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Case Report

Intrahepatic splenosis clinically diagnosed as a Carcinoma: a case report

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Abstract: Splenosis is the autotransplantation of splenic tissue, mostly found in patient with the history of splenic trauma or splenectomy. Intrahepatic splenosis is exceptionally rare. Thus, it is often clinically misdiagnosed as a malignant tumor. Herein, we present a case of intrahepatic splenosis which was clinically misdiagnosed as a hepatocellular carcinoma in a 50-year-old Chinese male. The patient underwent a splenectomy due to traumatic splenic rupture 20 years ago. However, the history was absolutely overlooked before the pathological diagnosis was made. Splenosis should be an important differential diagnosis in the patients with the history of splenic rupture or splenectomy.

Keywords: Splenosis, hepatocellular carcinoma, liver

Introduction

Splenosis is the spontaneous ectopic transplantation of splenic tissue to unusual sites [1]. It can usually involve abdomen, peritoneum, mesentery, pelvic cavity and thoracic cavity [2-5]. Occurrence in the liver is relatively uncommon [6-10]. The majority of the patients have the history of previous operation for the splenic trauma. However, the latency between the occurrence of splenic trauma and the diagnosis of splenosis is usually very long [11], so the history may be overlooked, especially, if one is unfamiliar with the relationship between the splenic surgery and splenosis. Herein, we present a case of intrahepatic splenosis in a 50-year-old Chinese male. The patient underwent a splenectomy due to traumatic splenic rupture 20 years ago. However, the history was overlooked; therefore, a preoperative diagnosis of hepatocellular carcinoma was made.

Case report

Clinical history

A 50-year-old male referred to our hospital for complaining of presenting with a tumor in the left lobe of the liver. He reported that a lower echo mass measuring 5.71×2.71 cm was found by ultrasound on regular physical examination (**Figure 1**). The liver was slightly enlarged with the smooth capsule. Subsequent abdominal magnetic resonance imaging (MRI) confirmed the presence of the tumor. On the contrast enhancement *MRI, the mass showed earlier enhancement and fast failure. A malignancy was considered. On physical examination, no positive sign was found. Lab*oratory examination revealed values of serum alpha-fetoprotein (AFP), CA19-9, CA125 and prostate specific antigen (PSA) were in normal level. No lesions in other organs including lung, prostate and rectum were detected. Although the patient had a history of splenectomy due to traumatic splenic rupture 20 years ago. In fact, the history was absolutely overlooked prior to operation. Then the patient underwent the excision of the tumor, since the hepatocellular carcinoma was considered.

At surgery, there was a gray-red mass in the left liver lobe. The mass was relatively well circumscribed; then the mass with little liver tissue was removed, and underwent diagnostic examination.

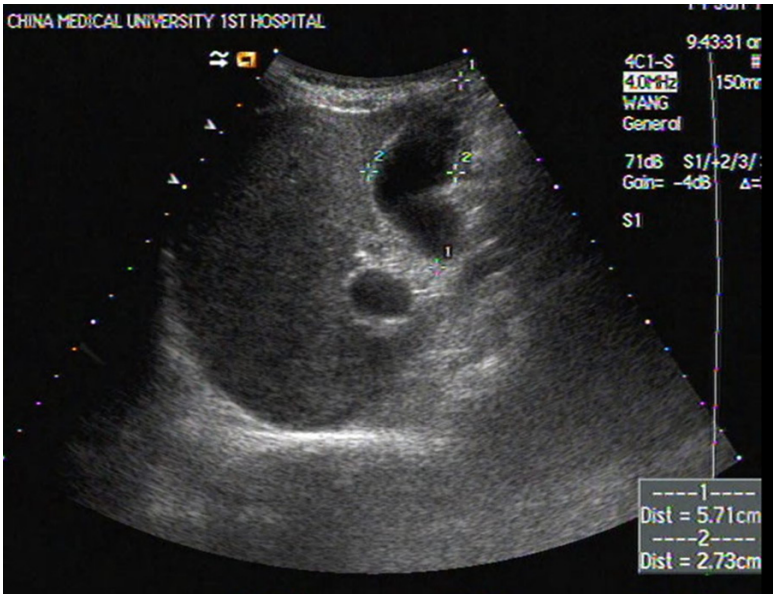

Figure 1. The ultrasonography showed a lower echo nodule measuring 5.71×2.71 cm in the liver. The nodule was relatively well circumscribed.

Results

Grossly, the resected tissue was approximately 7.5×6.5×4.8 cm. There was a nodular lesion measuring 4.9×3.6×3.0 cm in the liver tissue. The lesion was relatively well circumscribed. The cut face of the tumor was grey-red in color.

Histologically, a nodule consisting of red pulp and white pulp resembling the normal spleen tissue was found in the liver tissue. There was a thick fibrous capsule in the peripheral area of the splenic tissue, so the nodule was well circumscribed, and the splenic tissue could be easily distinguished from hepatic tissue. The red pulp was mainly composed of reticular meshwork with cordal macrophages and the venous sinuses with abundant red cells. The white pulp was consisted of the splenic lymphoid follicles (**Figure 2**).

According to the morphological findings and the past history of splenic trauma, the tumor was diagnosed as intrahepatic splenosis.

The patient did not undergo additional therapy after excision, and there was no evidence of disease 36 months later.

Discussion

Splenosis is a rare condition of ectopic splenic tissue mostly found in the patients with the history of splenic trauma or surgery [1]. It can occur in 16 to 67% of the patients after traumatic spleen rupture or spleen surgery [12]. It can occur in abdomen, peritoneum, mesentery, pelvic cavity and thoracic cavity [2-5]. The most frequent sites of involvement are abdominal cavity and pelvic cavity. However, the occurrence in the liver is exceptionally rare [6-10]. In China, primary hepatocellular carcinoma is the most common hepatic tumor. In addition, hepatic splenosis showed earlier enhancement and fast failure, so it could hardly differentiate from hepatocellular carcinoma by CT and MRI. Fortunately, some studies reported that scintigraphy with (99 m) Tc labelled heat-denatured erythrocyte was useful to correctly diagnose splenosis [13]. In addition to hepatocellular carcinoma, the differential diagnosis also includes focal nodular hyperplasia, hepatic adenoma, lymphoma, haemangioma and PEComa [14-16].

Moreover, the latency between the occurrence of splenic trauma and the diagnosis of splenosis is usually very long [11]. Our case underwent a splenectomy due to traumatic splenic rupture 20 years ago. The latency is so long, that the history was overlooked. If one is unfamiliar with the relationship between the splenosis and splenic rupture, the misdiagnosis was more easily made. Intrahepatic splenosis is generally asystematic entity, mostly found incidentally, this may be why the latency is so long in most cases.

The mechanism causing intrahepatic splenosis is still unclear. Knostman et al. [17] claimed that the portal vein entrance of isolated cells originated from the spleen is the cause of splenosis inside the liver that may be why intrahepatic splenosis more easily occur in the left lobes of the liver. Kwok et al. hypothesized that based on the susceptibility of the splenic erythropoiesis response to hypoxia and the inevitability of hypoxia caused by aging or pathological changes, the two events caused the occurrence of the intrahepatic splenosis [11]. But,

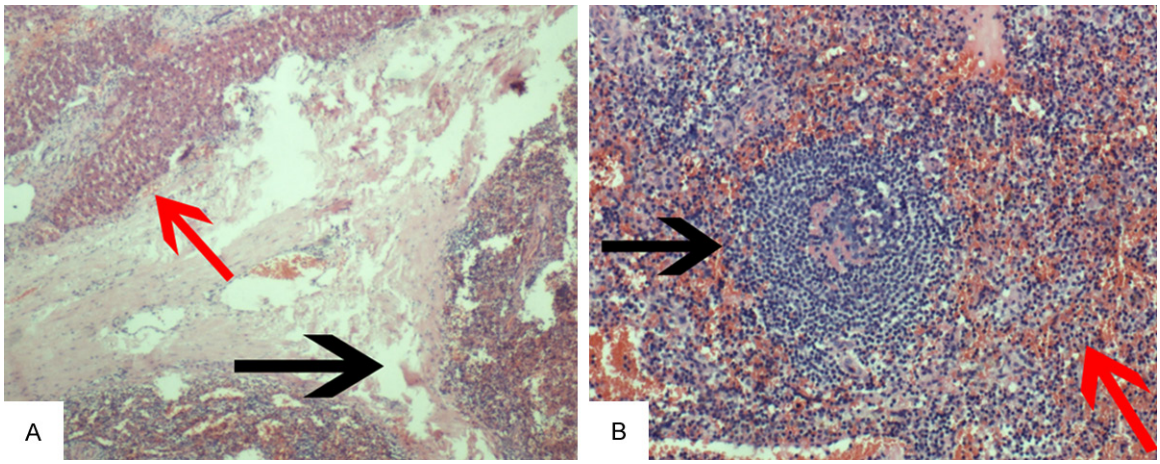

Figure 2. The pathological examination showed a nodule consisting of red pulp and white pulp reminiscent of normal splenic architecture, confirming the diagnosis of intrahepatic splenosis. A. The left area was normal hepatic tissue (red arrow), the right area was splenic architecture (black arrow). B. The splenic tissue consisted of red pulp (red arrow) and white pulp (black arrow).

this hypothesis should be further testified by additional studies.

Conclusion

Although intrahepatic splenosis is rare, it must be an important differential diagnosis in the patient with the history of splenic trauma or surgery. Our reported case was clinically misdiagnosed as a carcinoma, as the history of splenectomy was neglected. Therefore, it is crucial for the clinicians to know the past history of the patient well.

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Disclosure of conflict of interest

None.

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References

[1] Fremont RD and Rice TW. Splenosis. a review. South Med J 2007; 100: 589-93.

[2] Ferraz R, Miranda J, Vieira MM and Carlos Mota J. Thoracic splenosis. Rev Port Cir Cardiotorac Vasc 2010; 17: 153-5.

[3] Ekong E, Goojha C and Kamencic H. Expect the unexpected: a case of pelvic splenosis. J Obstet Gynaecol Can 2011; 33: 787.

[4] Short NJ, Hayes TG and Bhargava P. Intra-abdominal splenosis mimicking metastatic cancer. Am J Med Sci 2011; 341: 246-9.

[5] Ksiadzyna D and Pena AS. Abdominal splenosis. Rev Esp Enferm Dig 2011; 103: 421-426.

[6] Gruen DR and Gollub MJ. Intrahepatic splenosis mimicking hepatic adenoma. AJR 1997; 168: 725-726.

[7] DiConstanzo GG, Picciotto FP, Marsilia GM and Ascione A. Hepatic splenosis misinterpreted as hepatocellular carcinoma in cirrhotic patients referred for liver transplantation: report of two cases. Liver Transpl 2004; 10: 706-709.

[8] Lok AS, Seeff LB, Morgan TR, di Bisceglie AM, Sterling RK, Curto TM, Everson GT, Lindsay KL, Lee WM, Bonkovsky HL, Dienstag JL, Ghany MG, Morishima C and Goodman ZD. HALT-C Trial Group. Incidence of hepatocellular carcinoma and associated risk factors in hepatitis C-related a dvanced liver disease. Gastroenterology 2009; 136: 138-148.

[9] Liu K, Liang Y, Liang X, Yu H, Wang Y and Cai X. Laparoscopic resection of isolated hepatic splenosis mimicking liver tumors: case report with a literature review. Surg Laparosc Endosc Percutan Tech 2012; 22: e307-11.

[10] Abu Hilal M, Harb A, Zeidan B, Steadman B, Primrose JN and Pearce NW. Hepatic splenosis mimicking HCC in a patient with hepatitis C liver cirrhosis and mildly raised alpha feto protein; the important role of explorative laparoscopy. World J Surg Oncol 2009; 5; 7: 1.

[11] Kwok CM, Chen YT, Lin HT, Su CH, Liu YS and Chiu YC. Portal vein entrance of splenic eryth-

rocytic progenitor cells and local hypoxia of liver, two events cause intrahepatic splenosis. Med Hypotheses 2006; 67: 1330-2.
[12] Fleming CR, Dickson ER and Harrison EG. Splenosis: autotransplan-tation of splenic tissue. Am J Med 1976; 61: 414-419.
[13] Hagan I, Hopkins R and Lyburn I. Superior demonstration of splenosis by heat-denaturated tc-99m red blood cell scinti-graphy compared with tc-99m sulphur colloid scintigraphy. Clin Nucl Med 2006; 31: 463-466.
[14] Hong H, Patonay B and Finley J. Unusual reticulin staining pattern in well-differentiated hepatocellular carcinoma. Diagn Pathol 2011; 6: 15.
[15] Grieser C, Steffen IG, Seehofer D, Kramme IB, Uktolseya R, Scheurig-Muenkler C, Hamm B and Denecke T. Histopathologically confirmed focal nodular hyperplasia of the liver: gadoxetic acid-enhanced MRI characteristics. Magn Reson Imaging 2013; 31: 755-60.
[16] Liu Y, Wang J, Lin XY, Xu HT, Qiu XS and Wang EH. Inflammatory angiomyolipoma of the liver: a rare hepatic tumor. Diagn Pathol 2012; 7: 122.
[17] Knostman KA, Weisbrode SE, Marrie PA and Worman JL. Intrahepatic splenosis in a dog. Vet Pathol 2003; 40: 708-10.