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Case Report

Contribution of the single photon emission computed tomography with 99mTc red blood cells in splenosis

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ABSTRACT

The term splenosis refers to the presence of auto-transplanted splenic tissue in a heterotopic location. These foci can be localized to the liver simulating a malignant lesion. Sometimes these lesions are difficult to identify using conventional imaging techniques (ultrasound, CT and MR). Then, a scan with denatured erythrocytes marked with 99mTc has proven to be an effective technique to confirm the diagnosis of splenosis and to establish its extension. The incorporation of hybrid imaging techniques (SPECT-CT) into usual clinical practice has increased the precision of the localization of these foci of splenosis. We hereby report the cases of two patients diagnosed with splenosis, the first by laparotomy and the second after performing scintigraphy with red blood cells labeled with 99mTc. In the first case, the laparotomy revealed numerous reticulated nodules on the diaphragmatic peritoneal surface, the transverse colon and the right kidney. Finally, the anatomopathological diagnosis confirmed a case of splenosis. In the second case, the results of the 99mTc marked red blood cell gammagraphy and SPECT-CT were consistent with the diagnosis of splenosis in the patient. To obtain correct information in cases of lesions highly suspicious of splenosis, 99mTc marked red blood cell gammagraphy should be performed due to the high sensitivity and specificity of the test. Combined diagnostic imaging (SPECT-CT), have increased the specificity of this test due to improvements in the characterization of lesions. We believe that the use of this technique will help avoid unnecessary surgical procedures.

Key words: Splenosis, SPECT-CT, Ultrasound, Spleen, Hepatocellular carcinoma

INTRODUCTION

Splenosis is caused by heterotopic auto-transplantation and the implantation of splenic tissue occurring in approximately 26% to 67% of patients with a traumatic rupture of the spleen or a splenectomy.[1-3] Splenic implants can be localized anywhere in the abdominal cavity, with the most frequently affected areas being the omentum and the parietal and visceral peritoneum. Nodules are characteristically multiple, diffusely spread throughout the peritoneal cavity and appear as small reddish blue nodules.[4] the blood supply to these splenic implants is derived from small penetrating vessels arising from donor surfaces.[4] The extra peritoneal foci (i.e., the retro peritoneum, liver, lung, pleural cavity or abdominal wall) are less commonly affected and usually related with more severe thoracic - abdominal trauma.[5,6]

On rare occasions, such as the occurrence of an intrahepatic lesion, the differential diagnosis applied for splenosis would be the same as that applied for the detection of a hepatocarcinoma.[7] This article discusses two cases of patients with a confirmed hepatic lesion identified by computed tomography (CT) and magnetic resonance imaging (MRI).

CASE REPORT

Case 1

The first case describes a thirty-year-old male with a 12-year-history of liver dysfunction and hepatitis C. With an abdominal ultrasound, a homogeneous and hipoechoic mass with a diameter of 2.8 x 2.0 cm was detected on the VII hepatic segment. During MRI analysis of the mass, the weighted images showed low intensity signals for T1 and high intensity signals for T2 (Figure 1).

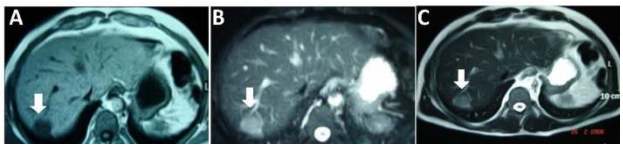

Figure 1: Different magnetic resonance (MR) images showing (a) gadolinium enhancement, (b) low intensity signals for T1 and (c) high intensity signals for T2.

In a dynamic study with gadolinium, the mass was more intensely highlighted in the arterial phase. Both of these findings strengthen the diagnosis of hepatocellular carcinoma.

On this basis, the patient underwent a laparotomy in which numerous reticulated nodules (ranging from 0.5 to 1 cm in size) were found on the diaphragmatic peritoneal surface, the transverse colon and the right kidney.

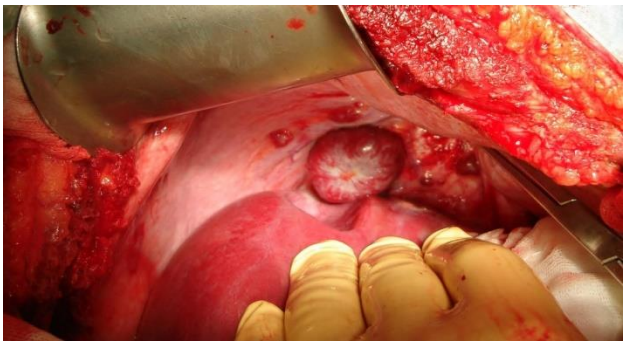

Figure 2: Intraoperative picture revealing splenic tissue 4 x 3 cm in size implanted in the diaphragm and invading the hepatic surface where a fovea can be observed.

The initially suspected diagnosis of hepatocellular carcinoma was the result of the implantation of a 4x3-cm splenic tissue in the diaphragm, pressing against the hepatic surface and causing its deformation (Figure 2). Definitive anatomopathological analysis confirmed a case of splenosis (Figure 3).

The patient had suffered from an abdominal trauma 27 years before and consequently underwent splenectomy and several blood transfusions.

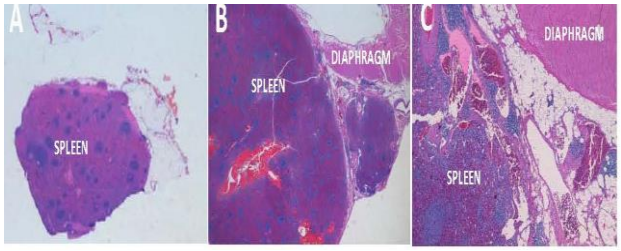

Figure 3: Microscopic image showing abnormally located spleen parenchyma in muscle tissue of the diaphragm.

Case 2

The second case describes a fifty-seven-year old male with high PSA values. During an ordinary abdominal ultrasonography, two hepatic space-occupying lesions were detected. The patient was asymptomatic, and no sign of chronic liver disease was detected. The patient was involved in two traffic accidents, one of which caused him to undergo a splenectomy.

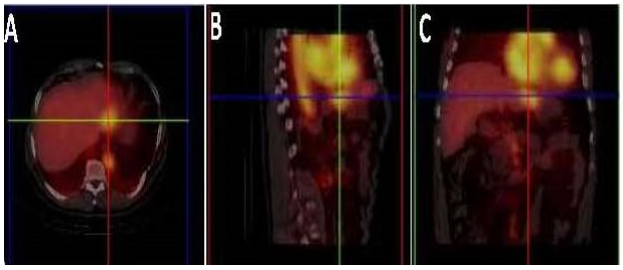

Figure 4: SPECT-TAC lesion located in the left hepatic lobe; a) Axial, b) sagittal and c) coronal planes.

During a hepatic MRI, two partially enucleated lesions were identified within the liver parenchyma. The first, 42x45 mm in size, was found in the I-II hepatic segments, and the second, 35x36 mm in size, was found in the V-VI hepatic segments. Both lesions suggested multifocal hepatocellular carcinoma. The images obtained show that, during standard hepatic gammagraphy with colloid 99mTc, an activity defect of the marker within the lesion found in the left hepatic lobe coincided with an area of ongoing hyperactivity of red blood cells marked with 99mTc. In the combined images (SPECT-CT), both areas were correlated with a lesion found in the left hepatic lobe (most likely the I-II segment). An area of hyperactivity was also detected in the right hepatic lobe (Figure 4). These lesions coincide with those previously found during CT and MRI detection.

DISCUSSION

Splenosis was first reported by Shaw and Shafi as an autopsy finding in 1937.[8] The term “splenosis” was firstly used by Buchbinder and Lipkoff in 1939 to describe the heterotopic transplantation of splenic tissue.[9] Splenosis

after abdominal blunt trauma, it is not a rare condition and it is estimated to occur in up to 67% of patients with traumatic splenic rupture. Splenosis has been reported mainly in the left segment of the abdomen, but it can also occur in the chest when diaphragm is damaged after a trauma.[1,2,4] The average interval reported between trauma and abdominal or pelvic splenosis is 10 years, with a range of 5 months to 32 years.[10] In order of frequency, the most common sites are the serosal surface of the small bowel, greater omentum, parietal peritoneum, serosal surface of the large intestine and mesentery.[2] However, unusual sites of splenic tissue, such as extensive involvement in the liver, pancreas, stomach, kidneys, ureters, bladder, uterus, or fallopian tubes have also been previously described.[4] Splenosis is generally observed in multiple rather than solitary locations, as its primary mechanism of auto transplantation initiated with the splenic rupture is the seeding of damaged splenic pulp into adjacent cavities, similar to endometriosis or malignancy (Figure 1).[2] An alternative probability of intrahepatic splenosis is that the splenic tissue and cells migration to the subcapsular location in the liver through the blood or lymphatic vessels after trauma.[11] In this regards, Kwok et al[12] hypothesized that the migration of erythrocytic progenitor cells through the portal vein following traumatic splenic rupture could promote intrahepatic splenosis development. The presentation of splenosis after liver injury is unusual. In fact, there are fewer than 30 reported cases in the literature.[7,13] In these cases, splenosis mimicking solid lesions is likely to be mistaken for malignant tumors. Before a liver injury, differential diagnosis usually includes hemangioma, adenoma, focal nodular hyperplasia, regenerative nodules, dysplastic nodules, hepatocellular carcinoma metastatic cancer, endometriosis, hemangiomas and accessory spleens.[1,4] However, due to its low rate of occurrence, liver splenosis is not included in the initial differential diagnosis. Thus, Wu C *et al*, through a pubmed based literature review for liver splenosis case reports published between 1900 and 2014, showed that approximately 76% (26/34) of patients diagnosed with liver splenosis had as initial diagnosis a hepatic neoplasia.[14] Liver splenosis must be suspected in patients with hepatic mass who have a history of abdominal trauma involving the spleen or have had a splenectomy. However, Sato N *et al* recently described the first case of intrahepatic splenosis in a chronic hepatitis C patient without a history of splenic trauma or surgery.[7] In this sense, the majority of the reported intrahepatic splenosis are associated to hepatitis virus infection and/or liver cirrhosis. Probably, such liver complications promote the intrahepatic splenosis growth.[13] Splenosis rarely causes symptoms and is, in most cases, an incidental finding.[1] However, some clinical features have been reported, which includes abdominal pain or bowel obstruction associated with compression or sudden torsion of the solid lesion.[13] In some cases of splenectomy for hematological diseases, it leads to recurrence of hematological manifestations.[15]

In intrahepatic splenosis, the characteristic imaging of abdominal splenosis are not specific and almost indistinguishable from those of other hepatic neoplasms, especially hepatocellular carcinoma. For the differential diagnosis of liver lesions, commonly used imaging techniques are ultrasound, helical CT and magnetic resonance imaging with gadolinium-DTPA.[6] In nuclear medicine, when presented with a liver lesion that is suspected to be malignant, a liver scintigraphy with 99mTc-colloid is performed. In this test, space-occupying lesions often behave as defects in the activity of the plotter.[14] Subsequently, a scan is performed with erythrocytes marked with 99mTc to differentiate between hepatocellular carcinoma and hemangioma; highly vascularized lesions, such as hemangioma, appear as a hyperactive focus with respect to the rest of the liver parenchyma (mainly late in the images, obtained 2-3 hours post-administration), while a hepatocellular carcinoma remains a focus of hypoactivity. Splenosis behaves similarly to hemangiomas.[4,14]

In cases of suspected splenosis, scintigraphy with red blood cells labeled with 99mTc has proven to be a useful technique with high sensitivity and specificity.[2,16] However, it has the disadvantage of poor anatomical location. This has been resolved with the addition to clinical practice of techniques of image fusion (SPECT-CT) that add anatomical accuracy to the localization of the foci of splenosis (increasing the specificity of the test) and will result in an improved approach to surgery.

In the event of an accidental detection of one or more hepatic lesions in patients with a past abdominal trauma affecting the spleen, splenosis diagnosis should be included in the differential diagnosis. To obtain correct information from the analysis of lesions in highly suspicious cases of splenosis, 99mTc marked red blood cell gammagraphy should be performed due to the high sensitivity and specificity of the test. With the introduction of combined diagnostic imaging (SPECT-CT), the specificity of this test is increased due to improvements in the characterization of lesions. Therefore, biopsy or operation could be avoided with the application of Tc-99mHDRS and SPECT-CT or SPIO-MRI in most situations.

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