Intrahepatic and intra-abdominal splenosis: A case report and review of literature

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**Abstract**

**BACKGROUND**

Splenosis is defined as the process by which tissue from the spleen disseminates through the body and grows in an ectopic location following trauma or a splenectomy. Visceral sites of splenosis are rare.

**CASE SUMMARY**

We report a case of intrahepatic splenosis in a 57-year-old man with a history of trauma over 40 years ago who initially presented with chest pain. Findings initially mimicked malignancy but a diagnosis of intrahepatic splenosis was confirmed using computed tomography and scintigraphy with technetium-99m heat-denatured red blood cells (Tc-99 DRBC).

**CONCLUSION**

Scintigraphy with Tc-99 DRBC is a reliable technique to diagnose splenosis and should be performed before using more invasive procedures are carried out. Splenosis should be considered as a possible differential diagnosis for a hepatic nodule in any patient with a history of abdominal trauma, previous splenectomy or atypical radiological features on imaging.

**Key words:** Intrahepatic splenosis; Abdominal splenosis; Computed tomography; Scintigraphy; Hepatocellular carcinoma; Case report

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Core tip: Intrahepatic splenosis is rare. On imaging it is difficult to distinguish splenosis from hepatic malignancy, particularly hepatocellular carcinoma. We report a case of a patient with intrahepatic and intra-abdominal splenosis diagnosed using scintigraphy with technetium-99m heat-denatured red blood cells. To the author’s knowledge, this is the first case where hepatic splenosis was confirmed without using invasive procedures such as biopsy or surgery. Splenosis should be considered as an important differential for a hepatic lesion in a patient with a history of trauma or splenectomy, particularly if the lesion is located near the capsule and associated with multiple abdominal deposits.

INTRODUCTION

Splenosis is a benign acquired condition. Following trauma or a splenectomy, splenic tissue may autotransplant in an ectopic location. Common sites include the serosal surface of the small or large intestine, greater omentum or the peritoneum[1]. Less frequently, splenic nodules may be found in the liver[2], stomach[3], pancreas[4] and following rupture of the diaphragm in the thorax[5]. The kidneys[6], ovaries[7] and subcutaneous tissue[8] are even rarer sites of splenosis. Splenosis is usually asymptomatic and when incidentally discovered can be difficult to distinguish from malignancy using computed tomography (CT) or magnetic resonance imaging (MRI).

CASE PRESENTATION

Chief complaints
A 57-year-old male presented to the Emergency Department with severe right-sided pleuritic chest pain radiating to his back. There was no associated breathlessness, fever, cough, haemoptysis, dizziness, syncope, numbness, paraesthesia or weakness.

History of present illness
The patient reported that the symptoms began abruptly two days ago and progressively worsened, without any triggers. The pain settled following the administration of morphine after admission to hospital.

History of past illness
In 2015 he was diagnosed with benign prostatic hypertrophy and had suffered traumatic injury following a road traffic accident over 40 years ago. Of note, he had no history of hepatic disease.

Physical examination
There was marked tenderness on inspiration on the right side of the chest, but otherwise physical examination was unremarkable. The patient’s vital signs were normal with a temperature of 37.0 °C, heart rate of 74 bpm, blood pressure of 125/75 mmHg, respiratory rate of 15 breaths/min and oxygen saturations of 98% in room air.

Laboratory testing
Routine blood tests were within normal ranges including liver function tests, alpha-fetoprotein, prothrombin time and a normal D-Dimer.

Imaging examinations
A CT angiogram was performed to rule out aortic dissection due to his acute presentation. No evidence of the latter was seen but there was a 3-cm large arterially enhancing lesion in segment IV of the liver (Figure 1A). The lesion was arterialised with faint hypoenhancement in the portal venous phase (Figure 1B and C). Multiple arterially enhancing peritoneal, lesser sac and retroperitoneal nodules were additionally seen following the same enhancement pattern. The patient’s spleen was observed to be lobulated and the right inferior ribs and right iliac crest had an abnormal appearance suggestive of previous trauma.
Figure 1 Computed tomography axial images from a patient with intrahepatic splenosis. A: arterial phase; a heterogenously enhancing left lobe liver lesion is present 3 cm in diameter (thick arrow). Similar heterogeneously enhancing peritoneal and lesser sac nodules are seen (arrowhead). The deformed spleen shows typical heterogenous “zebra stripe” arterial enhancement; B and C: portal venous phase; the liver lesion (thick arrow) and the peritoneal and retroperitoneal nodules (arrowhead) are isodense and the same density as the spleen (thin arrow).

**FINAL DIAGNOSIS**

Following discussion of these imaging findings and the patient’s history in a specialist multidisciplinary team meeting, the possibility of intrahepatic splenosis with additional intra-abdominal splenosis was considered. This diagnosis was confirmed using scintigraphy with technetium-99m heat-denatured red blood cells (Tc-99 DRBC), which demonstrated uptake of the radiolabelled red blood cells by the multiple peritoneal nodules, as well as the lesion within the liver and the anterior abdominal wall (Figure 2).

**OUTCOME AND FOLLOW UP**

Due to the extensively reported benign nature of this condition, treatment was not required. The patient was informed of the incidental imaging finding and reassured. He was also informed that his chest pain was likely to be musculoskeletal in nature.

**DISCUSSION**

We describe a case of intrahepatic splenosis diagnosed radiologically, in a patient with a history of trauma. To the authors’ knowledge, this is the first case of intrahepatic splenosis diagnosed without the need for histological analysis, thereby avoiding the potential risk of complications secondary to invasive investigations such as a liver biopsy or laparoscopic surgery.

To date, 21 case reports of intrahepatic splenosis have been described in the literature. We specifically review 13 cases which include CT and MRI (Table 1). Nine cases describe solitary lesions[9-17], whilst four cases involved multiple lesions[18-21]. The nodules ranged in size from 1.5-5.0 cm and were primarily found in the left lobe of the liver. Additional abdominal splenic nodules were reported in four cases to be located in close proximity to the upper pole of the left kidney[19], pancreatic tail[19], mesentery of the colon[21], paravesical space[21], caecum[21] and abdominal wall[21].
All patients except one had undergone a splenectomy in the past. Notably, a wrong diagnosis was made in all but one case, primarily of hepatocellular carcinoma (HCC), leading to unnecessary surgery with the correct diagnosis only being made following post-operative histological analysis. This is partly due to the fact that splenosis is rare and hence is often not considered amongst the differential diagnosis. Additionally, six of the patients included in the literature review had chronic liver disease including hepatitis B, a major risk factor for the development of HCC and four patients had raised tumour markers. In such cases, HCC presents a more likely diagnosis rather than hepatic splenosis. In the isolated case where splenosis was correctly suspected, percutaneous biopsy was still carried out for confirmation.

Imaging is a useful diagnostic tool to distinguish splenosis from other lesions such as HCC, hepatic metastasis, haemangioma and focal nodular hyperplasia (FNH). CT and MRI provide panoramic imaging of the abdomen and can identify the size, location and enhancement characteristics of all lesions. Critically, all the splenic deposits exhibit an enhancement pattern identical to the native spleen on all imaging, with a heterogeneous classical striped arterial hyperenhancement. However, this may be difficult to characterise if the native spleen is small or has been removed.

Classically, on unenhanced CT splenic tissue appears hypointense relative to the liver, whilst on MRI it appears hypodense on T1 and hyperintense on T2-weighted images. Five and six cases in the literature review exhibited these CT and MRI findings respectively (Table 1). On administration of contrast, splenic nodules are hyperintense in the arterial phase often with a striated appearance as seen in our patient. They vary in appearance in the portal venous phase and may be hypointense, isointense or hyperintense.

HCC has a variable appearance on both CT and MRI depending on biological characteristics including their degree of differentiation. Since their blood supply is derived from the neoangiogenesis of non-triadal arteries, HCC, like splenosis typically appear hyperenhancing in the arterial phase with portal venous washout (Table 2).

Hepatic metastasis also varies widely in appearance depending on the location of the primary tumour. They may appear hypo or hypervascular but typically show portal venous washout. Haemangioma, FNH and adenoma are benign lesions which typically show arterial hyperenhancement and hence may be mimicked by splenosis.

Of note, MRI using superparamagnetic iron oxide contrast instead of gadopentetate dimeglumine has been used to distinguish hepatic splenosis from malignancy. Following intravenous administration, these particles are removed from the circulation specifically by the reticuloendothelial cells of the liver and spleen, leading to a reduction in signal intensity of the hepatic and splenic parenchyma on T2-weighted MRI. Such a reduction in signal intensity is however not seen in malignant lesions except some well differentiated HCCs. Splenic nodules still have a higher intensity than the hypointense liver as they take up more contrast. Nonetheless, uptake of contrast still occurs in FNH and so the specificity of this technique is limited in isolation.

Scintigraphy using Tc-99 DRBC is the current diagnostic tool of choice. This is due to its high specificity in identifying splenic tissue. It involves intravenous injection of heat denatured erythrocytes labelled with Tc-99. The majority, as many as 90% of the
In conclusion, splenosis should be considered as a possible differential diagnosis for a hepatic nodule in any patients with a history of abdominal trauma or previous splenectomy, especially when the nodules are located near the capsule of the liver and are associated with multiple intra-abdominal deposits. Scintigraphy using Tc-99m labelled erythrocytes are sequestered in splenic tissue, whilst normal liver tissue or malignant lesions have relatively modest uptake of the radioactive isotope\textsuperscript{[30]}. This technique is therefore a reliable means of distinguishing splenic tissue from other hepatic lesions and avoids subjecting a patient to invasive procedures such as biopsy or surgery which are associated with their own risks.

Patients with splenosis are typically asymptomatic. Hence, surgery is only indicated if rare complications such as infarction\textsuperscript{[31]}, bleeding\textsuperscript{[32] or adhesions resulting in bowel obstruction occur\textsuperscript{[30]}. It is suggested that splenosis may even be beneficial, providing some degree of immunological protection\textsuperscript{[33]}. As it is a benign condition, it is often only diagnosed incidentally decades after the initial trauma following imaging for an unrelated condition. It is therefore difficult to ascertain the time taken for splenosis to occur. However, the process of splenic cells seeding in and growing on the serosal surface of the liver after recruiting nearby hepatic vasculature is likely to take several years. In the literature cases of intra-hepatic splenosis were diagnosed on the serosal surface of the liver after recruiting nearby hepatic vasculature is likely for splenosis to occur. However, the process of splenic cells seeding in and growing on the serosal surface of the liver after recruiting nearby hepatic vasculature is likely to take several years. In the literature cases of intra-hepatic splenosis were diagnosed from a range of 5 to 46 years after trauma or splenectomy\textsuperscript{[17]}.\textsuperscript{[18]}

**CONCLUSION**

In conclusion, splenosis should be considered as a possible differential diagnosis for a hepatic nodule in any patients with a history of abdominal trauma or previous splenectomy, especially when the nodules are located near the capsule of the liver and are associated with multiple intra-abdominal deposits. Scintigraphy using Tc-99m labelled erythrocytes is often only diagnosed incidentally decades after the initial trauma following imaging for an unrelated condition. It is therefore difficult to ascertain the time taken for splenosis to occur. However, the process of splenic cells seeding in and growing on the serosal surface of the liver after recruiting nearby hepatic vasculature is likely to take several years. In the literature cases of intra-hepatic splenosis were diagnosed from a range of 5 to 46 years after trauma or splenectomy\textsuperscript{[17]}.
Table 2  Typical enhancement characteristics of ectopic splenic tissue and other hepatic lesions

| Lesion                  | Enhancement pattern                          |
|-------------------------|----------------------------------------------|
| Splenosis               | Arterially hyperenhancing; variable venous enhancement |
| HCC                     | Arterially hyperenhancing; venous hypoenhancement |
| Hepatic metastasis      | Arterially variable enhancement; venous hypoenhancement |
| Haemangioma             | Arterially peripheral nodular enhancement; venous infilling |
| FNH                     | Arterially hyperenhancing; venous iso/hyperenhancement with late enhancement of scar on MRI |
| Hepatic adenoma         | Arterially hyperenhancing; variable venous enhancement |

HCC: Hepatocellular carcinoma; FNH: Focal nodular hyperplasia; MRI: Magnetic resonance imaging.

DRBC is a reliable technique to diagnose splenosis and should be carried out in all patients suspected of the condition before more invasive diagnostic procedures are considered. Greater awareness of this condition could reduce the high incidence of unnecessary invasive interventions in these patients.

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