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Case Report

Intrahepatic splenosis demonstrated by diffusion weighted MRI with histologic confirmation

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ABSTRACT

Acquired ectopic splenic tissue is called splenosis, which is common after the history of trauma or surgical exploration. We present a rare case of intrahepatic splenosis in 36-year-old male patient mimicking a liver neoplasm on imaging however presented with left flank pain for 5 months and had remote history of splenectomy after splenic rupture from trauma. We discuss various imaging modalities and the role of various magnetic resonance imaging sequences and nuclear medicine examination. We also discuss the differentiating features to be kept to make the correct diagnosis along with a brief review of literature. We mentioned signal intensities of splenic lesions and normal signal intensity of spleen in different magnetic resonance imaging sequences and with high suspicion how we can diagnose splenosis and avoid unnecessary biopsy and its result related stress.

Introduction

Presence of ectopic splenic tissue can be congenital (accessory spleen) or may be acquired when it is known as splenosis. Splenosis by definition is autotransplantation or implantation of the normal splenic tissue at a different site from the usual site of spleen [1]. It generally occurs in around 65% of the patients with a history of splenic trauma, surgical exploration, and rarely splenic rupture due to tick borne disease [1–3]. Often, asymptomatic, however patients may present with non-specific pain, bleeding, and sometimes anemia due to red cell sequestration [1]. In this article, we have discussed the epidemiology, clinical presentation, and management of rare in-

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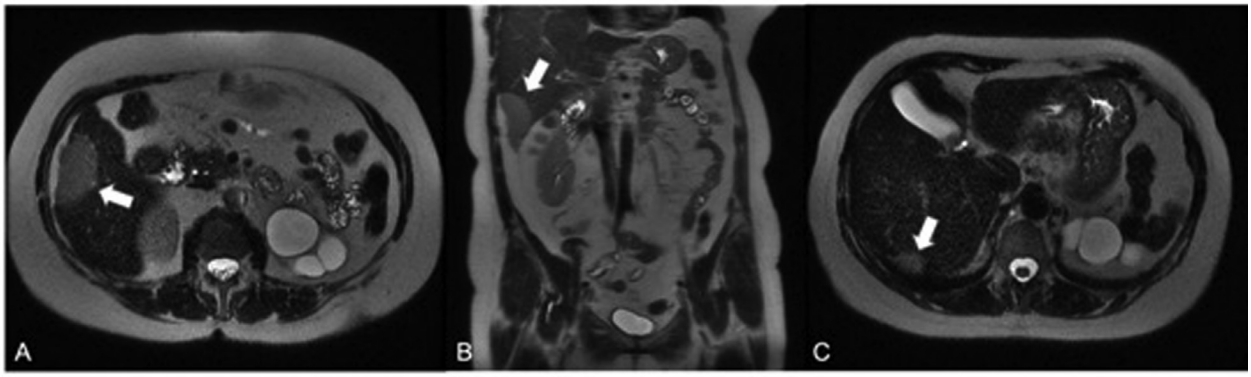

Fig. 1 – T2WIs showing relatively T2 hyperintense 5.0 × 5.0 × 3.0 cm well-circumscribed lesion in the inferior right hepatic lobe (white arrow in images A and B). The lesion shows well-defined borders with the liver parenchyma. Similar intensity but smaller lesion is also seen on posterior right hepatic lobe (white arrow in image C). Note absence of normal spleen in the left upper quadrant.

trahepatic splenosis, with stress on functional magnetic resonance imaging (MRI) using diffusion weighted imaging.

Case report

A 36-year male presented with intermittent left flank pain for 5 months. He underwent ultrasound on an outside institution, which showed left-sided gross hydronephrosis, which likely resulted in left flank pain. However, incidental note was made of a well-defined right hepatic mass lesion. Thus, the patient was referred to us for further evaluation with MRI. Patient had no history of unintended weight loss, anorexia, or weakness. Significant history of splenectomy several years back for traumatic splenic rupture following post motor vehicle accident. No personal history of alcohol abuse, substance abuse, and hepatitis infection.

Lab investigations including complete blood count, basic metabolic panel, liver function test, and alpha-fetoprotein levels were unremarkable. MRI abdomen without and with gadolinium-based contrast agent (liver mass protocol) showed a well-defined approximately 5 cm T2 hyperintense mass in the right hepatic lobe, with no distinct fat plane with the liver parenchyma (Fig. 1A and 1B). Smaller lesion with similar signal characteristics and border was also noted in posterior right hepatic lobe (Fig. 1C). These lesions showed avid enhancement on post contrast T1 fat saturated images (Fig. 2A and 2B). These lesions showed high signal on diffusion weighted images (Fig. 3A and 3B) with corresponding low signals on apparent diffusion coefficient (ADC) images (Fig. 3C and 3D). Based on the imaging characteristics and prior history of splenic rupture, these likely represented splenosis. Due to lack of other areas of peritoneal or extrahepatic splenosis, confirmation was obtained with the tissue sampling from the larger hepatic lesion through computed tomography (CT)-guided core biopsy, which confirmed normal splenic tissue on tissue sampling—thus confirming the diagnosis of intrahepatic splenosis. Patient was managed conservatively.

Discussion

Splenosis is a rare condition and it is crucial to differentiate intrahepatic splenosis from liver tumors, to avoid unnecessary surgical exploration [1]. Differentiation between accessory spleen and splenosis requires carefully noticing the location, size, capsule, internal characteristics, and feeding arteries [1,4]. Accessory spleens are usually located in the region of spleno-pancreatic or gastro-splenic ligaments and always on the left of the dorsal mesogastrium. On the other hand, splenosis implants are often unpredictable in location and can be seen intraperitoneally, along the serosal surface of the bowel, in the greater omentum, along the peritoneum, in the pelvic cavity, or along the surface of the diaphragm including occasional site within the liver parenchyma, pericardium and subcutaneous tissue [1,4].

Accessory spleens are generally round, well encapsulated and contain normal hilum within a normal splenic tissue and are supplied by small branches of the splenic artery. Accessory spleen is more common than splenosis and is often found during autopsy or intra-abdominal surgery for an unrelated pathology. Accessory spleens generally measure up to 1.5 cm in diameter and are maximum up to 6 in number and are a result of failure of fusion of the normal splenic analgen tissue during embryonic development [4]. However, splenosis are unencapsulated or poorly encapsulated, have no characteristic shape, and contain a portion of the distorted splenic tissue [4]. Splenosis recruit arteries from surrounding structures and can range from a few millimeters to a few centimeters in size with a reported maximum size of 12 cm [1,4,5].

One of the rare locations of the splenosis is intrahepatic, as in the present case, where it can mimic a hepatic mass and can be easily mistaken for lesions like hepatic adenoma, focal nodular hyperplasia, regenerative or dysplastic nodules, hemangioma, or hepatocellular carcinoma (HCC) [1,4,6]. It has been postulated that intrahepatic splenosis is secondary to hematogenous spread of splenic tissue fragments after splenic surgery or splenic trauma [1,2]. Another hypothesis

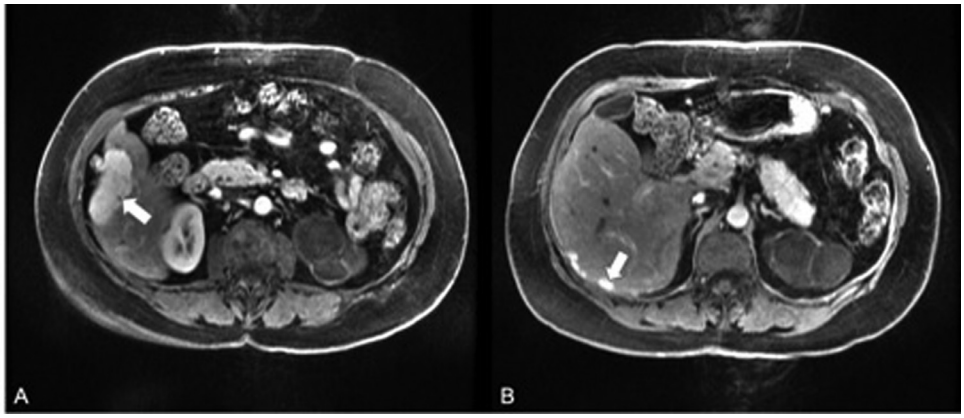

Fig. 2 – T1 post contrast fat-sat axial T1 weighted images show avid enhancement of the bigger lesion (white arrow in image A) and smaller lesions (white arrow in image B). Note that the spleen is absent. Incidental left sided gross hydronephrosis.

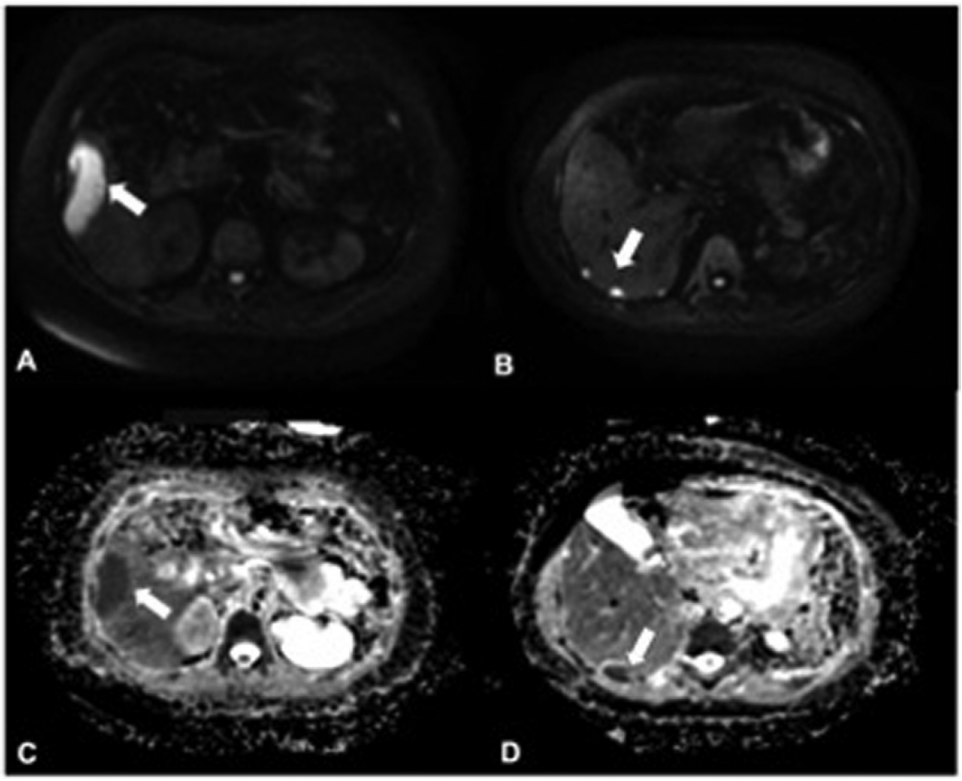

Fig. 3 – DWI and ADC maps of the lesions. The bigger and smaller lesions show high signal on DWI (images A and B, respectively) with corresponding low signal on ADC map (images C and D, respectively), showing true restricted diffusion. These imaging features show that the lesions follow splenic signal on all sequences.

states that gradual invasion of the subcapsular perihepatic splenic implants within the hepatic parenchyma due to constant compression and contraction of the diaphragmatic muscles leads to intrahepatic splenosis [7]. Splenosis is usually asymptomatic and found incidentally during imaging or surgical exploration for the other conditions, however can sometimes also present with nonspecific abdominal pain, diarrhea or small bowel obstruction due to formation of adhesions [1,4,5]. The lesion seen in our patient could be intracapsular or extracapsular as it is along the hepatic capsule on imaging and no surgery performed to directly visualize the exact location of the hepatic lesion.

A rare case of intracranial splenosis has also been previously reported [7]. Intrathoracic splenosis have been known to be misinterpreted as primary lung or mediastinal neoplasms [1,4,9]. In a patient with pleural-based nodules, and a history of the thoracoabdominal trauma with diaphragmatic rupture should include thoracic splenosis high up on the list of

differential diagnosis [9]. The average interval between trauma and abdominal splenosis is approximately 10 years, ranging from 5 months to 32 years [8,9].

The specific diagnosis of splenosis can be made via MRI or nuclear medicine exams. Scintigraphy studies include Tc-99m heat damaged autologous red blood cell (RBC) scintigraphy and Tc-99m sulfur colloid scintigraphy. Tc99m RBC imaging is more specific, as 90% of Tc-99m labelled autologous RBCs are taken by reticuloendothelial cells, which is helpful to identify the splenic tissue [1]. Overlap of uptake within liver and spleen or poor splenic uptake altogether makes sulfur colloid imaging less specific and inconclusive for the diagnosis of splenosis [4]. Another less common scintigraphic agent In-111 labeled platelets are more sensitive than Tc-99m sulfur colloid scintigraphy, as they have also higher predilection toward reticuloendothelial cells in the liver, spleen, and bone marrow [8]. CT findings of splenosis are nonspecific as well. It is seen as a discrete noncalcified hypodensity on a noncontrast images, with homogenous enhancement on delayed arterial phase and slight enhancement on venous phase. This can be confused with a neoplastic process with similar enhancement pattern, especially if splenosis is present in the liver or anywhere in the abdominal or thoracic cavity [4,6]. Ultrasound imaging, even though being cost effective nature and with no ionizing radiation exposure to the patient, is also nonspecific, and demonstrates a hypoechoic to isoechoic mass, with venous and arterial flow if located intrahepatically, this again is very concerning for a neoplastic process [4,7,8,16].

MRI is widely used for staging of intra-abdominal and pelvic tumors, as the findings are more specific [15]. MRI has higher spatial resolution as compared to scintigraphic studies and higher contrast resolution as compared to CT imaging [5]. On T1 weighted image splenosis is seen as a homogeneous low signal structure with a thin hypointense rim, which represents the fibrous capsule which is also showing hypointensity on T2 weighted image (T2WI) and by far the fibrous capsule is more common in splenosis as compared to primary hepatic neoplasm [6,8]. On T2WI, it appears as a homogeneous isointense to hyperintense structure, just like normal splenic tissue and can be easily compared to a normal spleen [4]. Diffusion imaging is a promising sequence for diagnosis of tumors, infarcts, and abscesses and is also useful in diagnosis of splenosis [14]. Diffusion imaging is based on spin echo T2 weighted sequence with bipolar rephasing and dephasing gradients situated at 180° of refocusing pulse gradient [10,11]. If the water molecule is not moving in the imaged field there is production of an additional phase shift during dephasing gradient, which cancels the effect of rephasing gradient and that leads to a loss of signal of the static water molecule [10,11]. On the contrary, in restricted diffusion environment, water molecules are not static and so every time during exposure to rephasing and dephasing gradients, these moving water molecules retain their signal intensity. At least 2 different b values are used in DWI for the quantitative analysis of restricted diffusion which is known as ADC mapping [10,11]. The “b values” are basically strength and duration of rephasing and dephasing gradients and time between them. Generally, 1 smaller b value (<100 s/mm^2) and 1 larger b value (>500 s/mm^2) is used which gives increased sensitivity to diffusion [11]. One interesting and commonly seen feature of the normal splenic tissue is that it has the most restricted diffusion with lowest ADC value as compared to other normal intra-abdominal organs [5,10]. Even though it can be difficult to differentiate between intrahepatic splenosis from hepatic tumors on the basis of contrast enhancement, restricted diffusion of the splenic tissue is quite helpful [10]. Diffusion imaging is also helpful when gadolinium administration is restricted due to its contraindications in renal failure patients.

Superparamagnetic iron oxide (SPIO) contrast-enhanced MRI is rarely performed, however, can be used to differentiate between splenosis and well-defined HCC. SPIO particles are taken up by the normal reticuloendothelial cells including the splenic tissue and cause signal loss on the T2WI, making splenosis T2 hypointense on SPIO-enhanced MRI. On the other hand, a HCC does not lose signal on T2WI [5,7]. Another less commonly used sequence for diagnosing splenosis is chemical shift imaging, where hemosiderin deposition in the normal splenic tissue causes signal drop and hypointensity on in-phase T1 imaging as compared to out of phase imaging [12].

Diagnosis of not so common intrahepatic splenosis requires a high level of suspicion, which includes thorough history taking including past trauma or surgical exploration. In a background of hepatitis or cirrhosis, correlation with alpha fetoprotein level should be done to exclude a possible HCC [1,13]. If alpha fetoprotein is normal, further evaluation with nuclear scintigraphy studies with heat denatured RBCs or special MRI sequences like SPIO-enhanced or diffusion weighted MRI imaging can be performed to avoid unnecessary percutaneous biopsy or surgical removal. Accessory spleen is a common mimicker, which should always be considered in the differential diagnosis, and correlation with clinical history and past imaging should be performed.

Conclusion

- Knowledge of splenic lesions and normal signal intensity of spleen in different MRI sequences are very important facets to diagnose splenosis, which can easily helpful to avoid unnecessary intervention and pathology result related stress. High level of awareness is important in such scenarios especially proper history has been taken including remote history of trauma.
- Even though pulse sequences like chemical shift imaging and SPIO contrast-enhanced MRI are helpful in making the diagnosis of splenosis they are not frequently used on the contrary DWI is commonly used in almost all the patients undergoing MRI.

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