the management of gastrosplenic fistula, and the patient could be managed by less radical, simple laparoscopic fistulectomy, with partial gastric resection. If the fistula is caused by a malignant process, the surgical repair should be followed by definitive treatment with chemotherapy and radiotherapy.

Saudi Med J 2007; Vol. 28 (12): 1898-1900

From the Departments of Medicine, Division of Gastroenterology (Al-Ashgar, Khan, Ghamdi), General Surgery (Bamehriz), and Oncology (Maghfoor), King Faisal Specialist Hospital and Research Centre, Riyadh, Kingdom of Saudi Arabia.

Received 26th November 2006. Accepted 15th April 2007.

Address correspondence and reprint request to: Dr. Hamad I. Al-Ashgar, Division of Gastroenterology, Department of Medicine, King Faisal Specialist Hospital and Research Centre, PO Box 3354, Riyadh 11211, Kingdom of Saudi Arabia. Tel. +966 (1) 4424729. Fax. +966 (1) 442-7499. E-mail: alashgar@kfshrc.edu.sa

Gastrosplenic fistulas are extremely rare, usually arising as a complication of diseases of the spleen or the stomach. Malignancy is the primary cause reported in >50% of the patients, followed by perforated peptic ulcer (30%), Crohn's disease, and posttraumatic splenic abscess. Among the different histologic types of lymphoma, histiocytic lymphoma (diffuse large cell) appears to have a greater propensity to produce perforation or fistula formation.[1,2] Splenic malignant lymphoma, especially the diffuse, large cell type may be one cause of this distinctive complication as a tendency to gastric wall invasion and extensive tumor necrosis.[3-6] Gastrosplenic fistulas after chemotherapy for splenic lymphoma, and disseminated histocytic lymphoma have also been reported.[7,8] We report a case of gastrosplenic fistula secondary to Hodgkin's lymphoma (nodular sclerosis) highlighting the successful treatment by laparoscopic excision of fistula followed by chemotherapy.

Case Report. A 16-year-old girl was referred to our hospital in 2004 with a history of dysphagia, and a diagnosis of achalasia. Barium swallow, esophageal manometry, and endoscopy confirmed the diagnosis of achalasia, and she underwent endoscopic balloon dilatation. Post dilatation barium swallow showed no perforation and she was discharged home. During follow up visits to her referring hospital, she observed marked improvement in her dysphagia, however, she continued to lose weight, developed malaise and low grade fever. Four months after, she was referred back to our hospital with severe left sided chest pain, fever, with chills and rigors of one-week duration. On examination she was pale, febrile, tachycardic, and slightly tachypneic. She was found to have several enlarged lymph nodes in the left cervical region. Examination of the respiratory system revealed clinical signs of left sided pleural effusion. There was mild tenderness in the left upper quadrant of abdomen and splenomegaly. She underwent urgent upper gastrointestinal (GI) endoscopy to rule out esophago-pleural fistula, secondary to esophageal rupture caused by previous balloon dilatation. Endoscopy revealed an opening in the lateral part of the fundus with a blind end (**Figure 1**). Subsequent barium swallow reported a marked radiological improvement in her achalasia, however, barium meal (**Figure 2**) confirmed a communicating gastrosplenic fistula. Computed tomographic (CT) scan of

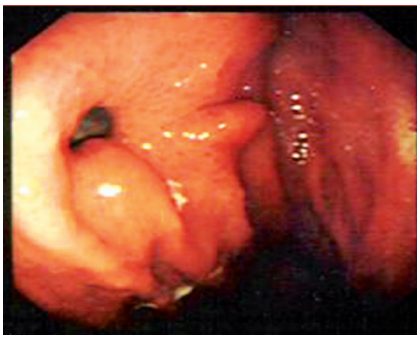

Figure 1 - Endoscopic view of gastrosplenic-fistula at the gastric fundus.

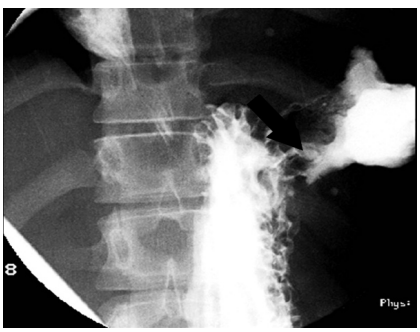

Figure 2 - Barium meal demonstrating gastro-splenic fistula (before surgery).

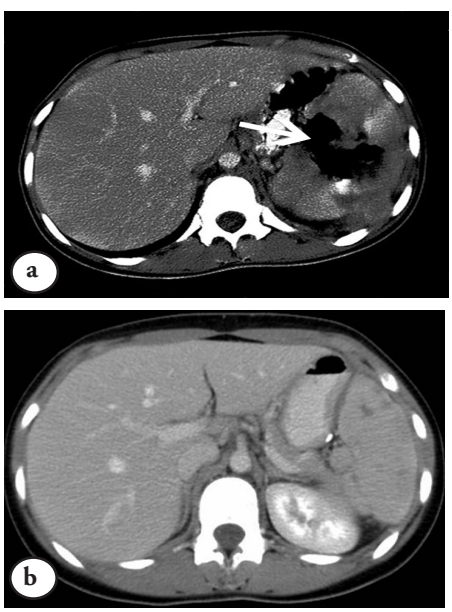

Figure 3 - Computed tomography of the abdomen showing **a)** fistula before surgery with central splenic necrosis (arrow showing gastrosplenic fistula), and **b)** complete resolution of gastro-splenic fistula after surgery and chemotherapy.

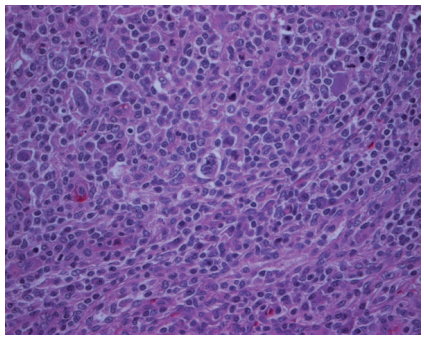

Figure 4 - Cervical lymph node biopsy (Hematoxylin and Eosin stain), showing classical Reed-Sternberg cell and mononuclear variant in polymorphous background in Hodgkin's disease.

abdomen and chest revealed para-aortic and hilar lymphadenopathy, marked splenomegaly, and presence of a large gastrosplenic fistula with air fluid levels at the splenic hilum (**Figure 3a**) suggestive of a splenic collection and abscess. Excision biopsy of a left cervical lymph node (**Figure 4**) revealed classical Hodgkin's lymphoma (nodular sclerosis type). She was staged as IIIS Hodgkin's lymphoma. She underwent laparoscopic exploration, which showed large retroperitoneal lymph nodes. The enlarged portion of the spleen, left lobe of the liver, and fundus of the stomach were matted together with fibrous adhesions and formed a fistula tract between fundus and the necrotic splenic hilum. The gastrosplenic fistula was around 2x2 cm in size. The fistula was excised from the stomach wall with partial removal of a part of the gastric fundus. The fluid from the splenic cavity was aspirated. The cavity in the spleen was properly irrigated and left with an outside drain. The cultures from the splenic necrotic fluids were negative. The postoperative period was uneventful. Laparoscopic surgical repair was followed by 7 cycles of doxorubicin, bleomycin, vinblastine, and dacarbazine chemotherapy. The chemotherapy was well tolerated, and she had a full recovery. The abdominal CT after surgery, and chemotherapy showed complete resolution of fistula (**Figure 3b**). She remains in complete clinical, and radiologic remission one year after completion of chemotherapy.

Discussion. Gastrosplenic fistula was first reported by Scoville et al[5] in 1962, from Belgium. They reported 2 patients with lymphoma of the spleen who developed gastrosplenic fistulas. One had a double fistula from the spleen to the colon and jejunum discovered at autopsy. The other was diagnosed radiographically by the presence of air within an enlarged spleen, termed "aerosplenomegalie" by the author.[5] The report highlighted that lymphomas causing splenomegaly with central necrosis have a tendency to adhere to, or develop

fistula to other organs. Spontaneous fistula formation may also occur after the successful treatment of lymphoma by radiation, chemotherapy, or combination. A review of available publications identified only 16 case reports of gastrosplenic fistula in the literature. In 6 of these cases, the fistula was associated with splenic lymphoma,[3-6] in 3 with chemotherapy for lymphoma,[7,8] in 4 cases with peptic ulcer disease,[9] and in one each with Crohn's disease,[10] adeno-carcinoma of stomach,[11] and post traumatic splenic abscess.[12] Fistula formation is a rare complication of primary and secondary lymphomas of the gastrointestinal tract. Among the different histological types, diffuse large cell (histocytic) non-Hodgkin's lymphomas appear to have a greater propensity to produce perforation or fistula formation. The common findings among all 9 reported cases of lymphomas were a moderate to massive splenomegaly with extensive central necrosis, involving >90% of the splenic parenchyma. As the malignant process invaded the wall of the stomach, subsequent necrosis of the common wall occurred with establishment of an abnormal communication through the necrotic center of the tumor. Similarly, in our case the gastrosplenic fistula occurred as a complication of splenomegaly with central necrosis secondary to Hodgkin's lymphoma (nodular sclerosis). Previous history of balloon dilatation for achalasia was not related to the formation of gastrosplenic fistula, and it was clearly a coincidental finding.

Management of gastrosplenic fistula requires urgent surgical intervention, as the cavity within the splenic parenchyma is being bathed by gastric juice, and erosion of the splenic vessels can lead to massive GI hemorrhage.[13] The patient of Bubenik et al,[4] with histocytic lymphoma and gastrosplenic fistula underwent an en bloc resection of the greater curvature of the stomach, splenectomy, and distal pancreatectomy, while in another patient of Bird et al,[13] splenic embolization was followed by near-total gastrectomy, splenectomy, and esophagogastrostomy for a splenic mass with erosion into the stomach. Harris et al[14] concluded that improved survival may be predicted by a less radical operation following chemotherapy and radiation treatment. In our case, we excised the fistula and performed partial gastrectomy laparoscopically. The splenic cavity was properly washed repeatedly and left with a drain. Carolin et al[8] recommended splenectomy in every case of gastrosplenic fistula to prevent potential bleeding. In our patient, however, splenectomy was not performed and the spleen size was markedly reduced after chemotherapy.

In conclusion, only a few cases of gastrosplenic fistula have been reported in non-Hodgkin's lymphoma, however, it could also occur in Hodgkin's disease involving the spleen. Gastrosplenic fistula due to malignant disease may not need radical surgery, as laparoscopic surgical closure of the fistula may be a better and less invasive option, and may allow definitive therapy of the malignant process to start earlier than after a conventional surgical exploration.

References

1. Allison JE. Gastrocolic fistula as a complication of gastric lymphoma. ***Am J Gastroenterol*** 1973; 59: 499-504.
2. Ehrlich AN, Stalder G, Geller W, Sherlock P. Gastrointestinal manifestations of malignant lymphoma. ***Gastroenterology*** 1968; 54: 1115-1121.
3. Blanchi A, Bour B, Alami O. Spontaneous gastrosplenic fistula revealing high-grade centroblastic lymphoma: endoscopic findings. ***Gastrointest Endosc*** 1995; 42: 587-589.
4. Bubenik O, Lopez MJ, Greco AO, Kraybill WG, Cherwitz DL. Gastrosplenic fistula following successful chemotherapy for disseminated histiocytic lymphoma. ***Cancer*** 1983; 52: 994-996.
5. de Scoville A, Bovy P, Demeester P. [Radiologic "aerosplenomegaly" caused by necrotizing splenic lymphosarcoma with double fistulization into the digestive tract]. ***Acta Gastroenterol Belg*** 1967; 30: 840-846. French
6. Choi JE, Chung HJ, Lee HG. Spontaneous gastrosplenic fistula: a rare complication of splenic diffuse large cell lymphoma. ***Abdom Imaging*** 2002; 27: 728-730.
7. Hiltunen KM, Airo I, Mattila J, Helve O. Massively bleeding gastrosplenic fistula following cytostatic chemotherapy of a malignant lymphoma. ***J Clin Gastroenterol*** 1991; 13: 478-481.
8. Carolin KA, Prakash SH, Silva YJ. Gastrosplenic fistulas: a case report and review of the literature. ***Am Surg*** 1997; 63: 1007-1010.
9. Joffe N, Antonioli DA. Penetration into spleen by benign gastric ulcers. ***Clin Radiol*** 1981; 32: 177-181.
10. Cary ER, Tremaine WJ, Banks PM, Nagorney DM. Isolated Crohn's disease of the stomach. ***Mayo Clin Proc*** 1989; 64: 776-779.
11. Krause R, Larsen CR, Scholz FJ. Gastrosplenic fistula: complication of adenocarcinoma of stomach. ***Comput Med Imaging Graph*** 1990; 14: 273-276.
12. Nikolaidis N, Giouleme O, Gkisakis D, Grammatikos N. Posttraumatic splenic abscess with gastrosplenic fistula. ***Gastrointest Endosc*** 2005; 61: 771-772.
13. Bird MA, Amjadi D, Behrns KE. Primary splenic lymphoma complicated by hematemesis and gastric erosion. ***South Med J*** 2002; 95: 941-942.
14. 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: 2460-2467.