ACTA RADIOLÓGICA PORTUGUESA
January-April 2019 | Vol 31 nº 1 | 31-35

Radiological Case Report / Caso Clínico

Intrahepatic Splenosis Mimicking Multifocal Hepatocarcinoma in a Patient with Chronic Hepatitis

Esplenose intra-hepática simulando Hepatocarcinoma Multifocal num doente com Hepatite Crónica

Adriana Moreira[1], Pedro Canão[2], Adriana Sá Pinto[3], Rui Cunha[1]

[1]Department of Radiology, Centro Hospitalar de São João, Porto, Portugal
[2]Department of Anatomical Pathology, Centro Hospitalar de São João, Porto, Portugal
[3]Department of Nuclear Medicine, Centro Hospitalar de São João, Porto, Portugal

Address

Adriana Moreira
Centro Hospitalar de São João
Serviço de Radiologia
Alameda Prof. Hernâni Monteiro
4200-319 Porto, Portugal
e-mail: adriana.clmoreira25@gmail.com

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.

Resumo

A esplenose intra-hepática é um autotransplante heterotópico de tecido esplénico no fígado.
Os autores apresentam o caso de um homem de 48 anos com doença hepática crónica que apresentou dois nódulos hepáticos em exames de rotina. As características dos nódulos em RM levaram os autores a levantar a hipótese de esplenose intra-hepática (EIH) mas um hepatocarcinoma não podia ser excluído. Os resultados da cíntigrafia com Tc-99m-DRBC não favoreceram a hipótese de EIH e o paciente foi submetido a cirurgia para resseção dos nódulos. A histologia confirmou o diagnóstico de EIH.
Este caso demonstra a importância de considerar a EIH no diagnóstico diferencial de nódulos hepáticos.

Palavras-chave

Abdómen; Fígado; Esplenose; Tomografia computorizada; Ressonância magnética.

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.[1,2] 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.[3]
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.[4]
There are few reports in the literature regarding intrahepatic splenosis (IHS), which can be mistaken for a hepatic tumor.[5-7]
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.[7]

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.
He had a medical history of hypertension and type 2 diabetes, dilated miocardiopathy associated with the alcohol consumption and had undergone splenectomy for traumatic splenic rupture 30 years ago. His family history was non-specific.
The physical examination showed that the patient was in good overall health, anicteric, with no pain in the abdomen, and had no palpable masses.
Blood examination revealed normal levels of hepatic enzymes and of the alpha-fetoprotein.
An abdominal ultrasound was requested to evaluate the chronic hepatitis and showed two solid and hipoecoic liver nodules (Figure 1).
Abdominal contrasted CT revealed lobulated hepatic contour suggesting chronic hepatic disease and two well-circumscribed, low density nodules, measuring 20 mm in the subcapsular region of the segment 7 and 25 mm in the segment 2. The dynamic study showed that both lesions had homogeneous enhancement in the arterial phase and "fading" during portal and equilibrium phases (Figures 2 and 3).
Further characterization was made by magnetic resonance imaging (MRI) (Figures 4 - 5), and both lesions had homogeneous arterial enhancement and washout in the subsequent phases of the dynamic contrasted study. However, although they were difficult to depict on T2-weighted images, there was slight hypointensity. The nodule

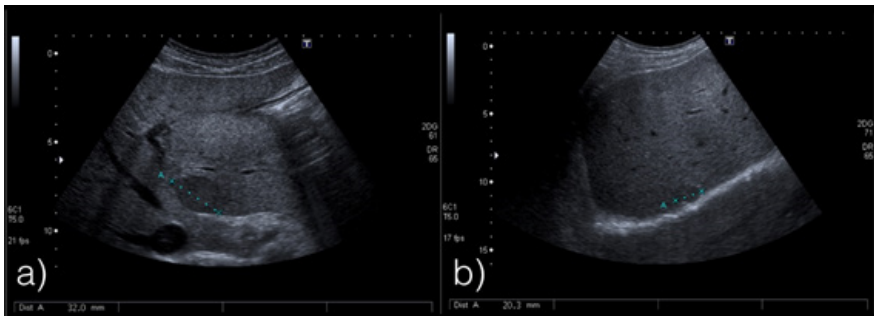

Fig. 1 – Abdominal ultrasound shows two solid, hypoecoic liver nodules: nodule one (a) - measuring about 3 cm in the subcapsular region of segment 2; nodule two (b) - measuring 2 cm in segment 7.

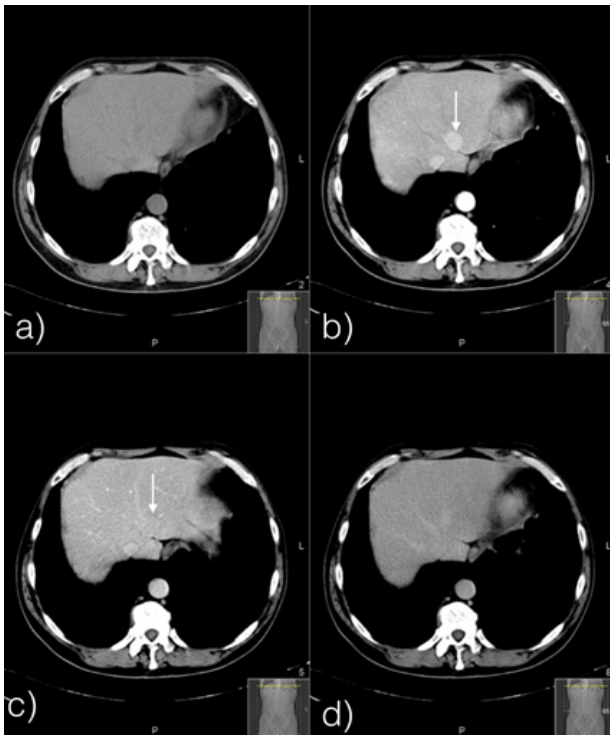

Fig. 2 – Contrast enhanced abdominal CT depicting the nodule (arrow) in segment 2 – a) on the unenhanced image the nodule is hard to identify; b) arterial phase shows clear homogenous enhancement of the nodule that fades on portal (c) and late (d) phases.

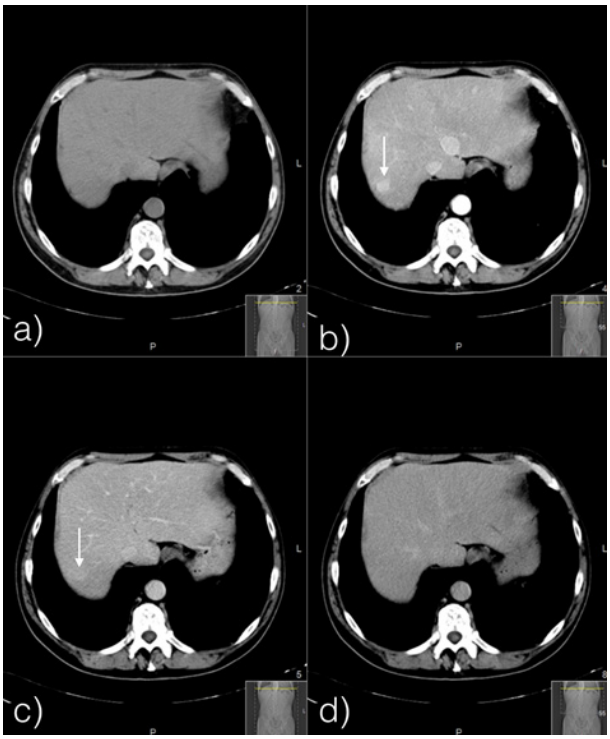

Fig. 3 – Contrast enhanced abdominal CT depicting the nodule in segment 7 – the nodule (arrow) has similar behavior to the nodule in figure 2.

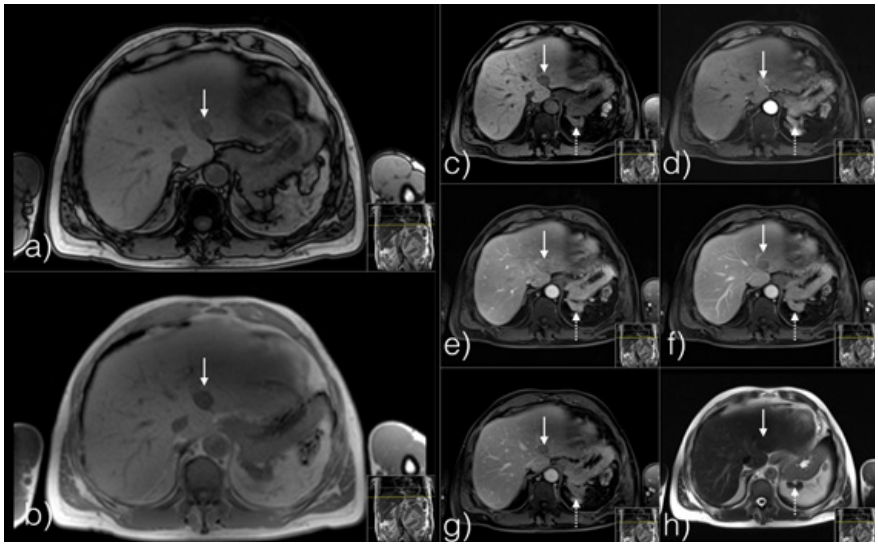

Fig. 4 – MRI of the abdomen – out (a) and in (b) phases show the hypointense nodule (arrow) in the segment 2. There is no loss of signal in the out-of-phase image to suggest fat content. Unenhanced T1-weighted image (c) shows the hipointense nodule (arrow). There is homogeneous enhancement of the nodule 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.

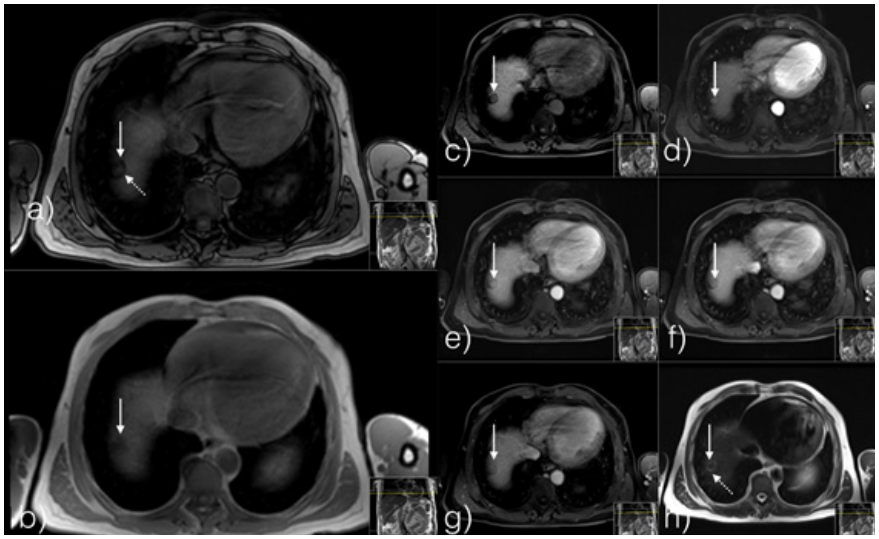

Fig. 5 – MRI of the abdomen – out (a) and in (b) phases show the hypointense nodule (arrow) in the segment 2. There is a halo of signal loss in the out-of-phase image surrounding the nodule (dashed arrow), suggesting the existence of a capsule with fat. There is no signal loss inside the nodule. Unenhanced T1-weighted image (c) shows the hipointense 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.

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.

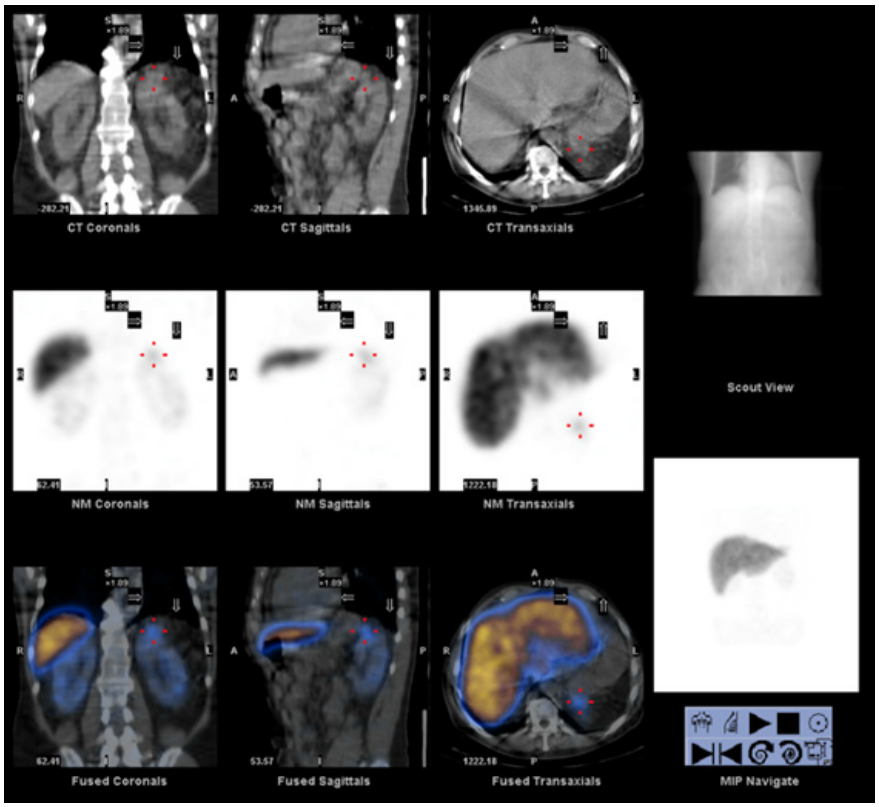

Fig. 6 – SPECT-CT fusion images of the abdominal region- mild uptake in the post-splenectomy bed at the Scintigraphy suggesting splenosis focus. Liver uptake was globally homogenous with no focal uptake at the site of the lesions depicted on MRI.

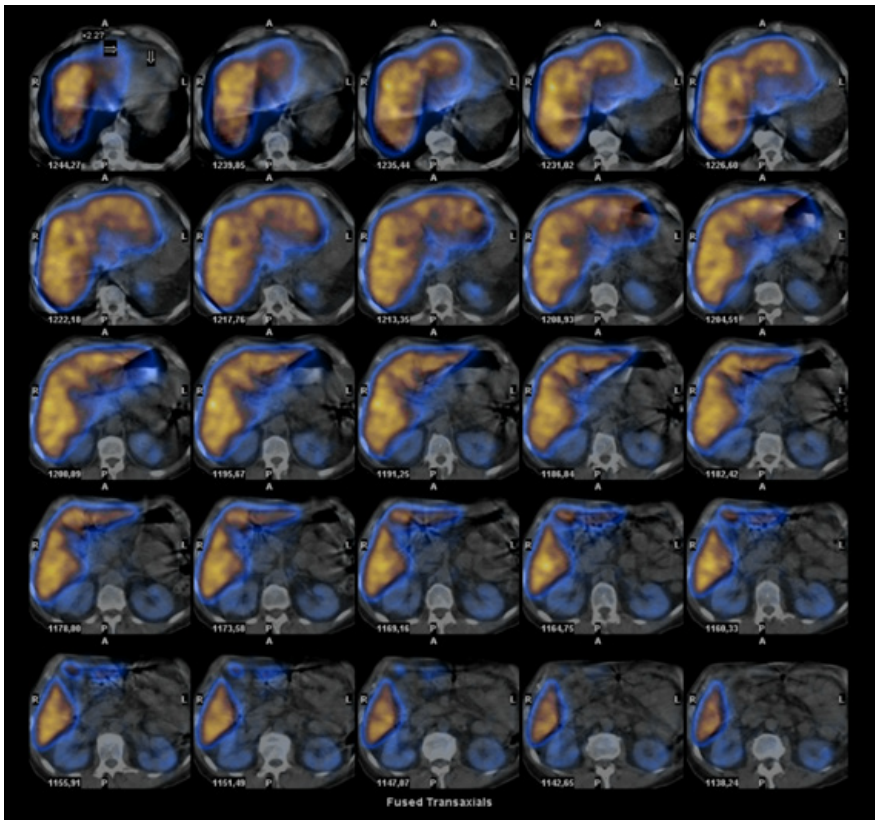

Fig. 7 – SPECT-CT fusion images of the abdominal region- liver uptake was globally homogenous with no focal uptake at the site of the lesions depicted on MRI.

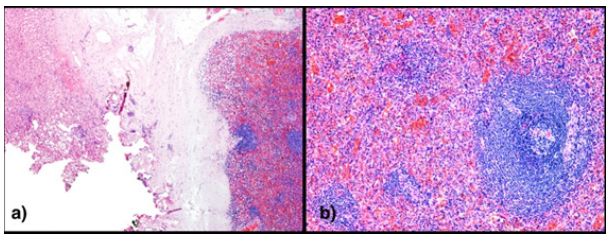

Fig. 8 – Histological features of splenosis. (A) Photomicrography showing the splenic tissue surrounded by a fibrous capsule in the visceral peritoneal surface of the liver (H&E, x40); (B) Higher magnification view of splenic tissue showing red pulp composed of vascular sinuses with congestion and white pulp composed of lymphoid aggregates (H&E, x100).

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.[7,8] 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.[9]

Although splenosis can occur anywhere, the implants are found most commonly in the peritoneal cavity, such as mesentery, omentum and other peritoneal surfaces.[10]

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.[11]

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.[8,12]

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.[11]

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.[13]

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.[14] Due to this, this method may potentially avoid invasive interventions.[4] 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.

Received / Recebido 18/12/2018
Acceptance / Aceite 26/01/2019

Ethical disclosures / Divulgações Éticas

Conflicts of interest: The authors have no conflicts of interest to declare.
Conflitos de interesse: Os autores declaram não possuir conflitos de interesse.
Financing Support: This work has not received any contribution, grant or scholarship.
Suporte financeiro: O presente trabalho não foi suportado por nenhum subsídio ou bolsa.
Confidentiality of data: The authors declare that they have followed the protocols of their work center on the publication of data from patients.
Confidencialidade dos dados: Os autores declaram ter seguido os protocolos do seu centro de trabalho acerca da publicação dos dados de doentes.
Protection of human and animal subjects: The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).
Protecção de pessoas e animais: Os autores declaram que os procedimentos seguidos estavam de acordo com os regulamentos estabelecidos pelos responsáveis da Comissão de Investigação Clínica e Ética e de acordo com a Declaração de Helsínquia da Associação Médica Mundial.

References

1. Abu Hilal M, Harb A, Zeidan B, et al. Hepatic splenosis mimicking HCC in a patient with hepatitis C liver cirrhosis and mildly raised alpha feto protein; the important role of explorative laparoscopy. World J Surg Oncol. 2009;7:1.
2. Fleming CR, Dickson ER, Harrison EG. Splenosis: autotransplantation of splenic tissue. Am J Med. 1976;61:414-19.
3. Fremont RD, Rice TW. Splenosis a review. South. Med. J. 2007;100:589-93.
4. Wang WC, Li XF, Yan ZL, et al. Intrahepatic splenosis mimics hepatocellular carcinoma in a patient with chronic hepatitis B: A case report and literature review. Augustin. G, ed. Medicine. 2017;96:e8680.
5. Yoshimitsu K, Aibe H, Nobe T, et al. Intrahepatic splenosis mimicking a liver tumor. Abdom Imaging, 1993;18:156-8.
6. Lee JB, Ryu KW, Song TJ, et al. Hepatic splenosis diagnosed as hepatocellular carcinoma: report of a case. Surg Today. 2002;32:180-2.
7. Yu H, Xia L, Li T, et al. Intrahepatic splenosis mimicking hepatoma. BMJ Case Reports. 2009.
8. Wu C, Zhang B, Chen L, Zhang B, Chen X. Solitary perihepatic splenosis mimicking liver lesion: a case report and literature review. Coito. AJ, ed. Medicine. 2015;94:e586.
9. Grande M, Lapecorella M, Ianora AA, et al. Intrahepatic and widely distributed intraabdominal splenosis: multidetector CT, US and scintigraphic findings. Intern Emerg Med. 2008;3:265-7.
10. Turkmen C, Yormaz E. Incidental detection of multiple abdominopelvic splenosis sites during Tc-99m HMPAO-labeled leukocyte scintigraphy. Clin Nucl Med. 2006;31:572-4.
11. Weiand G, Mangold G. Accessory spleen in the pancreatic tail – a neglected entity? A contribution to embryology, topography and pathology of ectopic splenic tissue. Chirurg. 2003;74:1170-1.
12. Kwok CM, Chen YT, Lin HT, et al. Portal vein entrance of splenic erythrocytic progenitor cells and local hypoxia of liver, two events cause intrahepatic splenosis. Med Hypotheses. 2006;67:1330-2.
13. Sato N, Abe T, Suzuki N, et al. Intrahepatic splenosis in a chronic hepatitis C patient with no history of splenic trauma mimicking hepatocellular carcinoma. The American Journal of Case Reports. 2014;15:416-20.
14. Wang CS, Lin CL, Lee HC, et al. Usefulness of serum des-gammacarboxy prothrombin in detection of hepatocellular carcinoma. World J Gastroenterol. 2005;11:6115-9.