

Original Article

Splenosis in gastric fundus mimicking gastrointestinal stromal tumor: a report of two cases and review of the literature

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Abstract: Splenosis refers to heterotopic autotransplantation and implantation of splenic tissue following splenic trauma or surgery. Splenosis in gastric fundus is rare and difficult to diagnose, since splenosis has similar manifestation with gastrointestinal stromal tumor (GIST) under routine endoscopy examination. In this report, we present two quite rare case of splenosis. Both of their pre-operative diagnose under endoscopic ultrasonography was considered as GIST. Finally, one in the abdominal cavity, adhering closely to the gastric fundus, measuring 20 mm × 15 mm, was resected by surgical operation, and one in the gastric fundus, measuring 20 mm × 20 mm, was resected by endoscopic surgery. The precise diagnosis of splenosis was distinct by post-operative histopathologic examination. In addition, we also made a mini review of previously published articles, in order to provide indication to solve future doubts in diagnosing and treating splenosis.

Keywords: Splenosis, gastrointestinal stromal tumor, endoscopic ultrasonography

Introduction

Splenosis is a relatively uncommon secondary tumor caused by an ectopic autotransplantation of splenic tissues after splenic trauma or surgery. The true incidence is unknown because splenosis is usually an incidental finding at surgery or autopsy [1, 2]. Most of the patients are clinically asymptomatic or have slight abdominal discomfort, but severe symptoms such as gastrointestinal bleeding are also reported [3-5]. So the precise diagnosis and proper treatment are significant.

Splenosis in gastric fundus is quite rare. Only one case has been reported by K. Yang et al. in 2013 [6]. In this report, we present two cases of splenosis in gastric fundus. Both of the cases are firstly suspected as a gastrointestinal stromal tumor (GIST) according to its representation under the endoscopic ultrasonography, but finally diagnosed as splenosis according to their post-operative histopathological examinations.

Case report

Case one

A 40-year-old woman complained of abdominal pain and pharyngalgia initially. The symptoms lasted for half a year and aggravated for one month, mainly triggered by eating and drinking. The patient had undergone splenectomy for a traumatic splenic injury twenty years ago, and the scar on the left superior abdominal wall was well-healed. There was tender pain in her upper and mediate abdomen. No other abnormal signs were observed, and her laboratory tests are normal. The abdominal B ultrasound showed cholecystitis and cystic polyp. The gastroscopy found the presence of a mass in the gastric fundus, measuring 20 mm × 15 mm (**Figure 1A**), with varicose veins around it. The mass was glossy and was suspected as a GIST according to its representation under the endoscopic ultrasonography (**Figure 1B**). The contrast-enhanced computed tomography (CT) was performed (**Figure 2**), without any positive findings.

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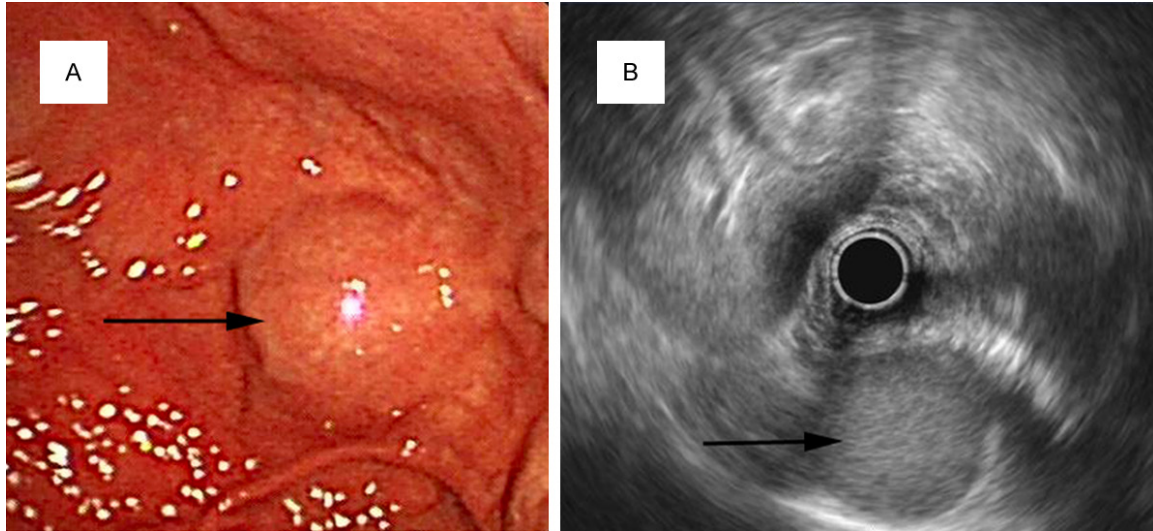


Figure 1. A. Presence of a mass measuring 20 mm × 15 mm with varicose veins around it can be found in the gastric fundus by gastroscopy. B. A smooth mass with homogeneous low echo derived from muscularis propria can be seen under the endoscopic ultrasonography.

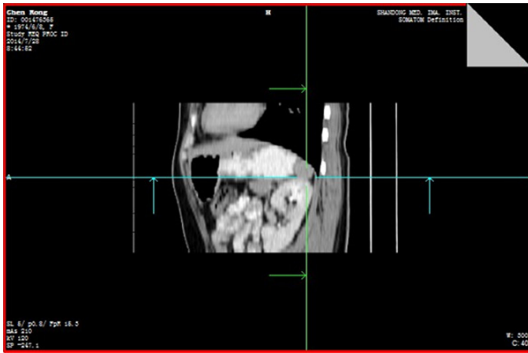


Figure 2. Features under the contrast-enhanced computed tomography (CT).

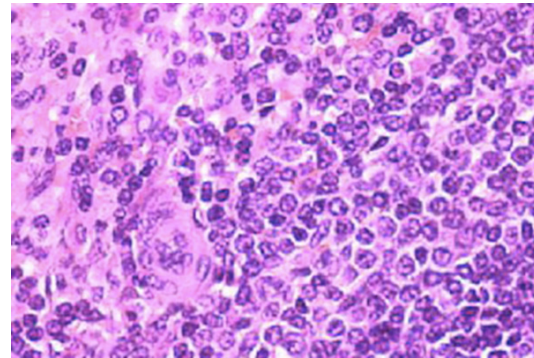


Figure 3. Pathological characteristics of splenisosis.

The patient was transferred to a gastrointestinal surgery. After the incision was made along the subcostal border, we unexpectedly found two nodular goiters on the greater omentum, each measuring 30 mm × 20 mm × 20 mm. Considering the patient's history of splenectomy, the possibility of splenisosis was first suspected at that time. The goiters were resected along with the greater omentum. After the gastrocolic ligament and the splenocolic ligament were cut off, a mass growing in the abdominal cavity, attached closely to the gastric wall, was found. The mass was peeled off carefully using scissors and suction, and the gastric wall was kept well.

The histopathologic examinations for all three specimens are consistent with splenisosis.

(**Figure 3**). The patient's postoperative recovery was uneventful and she was discharged after six days. No signs of recurrence were detected on CT during a five-month follow-up period.

Case two

A 32-year-old male presented with a 6-month history of reduplicated regurgitation of sourness and belching. Past medical history included a splenectomy for splenic injury in a traffic accident 4 years ago. No other abnormality was found in the physical examination except a 11 cm × 1 cm scar on the left superior abdominal wall. His laboratory test results were normal. The gastroscopy revealed that there was a smooth and round mass in the gastric fundus, about 20 mm × 20 mm (**Figure 4A**). The endoscopic ultrasonography showed that the low

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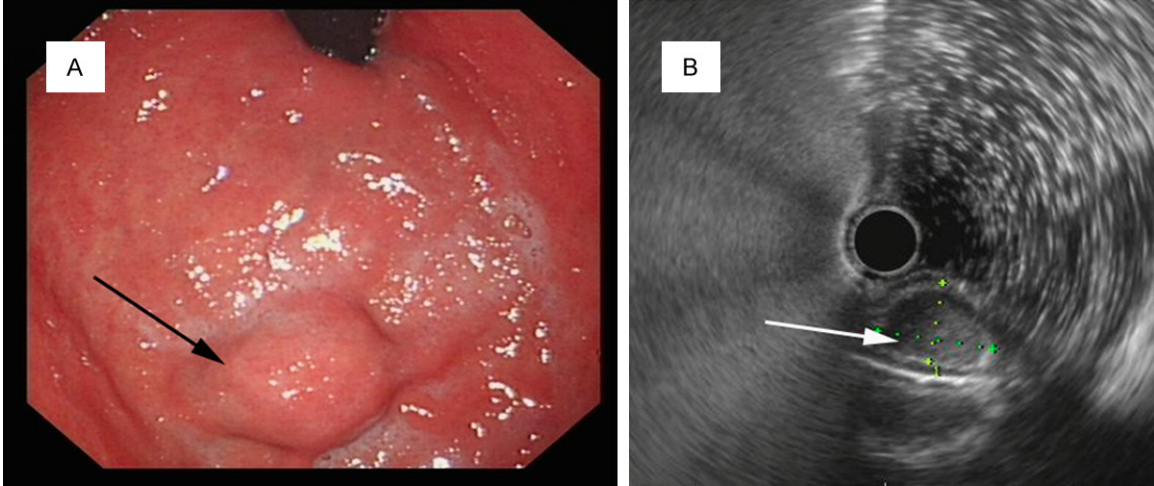


Figure 4. A. The Gastroscopy revealed that there was a smooth and round mass in the gastric funds, about 20 mm × 20 mm. B. The endoscopic ultrasonography showed that, the low echo level mass was homogeneous, and it seemed derived from muscularis propria.

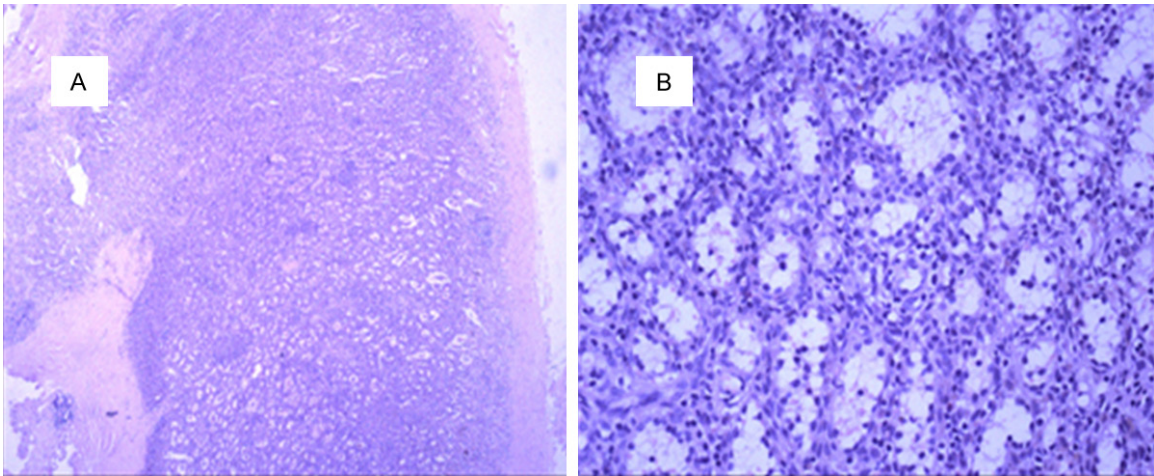


Figure 5. A. Heterotopic spleen transplantation immunohistochemistry (× 100 magnification microscope) B. Visible abnormal spleen tissues (× 400 magnification microscope).

echo level mass was homogeneous, and derived from muscularis propria (**Figure 4B**). The diagnosis of GIST was made. The following contrast-enhanced CT was rejected by the patient for personal reasons.

A endoscopic full-thickness resecting (EFR) surgery was performed. Methylene blue and glycerol fructose mixture was injected in submucosa, and a insulated tip diathermic knife (IT knife) or Dual knife was used to help dissecting the tumor. The tumor was located in muscularis propria, growing towards the gastric cavity. It adhered closely to the neighboring normal gastric tissue. The whole tumor was resected and

sent for pathological examination. Unexpectedly, the pathological examination showed clearly that the tumor was splenosis (**Figure 5**). The patient's history of splenectomy further confirmed the possibility of this diagnosis.

The patient's postoperative recovery was fine and he was charged after 10 days. Follow-up period was un-eventful.

Discussion

Splenosis refers to heterotopic autotransplantation and implantation of splenic tissue in human body cavity or any parenchymal organs.

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Splenic pulp mainly seed throughout the abdominal cavity during splenic trauma or surgery, so mostly of the patients suffered relevant medical history [7]. In 1910, Von Kutter firstly presented the definition of splenosis at autopsy, and in 1937, Shaw and Shaft H firstly reported six clinical cases of splenosis after splenic surgeries.

Animal experiments have demonstrated that the isolated splenic pulp may survive after seeded into any position of the abdominal cavity [8, 9]. Splenosis does not include all kinds of cells in normal spleen. The undifferentiated reticular cells achieve dominant position, while other cells change to ischemia or necrosis. Once the support structures are built by reticular cells and fibrous tissue, reticular cells are induced to differentiate into endothelial antrum, capillary vessel and lymphocytes, which finally create spleen tissue [10]. However, compared with normal spleen tissue. Splenosis is lack of columnar structure, follicle and germinal center, and they are surrounded by granular hemosiderin. Due to the distinct condition of differentiation, splenosis mostly present as tubercles. The number of the tubercles mainly decided by the grade of splenic injury, several or several tens. The enveloped tiny artery nourish the tubercles, but sometimes the blood and nutrient is not enough, so most of the tubercles are less than 3 cm [3, 11].

Previous articles have reported that the patients with histology of splenic injury or surgery have 67% possibility of splenosis. Most of the patients are clinically asymptomatic or with slight abdominal discomfort, with no harm to their physiological function. Other patients' symptom is decided by the position of the splenosis. Abdominal cavity is the most common position, including the liver, adrenal gland, omentum majus, mesenterium. The pelvic cavity, subcutaneous tissue and other body cavity or parenchymal organs may also be involved [12-15]. Splenosis at specific position can cause specific clinical symptoms. For example, gastroenterological splenosis may lead to gastrointestinal bleeding; multiple splenosis tubercles in abdominal cavity cause intestinal adhesion combining acute intestinal obstruction; and if mesenteric vessels supplying massive blood to the splenosis tubercles may cause the secondary hypersplenism [4, 5].

Splenosis in gastric fundus is quite rare. Only one case has been reported by K. Yang et al. in 2013 [6]. In this report, we present two cases of splenosis in gastric fundus mimicking gastrointestinal stromal tumor. Both of the two patients have histology of splenic injury and surgery. They were admitted to the hospital for unspecific gastrointestinal symptom, and received gastroscopy and endoscopic ultrasonography examination. However, the manifestations of splenosis and GIST are quite similar: a smooth or round mass in the gastric funds, with homogeneous low echo, and it seemed derived from muscularis propria. Here by the diagnosis of splenosis until it is proved by histopathological can not be made examination.

For patients with histology of splenic injury or surgery, when widely available imaging modalities like type-B ultrasonic inspection, Computed Tomography, Magnetic Resonance Imaging or other imaging examinations find parenchymal mass in abdominal cavity, the diagnosis of splenosis should be considered. Under the CT examination, the splenosis have similar density with normal spleen tissue, but in arterial phase, normal spleen tissue has the sign of graniphryic enhancement, but splenosis has homogeneous enhancement. The type-B ultrasonic inspection, Computed Tomography and Magnetic Resonance Imaging have limited value in diagnosing splenosis [3, 11], percutaneous tumor puncture and cytological microscopic examinations only can find abundant lymphocytes, which cannot distinguish it with lymphoproliferative disease. Taken together the above examination have little value in diagnosing splenosis. Only combining disease histology, imaging examination and radioisotope scanning are available. Once the diagnosis of splenosis is made, if the patient is asymptomatic, surgical resection is not necessary. But if the splenosis in specific position have caused acute intestinal obstruction, gastrointestinal hemorrhage, or it cannot be distinguished with abdominal malignant tumor, surgery is required. The inter-operative frozen section pathological examination is the gold standard, the diagnosis of splenosis should avoid excessively wide excision, in case unnecessary injury are made to the body.

Disclosure of conflict of interest

None.

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