

Case Report

Upper Gastrointestinal Bleeding from Gastric Splenosis; A Case Report and Literature Review

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Splenosis is a common condition found in a case that has a history of splenic trauma or splenectomy. It is usually a non-significant condition in clinical practice. However, splenosis can give rise to some complications including gastrointestinal hemorrhage as in the present case. The authors report here a case of gastric splenosis presenting with active upper gastrointestinal hemorrhage that was eventually managed with surgical resection, and the literature regarding splenosis was reviewed.

Keywords: Splenosis, Upper gastrointestinal bleeding, Gastric

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Splenosis is the autotransplantation of splenic tissue, usually following splenic trauma or splenectomy⁽¹⁾. Although, this condition is common in postsplenectomized patients⁽²⁾, it has rarely been seen in general standard textbooks. Most splenotic cases are asymptomatic. However, splenosis can give rise to some complications including gastrointestinal hemorrhage⁽³⁾. Herein, the authors describe a patient who presented with active upper gastrointestinal bleeding from intramural splenosis of the stomach.

Case Report

A 16-year-old Thai woman presented at the emergency room with a chief complaint of hematemesis during the past two hours. She developed hematemesis about 200 ml. At the emergency room, she vomited another 150 ml of fresh blood. She had been diagnosed as beta-thalassemia/hemoglobin E since she was a preschool child and had received blood transfusion one to two times a month. Eleven years ago, she underwent splenectomy because of splenomagaly. However, she still needed blood transfusion monthly afterward. In the past one year, she was admitted in a provincial hospital because of an episode of non-active hematemesis, but no further endoscopy or other specific investigation was done.

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Pertinent physical examination revealed a blood pressure of 110/70 mmHg, and a pulse rate of 84/min. The respiratory rate and temperature were in the normal range. Her eyes showed moderately pale conjunctivae. There was a surgical scar on the left subcostal area. The abdomen was soft and non-tender. The liver could be palpable, two fingerbreadths below right costal margin. Rectal examination revealed melena. Other findings were unremarkable. A nasogastric tube was put into the stomach and 150 ml of fresh blood was obtained. Laboratory evaluation showed the hematocrit of 21%. The blood smear findings showed moderately hypochromic and microcytic with marked anisocytosis, but no Howell-Jolly body was seen. Other laboratory values were unremarkable.

Esophagogastroduodenoscopy (EGD) was performed, revealing a submucosal mass, 4 cm in diameter with a small ulcer at its center, locating at the proximal part of the gastric body. CT scan of the abdomen showed a lobulated well-defined enhancing soft tissue with internal curvilinear calcification at left upper quadrant causing mass effect to the gastric body and fundus as seen in Fig. 1 and 2. This mass was well enhanced on arterial phase and becoming slight hyperdense on venous phase.

Exploratory laparotomy was performed in order to get rid of the mass that was supposed to be the cause of bleeding. During the operation, a large, irregular surface, dark-brown color, intraperitoneal mass, consistent with splenic tissue, was seen in the

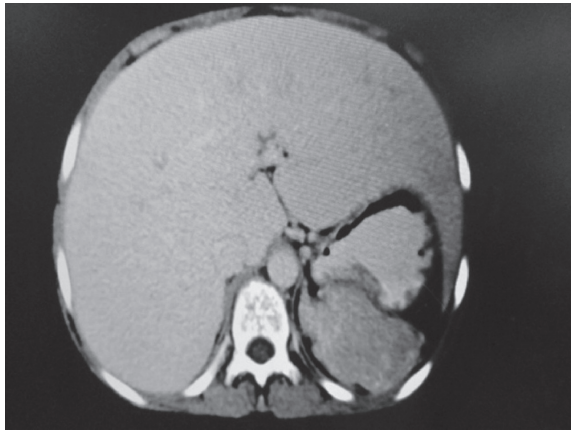


Fig. 1 An axial view from the contrast-enhanced CT scan on the portovenous phase shows a slight hyperdense mass, 7 cm in diameter, at left upper quadrant, causing mass effect to gastric fundus.

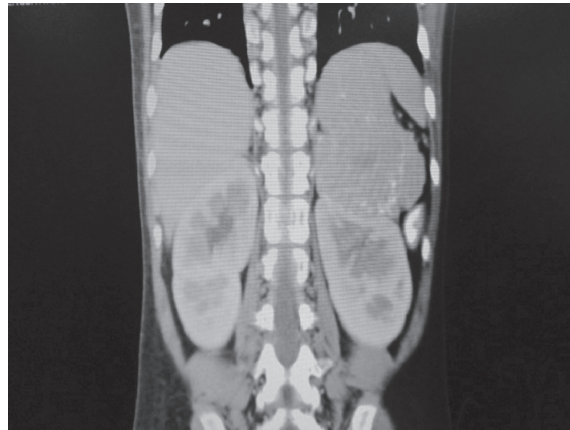


Fig. 2 A coronal view from the contrast-enhanced CT scan on the portovenous phase, shows this huge mass at the posterior left upper quadrant, pressing the upper pole of left kidney.

splenic fossa, at the left upper abdomen, and attaching the diaphragm. Some part of this mass adhered to the proximal greater curvature of the stomach. Wedge resection was done at the involved stomach by a linear stapler and partial removal of the adhering mass was performed. The gross specimen showed a dark-red mass occupying into the gastric wall. The histopathologic examination revealed a well-encapsulated mass containing red and white pulp, confirming splenic heterotopia.

The patient had an uneventful postoperative recovery. She has been symptom-free with no episode of hematemesis or melena for three years after the operation.

Discussion

This patient presented with an episode of active upper gastrointestinal (GI) hemorrhage. EGD was done and found a submucosal mass with an ulcer at the gastric body. CT scan of the abdomen showed a large lobulated mass at the left subdiaphragmatic area, adhering and pressing the stomach. The differential diagnosis included gastrointestinal stromal tumor, lymphoma, or leiomyoma. However, with a history of splenectomy, splenosis should be included in the differential diagnosis as well.

Post traumatic ectopic splenic tissue implantation, though initially reported in humans by Von Kuttner in an autopsy case with a history of splenectomy for trauma⁽¹⁾, was first reported with the term of “splenosis” by Buchbinder and Lipkoff in 1939⁽⁴⁾. Up to now, there is no epidemiological data

regarding the exact incidence of splenosis, however its incidence in patients who underwent splenectomy from splenic injury has been reported to be as high as 65 to 75%^(2,5,6). Its gross appearance can be found as reddish-blue color nodules, with different sizes and shapes⁽⁷⁾.

Although splenosis is usually found on the visceral and parietal peritoneum in the abdominal cavity, particularly in the left upper quadrant and pelvis^(8,9), it has also been reported elsewhere even in the thorax⁽¹⁰⁾, pericardium⁽¹¹⁾, subcutaneous tissue⁽¹²⁾, and brain⁽¹³⁾. There are two possible mechanisms regarding autotransplantation and formation of splenosis. The more common one is the result of dislodging of the splenic tissue from the ruptured spleen and then seeding on the peritoneal surface of adjacent organs or parietal peritoneum⁽¹⁴⁾. The other pathway is from hematogenous spreading to more distant organs⁽¹⁵⁾.

Some confusion may exist between the terms “splenosis” and “accessory spleen”. The former is an acquired condition whereas the latter is a congenital one. The number of accessory spleen usually found are less than 6, but splenosis is largely varied in number, ranging from a single to hundreds lesions. An accessory spleen has normal splenic histology and is supplied by branches of splenic artery. On the other hand, splenosis derives the blood supply from surrounding tissue and its histology can be varied, ranging from the same as a normal spleen to distorted architecture, lacking trabecular structure and poorly formed white pulp. The location of an accessory spleen is usually near the

splenopancreatic or gastrosplenic ligament whereas splenosis can be found anywhere. Although accessory spleen can be a cause of submucosal mass of the stomach⁽¹⁶⁾, until now there has been no report indicating its association with gastric ulcer or upper gastrointestinal hemorrhage, which on the other hand, can result from splenosis.

Because of the similarity of their histology, splenosis has been thought to be capable of normal splenic function. An evidence supporting this is the disappearance of Howell-Jolly bodies in the peripheral blood smear of the splenotic patients who have a history of splenectomy^(7,8). For the immunologic function, the data from some animal studies showed that some amount of splenic tissue implantation after splenectomy could restore the bacterial protective function, however, the evidence from case series in humans is still inconclusive⁽¹⁷⁾. Despite the fact that there is some level of immunologic function in splenotic patients, it cannot be showed that this function is associated with a decrease of mortality related to infection⁽¹⁸⁾.

Splenosis can be symptomatic many years after the onset of trauma. The pathologic conditions reported in various organs include hemorrhage from stomach, small bowel, and colon, spontaneous intraabdominal hemorrhage, intestinal obstruction, and relapse of hematologic disease, as shown in Table 1^(1,9,12,19-40). Some patients may develop abdominal pain that possibly results from a twisted pedicle or a lymphoid response to infection, though this has never been proved yet⁽¹⁾.

In cases of upper GI hemorrhage, the location of splenosis that is highly associated with the incident of bleeding is in the wall of the gastric fundus⁽³⁾. The mechanism of bleeding in these cases was the same as gastric leiomyoma that causes gastric bleeding⁽¹⁾. Although a case of massive upper GI bleeding from splenosis has been reported⁽³⁶⁾, most GI hemorrhage cases from splenosis were not massive⁽³⁾ and no death case associated with splenosis has been reported up to now.

Most splenosis patients are asymptomatic. The lesion can be found incidentally during computed tomography (CT) scan or magnetic resonance imaging (MRI) examination⁽⁴¹⁾. On non-contrast CT scan, splenosis can be seen as single or multiple, well lobulated hypodense masses. After contrast administration, they will show heterogeneous enhancement in the arterial phase and iso- or slightly hypodense in the portal phase⁽⁴²⁾. The pre-contrast

MRI reveals its being homogeneously hypointense on T1-weighted images, and hyperintense on T2-weighted images, and after contrast administration, it will be hyperintense compared to the liver⁽⁴¹⁾. However, these findings are not specific enough to provide the definitive diagnosis⁽⁷⁾. More specific examination with MRI was reported by using superparamagnetic iron oxide (SPIO) intravenous administration. This agent is more specific to the phagocytic reticuloendothelial cells of the liver and spleen, providing specific signal intensity on MRI⁽⁴¹⁾, thus it has been used for detecting splenosis⁽⁴³⁾. Another diagnostic modality that has been supposed to be the non-invasive method of choice for the diagnosis of splenosis because of its high sensitivity and specificity is the nuclear scintigraphy by using Technetium-99m heat-damaged red blood cell (RBC)⁽⁴³⁾. It is more sensitive in detection of splenosis than CT or MRI, particularly in case of multiple, small lesions⁽⁴¹⁾. For the intramural lesion of gastrointestinal tract, like in the present case, localization of it can be done with the endoscopy in which it may be seen as a submucosal mass. However, this cannot be clearly defined as distinct from other submucosal tumors.

A tissue biopsy may be needed when the diagnosis is still uncertain or malignancy is concerned. CT-guided needle biopsy can be used in cases of peritoneal, pelvic, or intrahepatic splenosis. Endoluminal ultrasonography (EUS) with fine needle aspiration (FNA) of the targeted tissue may be helpful in the case of submucosal splenosis particularly, of the stomach⁽⁴⁴⁾.

Because it is a benign condition, excision of splenosis in asymptomatic patients is not mandatory. Removal of splenosis is indicated in symptomatic cases or those who develop a complication such as hemorrhage, obstruction, and abdominal pain^(29,34). In postsplenectomized patients found to have splenosis with some hematologic diseases such as Felty's syndrome, idiopathic thrombocytopenia purpura and autoimmune hemolytic anemia, removal of splenosis could resolve the disease⁽¹⁴⁾. In the case of asymptomatic intramural gastric splenosis, there has been no consensus whether it should be removed to prevent the risk for developing any complications in the future such as hemorrhage. However, there was a report of the asymptomatic intramural gastric splenosis that subsequently became larger and developed an ulcer over the mass⁽³⁵⁾. Thus, if the removal is not performed in such a case, follow-up examination will be required.

Table 1. Pathologic conditions of splenosis in various organs

Organ involved	Authors	Presenting problem	Diagnostic method	Treatment
Appendix	Brewster et al. ⁽¹⁹⁾	Acute abdominal pain	Laparotomy	Appendectomy with tumor removal
Colon and rectum	Gineu et al. ⁽²⁰⁾	Colonic obstruction (from extraluminal mass compression)	CT scan, MRI, colonoscopy	Tumor removal
	Kravarusic et al. ⁽²¹⁾	Relapse of spheroctytosis	Ultrasonography, RBC scan	Colectomy with tumor removal
Liver	Obokhare et al. ⁽²²⁾	Chronic abdominal pain and hemorrhage	CT scan, colonoscopy	Colectomy with tumor removal
	Brancatelli et al. ⁽²³⁾	Chronic abdominal pain	CT scan, MRI, RBC scan, and core biopsy	No report
Pelvis and gynecologic organ	Imbriaco et al. ⁽²⁴⁾	Chronic abdominal pain	Ultrasonography, CT scan, MRI	Tumor removal
	Desai et al. ⁽²⁵⁾	Acute abdominal pain	Ultrasonography, laparoscopy	Tumor removal
	Feferman et al. ⁽²⁶⁾	Acute abdominal pain with intraperitoneal hemorrhage	Laparotomy	Tumor removal
	Zitzer et al. ⁽²⁷⁾	Chronic pelvic pain	Ultrasonography	Salpingo-oophorectomy with tumor removal
Peritonium and retroperitonium	Depypere et al. ⁽²⁸⁾	Acute abdominal pain with intraperitoneal hemorrhage	CT scan, laparoscopy	Tumor removal
	Katz et al. ⁽²⁹⁾	Acute abdominal pain with intraperitoneal hemorrhage	CT scan	Medication
Small bowel	Lansdale et al. ⁽³⁰⁾	Relapse of thrombocytopenia	Ultrasonography, RBC scan	Tumor removal
	Sitrnek et al. ⁽³¹⁾	Colonic obstruction (from extraluminal mass compression)	Barrium enema and CT scan	Tumor removal
	Valli et al. ⁽³²⁾	Chronic abdominal pain	Ultrasonography, MRI	Tumor removal
Stomach	Abeles et al. ⁽³³⁾	Occult hemorrhage with intussusceptions	CT scan	Small bowel resection with tumor removal
	Bastile et al. ⁽¹⁾	Hemorrhage	EGD, RBC scan, angiography, laparotomy	Small bowel resection with tumor removal
Subcutaneous tissue	Arroja et al. ⁽³⁴⁾	Hemorrhage	CT scan, EGD and RBC scan	Medication
	Deutsch et al. ⁽³⁵⁾	Dyspepsia	EGD, EUS	Tumor removal
	Margari et al. ⁽³⁶⁾	Hemorrhage (massive)	CT scan, EGD	Tumor removal
Thorax	Boudova et al. ⁽³⁷⁾	Mass at the surgical scar	Excision biopsy	Tumor removal
	Chang et al. ⁽¹²⁾	Mass at the gun shot wound scar	Excision biopsy	Tumor removal
Multiple organs	Cordier et al. ⁽³⁸⁾	Hemoptysis	CT scan, MRI, RBC scan	Tumor removal
	Fukuhara et al. ⁽³⁹⁾	Chronic chest pain	CT scan, RBC scan	Tumor removal
Liver and peritonium	Sikov et al. ⁽⁹⁾	Occult hemorrhage	EGD, colonoscopy, CT scan, RBC scan, angiography	Small bowel resection and subtotal colectomy
	Yasar et al. ⁽⁴⁰⁾	Abdominal discomfort	CT scan, laparoscopy with biopsy	No report

CT scan = compute tomographic scan; EGD = esophagogastroduodenoscopy; MRI = magnetic resonance imaging; RBC scan = Technetium 99m radionuclide red blood cell scan

Conclusion

A rare case of gastric splenosis causing upper gastrointestinal bleeding which was successfully treated with surgery is presented. Splenosis should be included in the differential diagnosis in patients who have a history of splenic trauma or splenectomy presenting with gastrointestinal hemorrhage. Preoperative diagnosis of this condition is difficult; however, it can be made by using important data from the patients' clinical history and appropriate investigative imaging.

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Potential conflicts of interest

None.

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ภาวะเลือดออกจากทางเดินอาหารส่วนบนที่มีสาเหตุจากภาวะ splenosis ในกระเพาะอาหาร: รายงานผู้ป่วยหนึ่งราย และการทบทวนวารสารทางการแพทย์

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Splenosis เป็นภาวะที่พบบ่อยในผู้ป่วยที่มีประวัติการบาดเจ็บของม้ามหรือเคยได้รับการตัดม้าม ภาวะนี้มักจะไม่มีความสำคัญในเวชปฏิบัติทั่วไป อย่างไรก็ตาม splenosis สามารถทำให้เกิดภาวะแทรกซ้อนต่างๆ ได้แก่ ภาวะเลือดออกในทางเดินอาหาร ดังเช่นในผู้ป่วยรายนี้ คณะผู้นิพนธ์ได้รายงานผู้ป่วยหนึ่งรายที่เป็น splenosis บริเวณกระเพาะอาหารที่มาด้วยอาการเลือดออก จากทางเดินอาหารส่วนบนและได้รับการรักษาโดยการผ่าตัด คณะผู้นิพนธ์ยังได้ทบทวนรายงานจากวารสารต่างๆ ที่เกี่ยวข้องกับ ภาวะ splenosis
