European Review for Medical and Pharmacological Sciences 2013; 17: 2853-2856

Hepatic splenosis presenting as arterialised liver lesion in a patient with NASH

R. INCHINGOLO, P. PEDDU, J. KARANI

Department of Radiology, King's College Hospital, Denmark Hill, London, UK

Abstract. – INTRODUCTION: Splenosis represents the heterotopic autotransplantation of splenic tissue after a traumatic splenic rupture and splenectomy. It is not a rare condition and it is estimated to occur in up to 67% of patients with traumatic splenic rupture.

CASE REPORT: We report one case of patient, affected by non alcoholic steatohepatitis (NASH), with a hypervascularised liver lesion, that the final histological examination revealed hepatic splenosis. This is a rare condition that may be misinterpreted as adenoma or hepatocellular carcinoma (HCC). Imaging techniques and features that might contribute to the diagnosis and may avoid invasive treatment are also discussed. Although hepatic splenosis is a rare condition, this diagnosis should be considered in patients with previous history of abdominal trauma and then the diagnosis of splenosis may be confirmed by Tc-99m-DRBC scintigraphy, avoiding biopsy or further surgery.

Key Words:
Splenosis, Non-alcoholic steatohepatitis (NASH), Hepatocellular carcinoma (HCC), Imaging.

Introduction

Splenosis represents the heterotopic autotransplantation of splenic tissue after a traumatic splenic rupture and splenectomy. It is not a rare condition and it is estimated to occur in up to 67% of patients with traumatic splenic rupture[1]. Splenosis has been reported mainly in the abdominal cavity, but it can also occur in the chest when diaphragm is damaged after a trauma[2,3].

Hepatic splenosis is a rare event. In most of the cases reported in literature a fibrous capsule separated the liver from the splenic tissue which suggested implantation of the splenic tissue on capsular surface of the liver. We report a case of a intra-hepatic splenosis in a patient with history of splenic rupture several years ago, who has chronic liver disease in the form of NASH. The patient presented with a hypervascular liver during routine screening. He underwent surgical resection on the presumption that this solitary lesion could represent HCC (Hepatocellular carcinoma) and it was the postoperative histology that confirmed the diagnosis of intrahepatic splenosis.

In this report we review the literature and discuss the potential aetiological factors for this rare abnormality. Imaging techniques and features that might contribute to the diagnosis and may avoid invasive treatment are also discussed.

Case Report

A 53 year-old man with NASH was found to have a focal liver lesion during routine screening with ultrasound (US). He underwent Computed Tomography (CT) and Magnetic Resonance Imaging (MRI) for further characterization. The lesion demonstrated marked arterial enhancement. A working diagnosis of HCC was made and the patient was referred to our tertiary hepatobiliary unit for further management and consideration of liver resection.

The patient had no family history of malignancy. Abdominal examinations revealed soft palpable liver edge. The laboratory test results were within normal range except for mildly elevated gamma glutamyl transferase 141 IU/L (normal range 1-55 IU/L). Serology for hepatitis B and C was negative. Serum alpha-feto protein was less than 3 ng/mL. He had undergone splenectomy at the age of 20 following a road traffic accident. His other significant medical history included type 2 diabetes mellitus, hypertension, gout and sleep apnoea.

CT scan performed at the referring hospital demonstrated marked hepatic steatosis and a 35 mm well circumscribed exophytic nodule in segment 3. The lesion demonstrated marked enhancement on the arterial phase scans and remained hyperdense relative to the surrounding liver parenchyma on portal venous phase images (Figure 1). MRI confirmed the presence of the lesion in segment 3, 35 mm in maximum diameter, with increased arterialization after gadolinium injection.

Corresponding Author: Riccardo Inchingolo, MD; e-mail: riccardoin@hotmail.it

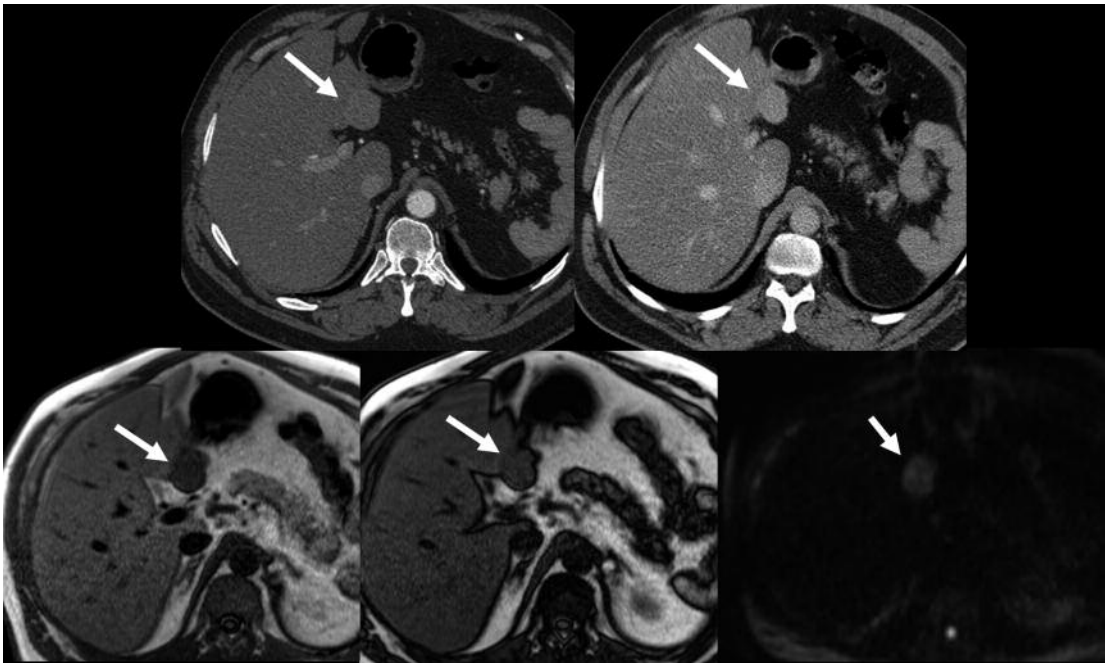

Figure 1. Top row left to right: CT arterial and venous phase scans demonstrating a well circumscribed exophytic lesion (*arrow*) arising from the posterior surface of the left liver. The lesion enhances avidly on the arterial phase compared to adjacent liver parenchyma and remains hyperdense on the venous phase scans. Bottom row: left to right: MRI T1 weighted in and out of phase scans demonstrating variable signal intensity within the lesion compared to the adjacent liver parenchyma (*arrows*). There is some loss of signal in the in-phase images, when compared to the out of phase, indicating hemosiderin accumulation within the lesion. The far right image demonstrates marked restriction in diffusion on the DWI sequence.

He was reviewed in the Outpatient Clinic in our Hospital and repeat MRI of liver and staging CT of the chest was performed for accurate staging and suitability for resection. The interval between the two sets of imaging was four months. The lesion was solitary with unchanged morphology (size and vascular characteristics). There was some loss of signal in the in-phase images, when compared to the out of phase, indicating a degree of hemosiderin accumulation in the tissue. The diffusion weighted (DWI) sequences demonstrated restricted diffusion within the lesion (Figure 1).

The working diagnosis of HCC or hepatic adenoma was made. Following fully informed discussion with the patient a left lateral segmentectomy was performed.

At laparoscopy, the surgeon reported to see an exhophytic liver lesion in the posteromedial surface of segment 3. The lesion was adherent to Iesser omentum and extended laterally into segment 4. He considered unsafe to proceed laparoscopically and he converted to open left lateral segmentectomy, with partial resection of segment 4.

The resected specimen was a soft brown-red portion of liver consistent with left lateral segment. Adjacent to the amputation margin was a slightly darker bosselation. Incision exposed uniform, soft, brown-red surfaces interrupted at the site of the bosselation by a well-demarcated, dark brown-red region 3.5 cm in greatest dimensions and separated from the amputation margin by a cuff of non-lesional tissue at least 0.2 cm wide. Microscopical examination showed intrahepatic splenic tissue, with abundant ceroid- and haemosiderin-laden macrophages. Separated from adjacent liver by compressed portal-tract structures. Steatosis of hepatocytes affected 90% of the lobule (Figure 2).

Discussion

In 1939, Buchbinder and Lipkoff[4] introduced the term "splenosis" to describe the first case of intraabdominal splenic deposit in a young woman following splenectomy for splenic rupture. Although splenosis is considered a rare event, it is actually estimated to occur in up to 67% of pa-

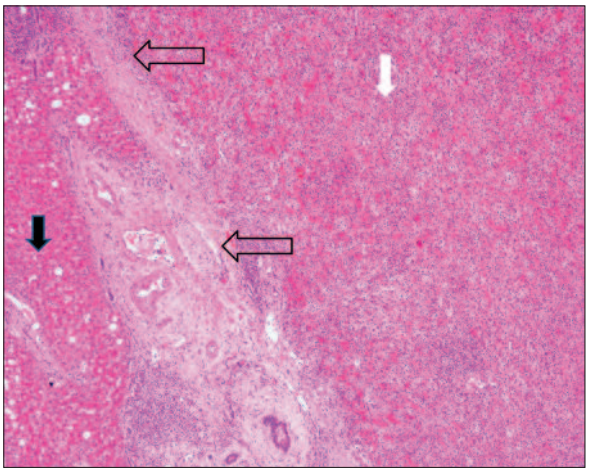

Figure 2. Histological examination showed splenic tissue (*white arrow*) divided from the liver parenchyma (*black arrow*) by a variably thickened pseudocapsule (*empty arrow*); a degree of fatty infiltration within the liver was noted. (Haematoxylin/eosin; original magnification 40×).

tients with traumatic spleen rupture. Since then, a number of intrabdominal splenosis cases have been reported in the literature to date[5-7].

Intrahepatic splenosis is unusual location. Usually splenic tissue gets deposited on the capsular surface of the liver following traumatic rupture and subsequently hypertrophies. A clear fibrous capsule separating the liver parenchyma from the splenic tissue is identified in those cases at histology. To our knowledge there are few reports describing intra-hepatic splenosis[8]. The exact aetiology is not clearly understood. The potential explanations include congenital hyperplasia of ectopic splenic tissue following splenectomy[8] or haematogenous spread of splenic pulp through the splenic vein following traumatic rupture and subsequent splenectomy[9].

Choi et al[10], reviewed ten cases of hepatic splenosis that were reported in the literature between 1997 and 2007. Nine out of 10 patients had a history of splenectomy due to traumatic splenic rupture. The mean age of these patients was 46. The mean interval between splenectomy and diagnosis of hepatic splenosis was 29 years (range 17-46). All cases were asymptomatic and were found incidentally. The mean size of the hepatic nodule was 4.3 cm. All nodules were located near the liver capsule on imaging studies, either invaginating into the liver parenchyma or growing exophytically. In seven cases, the lesion was found in the left liver, whereas in five cases, the nodule was founded in the space between the diaphragm and the surface of segment II, growing into the liver parenchyma. One patient had three lesions. In all these patients the initial working diagnosis was HCC, hepatic adenoma, or FNH (focal nodular hyperplasia) depending on the presence or absence of pre existing liver disease. In all these patients the final diagnosis was made at histology which was obtained either after surgery or after biopsy. No haemorrhagic complications were described in those patients who under went biopsy.

With the standard imaging techniques (US, CT or MRI) hepatic splenosis may be confused with adenoma, HCC or hypervascular metastases[11-14] and it can lead to surgery or other invasive treatments such as trans-catheter arterial chemoembolisation (TACE) or radiofrequency ablation (RFA).

In our case the lesion was found to be well circumscribed nodule in segment 2 with typical arterial enhancement on CT and MRI. Given the existence of background chronic liver disease, differential diagnosis of hepatoma/hepatic adenoma was made although serum Alfa fetoprotein was within normal range. We decide to not perform percutaneous biopsy because of the risk of peritoneal dissemination in case of HCC[15,16] and the risk of bleeding given its location and exhophytic morphology. After an informed discussion with the patient we decided to perform surgical resection as diagnostic and therapeutic option.

Some Centres have described radionuclide scintigraphy using a sensitive, heat-denaturated technetium-99m-labeled red blood cells[17,18] as highly sensitive and highly specific diagnostic test for hepatic splenosis due to the significantly increased tracer uptake in the splenic tissue[10].

Conclusions

In patients with a history of splenic trauma and splenectomy, hepatic splenosis should be considered in the differential diagnosis of arterialized focal lesion particularly if the lesion is close to the capsule. A scintigraphy with sensitive heat-denatured technetium-99m-labeled red blood cells should be performed following standard imaging to confirm or exclude the diagnosis. This might avoid further invasive treatment.

Acknowledgements

We thank A. Knisely, Institute of Liver Studies, King's College Hospital, London, UK, for photomicrographs.

Conflict of Interest

The Authors declare that there are no conflicts of interest.

References

1) Case records of the Massachusetts General Hospital. Weekly clinicopathological exercises. Case 29-1995. A 65 year-old man with mediastinal Hodgkin's disease and a pelvic mass. N Engl J Med 1995; 333: 784-791.
2) Jackson HD, Carney KJ, Knautz MA, Tenholder MF. Left upper lobe mass and diffuse reticular-nodular infiltrate. Chest 1994; 105:1864-1865.
3) Toncini C, Vota L. Splenosis. Description of a case and review of literature. Minerva Chir 1981; 36: 1259-1266.
4) Buchbinder JH, Lipkoff CJ. Splenosis: multiple peritoneal splenic implants following abdominal injury: a report of a case and review of the literature. Surgery 1939; 6: 927-934.
5) Bock DB, King BF, Hezmall HP, Oesterling JE. Splenosis presenting as a left renal mass indistinguishable from renal cell carcinoma. J Urol 1991; 146: 152-154.
6) Gunes I, Yilmazlar T, Sarikaya I, Akbunar T, Irgil C. Scintigraphic detection of splenosis: Superiority of tomographic selective spleen scintigraphy. Clin Radiol 1994; 49: 115-117.
7) Livingston CD, Levine BA, Lecklitner ML, Sirinek KR. Incidence and function of residual splenic tissue following splenectomy for trauma in adults. Arch Surg 1983; 118: 617-620.
8) Davidson LA, Reid IN. Intrahepatic splenic tissue. J Clin Pathol 1997; 50: 532-533.
9) Menth M, Herrmann K, Haug A, Raziorrouh B, Zachoval R, Jung CM, Otto C. Intra-hepatic splenosis as an unexpected cause of a focal liver lesion in a patient with hepatitis C and liver cirrhosis: a case report. Cases J 2009; 2: 8335.
10) Choi GH, Ju MK, Kim JY, Kang CM, Kim KS, Choi JS, Han KH, Park MS, Park YN, Lee WJ, Kim BR. Hepatic splenosis preoperatively diagnosed as hepatocellular carcinoma in a patient with chronic hepatitis B: a case report. J Korean Med Sci 2008; 23: 336-341.
11) Yoshimitsu K, Aibe H, Nobe T, Ezaki T, Tomoda H, Hayashi I, Koga M. Intrahepatic splenosis mimicking a liver tumor. Abdom Imaging 1993; 18: 156-158.
12) Gruen DR, Gollub MJ. Intrahepatic splenosis mimicking hepatic adenoma. Am J Roentgenol 1997; 168: 725-726.
13) Foroudi F, Ahern V, Peduto A. Splenosis mimicking metastases from breast carcinoma. Clin Oncol (R Coll Radiol) 1999; 11: 190-192.
14) Gamulin A, Oberholzer J, Rubbia-Brandt L, Mentha G. An unusual, presumably hepatic mass. Lancet 2002; 360: 2066.
15) Takamori R, Wong LL, Dang C, Wong L. Needle tract implantation from hepatocellular cancer: is needle biopsy of the liver always necessary? Liver Transpl 2000; 6: 67-72.
16) Chang S, Kim SH, Lim HK, Lee WJ, Choi D, Lim JH. Needle tract implantation after sonographically guided percutaneous biopsy ofhepatocellular carcinoma: evaluation of doubling time, frequency, and features on CT. AJR Am J Roentgenol 2005; 185: 400-405.
17) Brancatelli G, Vilgrain V, Zappa M, Lagalla R. Case 80: splenosis. Radiology 2005; 234: 728-732
18) D'Angelica M, Fong Y, Blumgart LH. Isolated hepatic splenosis: first reported case. HPB Surg 1998; 11: 39-42.