

A case report of Abdominal Splenosis – a Practical Mini-Review for a Gastroenterologist

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Abstract

Splenosis is a benign condition caused by ectopic autotransplantation of splenic tissues after splenic trauma or surgery. It is usually diagnosed accidentally. However, occasionally splenosis poses a significant diagnostic dilemma, especially when this condition presents as a metastatic malignant disease on abdominal imaging.

This is the case report of a 54-year-old woman, who underwent post-traumatic splenectomy at the age of 12 years. The physical examination of this patient complaining of epigastric and low back pain, except for postoperative abdominal scar, as well as laboratory tests were normal. Esophagogastroduodenoscopy showed chronic gastritis. Abdominal ultrasound revealed no spleen, enlarged lymph nodes in the right retroperitoneum and a focal lesion in the uterus. In addition, the CT scan disclosed enlarged left hepatic lobe, numerous oval tumors between intestinal loops, in the caecal region, under the diaphragm and abdominal wall, periaortic enlarged lymph nodes and an osteolytic lesion in the first lumbar vertebra, suggesting disseminated malignant disease. The exploratory laparotomy showed multiple abdominal nodules, a tumor of the ileum, the greater omentum and the uterus. Except for the last one (leiomyoma), all excised tumors proved to have histological structure typical for the spleen and, finally, abdominal splenosis was diagnosed.

Splenosis should be considered when a differential diagnosis of tumor-like lesions disclosed on abdominal imaging occurs in a patient with a splenic injury in the past.

Key words

Abdominal splenosis – metastatic disease – diagnosis.

Case report

A 54-year-old female with an unremarkable family history was referred to the out-patient gastroenterology department with complaints of occasional epigastric pain with positive IgG antibodies against *H. pylori*. She had undergone post-traumatic splenectomy and blood transfusions after a blunt injury in a road accident at the age of 12. Except for degenerative bone disease the patient had been healthy until 12 years ago, when she was diagnosed with sideropenic anemia secondary to abundant menses. Temporal treatment with oral iron and vitamin preparations proved to be successful. After menopause her blood count normalized and she demonstrated stable blood parameters in the 2 years follow-up.

The patient denied any symptoms except for occasional dysgeusia, heartburn, mild epigastric and low back pain. She also denied drug and alcohol abuse as well as smoking throughout her life and had not been taking any pharmacological agents in the last few months. The physical examination was remarkable only for the presence of a postoperative abdominal scar in the left upper quadrant and overweight (BMI 28.7 kg/m²). Initial routine laboratory tests including ESR, complete blood count, total bilirubin, GGT, alkaline phosphatase, aminotransferases, amylase, glucose, electrolytes, creatinine, blood lipids did not reveal any abnormalities. Esophagogastroduodenoscopy showed mild gastritis with positive rapid urease test for *H. pylori*. Abdominal ultrasound revealed no spleen, a few enlarged lymph nodes in the right retroperitoneum and a focal lesion in the uterus. CT scan disclosed no spleen, enlarged left hepatic lobe and numerous oval soft-tissue nodules between intestinal loops, in the caecal region, under the diaphragm and abdominal wall, with a maximum size of 2.2 x 1.6 x 2.6 cm, some of them infiltrating the diaphragm. In addition, there were a few lymph nodes up to 1.2 cm in diameter along the abdominal aorta and iliac arteries and an osteolytic lesion in the first lumbar vertebra suggesting disseminated metastatic malignant disease. As the results of additional laboratory tests including tumor markers, gynecological examination, mammography, chest X-ray, colonoscopy,

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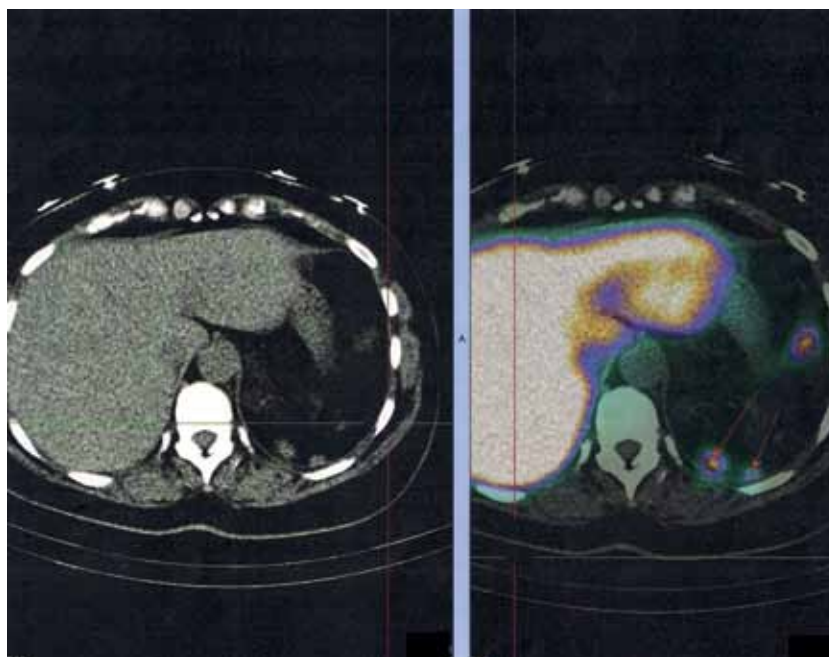


Fig 1. Abdominal splenosis - axial SPECT-CT and Tc99m scintigraphy of the abdominal cavity in a 54-year-old female with post-traumatic splenectomy in the past showing multiple oval splenic implants in the abdomen, some of them adjacent to the diaphragm (arrows).

and neck ultrasound were irrelevant, the patient underwent an exploratory laparotomy that showed disseminated, multiple, small, soft, bluish-red peritoneal nodules, similar tumors in the greater omentum (7 mm in diameter) and on the serosal surface of the ileal wall (17 mm). An additional mass (27 mm) was noticed in the uterus. The rest of the abdominal and pelvic cavity, except for no presence of the spleen, was normal. Surprisingly, except for the last one (leiomyoma hialinisans, SMA +, K167 <1%), all excised tumors proved to have a histological structure typical for the spleen without evidence of neoplasia. Postoperative Tc99m scintigraphy showed multiple oval splenic implants in the abdomen, some of them adjacent to the diaphragm (Fig. 1). Further evaluation of the spinal column with MRI showed degenerative changes and a focal lesion 10 mm in diameter in the first lumbar vertebra of increased intensity on T1- and T2-weighted scans corresponding to vascular malformation (angioma).

Subsequently, abdominal splenosis following the post-traumatic splenectomy was diagnosed. The postoperative course was uneventful, leading to the complete recovery of the patient who remains asymptomatic 12 months following the procedure.

Discussion

Unexpected findings on explorative laparotomy draw attention to splenosis, a benign condition caused by an ectopic autotransplantation of splenic tissues onto exposed vascularised intra- and extraperitoneal surfaces following splenic injury or elective splenectomy [1]. H. Albrecht is the author of the first recorded case of splenosis (1896)

[2], but the term “splenosis” was coined by Buchbinder and Lipkoff in 1939 [3]. Splenosis usually occurs in the left upper quadrant of the abdomen [1]; however, other intraabdominal locations have been also described [4-6]. Splenic autotransplants are also found in the thoracic cavity [7-9], subcutaneously [9-10] or even intracranially [11].

The incidence of abdominal splenosis in the general population remains unknown. According to Muller and Ruthlin, who performed ultrasonographic follow-up studies in patients after post-traumatic splenectomy, presumed abdominal splenosis (without histological confirmation) occurred in one-third of the patients [12].

Splenosis usually results from trauma with subsequent splenectomy, mostly in teenage years [13]. An average interval between post-traumatic splenectomy and the diagnosis of abdominal splenosis is 10 years (5 months - 36 years), shorter than in our patient (42 years).

Splenic remnants implant easily on the serosal surfaces, parasitize adjacent blood vessels, and grow into mature splenic tissue, therefore seeding of damaged splenic pulp is the crucial mechanism behind autotransplantation after the splenic rupture [14]. Additional mechanisms are the hematogenous spread of splenic pulp or splenic erythrocyte progenitor cells and their subsequent growth in response to tissue hypoxia [14].

Splenosis is usually asymptomatic and splenic implants are found accidentally during unrelated diagnostic imaging or surgery. Occasionally, abdominal splenosis can cause gastrointestinal bleeding [15, 16] or abdominal pain due to bowel obstruction, intraperitoneal nodule infarction, hematoma, enlarging abdominal or pelvic masses, ureteral

compression and hydronephrosis [17-20] or recurrence of hematologic diseases treated with splenectomy [21].

A definite preoperative diagnosis of splenosis requires a high index of suspicion. A detailed medical history, thorough physical examination, lack of typical changes in the blood smear often present after splenectomy (Howell-Jolly bodies, reticulocytosis), and protective levels of antipneumococcal antibodies in a non-vaccinated patient should make the physician consider this rare condition.

Abdominal ultrasound and radiological studies show only limited diagnostic value in abdominal splenosis. Sonographic findings are not specific and reveal round and oval soft-tissue masses in the various locations. Low density of splenic tissue makes it difficult to visualize on standard x-rays. CT reveals the number, shape, size, location but not identity of the nodules. Non-characteristic sonographic and radiological picture of abdominal splenosis may be confused with numerous conditions such as metastatic disease, abdominal lymphoma, carcinomatosis, hemangiomas, peritoneal mesothelioma, multifocal endometriosis, adenomas, primary renal or hepatic malignancy, gliomatosis peritonei, granulomatous peritonitis, accessory spleens or reactive adenopathy.

Standard MRI showing hypointense or hyperintense splenic implants depending on presentation (T1 and T2, respectively) with heterogeneous enhancement C+ (GAD) is not very useful in differential diagnosis [22]. Several papers have proposed ferumoxides-enhanced MRI after IV administration of supramagnetic iron oxide particles that are removed from the circulation by the phagocytic reticuloendothelial system [23, 24].

At present, noninvasive Technetium (Tc) 99m radionuclide scanning is the mainstay in the diagnosis of splenosis. Tc-99m sulphur colloid is sequestered in the reticuloendothelial system and detects heterotopic splenic tissue [8, 25, 26]. If differentiation from hepatic tissue is necessary, a 5 mCi (185 MBq) Tc 99m-tagged heat-damaged autologous red blood cells (RBCs) or Indium 111-labeled platelets scintigraphy can be performed [27]. RBCs scintigraphy, although not free from the risk of adverse effects seems to be more sensitive in early splenosis, functional hyposplenism or poor splenic uptake as well as when the spleen and the liver overlap, causing poor visualization of splenic tissue by the sulphur colloid test. If the diagnosis is confirmed preoperatively by appropriate radionuclide modalities in a patient with history of abdominal trauma, laparotomy can be avoided.

In the presented case, due to an alarming radiologist's misinterpretation of the CT findings, splenic implants were mistaken for neoplastic changes and the patient was subjected to the prompt exploratory laparotomy. Nowadays, laparoscopy provides a minimally invasive entry for the visualization, biopsy or resection of suspected masses [28]. Splenic implants are often bluish, but may vary in color, have no hilum and are supplied by local arteries that penetrate their fibrotic capsule. Lack of adhesions within the abdominal cavity is quite a characteristic feature of splenosis.

If the biopsy is performed without a preoperative

suspicion of splenosis, the frozen section should be carefully examined for evidence of splenosis. Macroscopically and histologically, two adjacent implants may differ markedly. The tissue in splenosis often reveals distorted architecture with no hilum and a poorly formed capsule but splenosis with histology and immunohistochemistry indistinguishable from the normal spleen has been also described [10, 29].

Case reports describing a recurrence of hematological disorders in splenectomized patients suggest a functional reticuloendothelial system within the implants [21, 30]. Significant increase in the serum levels of antipneumococcal antibodies in patients with splenic autotransplantation indicate that implanted splenic tissue may be beneficial and protects against systemic encapsulated bacterial infection in asplenic individuals.

Therefore, the current opinion is that when splenosis is diagnosed, surgical removal is recommended only in the case of symptomatic/complicated splenosis and in patients with hematological disease for whom splenectomy is beneficial. Surgical approach should be also indicated in the case of an uncertain diagnosis, especially when scintigraphic methods are not readily available and there is a suspicion of a malignant disease, such as in the patient described.

Conclusions

With the increased prevalence of abdominal trauma due to road accidents and the growing armamentarium of imaging modalities abdominal splenosis may turn up more often than ever. Occasionally, it leads to a great diagnostic dilemma because splenic implants may be misinterpreted as neoplastic lesions or adenopathies. Presumed diagnosis can be made in a patient with absence of siderocytosis and Howell-Jolly bodies in the blood smear and a history of splenectomy or abdominal trauma in the past. Tc99m scintigraphy is a reliable noninvasive diagnostic method of choice in this rare condition and helps to avoid unnecessary abdominal surgery. In order to prevent any possible diagnostic doubts and invasive examinations in the future, confirmed splenosis should be recorded in the medical documentation of the patient.

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