# Multiple intra-hepatic and abdominal splenosis: an easy call if you know about it

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# Abstract

Hepatic splenosis represents the heterotopic implantation of splenic tissue caused by the spillage of cells from the spleen usually after splenectomy or splenic trauma. This condition is usually an incidental finding during surgery and its real incidence is unknown. Splenic implants, which can be placed anywhere in the abdominal cavity, are usually multiple and may be confused with different benign and malignant conditions such as renal tumors, abdominal lymphomas, and endometriosis. We hereby report an unusual case of multiple abdominal splenosis, with a particular intra-hepatic location, that could be misinterpreted as an hepato-cellular carcinoma.

#### **Keywords**

Abdomen, computed tomography (CT), magnetic resonance imaging (MRI), liver, spleen

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# Introduction

Splenosis is the auto-transplantation of splenic tissue in a heterotopic location that mostly develops after abdominal injuries (1). It is presumed that the spillage of the damaged splenic pulp into the adjacent cavities causes the beginning of the seeding process (2). There are other hypotheses such as the hematogenous spread of splenic pulp, which claims that the splenic erythrocyte progenitor cells enter the liver through the portal vein and then grow as a consequence of tissue hypoxia (3). The term "splenosis" was first used by Buchbinder and Lipkoff in 1939, to describe the heterotopic transplantation of splenic tissue as a consequence of splenic trauma or surgery (4). Nowadays the incidence rate for patients with a history of splenic trauma or splenic surgery is reported to be 67% (5,6). The appropriate use of commonly available diagnostic tools can facilitate accurate diagnosis, avoiding unnecessary surgical procedures.

This case report shows an unusual case of multiple abdominal splenosis, with a particular intra-hepatic location, that could be misinterpreted as an hepatocellular carcinoma.

# **Case report**

A 69-year-old man, was admitted to our institution because of dyspnea, severe lower extremity edema, and right upper quadrant abdominal pain. Physical examination and past medical history were unremarkable except for the history of ventricular tachycardia. The sister had died because of breast cancer. He had a post-traumatic splenectomy as a result of a car accident that had happened when he was a child. An abdominal ultrasound was performed, showing no significant abdominal abnormalities except for the presence of a 4-cm solid echogenic mass located in the sixth liver segment. The patient had no history of alcohol abuse. Initial routine laboratory tests, including complete blood count and serum chemistry, did not reveal any

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abnormalities except for a mild anemia and a slight alteration of C-reactive protein. Moreover, the patient's serology for hepatitis B and C was negative. Additionally, specific laboratory tests, including tumor markers such as carcino-embryonic antigen and alpha-fetoprotein, were unremarkable. Contrastenhanced helical computed tomography (CT) examination of the abdomen was subsequently performed and it showed multiple intra-abdominal nodular lesions. The largest of these lesions was found adjacent to the sixth liver segment with a maximum diameter of  $6.5 \times 4.6$  cm and  $4.0 \times 1.8$  cm and near the falciform ligament (Figs. 1 and 2). Additional lesions were found next to the mesenteric bowels, in the left paravesical space (Figs. 3 and 4), scattered throughout the peritoneal cavity, in the splenic loggia and near the



**Fig. 1.** Contrast-enhanced arterial phase CT of the abdomen showing one mildly enhancing lesion contiguous to the sixth liver segment (arrow) measuring  $6.5 \times 4.6$  cm.

posterior cecal wall. A contrast-enhanced magnetic resonance (MR) scan, which confirmed the presence of the multiple abdominal and intra-hepatic nodular lesions, was obtained. These masses displayed low signal intensity on unenhanced T1-weighted (T1W) images, and were slightly hyperintense on T2-weighted (T2W) sequences, showing lower enhancement on contrastenhanced T1W images compared to the normal surrounding liver parenchyma. Considering the CT and MR imaging (MRI) findings, and according to the history of splenectomy, the absence of hepatic liver diseases, and normal lab examination, the possibility of multiple abdominal splenosis with particular intrahepatic locations was claimed. The patient subsequently underwent fine needle aspiration biopsy of the largest liver mass at the sixth segment. Histopathologic examination confirmed the benign nature of the lesion, which consisted of splenic tissue. Thus, a final diagnosis of multiple splenosis was made and the patient was discharged without any other complications.



**Fig. 3.** Contrast-enhanced arterial phase CT of the abdomen showing a  $4.6 \times 3.6$  cm lesion adjacent to the mesenteric bowels, in the right anterior para-renal space (arrow).



**Fig. 2.** Contrast-enhanced arterial phase CT of the abdomen showing two additional lesions located within the sixth liver segment of  $4.0 \times 1.8$  cm and along the falciform ligament of  $2.6 \times 1.2$  cm (arrows).



**Fig. 4.** Contrast-enhanced arterial phase CT of the abdomen showing a lesion of  $4.4 \times 3.2$  cm in the left anterior para-vesical space (arrow).

# Discussion

We describe an unusual case of multiple abdominal splenosis with particular intra-hepatic locations, diagnosed with combined CT and MRI, that could be misinterpreted as a primitive hepatocellular carcinoma and histologically confirmed as benign splenic tissue. This case report highlights the rarity of this condition and the difficult differential diagnosis. A careful clinical history evaluation and an appropriate CT and MRI interpretations should be made by clinicians to distinguish splenosis from other similar benign and malignant lesions, in order to select the most appropriate diagnostic and therapeutic approach.

Splenosis is a condition with several cases described in the literature (5-9). Ectopic splenic tissue can be found in the body as two distinct forms: accessory spleens and splenosis. Accessory spleens are congenital, arising from the dorsal mesogastrium during the embryonic period (10), usually solitary, located near the spleen, in the region of gastrosplenic ligaments and receiving blood supply from the splenic artery. Histologically these resemble the normal spleen with a hilus, normal pulp, and splenic capsule. On the other hand, splenic implants are usually multiple and small (11), with a distorted architecture with no hilum, poorly formed capsules, and variable in shape and size (10). They receive blood supply from nearby arteries at the site of implantation that can occur in any part of the body: they may be localized in the peritoneum (10-12), in the thorax (13), in the pelvis (14,15), in the skin (16,17), or even cerebrum (18). In particular, hepatic splenosis refers to a heterotopic implantation of splenic tissues in the liver. Mechanism of splenic tissue spread is not completely understood. One of the hypotheses is that after rupture or capsule compromise, splenic pulp displaces and "seeds" on the liver surface and then penetrates into the depth. Alternatively, spleen cells may hematogenously spread through the splenic vein and subsequently implant in the liver (9,10,19-21). Splenic implants can be partially or fully functioning and usually are asymptomatic; nevertheless, some clinical features have been reported. They mainly depend on size and localization of the ectopic spleen tissues and can include abdominal pain, gastro-intestinal bleeding, bowel obstruction, or appendicitis secondary to adhesive bands of splenic implants associated with compression or sudden torsion of the solid lesion. When it happens, they represent an indication for surgical resection of the ectopic splenic tissue. Around 18 cases of intra-hepatic splenosis have been reported in the literature during the last ten years (22). According to published reports, intra-hepatic splenosis usually manifests as nodular and solitary lesions, in the size range of 2-6 cm (19). In our case, we detected three

intra-hepatic nodules with a maximum diameter of  $6.5 \times 4.6$  cm. In addition, several other intra-abdominal locations were identified near the mesenteric bowels, posteriorly to the cecum and in the left para-vesical space. Intra-hepatic splenosis, in absence of any clinical symptoms, do not require treatments. Thus, the correct identification of this rare condition is particularly important to avoid unnecessary procedures. The differential diagnosis of splenosis is difficult to realize, due to lack of typical radiological features using conventional ultrasound, CT, and MRI and include hepatic or renal malignancies, metastatic disease, pheochromocytoma, lymphoma, carcinomatosis, endometriosis, or adenopathy. Fine needle aspiration is actually considered the gold standard to make a definitive diagnosis of splenosis (9), but it is an invasive procedure which is not immune to complications especially bleeding. Nowadays, the use of <sup>99m</sup>Tc labelled heat-denatured erythrocyte scintigraphy (99mTc-Pyperythrocyte SPECT) allows the diagnosis to be confirmed. It should be considered as the technique of choice due to its great sensitivity and specificity, as previously described (23). Biopsy or operation could be avoided with the application of <sup>99m</sup>Tc-Pyperythrocyte SPECT scintigraphy (24). A missed diagnosis of intra-hepatic splenosis usually has a negative influence on patient's management. Besides, the most difficult differential diagnosis is of accessory spleen. Splenosis may also have some immunologic values and splenic filtering functions, which could be beneficial for organism. Therefore, hepatic splenosis requires no treatment in most cases. When the diagnosis remains unclear, further biopsy or laparoscopy is recommended. If hepatic splenosis is confirmed, careful clinical and diagnostic follow-up with CT or MRI is beneficial.

In conclusion, considering the patient's clinical history of splenectomy or splenic trauma, splenosis should be added to the list of possible differential diagnosis of multiple incidental intra-abdominal or intra-hepatic masses, in order to avoid unnecessary surgical intervention. When imaging features of a lesion are compatible with normal splenic tissue in a patient with splenic trauma or splenectomy, the diagnosis of splenosis should be taken into account.

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