Intrahepatic Splenosis Mimicking Multifocal Hepatocarcinoma in a Patient with Chronic Hepatitis

Adriana Moreira, Pedro Canão, Adriana Sá Pinto, Rui Cunha

Abstract

Intrahepatic splenosis is the heterotopic autotransplantation of splenic tissue in the liver. We present a case of a 48-year-old male with chronic hepatic disease with two hepatic nodules on routine imaging. The nodule's characteristics on MRI made us raise the hypothesis of intrahepatic splenosis (IHS) but hepatocellular carcinoma could not be ruled out. Tc-99m-DRBC scintigraphy results did not favour the IHS hypothesis and the patient underwent surgery to resect the nodules. Histology confirmed the diagnosis of IHS. This report demonstrates that IHS should be considered in the differential diagnosis of hepatic nodules.

Keywords

Abdomen; Liver; Splenosis; Computed tomography; Magnetic resonance imaging.

Background

Splenosis is defined as the heterotopic autotransplantation of splenic tissue throughout the peritoneal cavity, which frequently seeds onto exposed vascularized peritoneal surfaces in up to 67% of patients following splenic trauma or surgery. Most of the reported cases of splenosis occurred as a result of trauma or iatrogenesis involving the spleen early in life. The average interval between the initial trauma or surgery and the onset of splenosis ranges from 5 months to 32 years and lasts an average of 10 years. Because there are various sites of splenosis and it can occur in variable shapes and sizes, it is often misdiagnosed by clinicians. Particularly, it can be misdiagnosed as a malignant tumor. There are few reports in the literature regarding intrahepatic splenosis (IHS), which can be mistaken for a hepatic tumor.

The frequency of IHT is likely underestimated because most splenic implants are asymptomatic and only found either incidentally or after symptomatic complications. Surgical intervention has been reported to be unnecessarily performed to arrive at the correct diagnosis.

Case Presentation

A 48-year-old male was followed in our hospital internal medicine consultation because of chronic heart failure and chronic hepatic disease of alcoholic origin.
Fig. 1 – Abdominal ultrasound shows two solid, hypoechoic liver nodules: nodule one (a) – measuring about 3 cm in the subcapsular region of segment 2; nodule two (b) – measuring 2 cm in segment 7.

Fig. 2 – Contrast enhanced abdominal CT depicting the nodule (arrow) in segment 2 – a) on the unenhanced image the nodule is hard to identify; b) arterial phase shows clear homogenous enhancement of the nodule that fades on portal (c) and late (d) phases.

Fig. 3 – Contrast enhanced abdominal CT depicting the nodule in segment 7 – the nodule (arrow) has similar behavior to the nodule in figure 2.

Fig. 4 – MRI of the abdomen – out (a) and in (b) phases show the hypointense nodule (arrow) in the segment 2. There is no loss of signal in the out-of-phase image to suggest fat content. Unenhanced T1-weighted image (c) shows the hypointense nodule (arrow). There is homogeneous enhancement of the nodule in the arterial phase (d) and washout during portal (e) and subsequent phases (f and g). These characteristics are concerning for hepatocellular carcinoma (HCC). On T2-weighted image without fat saturation (h) the nodule is isointense to the remaining hepatic parenchyma, which is not the most typical behavior of HCC. Note that the spleen is absent and that there is an image in the splenic bed (dashed arrow) with similar characteristics to the hepatic nodule described.
on segment 7 showed a halo of fat in the out-of-phase images but no fat was seen inside the lesion. Based on these characteristics, hepatocellular carcinoma could not be ruled out but there was an image on the post-splenectomy bed that had similar behavior so we raised the hypothesis of these hepatic nodules being nodules of intra-hepatic splenosis.

Tc-99m-labelled heat-denaturated autologous red blood cells scintigraphy (Tc-99m-DRBC scintigraphy) was performed and planar images, single photon emission computed tomography (SPECT) and a low-dose CT were acquired for attenuation correction and anatomical location, 2 hours after administration of the radiopharmaceutical. A mild uptake in the post-splenectomy bed was observed, suggestive of a splenosis implant (Figure 6).

The liver uptake was globally homogenous with no focal uptake in the region of the previously mentioned lesions (Figure 7). These findings did not favor the hypothesis of IHS.

Given the findings described, the patient underwent surgery for resection of the hepatic nodules.

Histopathologic examination showed well-demarcated nodules of splenic tissue in the visceral peritoneal surface of the liver (Figure 8a), which were surrounded by a fibrous capsule and composed by sinusoidal structures with congestion and lymphoid sheaths with scattered follicular aggregates, denoting red and white pulp, respectively (Figure 8b). No hilus was identified. No evidence of malignancy was found. Thus, these features allowed the final diagnosis of splenosis.

Fig. 5 – MRI of the abdomen – out (a) and in (b) phases show the hypointense nodule (arrow) in the segment 2. There is a halo of signal loss in the out-of-phase image surrounding the nodule (dashed arrow), suggesting the existence of a capsule with fat. There is no signal loss inside the nodule. Unenhanced T1-weighted image (c) shows the hypointense nodule (arrow). There is homogeneous enhancement of the nodule in the arterial phase (d) and washout during portal (e) and subsequent phases (f and g). These characteristics are similar to the nodule described in segment 2. On T2-weighted image without fat saturation (h) the nodule is isointense to the remaining hepatic parenchyma (arrow) and the hyperintense capsule (dashed arrow) is noted, suggesting fat content.

Fig. 6 – SPECT-CT fusion images of the abdominal region – mild uptake in the post-splenectomy bed at the Scintigraphy suggesting splenosis focus. Liver uptake was globally homogenous with no focal uptake at the site of the lesions depicted on MRI.
Discussion

Splenosis is recognized as a heterotopic autotransplantation of splenic tissue following splenic trauma or surgery. The disrupted splenic fragments seed the peritoneal surface, acquire a vascular supply, and become clinically evident as a result of regrowth at the implantation sites. Splenosis implants are usually multiple, supplied by arteries from the surrounding tissue and typically have neither hilus nor normal elastic muscular capsule.

The case herein reported is characterized by similar features, demonstrating splenosis implants on the liver surface, surrounded only by a fibrous capsule. Most commonly, these implants are not of clinical significance and are often incidentally detected at autopsy or during abdominal surgery. Although splenosis can occur anywhere, the implants are found most commonly in the peritoneal cavity, such as mesentery, omentum and other peritoneal surfaces. Splenosis is a distinct entity from accessory spleens, which represent failure of the splenic anlagen to fuse during embryogenesis and can be found in 10–40% of patients at autopsy and rarely exceed 1.5 cm in diameter. Accessory spleens are usually solitary, rarely exceed six in number, and are most commonly located on the left side of the dorsal mesogastrium in the region of the splenopancreatic or gastrosplenic ligaments. Histologically, accessory spleens resemble the normal spleen which contain a hilus, a capsule, parenchyma with normal pulp and always receive blood supply from branches of the splenic artery.

Intrahepatic splenosis is the heterotopic autotransplantation of splenic tissue in the liver. The pathogenesis of intrahepatic splenosis is still unclear. However, it is considered to develop from seeding of splenic fragments into serosal surfaces at the time of splenic trauma or splenectomy. Moreover, based on the susceptibility of the splenic erythropoiesis response to hypoxia and the inevitability of hypoxia caused by aging or pathological changes, the two events, 1) the migration of the erythrocytic progenitor cells via the portal vein following traumatic splenic rupture and 2) the induction of erythropoiesis by local hypoxia of liver, might cause the occurrence of the intrahepatic splenosis. Fewer than 20 cases of intrahepatic splenosis have been reported until now. However, IHS is not widely recognized by most physicians; therefore, its incidence may be underestimated. IHS can be misdiagnosed as liver cancer and have patients undergo surgical resection, in which case it is only intraoperatively that the splenic tissue is found and confirmed by subsequent surgical pathology.

AFP is the most commonly used indicator of HCC screening and it plays an important role in the diagnosis, the evaluation of clinical efficacy, and the monitoring of tumor relapse. However, the AFP level may be normal in 35% to 45% of HCC patients, indicating the diagnostic limits of single HCC biomarker and nearly 40% of HCC patients may be missed. There are no typical radiological features of intrahepatic splenosis and it is usually difficult to distinguish this
condition from other liver tumors. In the presence of chronic liver injury, establishing the correct diagnosis can prove to be difficult.

The primary reason for misdiagnosis is that HIS often develops in patients with chronic hepatitis, and the radiographic features are quite similar to those of HCC, showing early enhancement in the arterial phase and washout in the equilibrium phase.\(^5\)\(^7\)

Tc-99m-DRBC scintigraphy is considered the technique of choice to access IHS. It is a non-invasive nuclear medicine procedure with great sensitivity and specificity.\(^4\) 90% of the damaged cells will be trapped by the splenic tissue, allowing a good uptake of the radioactive compound and an excellent diagnostic accuracy, even considering the normal liver tissue uptake.\(^9\) Due to this, this method may potentially avoid invasive interventions.\(^3\) In this case, the Tc-99m-DRBC scintigraphy was not consistent with the splenosis hypothesis. This was probably due to the dimensions of the lesions, which might be too small to the SPECT resolution.

In most cases in the literature of intrahepatic splenosis mimicking HCC, the correct diagnosis was only possible on histological examination after a laparotomy and open liver resection.

Standard imaging modalities, such as CT or and Gd-EOB-MRI, often cannot differentiate IHS from HCC. Unfortunately, in our case, there was no sufficient evidence to support the hypothesis of HIS and so liver resection was performed.

When confronted with a suspicious hepatic nodule, the clinical manifestations of patients combined with the auxiliary examination results, and the special history of splenectomy or traumatic rupture of the spleen, clinicians should be alert to the possibility of an splenosis nodule. If this diagnosis is confirmed before surgery, it will be possible to avoid an unnecessary procedure.

**Conclusion**

IHS is not widely recognized by most physicians and radiologists and can be misdiagnosed as liver cancer, subjecting patients to surgical resection. When confronted with a suspicious hepatic nodule, in a patient with prior history of splenectomy or traumatic rupture of the spleen, the radiologist should have the possibility of splenosis nodule in mind when making the differential diagnosis to avoid unnecessary surgery.

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