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Gastric splenosis mimicking a gastrointestinal stromal tumor: A case report

Claudio Isopi, Giulia Vitali, Federica Pieri, Leonardo Solaini, Giorgio Ercolani

Abstract

BACKGROUND

Mass lesions located in the wall of the stomach (and also of the bowel) are referred to as “intramural.” The differential diagnosis of such lesions can be challenging in some cases. As such, it may occur that an inconclusive fine needle aspiration (FNA) result give way to an unexpected diagnosis upon final surgical pathology. Herein, we present a case of an intramural gastric nodule mimicking a gastric gastrointestinal stromal tumor (GIST).

CASE SUMMARY

A 47-year-old Caucasian woman, who had undergone splenectomy for trauma at the age of 16, underwent gastroscopy for long-lasting epigastric pain and dyspepsia. It revealed a 15 mm submucosal nodule bulging into the gastric lumen with smooth margins and normal overlying mucosa. A thoraco-abdominal computed tomography scan showed in the gastric fundus a rounded mass (30 mm in diameter) with an exophytic growth and intense enhancement after administration of intravenous contrast. Endoscopic ultrasound scan showed a hypoechoic nodule, and fine needle FNA was inconclusive. Gastric GIST was considered the most probable diagnosis, and surgical resection was proposed due to symptoms. A laparoscopic gastric wedge resection was performed. The postoperative course was uneventful, and the patient was discharged on the seventh postoperative day. The final pathology report described a rounded encapsulated accumulation of lymphoid tissue of about 4 cm in diameter consistent with spleen parenchyma implanted during the previous splenectomy.

CONCLUSION

Splenosis is a rare condition that should always be considered as a possible alternative diagnosis for gastric intramural mass lesions.
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Core Tip: Intramural gastric nodules are rare, but all differential diagnoses must always be considered. If feasible, a preoperative fine needle aspiration can help the surgeon in selecting the best treatment option. Splenosis is uncommon in the general population, but it must be considered in each patient with a history of splenectomy (especially after trauma). In this specific cluster it is reasonable to insist on ruling out splenosis even making a second histologic sampling after a first failure.

INTRODUCTION

The masses arising from the wall of the stomach are referred to as “intramural”. In these cases the endoscopic and radiologic features may lead to several differential diagnoses because several overlapping characteristics have been shown to exist among the various gastric masses. Intramural lesions can be benign or malignant, and the most common diagnosis is gastrointestinal stromal tumors (GISTs).

Only a preoperative sampling allows planning the best therapeutic approach, but when the nature of the nodule cannot be preoperatively determined, an assessment about size, possible diagnoses, patient’s characteristics and clinical symptoms should be done before considering an upfront surgical approach.

Herein, we present a case of an intramural gastric nodule mimicking gastric gastrointestinal stromal tumor, whose nature could be defined only after surgery.

CASE PRESENTATION

Chief complaints
A 47-year-old Caucasian woman was referred to our unit for an intragastric nodule detected during a gastroscopy.

History of present illness
The gastroscopy was performed for long lasting epigastric pain and dyspepsia.

History of past illness
Patient’s past medical history included: Asthma, hypothyroidism, migraine and a splenectomy for trauma.

Personal and family history
No family histories were identified.

Physical examination
The patient was in good general condition and slightly overweight (body mass index: 25.6). There were no abdominal mass and no pain on palpation.

Laboratory examinations
Routine laboratory tests revealed no abnormalities.
Imaging examinations
Endoscopy showed a 15 mm submucosal nodule bulging into the gastric lumen with smooth margins and macroscopically normal overlying mucosa. Biopsies were negative for malignancy and showed superficial chronic gastritis.

Consequently, a thoraco-abdominal computed tomography scan (Figure 1A and 1B) and an endoscopic ultrasound with a fine needle aspiration were planned. Those investigations found a roundish formation on the gastric fundus of about 30 mm in diameter with an exophytic development. The mass was in close contiguity with the left adrenal gland and the left pillar of the diaphragm with no signs of infiltration. The ultrasound appearance was of a solid mass with well-defined margins with a homogeneous and well vascularized internal texture in the absence of calcified or necrotic areas. The fine needle aspiration (FNA) was performed without complications, but the result was nondiagnostic due to inadequate tissue yield.

FINAL DIAGNOSIS
Our main diagnostic suspect remained a gastric GIST and the symptoms could be related to the location of the mass. After a careful evaluation of the risks and benefits and according to the European Society for Medical Oncology guidelines[1], the surgical excision was planned.

TREATMENT
The laparoscopic resection was performed with a three trocars technique (10 mm supraumbilical and right hypochondrium and 5 mm left hypochondrium). After a careful lysis of the adhesions related to the previous splenectomy, the exophytic mass of the fundus was identified. The perigastric vessels were dissected in order to expose the nodule; the resection was performed with a linear stapler.

OUTCOME AND FOLLOW-UP
The postoperative course was uneventful, and the patient was discharged on the seventh postoperative day. The final pathology of the specimen did not confirm our hypothesis but reported a rounded encapsulated accumulation of lymphoid tissue of 4 cm in diameter consistent with spleen parenchyma probably implanted during the previous splenectomy (Figure 2).

At the 6 mo follow-up the patient was symptom free.

DISCUSSION
Ectopic splenic tissue can be found in the body as accessory spleens and splenosis[2]. The former is congenital and receives blood supply from the splenic artery. The latter is a benign condition caused by the spillage upon the peritoneal surface of cells from the spleen after splenic trauma or surgical procedures.

Splenosis is usually considered to be a rare phenomenon, but its real prevalence is difficult to define. Pearson et al[3] showed that recurrent splenic activity after urgent splenectomy is frequent, and according to Sikov et al[4], its incidence could be as high as 76% in patients who had undergone splenectomy for trauma.

Splenosis is a benign condition, usually found incidentally and unless symptomatic surgery is not indicated[5]. In some cases the implantation could be responsible for serious conditions like gastrointestinal hemorrhage, pain from compression of the abdominal structures and bowel obstruction[6]. Splenosis may resemble several abdominal malignancies. As such several studies reported cases of splenosis mimicking a pancreatic mass[7], lymphomas[8], neuroendocrine tumors[9], intramural colonic masses[10], liver masses[11,12] and GISTs[13-16]. For this variability, the diagnosis of splenosis may be challenging. On a peripheral smear the absence of Howell-Jolly and Heinz bodies and siderocytes despite a history of splenectomy could mildly suggest the presence of a splenosis[17]. Imaging may not be accurate in defining this condition[18]. Differential diagnoses between benign[19-21] and malignant[22-23] forms and
the radiologic features of intramural gastric masses\(^{33,34}\) are presented in the Table 1.

Nowadays, there is a general consensus that the mainstay for the diagnosis of splenosis is the noninvasive scintigraphy using technetium-99m-labeled heat damaged red blood cell or indium 111-labeled platelets\(^{35}\). However, it must be highlighted that the real critical point in diagnosing splenosis is thinking about it in a suggestive past medical history.

During the assessment of a gastric intramural nodule, mass biopsy may help solving the diagnostic dilemma. However, in our case preoperative diagnosis was not possible, and the patient was submitted to surgery according to her symptoms and the most probable diagnosis.

**CONCLUSION**

Splenosis is a rare condition that should always be considered as a possible diagnosis in patients who had undergone splenectomy. If feasible, a preoperative FNA may be the best preoperative investigation to rule out other diagnoses and to plan the most appropriate treatment.
Table 1 Characteristics of intramural gastric masses

<table>
<thead>
<tr>
<th>Location in the stomach</th>
<th>CT special features</th>
<th>Special features</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lipoma[18]</td>
<td>Antrum</td>
<td>Attenuation values -70 HU to -120 HU Solitary, fibrous capsulated, soft (change in size and shape with peristalsis), no vessels</td>
</tr>
<tr>
<td>Leiomyoma[26]</td>
<td>Cardia</td>
<td>Low attenuation, endoluminal Low attenuation, endoluminal Negative for c-kit, positive for desmin and smooth muscle actin</td>
</tr>
<tr>
<td>Schwannoma[23]</td>
<td>Body</td>
<td>Minimal enhancement on the arterial phase Absence of calcification, hemorrhage, necrosis; not encapsulated; positive for S-100</td>
</tr>
<tr>
<td>Glomus tumor[20]</td>
<td>Antrum</td>
<td>Strong enhancement on early-phase Highly vascular; positive for calponin and smooth muscle actin</td>
</tr>
<tr>
<td>Inflammatory fibroid polyyp[22]</td>
<td>Antrum</td>
<td>Enhancement on arterial phase Positive for CD34 and vimentin</td>
</tr>
<tr>
<td>Hemangioma[29]</td>
<td>-</td>
<td>Strong enhancement on early-phase Phleboliths are pathognomonic</td>
</tr>
<tr>
<td>Plexiform fibroxyoma[19]</td>
<td>Antrum</td>
<td>Myxoid tissue interspersed with vessels Unique to the stomach, size from 2 cm to 15 cm</td>
</tr>
<tr>
<td>Ectopic pancreas[9]</td>
<td>Greater curvature</td>
<td>Similar to normal pancreas -</td>
</tr>
<tr>
<td>Splenosis[3]</td>
<td>-</td>
<td>Enhancement on arterial phase Splectomized patients</td>
</tr>
</tbody>
</table>

**Malignant lesions**

<table>
<thead>
<tr>
<th>Location in the stomach</th>
<th>CT special features</th>
<th>Special features</th>
</tr>
</thead>
<tbody>
<tr>
<td>GIST[9]</td>
<td>Body</td>
<td>Smoothly circumscribed, bullseye sign Positive for c-kit or dog-1; 50% greater than 2 cm</td>
</tr>
<tr>
<td>Non-GIST sarcoma (liposarcoma, leiomyosarcoma, unclassified sarcoma)[27]</td>
<td>-</td>
<td>Usually large, heterogeneous enhancement Positive for desmin and smooth muscle actin, negative for c-kit</td>
</tr>
<tr>
<td>Lymphoma[13]</td>
<td>-</td>
<td>Wall thickening Distant (more than close) and large adenopathy</td>
</tr>
<tr>
<td>Carcinoid[29]</td>
<td>-</td>
<td>Multiple small lesions Reactive to synaptophysin and chromogranin A, hypergastrinemia related symptoms</td>
</tr>
<tr>
<td>Inflammatory myofibroblastic tumor[31]</td>
<td>-</td>
<td>Heterogeneously enhancing tumor (malignant appearance) Borderline tumor, more frequent in young adults and children; reactivity for ALK</td>
</tr>
<tr>
<td>Metastasis[30-32]</td>
<td>-</td>
<td>-               &quot;Homomorphic&quot; endoscopic features; dyschromic lesions</td>
</tr>
</tbody>
</table>

CT: Computed tomography; GIST: Gastrointestinal stromal tumor; ALK: Anaplastic lymphoma kinase.

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