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Case Report

Hepatic splenosis mimicking a tumor: a rare case and literature review

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Abstract: Splenosis is the autotransplantation of normal splenic tissue mostly following splenic rupture or splenectomy. Isolated hepatic splenosis is rare and frequently misinterpreted as a neoplasm. Despite its rarity, hepatic splenosis should always be considered because its clinical management diverges from that of a neoplasm. Here, we report a case of a 59-year-old man with an occupied lesion mimicking a tumor in the right lobe of the liver. Physical examination and laboratory tests revealed uneventful findings except for the postoperative abdominal scar. The patient had a history of acute icteric hepatitis more than 10 years ago and a splenectomy after trauma 40 years ago. We describe the computed tomography and magnetic resonance imaging findings in this case and also retrospective reports of hepatic splenosis to summarize some features assisting the diagnosis.

Keywords: Splenosis, neoplasm, ultrasound, MR imaging

Introduction

Hepatic splenosis was first defined in 1939 and referred to isolated splenic tissue within the liver. It is a rare condition that results from the autotransplantation of splenic tissue following either traumatic splenic rupture or splenectomy. Hepatic splenosis is usually a benign lump without characteristic signs, and most cases are diagnosed by accident because most patients are asymptomatic. The antidiastole of hepatic splenosis and hepatic tumors is difficult through images, but is important because the therapeutic strategies are very different. Imaging examination including methods such as ultrasound, liver magnetic resonance imaging (MRI), and computed tomography (CT) plays a crucial role in the identification of lesions, but no explicit diagnostic features have been proven. 99mTc-labeled heat-denatured erythrocyte scintigraphy (99mTc-Pyperythrocyte S-PECT) and super-paramagnetic iron oxide (SPIO) are deemed valuable for the diagnosis of splenosis, but they are not available in routine examinations. Because noninvasive examinations show limited value, we may not always obtain a certain diagnosis. When the imaging is inconclusive, invasive examinations, including needle aspiration biopsy and laparoscopy or laparotomy exploration and pathological diagnosis, are indispensable for obtaining a doubtless conclusion and arranging the proper therapies.

Here, we report a patient with a hepatic mass that was diagnosed as angiomyolipoma by ultrasound with contrast but as hepatocellular carcinoma (HCC) by MRI. The patient was scheduled for exploratory laparotomy, and the entity was proven to be splenosis through intraoperative frozen section analysis.

Case report

A 59-year-old man was admitted to our hospital with diabetes. During the health examination, an occupied lesion measuring 38 × 41 mm with a hypoecho and a rim of hyperecho was disclosed in the right lobe of the liver (**Figure 1A** and **1B**) through ultrasound. The patient did not complain of any abdominal discomfort, or show any weight loss. He had been diagnosed with acute icteric hepatitis over 10 years ago, but the therapy details are unclear. Over 40 years ago, a splenectomy was performed after abdominal trauma, and 700-800 ml of blood were transfused during the operation.

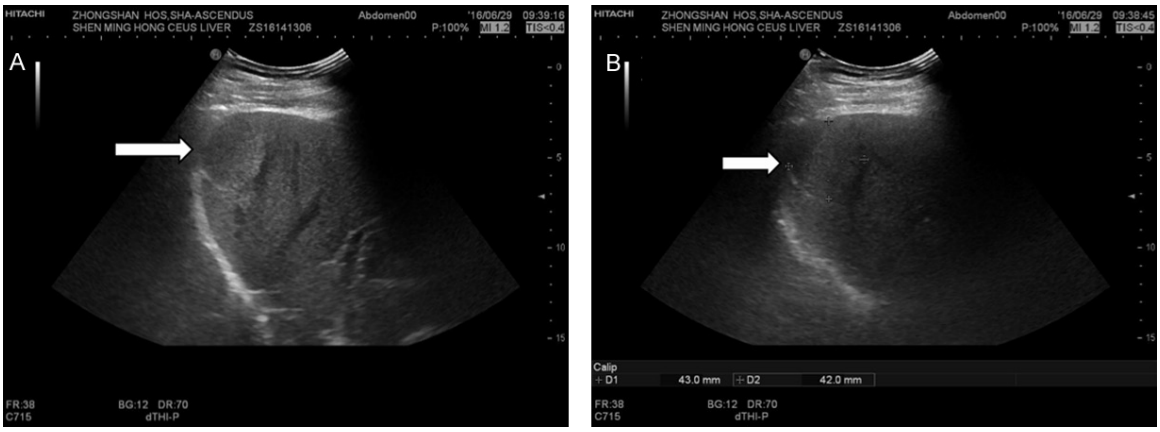

Figure 1. US of the liver. Ultrasound presents a hypoechoic lesion with a rim of hyperechoicity.

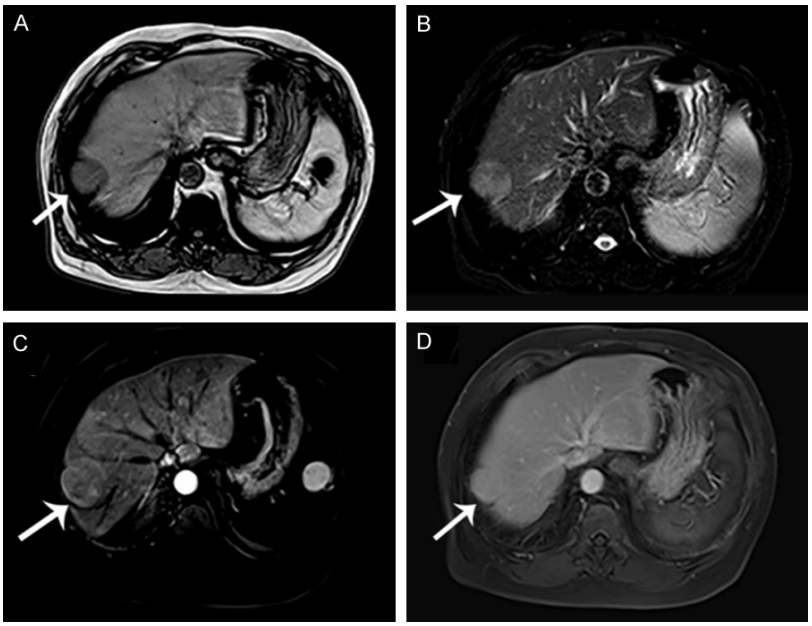

Figure 2. MR imaging of the liver. The entity presented low signal intensity on T1-weighted images (A), slightly higher on T2-weighted images (B). On dynamic contrast-enhanced imaging, the lesion manifested as "wash in" during the arterial phase (C) and "wash out" during the equilibrium phase (D).

The patient had no significant family history of malignancy or hepatitis, and the abdominal examination was unremarkable except for the postoperative abdominal scar. Laboratory tests at the time of hospitalization, including peripheral blood counts, biochemical investigation, and liver function tests were in the normal range. Tumor markers, including serum alpha-fetoprotein levels, revealed no abnormality. Serology results for hepatitis B and C were negative except for HBcAb.

Abdominal magnetic resonance imaging (MRI) was performed for further diagnosis. The lesion was 40 × 35 mm in size and displayed low signal intensity on T1-weighted images (**Figure 2A**), slightly higher on T2-weighted images (**Figure 2B**). On dynamic contrast-enhanced imaging, the lesion manifested as "wash in" during the arterial phase (**Figure 2C**) and "wash out" during the equilibrium phase (**Figure 2D**). The lesion was suspected to be hepatocellular carcinoma (HCC) by radiologists.

Contrast-enhanced ultrasound following MRI considered angiomyolipoma. After using SonoVue, the lesion in the right liver was enhanced at 20 s, reached a peak at 28 s (**Figure 3A**), and presented as isoecho at 38 s (**Figure 3B**). The lesion was slightly hyperechoic in the portal venous phase and delayed phase.

These ultrasound results could not exclude the suspicion of a hepatic tumor. Nest, an exploratory laparotomy was arranged for the patient to exclude hepatocellular carcinoma, adenoma, or other malignancy. During the exploration, we

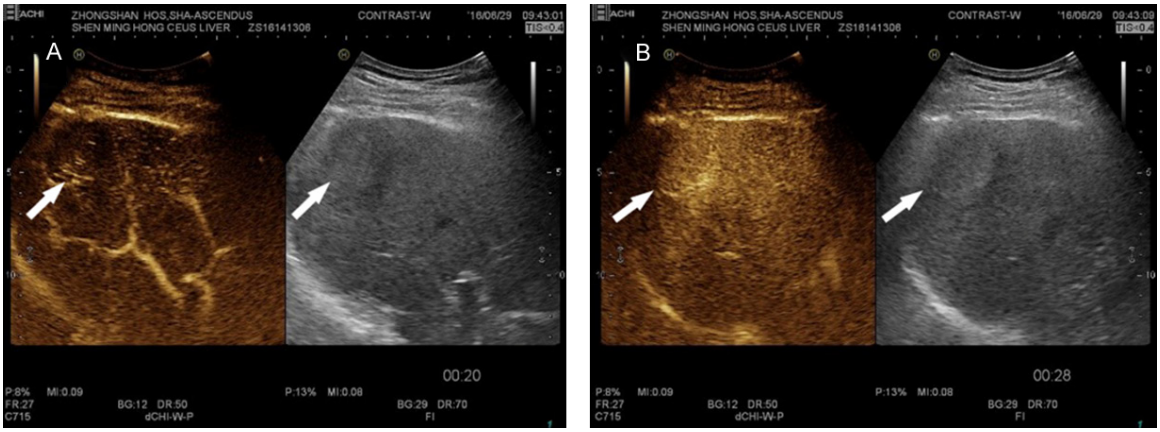

Figure 3. Contrast-enhanced ultrasound. After using SonoVue, the entity in the right liver was enhanced at 20 s (A) and reached a peak at 28 s (B).

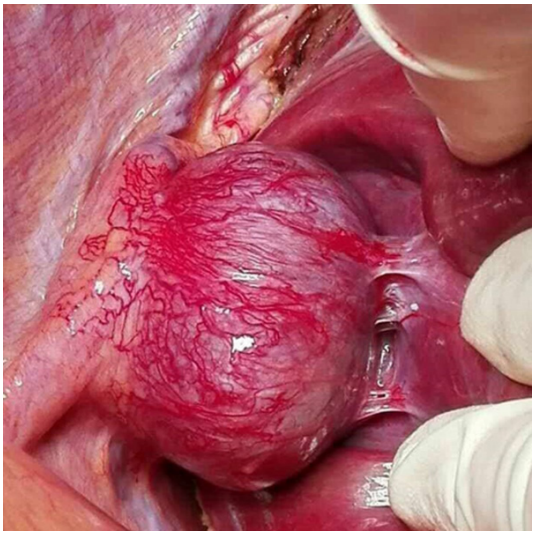

Figure 4. Operative exploration. Adhesion of the mass attached to the diaphragm in the liver.

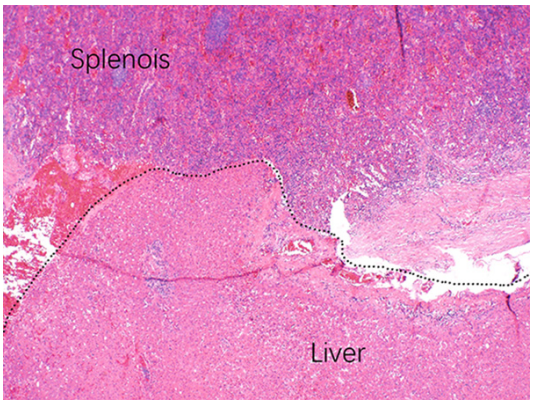

Figure 5. Pathological examination of this lesion exhibited abnormal architecture (H&E).

observed adhesion of the liver and a closed mass that was attached to the diaphragm (**Figure 4**), and intra-operative frozen section analysis of the mass indicated spleen tissue. The biopsy results of the sample confirmed hepatic splenosis (**Figure 5**). The entity was completely removed in the operation and no complications were observed at the time of hospitalization. Until the last following up on December 18th, 2017, the patients did not complain about any special discomfort relating to the operation.

Discussion

According to existing reports, splenosis occurs due to traumatic rupture of the spleen or splenectomy. It is generally symptomless and is often found accidentally during the diagnosis of another disease or routine examination, although abdominal pain and bowel obstruction are mentioned in a few cases [1-4]. Splenosis is not difficult to identify by conventional image examinations, but the mass is difficult to distinguish from other masses such as adenoma and HCC. Splenosis usually does not require surgery, unlike the other diseases it can be confused with. Awareness of splenosis is important for treatment decisions. In one patient with HCC, splenosis was mistakenly interpreted as a larger HCC lesion, and therefore the patient was considered to have unsuitable Milan criteria and lost the chance of liver transplantation [5]. The dilemma is how to ensure the diagnosis of splenosis while reducing invasive examinations and the risk of neoplasms.

Table 1. Literature Review of Hepatic Splenosis

Reference	Sex	Age	Time interval (y)	Symptom	Single or Multiple	Widest Diameter (mm)	Location (Left or Right)	Confirmed Method
Ishikawa M 2007 [9]	M	32	14	None	Unknown	40	R	Surgical excision
Labat-Debelleix V 2008 [10]	M	55	22	None	Unknown	unknown	Unknown	Unknown
Krawczyk M 2013 [3]	M	39	Unknown	Abdominal pain	M	32	L	Tc99
Tinoco González J 2014 [18]	M	60	36	None	S	48	L	Surgical excision
Kandil T.S 2014 [11]	F	45	20	Abdominal pain	Unknown	50	L	Surgical excision
Sandri Giovanni B. Levi 2014 [12]	M	54	25	None	S	45	L	Laparoscopy excision
Wu C 2014 [13]	M	33	12	Abdominal pain	M	40	L	Surgical excision
Sato N 2014 [14]	M	58	Unknown	None	S	37	R	Surgical excision
Tamm A 2015 [6]	M	43	Unknown	None	S	28	L	Tc99
Liu C 2015 [15]	M	33	30	None	M	45	L	Biopsy
Toktaş O 2015 [16]	F	40	33	None	Unknown	68	L	Surgical excision
Fung Ach 2016 [8]	M	55	37	None	M	47	R	Surgical excision
Jereb S 2016 [7]	M	22	18	None	M	26	L&R	Biopsy
De Riggi M. A 2016 [2]	M	31	13	Abdominal pain	S	54	L	Laparoscopy excision
He Zeng Lei 2016 [17]	M	51	20	None	M	33	L&R	Unknown
Our case	M	59	19	None	S	41	R	Surgical excision

The value of US, CT, and MRI is limited for splenosis. No cases of reported splenosis have been confirmed by these imaging modalities, but Tc99m scintigraphy and SPIO-enhanced MRI have been recommended as post-operation tests for patients who have a history of splenectomy. Tc99m scintigraphy has been recognized as the most sensitive and specific non-invasive examination. There are case reports diagnosed by Tc99m scintigraphy [3, 6].

Invasive examinations are the most often used techniques for a definite diagnosis. These diagnostic methods include needle aspiration biopsy, laparoscopy exploration, and laparotomy exploration. Fine-needle aspiration biopsy can have inconclusive results [7] and carries a risk of spreading a neoplasm. Laparotomy exploration is most utilized, although it results in an extensive wound [2, 7, 8]. Laparoscopy exploration has been mentioned more in recent years because of its reduced incision size compared with laparotomy exploration [2].

Sixteen cases of hepatic splenosis [2, 3, 6-18] have been described [**Table 1**] supplementary to two other studies containing 39 cases in total [13, 19]. Approximately 81.8% (45/55) of the patients had a history of splenectomy or splenic trauma. The mean diagnostic age was 49 years. Splenosis was found 24 years after splenectomy on average, with a range of 6 to 62.5 years. The diameter of the entity was 42.2 mm on average, ranging from 20 mm to 110 mm. Regarding location, 56.3% (27/48) of cases occurred in the left lobe of the liver, while 33.3% (16/48) occurred in the right lobe.

Conclusion

Although hepatic splenosis is thought to be a rare condition, it must be included in the differential diagnosis, especially in patients with a history of severe abdominal trauma or splenectomy.

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Disclosure of conflict of interest

None.

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