

Solitary Perihepatic Splenosis Mimicking Liver Lesion

A Case Report and Literature Review

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Abstract: Hepatic splenosis, one type of manifestation of ectopic spleen tissue, is rarely reported. It cannot be distinguished from hepatic malignancies because of lack of significant radiological features. By means of this case report and 31 literature reviews, potential treatment modalities concerning clinical diagnostics, patient's management could be discussed.

The report presents the case of a 33-year-old man with a liver lesion. Finally, after a mini-incision laparotomy, the lesion was resected and the diagnosis confirmed it as hepatic splenosis. A literature search for case reports published between January 1, 1900, and August 1, 2014, was performed on PubMed.

Approximately 80% (27/34) of patients diagnosed with hepatic splenosis had a history of splenectomy. The mean time interval between splenectomy and hepatic splenosis detection was 25 (1.5–47) years. The median size of reported hepatic splenosis is 30 mm in diameter. Technetium-99m-labeled heat denatured red-blood-cells scintigraphy or superparamagnetic iron oxide-enhanced magnetic resonance imaging is now considered to be the optimal method of diagnosing splenosis.

Hepatic splenosis requires no treatment in most cases. Operation should be performed if it is accompanied by hypersplenism in hematological diseases. When the diagnosis remains unclear, further biopsy or laparoscopy is recommended. If hepatic splenosis is confirmed, careful follow-up is beneficial.

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Abbreviations: CT = computed tomography, DWI = diffusion-weighted imaging, HAV = hepatitis A virus, HBV = hepatitis B virus, HCC = hepatocellular carcinoma, SPIO-MRI = superparamagnetic iron oxide-enhanced magnetic resonance imaging, Tc-99mHDS = Technetium-99m-labeled heat denatured red-blood-cells scintigraphy.

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INTRODUCTION

Splenosis is one manifestation of ectopic spleen tissue, which usually occurs after splenic trauma or splenectomy. The pathogenesis is possibly the autotransplantation of spleen fragment. It is mostly found in the peritoneal, pelvic, or thoracic cavity. However, hepatic splenosis, especially solitary perihepatic splenosis mimicking liver lesion, was rarely reported in the literature. With the deficiency of significant features, normal radiological examination cannot distinguish this from hepatic malignancies. Here, we report a patient with hepatic splenosis mimicking liver lesion. Literatures were reviewed and analyzed to provide information for clinicians to distinguish and treat this disease. Ethical approval was neither obliged nor sought because all management strategies and treatment procedures were part of routine health care. However, we have obtained the approval from the patient to report the case.

CASE PRESENTATION

A 33-year-old man presented to our hospital after detection of a hepatic lesion from a routine examination. He claimed no discomfort. His medical history included an urgent splenectomy due to traumatic rupture of spleen from a traffic accident 12 years ago and hepatitis A virus (HAV) infection in childhood. No family or genetic history was found. There was no positive sign on physical examination except a previous operation scar. Blood routine and liver–renal function laboratory tests remained normal except mild elevated platelet count $372 \times 10^9/L$ (normal range: $100\text{--}300 \times 10^9/L$) and total bilirubin $21.5 \mu\text{mol/L}$ (normal range: $3.4\text{--}20.5 \mu\text{mol/L}$). The Child–Pugh grade was A (score 5). Besides tumor markers including α -fetoprotein, carcinoembryonic antigen, and carbohydrate antigen 19-9, HAV immunoglobulin M and hepatitis B virus (HBV) deoxyribonucleic acid were unremarkable. Abdominal ultrasonography revealed a 3.5×2.2 cm diameter hypoechoic lesion in the left hepatic lobe near diaphragm (Figure 1A). Electrocardiogram and chest radiography were normal. Plain computed tomography (CT) scan demonstrated a homogeneous hypodensity mass relative to the surrounding liver parenchyma, measuring 2.1×3.4 cm in diameter (Figure 1B). MRI with perfusion-weighted and diffusion-weighted imaging confirmed the presence of a solitary 3.5×2.0 cm lesion, which suggests hepatocellular carcinoma (HCC), lying in segment II in a subcapsular position according to radiological features (Figure 2). Biopsy was recommended to the patient for clear diagnosis. However, being afraid of liver malignancy, the patient wanted to confirm the status of the lesion and chose surgery. With sufficient preoperative preparation, the patient underwent an exploratory surgery. Consulting with the previous operational adhesion, we performed the mini-incision laparotomy. Exploration showed that an extrahepatic $4 \times 2 \times 2$ cm splenosis between liver segment II and diaphragm, and dent on the surface of liver could be seen. Intraoperative ultrasound was performed and excluded any

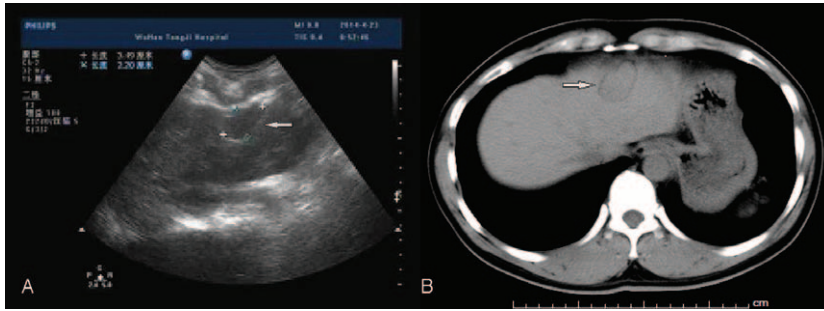


FIGURE 1. (A) Ultrasonography indicates a 3.5 × 2.2 cm diameter hypoechoic lesion, which envelope is integrated (white arrow). (B) Plain CT scan demonstrated a homogeneous hypodensity mass relative to the surrounding liver parenchyma, measuring 2.1 × 3.4 cm in diameter (white arrow). CT = computed tomography.

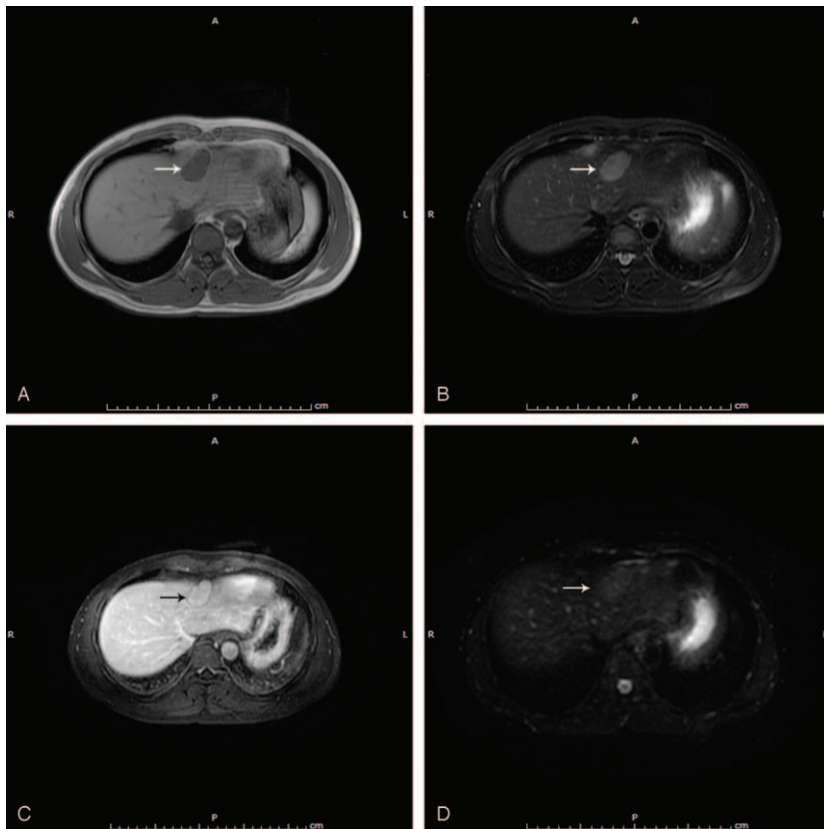


FIGURE 2. (A) T1-weighted MR image: the lesion is of low signal intensity (white arrow). (B) T2-weighted MR image: the lesion is of high signal intensity (white arrow). (C) Liver acceleration volume acquisition contrast: the lesion is of slight hypointensity (black arrow). (D) DWI: the lesion is of slight hyperintensity (white arrow). DWI = diffusion-weighted imaging, MR = magnetic resonance.

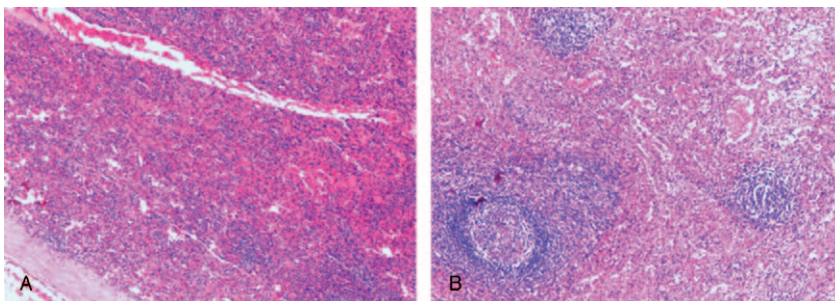


FIGURE 3. (A) Pathological result (hematoxylin–eosin staining, original magnification: 10×). (B) Pathological result (hematoxylin–eosin staining, original magnification: 40×).

TABLE 1. Literature Review of Hepatic Splenosis

Case	Age, y/Sex	Reason of Splenectomy	Time Interval, y	Risk of Liver Tumor	Number	Location	Size, mm	Primary Diagnosis	Confirmed Method
Galloro et al ⁹	56/M	Trauma	NM	Cirrhosis	1	NM	48	Hepatocarcinoma	FNA cytology
Yeh et al ¹⁰	64/M	Trauma	8	HCV	1	S6, extra	25	Liver tumor	Operation
Abu Hilal et al ¹¹	60/M	Trauma	46	HCV, cirrhosis, persistent alcoholic	Multiple	S7, extra	27 × 20	HCC	Laparoscopy
Choi et al ⁴	32/M	Trauma	26	HBV	Multiple	S4a and S6, extra	10–30	HCC	Operation
Inchingolo et al ¹²	53/M	Trauma	33	NASH	1	S3, exophytic	35	HCC, adenoma, FNH	Laparoscopy, Tc-99mHDRS
Menth et al ¹³	43/M	Trauma	25	HCV, cirrhosis	Multiple	S2, extra	4–36	HCC	CT-guided biopsy
Gruen and Gollub ⁶	38/F	Trauma	20	Fatty liver, oral contraceptive	1	S3 and S4, intra	39	FNH, hepatic adenoma	Operation
D'Angelica et al ³	38/F	Trauma	20	Oral contraceptive, alcohol abuse	1	Within the umbilical fissure, S3 and S4, extra	25	FNH, hepatic adenoma	Operation
Liu et al ¹⁴	38/M	Trauma	14	HBV	1	S2, extra	33 × 27	Hepatic malignancy	Laparoscopy
Li et al ¹⁵	61/M	NM	NM	Prostate cancer	Multiple	NM	NM	Splenosis	Needle biopsy
Kim et al ¹⁶	43/M	Trauma	21	HBV, cirrhosis	1	S6, intra	30	HCC	Operation
Di Costanzo et al ⁵	58/M	Trauma	46	HBV, cirrhosis	1	S2, intra	48	HCC	FNA biopsy, Tc-99mHDRS
Ferraioli et al ¹⁷	48/F	Trauma	41	HCV cirrhosis	1	S3, extra	31	HCC	US-guided biopsy
Lee et al ¹⁸	40/M	Trauma	28	HCV	1	S7, intra	60 × 31	Intrahepatic splenosis	US-guided biopsy
Lu et al ¹⁹	43/M	Trauma	20	HBV carrier	1	S6, intra	35	HCC	Operation
Lu et al ²⁰	59/M	Trauma	NM	HBV	3	S7, left lateral lobe, intra	12–22	Hepatic splenosis	Gd-DTPA, Tc-99mHDRS
Foroudi et al ²⁰	59/F	NM	47	Breast cancer	Multiple	S6, splenectomy bed	NM	Metastasis	FNA biopsy and CT
Davidson and Reid ²¹	54/M	Surgical injury	20	None	1	Left lobe, intra	20	NM	Necropsy
Grande et al ⁷	41/M	Trauma	35	None	Multiple	S7, intra and extra	5–45	Intrahepatic splenosis	Tc-99mHDRS
Tsitouridis et al ²²	63/M	Trauma	20	None	1	Left lobe, extra	80	Splenosis	Operation
Mescoli et al ²³	64/M	Gastric leiomyosarcoma	1.5	None	1	S4, extra	50	Peritoneal implantation	CT-guided biopsy
Kang et al ⁸	68/F	None	None	Cirrhosis	3	S3, 5, 7	62–110	Hemangioma or FNH	Biopsy
Yu et al ²⁴	54/M	Trauma	20	None	1	S2, intra	40	Hepatoma	Operation
Yoshimitsu et al ²⁵	51/F	Banti syndrome	23	Cirrhosis	1	S3, intra	25	HCC not excluded	Operation
Pekkafali et al ⁶	21/M	Trauma	15	None	1	Left lobe, intra	34 × 23	Intrahepatic splenosis	Tc-99mHDRS
De Vuysere et al ²⁷	50/M	Trauma	34	None	3	S2, intra	60	Intrahepatic splenosis	Open biopsy
Kondo et al ²⁸	55/M	Trauma	31	HCV	1	S7, intra	35	FNH, hemangioma, HCC	US-guided biopsy
Kashgari et al ²⁹	52/M	Trauma	30	HCV, cirrhosis	1	S7, intra	21 × 15	HCC	US-guided biopsy
Gamulin et al ³⁰	49/M	Trauma	37	None	1	Left lobe, extra	66 × 42	Lymphoma	Operation
Nakajima et al ³¹	41/M	Trauma	21	None	1	S6, intra	NM	Intrahepatic splenosis	US-guided biopsy
Zhao and Xu ³²	49/M	Trauma	17	None	1	Posterior right lobe, NM	50 × 30 × 30	Adenoma, FNH, hemangioma, HCC not excluded	Operation
Liu et al ³³	49/F	Trauma	20	NM	Multiple	S2, S4, NM	12 × 10–50 × 50	Metastasis	Operation

Extra = extrahepatic, F = female, FNA = fine-needle aspiration, FNH = focal nodular hyperplasia, Gd-DTPA = gadolinium–diethylene–triamine–pentaacetic acid, HBV = hepatitis B virus, HCV = hepatitis C virus, intra = intrahepatic, M = male, NASH = nonalcoholic steatohepatitis, NM = not mentioned, S2, 3, 4, 4a, 5, 6, 7 = hepatic segments II, III, IV, IVa, V, VI, VII, US = ultrasonography.

TABLE 2. Differential Diagnosis Between Splenosis and Accessory Spleen

	Splenosis	Accessory Spleen
Etiopathogenesis	Acquired	Congenital
Location	Anywhere, usually in the left upper quadrant abdomen	Near spleen, in the region of splenopancreatic or gastrosplenic ligaments
Number	Usually multiple	Usually solitary
Size in large	Usually small	Uncertain
Blood supply	From parasitized surrounding tissue	Branches of spleen artery
Hilum exist	No	Yes
Histology		
Trabecular network	Normal	Prominent
Capsular tissue	No smooth muscle component	Elastic muscular capsule

other mass on the liver. Then the lesion was rapidly resected with no need of hepatic resection. The histopathological result confirmed the lesion as spleen tissue, with no indication of neoplasia (Figure 3). After 7 days recovery, the patient was discharged without complications. Follow-up examination including blood routine, liver function, and liver CT scanning at 30 days did not show any abnormality. No adverse or unanticipated event was presented.

DISCUSSION

Albrecht¹ firstly described this disease in 1896, but till 1939, Buchbinder and Lipkoff² introduced the term “splenosis.” Splenosis was once considered to be rare because of its

asymptomatic clinical features. Recent articles^{3,4} show that incidence of splenosis varies from 26% to 67% in patients with traumatic splenic rupture. However, the accurate rate is unknown on account of no epidemiologic survey. Among the patients with splenosis, most have been performed splenectomy due to trauma or hematological diseases. Herein, the authors have searched literatures related to hepatic splenosis. All results can be seen in Table 1. Approximately 80% (27/34) of patients had a history of splenectomy because of splenic trauma. The mean time interval between splenectomy and hepatic splenosis detection was 25 years, with a range of 1.5 to 47 years. The median size of reported hepatic splenosis is 30 mm in diameter. Splenosis can occur anywhere within abdominal or pelvic

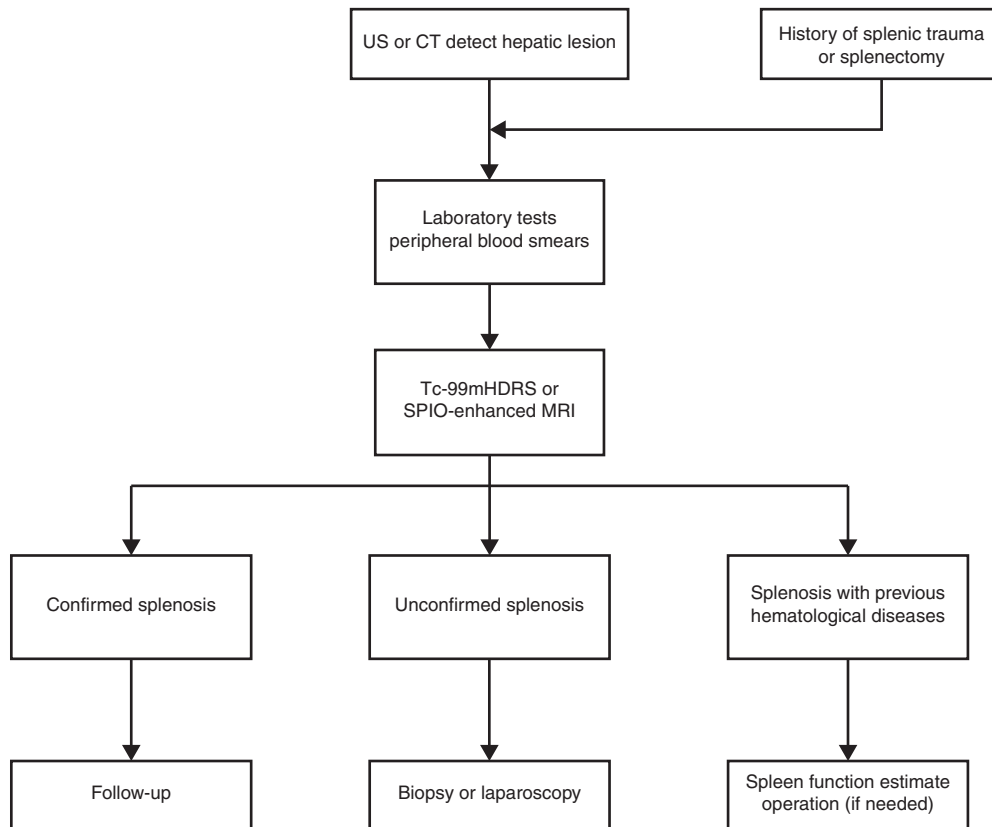


FIGURE 4. Clinical algorithms for the evaluation of perihepatic splenosis. CT = computed tomography, Tc-99mHRS = technetium-99m-labeled heat denatured red-blood-cells scintigraphy, MRI = magnetic resonance imaging, SPIO = superparamagnetic iron oxide, US, ultrasonography.

cavity, even in the chest when the diaphragm is damaged simultaneously.⁵ Even if the real pathogenesis remains unclear, splenosis is now considered of developing from seeding of splenic fragments into exposed serosal surfaces at the time of splenic trauma or splenectomy.⁴ Moreover, another mechanism, especially for intrahepatic splenosis, is hematogenous spread of splenic pulp or erythrocyte progenitor cells and secondary growth in response to tissue hypoxia.³⁴ In our case, perihepatic splenosis is probably because the space between the diaphragm and liver segment II is near the spleen and can be easily reached by splenic fragments during operation.

The limitation of this case is that we did not take splenosis as a possible diagnosis before operation, because of lack of experience. The splenic trauma was supposed to be essential to consider this diagnosis. We reviewed the related literatures to gain more knowledge about this disease. This case report and literature review would be helpful for clinicians to distinguish splenosis and select diagnostic methods and treatment.

Usually, splenosis is asymptomatic. However, some clinical features have been reported, which includes abdominal pain or bowel obstruction associated with compression or sudden torsion of the solid lesion.^{3,6} In addition, gastrointestinal bleeding was also reported. In some cases of splenectomy for hematological diseases, it leads to recurrence of hematological manifestations. It is probably because that splenosis overtakes some function of the spleen. Howell–Jolly bodies, which are normally retained by normal spleen, could be seen diminishing or absent on the peripheral blood smears in patients with functioning splenosis. Unfortunately, it is difficult to distinguish hepatic splenosis from liver tumor due to lack of typical radiological features using conventional ultrasound, CT and MRI. Up to now, technetium-99m-labeled heat denatured red-blood-cells scintigraphy (Tc-99mHDS) is considered to be the optimal method of diagnosing splenosis, but it fails to confirm the accurate anatomical localization.⁷ Furthermore, it has also been reported that superparamagnetic iron oxide-enhanced magnetic resonance imaging (SPIO-MRI) is a useful diagnostic tool to distinguish hepatic splenosis from malignant hepatic tumor. Normal reticuloendothelial tissue indicates loss of signal intensity on T2-weighted MRI because natural reticuloendothelial cells phagocytose SPIO particles.⁸ Tumor cells usually do not phagocytose particles, which results in a cancer-to-liver intensity difference, but well-differentiated HCCs is an exception. Therefore, biopsy or operation could be avoided with the application of Tc-99mHDS and SPIO-MRI in most situations. In our case, the young patient still wanted to confirm the diagnosis pathologically after he was informed that the lesion may be benign. In account of possible abdominal adhesion caused by the previous operation, finally, we chose a mini-incision laparotomy.

It is still challenging for clinicians to distinguish splenosis from hepatocellular carcinoma, hepatic adenoma, hemangioma, and focal nodular hyperplasia timely and correctly. A missed diagnosis of hepatic splenosis usually has a negative influence on patient's management. Besides, the most difficult differential diagnosis is of accessory spleen. We searched literatures and listed the main differences between these 2 ectopic spleen tissues in Table 2. The splenosis may have some immunologic value and splenic filtering function, which may be beneficial for organism. Thus, hepatic splenosis requires no treatment in most cases. In addition, splenosis with hematological diseases should be estimated on the basis of current spleen function. Operation should be performed if necessary. When the diagnosis remains unclear, further biopsy or laparoscopy is recommended. If

hepatic splenosis is confirmed, careful follow-up is beneficial (Figure 4).

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