Gastric fundus splenosis with hemangioma masquerading as a gastrointestinal stromal tumor in a patient with schistosomiasis and cirrhosis who underwent splenectomy

A case report and literature review

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Abstract

Rationale: Splenosis is the heterotopic auto-transplantation of the splenic tissues. Gastric splenosis in a rare location mimics a gastrointestinal stromal tumor (GIST). Gastric splenosis with hemangioma has not been reported throughout the literature.

Patient concerns: We report a case of a 74-year-old schistosomiasis cirrhosis splenectomy woman diagnosed with gastric fundus mass. Preoperative computed tomography and endoscopic ultrasonography revealed findings suggestive of a GIST.

Diagnoses: The mass located in the gastric fundus muscularis propria, measuring $3.9 \times 2.8 \times 2.4 \text{cm}$ with a dark red color, was removed by surgery. In the mass, a $1 \times 1 \text{cm}$ red-purple nodule was also found. On microscopic examination, a well-formed splenic tissue divided into two compartments–white pulp and red pulp–separated by an ill-defined interphase known as the marginal zone. However, a nodule in the heterotopic spleen was mainly composed of larger thin-walled muscular vessels. The final diagnosis was gastric splenosis with hemangioma.

Interventions: After discussion in a multidisciplinary conference, the patient was considered for a GIST resection under gastroscopy. In the process of peeling, the surface of the mucosal, submucosal, muscle layers and the tumor surface were diffusely oozing. The effect of electrocoagulation and hemostasis was extremely poor. Therefore, endoscopic surgery was arrested. After dealing with the patient’s family, a combination of laparoscopic-gastroscope double-mirror surgery was decided in accordance with the principle of minimally invasive surgery to preserve the stomach. Considering the great possibility of a malignant GIST, we still decided to continue to traditional surgical resection. The tumor was then removed via surgery.

Outcomes: The patient was favorable with healing and discharged on postoperative day 10.

Lessons: Gastric splenosis with an associated hemangioma is the first well-documented case. Its pathogenesis may be direct implantation. Appropriate medical history taking and Tc-99m heat-denatured RBC spleen scintigraphy (Tc-99mHDRS) are valuable for its diagnosis; however, pathology is the gold standard. Surgery is a reasonable treatment for gastric splenosis with hemangioma.

Abbreviations: CA = cancer antigen, CD = cluster of differentiation, CT = computed tomography, FNA = fine-needle aspiration, GIST = gastrointestinal stromal tumor, H&E = hematoxylin and eosin, MRI = magnetic resonance imaging, Tc-99mHDRS = Tc-99m heat-denatured RBC spleen scintigraphy.

Keywords: gastric fundus, hemangioma, schistosomiasis cirrhosis, splenectomy, splenosis

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1. Introduction

Splenosis is the heterotopic auto-transplantation of the splenic tissues arising from traumatic ruptures or iatrogenic splenectomy.\(^1\) Its incidence has been reported to be 67% for traumatic ruptures.\(^2\) Splenosis in the gastric area is a rare location for seeding or spreading. However, gastric splenosis with hemangioma has not been reported throughout the literature. Herein, we report a rare case of a schistosomiasis cirrhosis splenectomy in an asymptomatic elderly woman who had a splenosis with hemangioma located in the gastric fundus wall, which was misdiagnosed as a gastrointestinal stromal tumor (GIST) before on preoperative imaging.

2. Case report

A 74-year-old woman was admitted to the Gastroenterology Department of our hospital for an asymptomatic gastric mass. She had a schistosomiasis cirrhosis splenectomy at the age of 29 years.

The patient was initially submitted to a computed tomography (CT) scan for pneumonia in other hospitals, which revealed pipe stem cirrhosis (Fig. 1A), a well-demarcated 4-cm solid mass confined to the gastric wall suggestive of a GIST (Fig. 1B), and a 1-cm low-density lesion with a clear outline in the mass (Fig. 1B; red arrow). Thereafter, she was submitted to an upper gastrointestinal endoscopy in our hospitals, which revealed a smooth and rounded mass in the gastric wall without mucosal infiltration (Fig. 1C) at the level of the greater curvature. Endoscopic ultrasonography revealed a 3.95 × 2.82-cm slightly low-level echoic homogeneous mass derived from the muscularis propria (Fig. 1D) and a 1 × 1-cm lower level echoic area with a clear boundary in the mass (Fig. 1D; red arrow); these findings confirmed the diagnosis of a gastric GIST. The laboratory test findings were normal, except for the following: platelet count of 369 × 10^9/L, glutamyl transpeptidase level of 53.4 U/L, total bilirubin level of 22.4 μmol/L, serum creatinine level of 44.0 μmol/L, potassium level of 3.5 mmol/L, and levels of other serum tumor markers (cancer antigen [CA], cytokeratin 19, alpha fetoprotein, carcinoembryonic antigen, CA125, and CA15-3).

After discussion in a multidisciplinary conference, the patient was considered for a GIST resection under gastroscopy.

Under the gastroscopic, a large submucosal uplift was seen near the posterior wall of the gastric angle. The surface of the mucosa was hyperemic and edematous; the texture was hard; and the activity was poor. After dual-knife labeling, the mucosa and submucosa were cut open, and the mass was exposed in the muscularis propria of the stomach fundus wall. The surface of the mass was covered by larger blood vessels. CT = computed tomography, GIST = gastrointestinal stromal tumor.
preserve the stomach. In the process of laparoscopic umbilical puncture point incision, the intestinal mucosa was perforated and was thus subsequently repaired. Owing to the patient’s history of 2 abdominal surgeries, several adhesions were seen during laparoscopic surgery, which were then slowly separated. However, the tumor location was high and concealed (gastric angle near the posterior wall); even after following gastroscopy positioning instructions, the tumor still could not be found under laparoscopic direct vision. Therefore, we stopped the double-mirror combination surgery plan. Based on what was seen during the surgery, we communicated with the patient’s family again. Considering the great possibility of a malignant GIST, we still decided to continue the traditional surgical resection. The tumor was then removed via surgery; its size was approximately 3.5 × 5 cm, and its blood supply was extremely rich. The abdominal drainage tube and gastrointestinal decompression tube were indwelling. The patient’s vital signs were stable; she was then transferred to the intensive care unit and discharged on postoperative day 10.

On macroscopic examination, 3.9 × 2.8 × 2.4-cm dark red masses surrounded by a completely thin capsule were observed in the gastric fundus muscularis propria. On the cut surface, the mass appeared red to bluish with scattered white tiny nodules embedded in the muscularis propria. At the edge of the mass, an approximately 1 × 1-cm nodule appearing as a circumscribed, non-encapsulated, honeycomb-like, and red-purple nodule, which formed with dilated congested vascular space with bleeding, was also observed. On microscopic examination, a well-formed splenic tissue divided into 2 compartments—white pulp and red pulp—was separated by an ill-defined interphase known as the marginal zone (Fig. 2A). However, a nodule in the heterotopic spleen was mainly composed of larger thin-walled muscular vessels, which were variably dilated and occasionally displayed thrombosis. The widely dilated vessels showed attenuation of their walls, mimicking a cavernous hemangioma (Fig. 2B). Immuno-phenotypically, the endothelial lining cells of the vascular walls were immunoreactive for cluster of differentiation (CD) 31 (Fig. 2C), CD34 (Fig. 2D), and Factor VIII (Fig. 2E).

The final diagnosis was gastric fundus splenosis with an associated hemangioma.

3. Discussion
Splenosis was originally described in 1937 by Shaw and Shaf. However, the term “splenosis,” first used by Buchbinder and Lipkopf in 1939, refers to the dissemination with heterotopic auto-transplantation and implantation of the splenic tissues, which may follow disruption of the spleen’s capsule by trauma or iatrogenic splenectomy. Splenosis is commonly found in the abdominal and pelvic cavity, including the greater omentum, serosal surface of the small bowel, parietal peritoneum, mesentery, and diaphragm. Additional potential sites of implantation, including intrathoracic, intragastric, and intra-hepatic areas, lungs, kidneys, and brain, were reported.

The age of patients with gastric splenosis ranged from 17 to 68 years; the mean age was 44 years, and the median age was 42 years. The interval time from splenectomy to its diagnosis was from 4 to 38 years; the mean interval time was 14.36 years, and the median interval time was 12 years. The size ranged from 1.1 to 5 cm; the mean size was 2.31 cm, and the median size was 2.0 cm. The chief cause of splenectomy related to this condition was traumatic rupture (70%); the other cause was iatrogenic reasons (30%). The main primary suspected diagnosis was GISTs (47%); the other diagnoses included upper gastrointestinal bleeding (17%), gastric mass (12%), gastric smooth muscle tumor (6%; may be GISTs), dyspepsia (6%), gastric splenosis (6%), and gastric band...
ineffectiveness (6%). The leading method of confirmation was pathology (76%), followed by Tc-99m heat-denatured RBC spleen scintigraphy (Tc-99mHDRS) (12%), cytology (6%), and fine-needle aspiration (FNA) (6%) (Table 1).[1,2,4–6,11–21]

The pathogenesis of splenosis remains unclear. Some hypotheses to explain such have been proposed. The seeding/implantation hypothesis in which the isolated splenic pulp may survive after seeding into any position of the abdominal cavity has been demonstrated by animal experiments.[1] However, this hypothesis fails to explain cases found on other sites, such as the liver and lungs. A recent research showed that intra-organ splenosis could spread through the vasculature into the liver and the lungs and that the morphologic and immunologic structures formed in these regenerated autografts were influenced by the organ vasculature and extracellular matrix wherein the tissue fragments settle.[12,13] Although the hematogenous spread of gastric splenosis is possible, a direct implantation of splenic cells might be caused by needle transfixion during surgical hemoostatic maneuvers at the time of emergency splenectomy.[2]

Splenosis in the gastric area is an exceptional location for seeding or spreading. To date, no more than 20 cases have been reported. To the best of our knowledge, gastric splenosis with hemangiomia has not been reported throughout the literature. It is mainly composed of red and white pulps; however, it does not comprise all kinds of cells in the normal spleen. The implant’s or spread spleen’s undifferentiated reticular cells are induced to differentiate into the endothelial antrum, capillary vessel, and lymphocytes, which finally create the splenic tissues.[11] Splenic hemangiomia is the most common benign neoplasm of the spleen. Splenic hemangiomia are typically solitary lesions and appear as circumscribed, non-encapsulated, honeycomb-like, red-purple masses that frequently blend imperceptibly into the surrounding splenic parenchyma. Microscopically, the majority of splenic hemangiomia are cavernous in nature. A pure capillary architecture is less common, with many lesions containing varying proportions of both cavernous and capillary components. Immuno-phenotypically, splenic hemangiomia show reactivity of the endothelial lining cells of the vascular markers CD31, Von Willebrand factor, Ulex europaeus lectin I, and CD34.[23] Other types of splenic hemangiomia include venous hemangiomia, benign (infantile) hemangioendothelioma, and diffuse sinusoidal hemangiomatosis.

Gastric splenosis is usually asymptomatic and is only incidentally found in most circumstances. Only in a few cases, patients have upper gastrointestinal bleeding. It is a challenge to diagnose such. Based on the literature statistics, we found that most gastric splenosis cases had been misinterpreted as GISTs. The reason is that conventional ultrasound, CT, and magnetic resonance imaging (MRI) lack typical features to distinguish it from GISTs. Tc-99mHDRS is deemed to be the optimal method

Table 1

| Case                  | Age, y/Sex | Reason of splenectomy | Time interval, years | Number | Location                        | Size, cm | Primary suspected diagnosis | Confirmed method |
|-----------------------|------------|------------------------|----------------------|--------|---------------------------------|----------|-----------------------------|------------------|
| Wang et al[11]        | 40/M       | Traumatic rupture      | 6                    | 1      | Posterior wall of the stomach   | 2 × 1.5  | GIST                        | Pathology        |
| Reingles et al[12]    | 52/M       | Traumatic rupture      | 38                   | 1      | Gastric fundus                  | 1.3      | Gastrointestinal bleeding   | Pathology        |
| Nicolas et al[13]     | 30/F       | Traumatic rupture      | 10                   | 1      | Greater curvature of the stomach| NM       | Gastrointestinal bleeding   | Pathology        |
| Elwir et al[4]        | 20/F       | Splenopancreatectomy   | 4                    | 1      | Cardia                          | 2.1 × 1.8| Dyspepsia                   | Cytology and FNA |
| Carrera et al[6]      | 65/F       | Splenopancreatectomy   | 14                   | 1      | Gastric fundus                  | 1.8      | GIST                        | FNA              |
| Yang et al[2]         | 53/M       | Traumatic rupture      | 8                    | 1      | Anterior wall of the stomach    | 2.5 × 2  | GIST                        | Pathology        |
| Li et al[1]           | 40/F       | Traumatic rupture      | 20                   | 1      | Gastric fundus                  | 2 × 1.5  | GIST                        | Pathology        |
| Li et al[1]           | 32/M       | Traumatic rupture      | 4                    | 1      | Gastric fundus                  | 2 × 2    | GIST                        | Pathology        |
| Chung et al[15]       | 17/F       | Splenectomy for a giant epithelial splenic cyst | 8 | 2 | Posterior wall of the stomach | 1.1 × 0.9 × 1.1 and 1.9 × 1.6 × 1.7 | GIST | Pathology                  |
| Yang et al[4]         | 42/M       | Traumatic rupture      | 17                   | 1      | Gastric fundus                  | 5        | GIST                        | Pathology        |
| Mineccia et al[16]    | 17/F       | Traumatic rupture      | 5                    | 1      | Gastric fundus                  | 2.5 × 2.5 × 0.9 | GIST | Pathology                  |
| Alvite Canosa et al[17]| 49/M       | Traumatic rupture      | 21                   | 1      | Gastric fundus                  | 3 × 1    | Upper gastrointestinal bleeding | Pathology        |
| Amoja et al[18]       | 68/M       | Traumatic rupture      | 30                   | 1      | Greater curvature of the stomach | NM       | Gastric splenosis            | Tc-99mHDRS       |
| Falk et al[19]        | 64/F       | Splenopancreatectomy   | 7                    | 1      | Posterior wall of the stomach   | 3        | Gastric mass                 | Pathology        |
| Deutsch et al[20]     | 67/M       | Traumatic rupture      | 21                   | 1      | Lesser curve of the stomach     | NM       | Gastric smooth muscle tumor  | Pathology        |
| Laszewicz et al[1]    | 40/M       | NM                     | 1                    |        | Posterior wall of the stomach   | 1.5 × 1  | Upper gastrointestinal bleeding | Pathology        |
| Agha et al[21]        | 52/M       | Traumatic rupture      | 20                   | 1      | Gastric fundus                  | 3 × 2    | Gastric fundic mass          | Tc-99mHDRS       |

F = female, FNA = fine-needle aspiration, GIST = gastrointestinal stromal tumour, M = male, NM = not mentioned.
for diagnosis of splenosis because it has a higher uptake by the splenic tissues than Tc-99m sulfur colloid scan.\(^5\)\(^,\)\(^6\) Furthermore, superparamagnetic iron oxide-enhanced MRI is a useful diagnostic tool to distinguish gastric splenosis.\(^7\)\(^,\)\(^8\) However, a careful medical history inquiry may provide valuable clues for the differential diagnosis between gastric splenosis and GISTs. On pathology, it is easy to distinguish between the two. Gastric splenosis is mainly composed of red and white pulp. The cells of the red pulp sinusoids are bi-phenotypic immunoreactive for vascular (CD31 and Von Willebrand factor) and histiocytic markers (CD68 and lysozyme).\(^9\)\(^,\)\(^10\) The immunoreactivity for CD8 provides additional evidence for the presence of normal subsets of splenic red pulp lining cells.\(^11\) Most splenic hemangiomas are of cavernous or capillary in nature or of varying proportions of both and have immunoreactivity for vascular markers (CD31, Von Willebrand factor, and CD34). GISTs are mainly composed of spindle cells or epithelioid cells or both in different proportions. The immunoreactivity for CD34 and CD117 discovered on gastrointestinal stromal tumor-1 is helpful for the definitive diagnosis of GISTs. Splenosis is a benign disease and may have some immunologic and splenic-filtering functions, which may be favorable for the organism.\(^9\)\(^,\)\(^12\) Therefore, it is important to establish a correct diagnosis before preoperative imaging to avoid unnecessary surgery, and a thorough follow-up is beneficial in most circumstances. If the patient has clinical symptoms, and a surgery is inevitable, endoscopic or laparoscopy resection or traditional surgery can be selected on the basis of the size or location of the gastric splenosis. If the diagnosis is still unclear, further FNA and cytology examination are recommended.

## 4. Conclusion

In conclusion, gastric fundus splenosis with hemangioma is a unique case with a rare location for seeding or spreading; it is asymptomatic and an incidental finding. It has been misinterpreted as a GIST, and Tc-99mHDSR may be the optimal method for its diagnosis before preoperative imaging. Pathology confirmation is the gold standard. Based on the findings in this case, surgery is a reasonable treatment for gastric fundus splenosis with hemangioma.

### Author contributions

BG performed the pathology and histological examination. XHL, LW, MZ, ZWD, GJL, and LPM assessed the medical records and obtained information on the patient’s clinical history. JH and WYJ helped in the final drafting of the manuscript. All authors have read and approved the final manuscript.

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### Methodology

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### Software

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