



Intrahepatic Splenosis Mimicking Hepatocellular Carcinoma: A Case Report

간세포암으로 오인될 수 있는 간내 비장증: 증례 보고

Eunjin Hwang, MD , Seonjeong Min, MD* , Young Chul Kim, MD, Jung-Ah Choi, MD

Department of Radiology, Hallym University Dongtan Sacred Heart Hospital, Hwaseong, Korea

Splenosis refers to autotransplantation of splenic tissue into intraabdominal, intrathoracic or even any other parts of body and occurs after splenic trauma or splenectomy. Hepatic splenosis is usually incidental found during imaging workup. It can mimic other hypervascular mass because it presents vivid and heterogeneous enhancement representing that of splenic tissue. So far, about 20 cases of intrahepatic splenosis have been described in the literature. Herein we report a case of intrahepatic splenosis mimicking hepatocellular carcinoma in a patient with liver cirrhosis. The patient was finally diagnosed on the basis of specific radionuclide imaging.

Index terms Splenosis; Hepatocellular Carcinoma; Radionuclide Imaging; Technetium Tc 99m

Received April 24, 2018
Revised June 26, 2018
Accepted August 19, 2018

*Corresponding author
Seonjeong Min, MD
Department of Radiology,
Hallym University
Dongtan Sacred Heart Hospital,
7 Keunjaebong-gil,
Hwaseong 18450, Korea.

Tel 82-31-8086-2588
Fax 82-31-8086-2582
E-mail msj542@hallym.or.kr

This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<https://creativecommons.org/licenses/by-nc/4.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

ORCID iDs

Seonjeong Min
<https://orcid.org/0000-0002-1647-5671>
Eunjin Hwang
<https://orcid.org/0000-0002-9795-397X>

INTRODUCTION

Splenosis refers to heterotopic autotransplantation of the splenic tissue occurring after splenic trauma or splenectomy (1, 2). This condition is usually incidentally found without clinical symptoms. Mostly it is found in the intraperitoneal cavity but extraperitoneal locations have also been rarely reported as follows: intrathoracic, pericardium or subcutaneous tissue (3). In the literature, about 20 cases of splenosis involving the liver and their radiologic characteristics have been reported to date (4, 5).

We discuss a case in which a patient with liver cirrhosis (LC) and previous splenectomy due to trauma is diagnosed with intrahepatic splenosis. We describe the imaging findings of intrahepatic splenosis on ultrasonography (US), CT and MRI. The final diagnosis was confirmed by the radionuclide imaging.

CASE REPORT

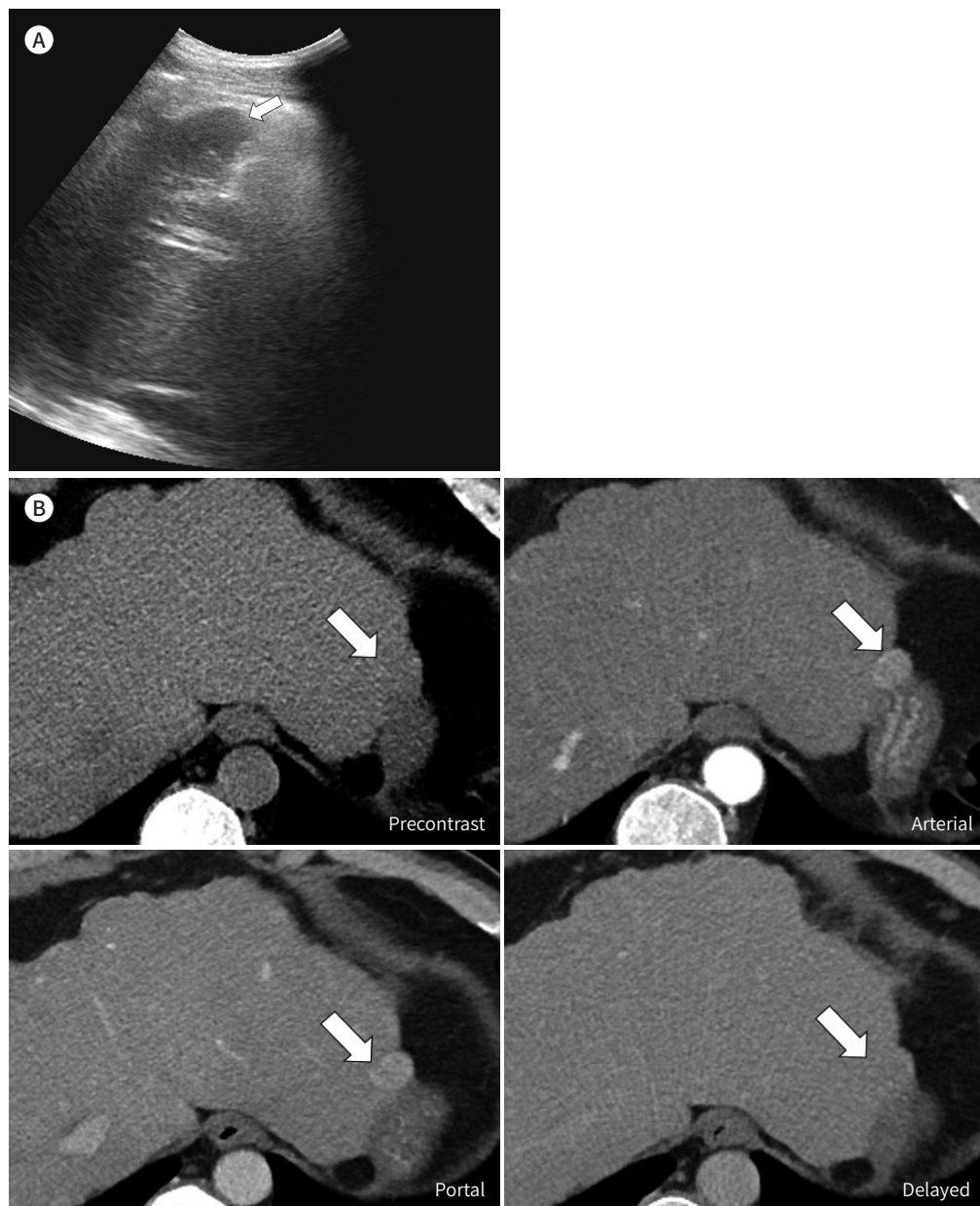
A 43-year-old man was on regular visit to the outpatient clinic after acute myocardial

infarction. He was a heavy alcoholic and the level of liver enzyme was elevated on screening blood tests. Initial laboratory results were as follows; albumin 3.4 g/dL (3.8–4.7), glutamic oxaloacetic transaminase 46 IU (11–37), alkaline phosphatase 226 IU (45–130). Although the Alpha-fetoprotein was within normal range, but the protein induced by absence of vitamin K (PIVKA II) was elevated as 20677 mAU/mL (9.1–26.8). Under diagnosis of alcoholic LC, the clinician examined him with US as an initial imaging workup.

Fig. 1. A 43-year-old man with a hepatic mass and liver cirrhosis.

A. Ultrasonography shows a homogeneous hypoechoic lesion located in the subcapsular region of the left hepatic lobe, measured 3.3×4.0 cm in dimension (arrow).

B. Pre-contrast CT image shows a slightly low-attenuated mass (arrows). The lesion shows homogeneous strong enhancement, hyper-attenuation to the liver and well demarcation on arterial phase of multiphase contrast enhanced CT images. It shows persistent enhancement on portal phase and iso- to slightly low attenuation comparing to the liver on delayed phase.



US showed typical characteristics of LC and a homogeneous hypoechoic lesion measuring 3.3×4.0 cm in dimension, located in the subcapsular region of the left hepatic lobe (Fig. 1A).

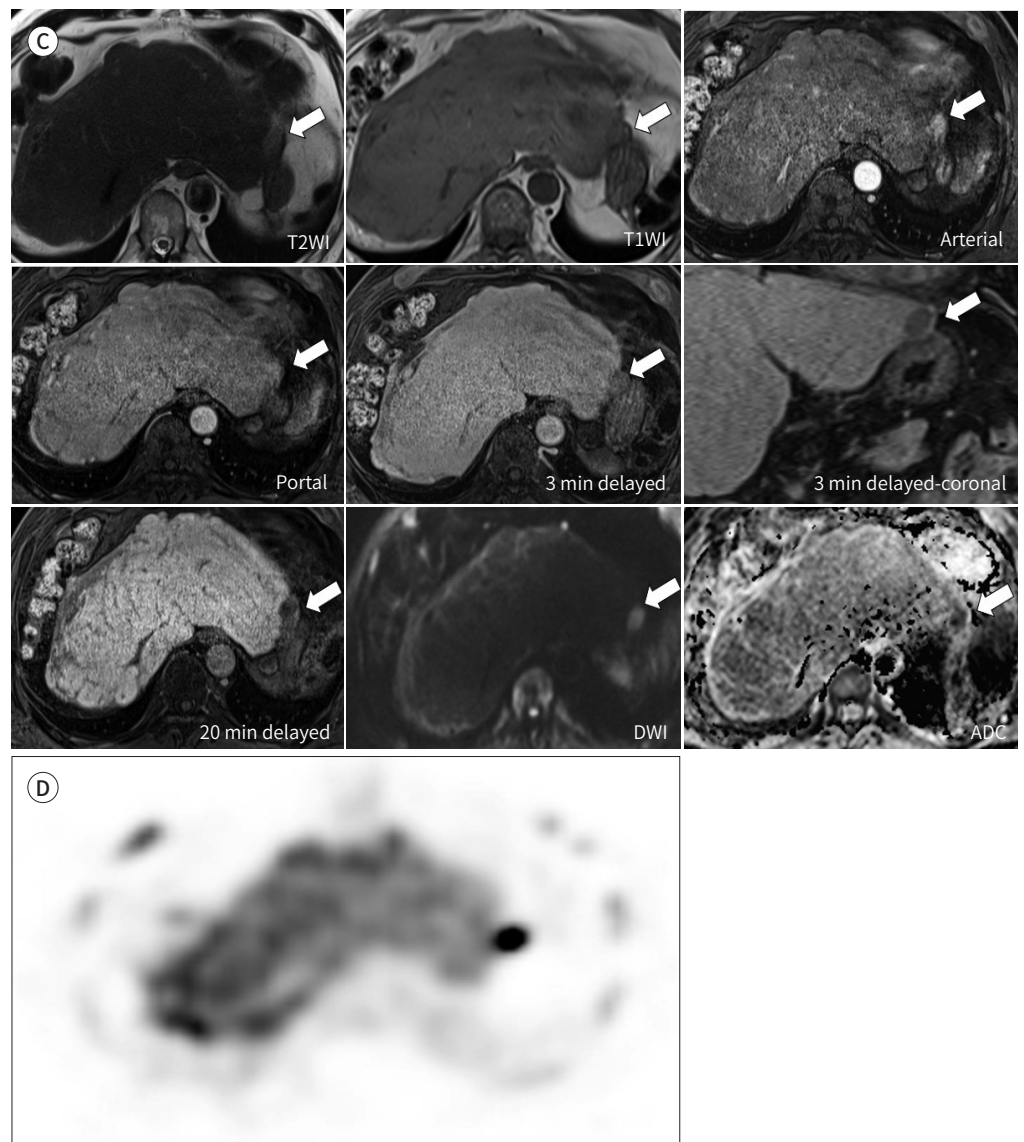
Multiphase contrast-enhanced CT scan was performed for further evaluation (Fig. 1B). The lesion appeared as a low-attenuated area compared to the surrounding liver parenchyma on pre-contrast image. After contrast administration, the lesion showed strong enhancement on the arterial and portal phases and mild washout on delayed phase.

Fig. 1. A 43-year-old man with a hepatic mass and liver cirrhosis.

C. The lesion (arrows) shows mild high SI on T2WI and mild low SI on T1WI. On hepatocyte-specific contrast-enhanced dynamic MRI, the lesion shows homogeneous and intense enhancement on arterial phase and persistent enhancement on portal phase. It shows washout on 3-minute delayed phase and low SI on 20-minute delayed hepatobiliary phase image. The lesion shows intrahepatic location with surrounding thin hepatic parenchyma on 3-minute delayed coronal image. The lesions shows diffusion restriction.

D. The heat damaged red blood cell scan with single photon emission computed tomography shows nodular and intense hot uptake at the corresponding area, suggesting intrahepatic splenosis.

ADC = apparent diffusion coefficient, DWI = diffusion-weighted imaging, SI = signal intensity, T1WI = T1-weighted image, T2WI = T2-weighted image



On MRI (Fig. 1C), the lesion showed homogeneous low signal intensity (SI) on T1-weighted image (T1WI) and high SI on T2WI. After intravenous administration of hepatocyte-specific contrast agent (Gadoxetic acid disodium, Primovist; Bayer-Schering, Berlin, Germany), the lesion showed strong enhancement on arterial phase, washout on 3-minute delayed image and low SI on 20-minute delayed image leading to high suspicion for hepatocellular carcinoma (HCC). The lesion seemed to be intrahepatic location surrounded by thin hepatic parenchyma.

However, the diagnosis of intrahepatic splenosis was more favored because he underwent splenectomy 20 years ago due to traffic accident. The patient was referred to the department of nuclear medicine to perform radionuclide scintigraphy for confirmative diagnosis. Heat damaged red blood cell (RBC) scintigraphy was performed with 2 mCi (37 MBq) 99m technetium (Tc)-labelled autologous RBCs and the uptake was observed at the corresponding area (Fig. 1D).

DISCUSSION

The term splenosis was first described in 1939 defined as auto-transplantation of the splenic tissue to other parts of body following splenic injury (4). Splenosis is believed to occur in up to 67% of patients who suffered from splenic trauma or splenectomy, but the true incidence is unknown because it is usually asymptomatic and incidentally found during imaging workup or surgery for an unrelated disease or at autopsy (2).

Splenosis rarely causes symptoms. Some lesions, however, can twist a long pedicle attached to the splenic nodule, causing acute abdominal pain. There also have been reports of spontaneous rupture causing intra-abdominal hemorrhage or gastrointestinal bleeding related to mucosal erosion as complications of splenosis (4, 6).

Hepatic splenosis is generally thought to occur with spillage and implantation of damaged splenic pulp into subcapsular area of liver. Alternatively, another proposed mechanism is the migration of the splenic pulp or erythrocyte progenitor cells through portal vein and proliferation and differentiation of splenic erythrocytic progenitor cells induction by local hypoxia of the liver (1, 7).

Choi et al. (2) reviewed 10 cases of hepatic splenosis. In 5 cases, the splenoses were between the diaphragm and segment II of liver as same as in our case. They proposed that the space between diaphragm and segment II of the liver is located close to the spleen so that it can be easily exposed during splenectomy. The chronic compression of the diaphragm might induce splenic tissue to invade into the liver parenchyma.

In previous reports (1), splenosis showed hypo- or isoechogenicity on US. The lesions mostly appeared as low attenuated area distinguished from the surrounding liver parenchyma on the pre-contrast CT scan, heterogeneously enhanced on the arterial phase and showed washout on the delayed phase. On MRI, the diagnosis was suggested by low SI on T1WI, high SI on T2WI, strong arterial enhancement and delayed washout after intravenous hepatocyte-specific contrast agent administration. Although such imaging findings are compatible with the characteristics of splenic tissue, these are indistinguishable from hepatic tumors, such as HCC, hepatic adenoma or focal nodular hyperplasia.

In our case, the lesion showed intrahepatic location surrounded by thin hepatic parenchyma on CT and MRI. Moreover, the clinical presentation, regarding hypervascular mass in cirrhotic liver and marked elevation of serum PIVKA II level, led to the primary suspicion for a HCC. But the possibility of hepatic splenosis was more favored based on the patient's history of previous splenectomy and the dynamic characteristics on CT and MRI. Unlike in a typical HCC, the mass did not show definite washout on portal phases on both CT and MRI. We recommended the radionuclide scintigraphy using heat damaged ^{99m}Tc -labeled RBCs for the confirmative diagnosis of intrahepatic splenosis because it was described as the most efficient and accurate diagnostic method on the previous literature (1, 5, 7).

After the confirmative diagnosis, the clinician supposed that the marked elevation of the level of PIVKA II was probably related to longstanding administration of oral warfarin due to the patient's past history of acute myocardial infarction and thrombus in the left ventricle of heart. On follow-up blood tests, the level of PIVKA II was normalized.

In conclusion, hepatic splenosis should be considered if the subcapsular mass is found in patients with a history of splenectomy. The imaging findings of US, CT and MRI can reveal characteristics of splenic tissue but still insufficient for diagnosis of splenosis. Thus, if the hepatic splenosis is suspected, the radiologist should recommend the most specific imaging technique, radionuclide scintigraphy, for the confirmative diagnosis. Unnecessary liver resection or invasive percutaneous liver biopsy in cases misdiagnosed as HCC should be avoided.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

REFERENCES

1. Wang WC, Li XF, Yan ZL, Wang Y, Ma JY, Shi LH, et al. Intrahepatic splenosis mimics hepatocellular carcinoma in a patient with chronic hepatitis B: a case report and literature review. *Medicine (Baltimore)* 2017;96:e8680
2. Choi GH, Ju MK, Kim JY, Kang CM, Kim KS, Choi JS, et al. 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
3. Chung WJ. Splenosis mimicking hepatocellular carcinoma. *Korean J Gastroenterol* 2014;64:173-175
4. Liu C, Liu J, Wang F. Intrahepatic splenosis mimicking liver cancer: report of a case and review of literature. *Int J Clin Exp Pathol* 2015;8:1031-1035
5. Sato N, Abe T, Suzuki N, Waragai M, Teranishi Y, Takano Y, et al. Intrahepatic splenosis in a chronic hepatitis C patient with no history of splenic trauma mimicking hepatocellular carcinoma. *Am J Case Rep* 2014;15:416-420
6. Pekkaali Z, Karsli AF, Silit E, Başekim CC, Narin Y, Mutlu H, et al. Intrahepatic splenosis: a case report. *Eur Radiol* 2002;12 Suppl 3:S62-S65
7. Li T, Yang XY, Tang ZY. Intrahepatic and intraperitoneal splenosis mimicking hepatocellular carcinoma with abdominal wall metastasis in a patient with hepatitis C cirrhotic liver. *Surgery* 2015;157:954-956

간세포암으로 오인될 수 있는 간내 비장증: 증례 보고

황은진 · 민선정* · 김영철 · 최정아

비장증은 비장조직이 복강 내, 흉강 내 또는 다른 신체 부위에서 발견되는 것으로 비장의 외상이나 비장절제술 후에 발생한다. 간내 비장증은 대개 영상의학적 검사에서 우연히 발견되며 비장조직과 같이 강하고 비균질한 조영증강을 보이기 때문에 과혈관성 종양으로 오인될 수 있다. 간내 비장증은 문헌에 약 20례가 보고되어 있다. 이에 우리는 환자에서 간암으로 오인될 수 있는 간내 비장증을 특정 핵의학 검사로 확진한 증례를 보고한다.

한림대학교 동탄성심병원 영상의학과