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

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ABSTRACT

A gastrospenic 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 gastrospenic 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 gastrospenic 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 gastrospenic 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.

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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 gastrospenic fistula. Computed tomographic (CT) scan of

Gastrospenic 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 Gastrospenic fistulas after chemotherapy for splenic lymphoma, and disseminated histocytic lymphoma have also been reported.7,8 We report a case of gastrospenic fistula secondary to Hodgkin’s lymphoma (nodular sclerosis) highlighting the successful treatment by laparoscopic excision of fistula followed by chemotherapy.
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 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. 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, in 3 with chemotherapy for lymphoma, in 4 cases with peptic ulcer disease, and in one each with Crohn’s disease, adeno-carcinoma of stomach, and post traumatic splenic abscess. Fistula formation is a rare complication of primary and secondary lymphomas of the gastrointestinal tract. Among the different histological types, diffuse large cell (histiocytic) 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. The patient of Bubenik et al., with histiocytic 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., splenic embolization was followed by near-total gastrectomy, splenectomy, and esophagogastronomy for a splenic mass with erosion into the stomach. Harris et al. 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. 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