

Case Report

Intrahepatic splenosis mimicking liver cancer: report of a case and review of literature

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Abstract: Intrahepatic splenosis is a rare situation occurred after traumatic splenic rupture or splenectomy. We report a 33 year old man with multiple isolated liver masses indistinguishable from primary and metastatic liver tumor by standard evaluation. The man underwent a splenectomy due to trauma at the age of three so that the hepatic splenosis was suspected. The final fine-needle biopsy pathological examination proved the diagnosis of intrahepatic splenosis. The importance of suspicion for this diagnosis should be highlighted when tumor-like lesions disclosed on imaging occurs in a patient with a splenic injury in the past. ^{99m}Tc labelled heat-denatured erythrocyte scintigraphy can be helpful to the diagnosis since it may avoid the performance of biopsies or surgical resections.

Keywords: Splenosis, hepatic cell carcinoma, computed tomography, magnetic resonance imaging, pathology

Introduction

Splenosis is a benign condition most commonly resulted from traumatic splenic rupture or splenectomy, in which splenic tissue has been autotransplanted to a heterotopic location [1]. Intrahepatic splenosis is a rare phenomenon and usually diagnosed accidentally [2]. It may cause a significant diagnostic dilemma, particularly when the mass presents as a malignant disease on imaging [3]. We report a patient who presented with abnormal liver enzymes and multiple intrahepatic mass that was clinically consistent with atypical hepatic cell carcinoma with intrahepatic metastasis or metastatic liver cancer. According to his history of splenectomy, the intrahepatic splenosis was also suspected. Finally, the mass was proved to be an intrahepatic splenosis by the pathological examination.

Case report

This is a case report of a 33-year-old man, who underwent post-traumatic splenectomy as a result of traffic accident at the age of 3 years old. The patient is a teacher of physical education and usually in a good health condition. He was admitted into our hospital for further work

up, for the mass in liver was found by ultrasound during the health examination in local hospital. At admission, the patient did not complain about any symptoms. He did not suffer from weight loss, fever or malnutrition and so on. The patient had no family history of cancer as well as other related diseases and no history of hepatitis and alcoholic abuse. On the physical examination, there was no remarkable finding except for a left para-median incisional scar owing to the previous splenectomy.

Then the patient underwent regular laboratory test in our hospital. Initial routine laboratory tests including ESR, complete blood count, serum chemistry did not reveal any abnormalities; moreover, the patient's serology for hepatitis B and C was negative. Esophagogastroduodenoscopy showed mild gastritis with positive rapid urease test for *H. pylori*. Additionally, specific laboratory tests including tumor markers as carcinoembryonic antigen (CEA), Alpha-fetoprotein (AFP) and carcinoma-antigen (CA19-9) were normal as well. Abdominal ultrasound revealed no spleen and big heterogenous intrahepatic lesion measuring 4.5×3.5 cm with clear margins in left segment of the liver (**Figure 1A**). Computed tomography also disclosed the absent of spleen and

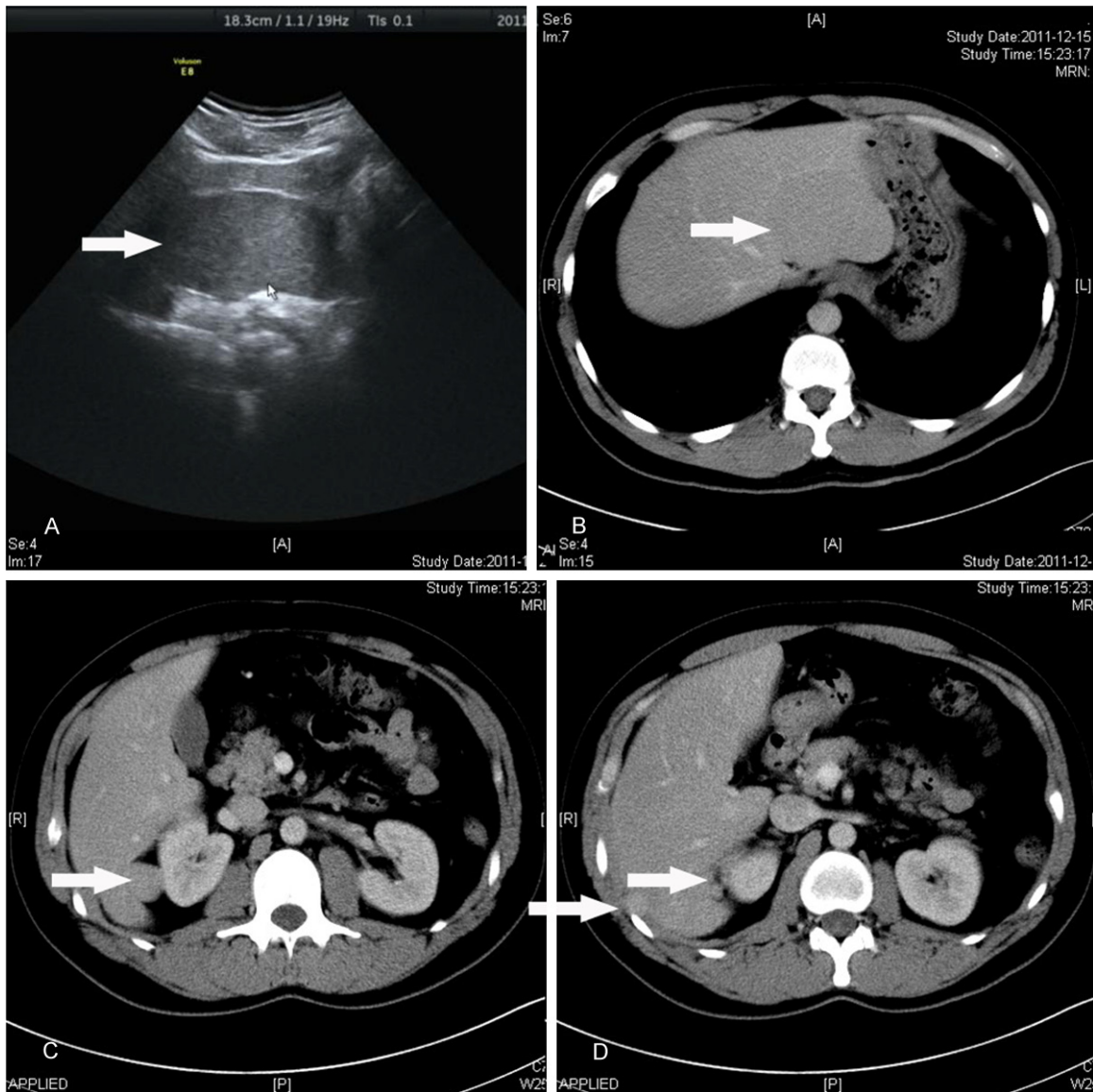


Figure 1. Ultrasound and CT of the liver. Ultrasonography (A) demonstrates a well-circumscribed, lobulated lesion with clear margins in the left lobe of the liver. The axial intravenous contrast enhanced computed tomography (B-D) note the multiple nodules in liver with the biggest 4.2×3.0 cm in the segment III and others in the segment X and XI.

demonstrated multiple nodules in liver with the biggest measuring 4.2×3.0 cm in the live segment III and others in the segment X and XI (Figure 1B-D). T2-weighted single-shot turbo spinecho magnetic resonance imaging revealed liver nodules consistent with computed tomography: whatever on axial (Figure 2B, 2D) or coronal (Figure 2A, 2C) imaging, a rounded and well circumscribed high density mass was seen in the segment III of the liver with two small ones in segment X and XI. Considering the imaging, the etiology of the mass was felt to be

atypical hepatic cell carcinoma with intrahepatic metastasis or metastatic liver cancer. However, according to his history of splenectomy, normal lab examination data and lack of clinic symptoms, the intrahepatic splenosis was suspected. To prove our suspicion, we did the fine needle aspiration biopsy of the left biggest liver mass. Pathologically, lymphoid hyperplasia was present with a small amount of each stage promyelocytic cell and fibrovascular tissue (Figure 3). The patient was diagnosed intrahepatic splenosis and discharged without com-

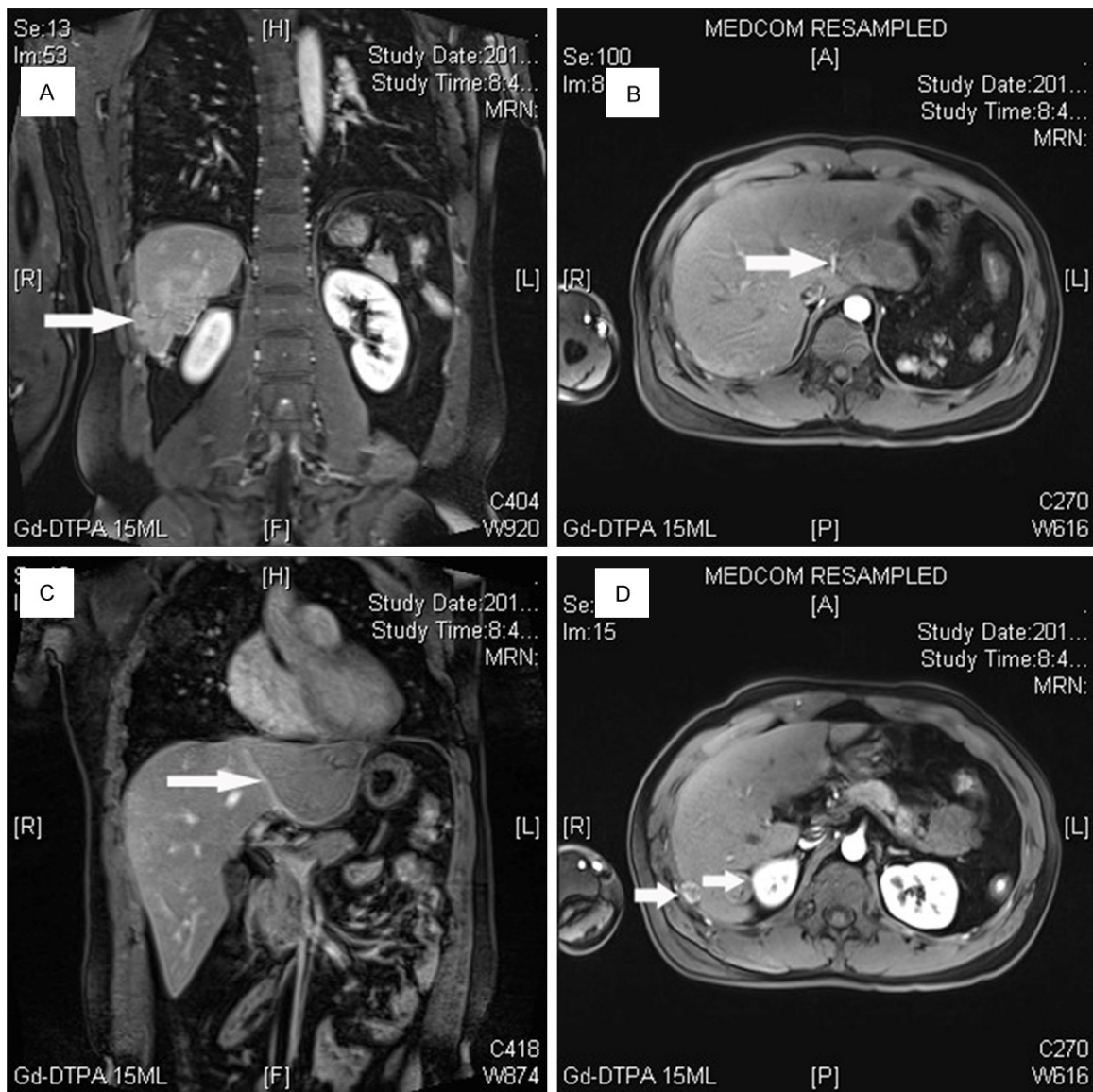


Figure 2. Axial and coronal liver MRI. Axial (B, D) and coronal (A, C) T2-weighted single-shot turbo spinecho imaging MRI shows the intermediate-to-high signal intensity of the lesion, with characteristics of an intrahepatic location.

plications. He was asymptomatic and his hepatic image examination had no change during the follow up period of 2 years. The patient is currently being followed up, as no more treatment is required unless symptoms occurred or worsen.

Discussion

Splenosis was first reported by Shaw and Shafi as an autopsy finding in 1937 [4]. The term "splenosis" is firstly used by Buchbinder and Lipkoff in 1939 to describe the heterotopic transplantation of splenic tissue as a conse-

quence of splenic trauma or surgery [5]. In fact, splenosis is now believed to be a fairly common phenomenon of splenic injury, and it is reported that splenosis occurs in 16%-67% of patients with a history of splenic trauma or splenic surgery [2, 6-8]. Splenosis occurred mainly in the left segment of the abdomen; furthermore, not only intra-abdominal locations as abdominal cavity and along surgical scars have been described [9, 10], but it can also occur in the thoracic cavity [11] or even cerebrum [12] when the diaphragm is damaged as a consequence of trauma [8]. But To our knowledge the intrahe-

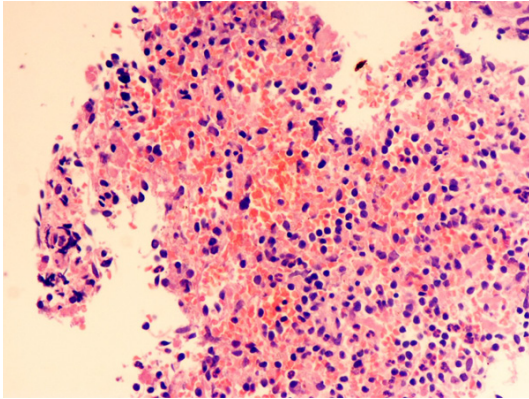


Figure 3. The pathological examination of this lesion found a full component of lymphoid hyperplasia was present with a small amount of each stage promyelocytic cell and fibrovascular tissue and this confirmed the diagnosis of splenosis, with no evidence of malignancy (H&E, $\times 200$).

patic splenosis is very rare and the reported hepatic splenosis is less than 20 cases [2, 8].

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 [13]. It may be based on the susceptibility of the splenic erythropoiesis response to hypoxia and the inevitability of hypoxia caused by aging or pathological changes [14]. In the vast majority of cases with splenosis, patients had no specific symptoms and the undetermined mass is found occasionally [15]. Sometimes, non-specific symptoms such as abdominal pain may lead to the diagnosis owing to infarction of heterotopic splenic tissue with its limited blood supply and increasing size [16]. Gastrointestinal bleeding after rupture of the splenic nodule and intestinal obstruction are rather rare complications associated with splenosis [15, 17].

In intrahepatic splenosis, the characteristic imaging of abdominal splenosis are not specific and almost indistinguishable from those of other hepatic neoplasms, especially hepatic cell carcinoma. Nevertheless, the gold standard for splenosis depends on pathological diagnosis which can be gained by needle aspiration prior to surgical operation [14]. Histologically, liver splenosis presents as nodular lesions rich in lymphoid tissue and hematopoietic cell. The liver biopsy performed in our hospital showed lymphocyte dysplasia with

hematopoietic cell and did not reveal typical splenic pulp as a consequence that the quantity and quality of the lesion was limited by fine needle aspiration.

At present, the use of ^{99m}Tc labelled heat-denatured erythrocyte scintigraphy (^{99m}Tc -Pypererythrocyte S-PECT) is considered as the technique of choice due to its great sensitivity and specificity, which allows the diagnosis to be confirmed and should be [7, 18, 19]. This is a non-invasive procedure that avoids the performance of biopsies or surgical resections which entail a high risk of complications, especially bleeding.

Since this splenic tissue may be partially or fully functioning it may have some beneficial immune function for the patient; indeed, the management of this entity should be conservative [14]. The current opinion is that when splenosis is diagnosed, surgical removal is recommended only in the case of symptomatic complicated splenosis and in patients with hematological disease for whom splenectomy is beneficial [7, 20]. A definite preoperative diagnosis of splenosis requires a high index of suspicion. A detailed medical history, thorough physical examination, and history of splenectomy and abdominal trauma should make the physician consider this rare condition [7].

Conclusions

With the increased prevalence of abdominal trauma due to all kinds of accidents abdominal splenosis may turn up more often than ever. Although intrahepatic splenosis represents an extremely rare condition, this diagnosis should always be taken into account in patients with history of abdominal trauma with splenic involvement presenting with an isolated liver lesion. Moreover, the ^{99m}Tc -labeled heat-denatured red blood cell scintigraphy can avoid unnecessary interventions such as biopsy, angiography, and operation by confirming the diagnosis of hepatic splenosis.

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Disclosure of conflict of interest

None.

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