

## Laparoscopic splenectomy for splenic hamartoma: Case management and clinical consequences

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### Abstract

Splenic hamartoma is a rare benign tumor, and although minimally invasive surgery may be suitable for this condition, there have only been 2 previous reports of laparoscopic surgery. Here we report the third case of splenic hamartoma managed by laparoscopic splenectomy. A 37-year-old male was incidentally diagnosed by abdominal ultrasonography with a hypoechoic mass measuring 2.5 cm × 2.4 cm in the spleen. Color Doppler sonography showed multiple flow signals within the mass and contrast-enhanced computed tomography revealed strong enhancement of the lesion. On T1- and T2-weighted magnetic resonance images, the splenic mass was demonstrated as isointense and hyperintense respectively. Although a malignant tumor could not be ruled out, a hand-assisted laparoscopic splenectomy was performed because the splenic mass was limited

in size and had not invaded adjacent organs. The pathological diagnosis was splenic hamartoma. The postoperative course was uneventful and the patient was discharged by the seventh postoperative day. Although splenic hamartomas have some specific imaging features, more reports and analyses of these cases are required to increase the reliability of the diagnosis and management. Hand-assisted laparoscopic splenectomy may play a pivotal role in the postoperative diagnosis and management of this condition.

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**Key words:** Splenic hamartoma; Splenoma; Splenic tumor; Laparoscopic splenectomy; Splenectomy

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### INTRODUCTION

Splenic tumors are relatively uncommon. Among them, lymphomatous tumors of the spleen are the most common with other tumors including angiosarcomas, plasmacytomas, primary malignant fibrous histiocytomas, and metastatic disease to the spleen often associated with diffuse carcinomatous dissemination. Benign splenic tumors are exceedingly rare with an incidence of 7 in 100 000 autopsy specimens<sup>[1]</sup> and most are either cysts or hemangiomas.

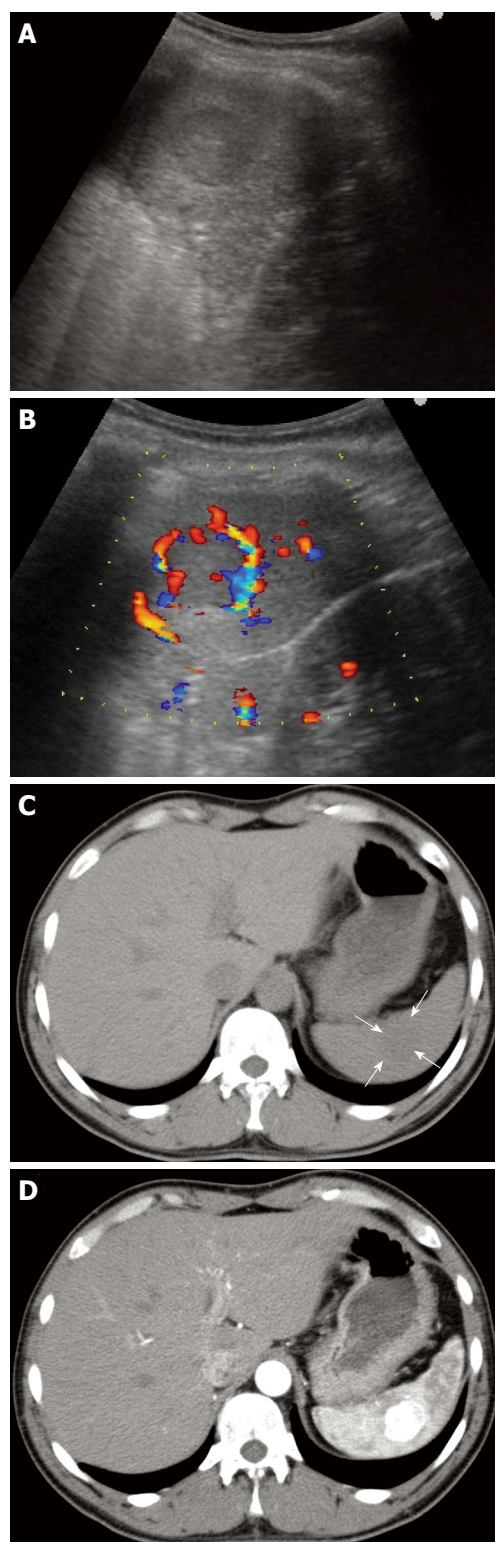
Splenic hamartoma is an extremely rare benign tumor with an incidence of 3 in 200 000 splenectomies<sup>[2]</sup>. Although splenic hamartomas are benign and usually asymptomatic<sup>[2]</sup>, it is important that they are not mistaken for malignant lesions which can sometimes occur due to diagnostic difficulties. Splenectomy has traditionally been performed through a generous laparotomy incision, requiring complete mobilization of the spleen for removal. Recently, laparoscopic surgery has become the standard technique for the surgical treatment of many disorders including malignant diseases. Although minimally invasive surgery may be suitable for splenic hamartomas, to the best of our knowledge, there have been only 2 previous reports of laparoscopic surgery for splenic hamartoma. Here we report a third case of splenic hamartoma that underwent laparoscopic splenectomy.

## CASE REPORT

A 37-year-old Japanese male was incidentally diagnosed with a splenic mass by abdominal ultrasonography (US) and visited our hospital for further investigation. On physical examination, he was afebrile with normal vital signs and no weight loss. The laboratory findings on admission were unremarkable: red blood cell count  $460 \times 10^4/\text{mm}^3$  (normal range  $370\text{--}490 \times 10^4/\text{mm}^3$ ); white blood cell count  $4.7 \times 10^3/\text{mm}^3$  (normal range  $4.0\text{--}8.0 \times 10^3/\text{mm}^3$ ); and platelet count  $25.3 \times 10^4/\text{mm}^3$  (normal range  $14.5\text{--}34.0 \times 10^4/\text{mm}^3$ ). Total protein, alanine aminotransferase, aspartate aminotransferase, total bilirubin and serum creatinine levels were all within normal limits, as were serum carcinoembryonic antigen and cancer antigen 19-9.

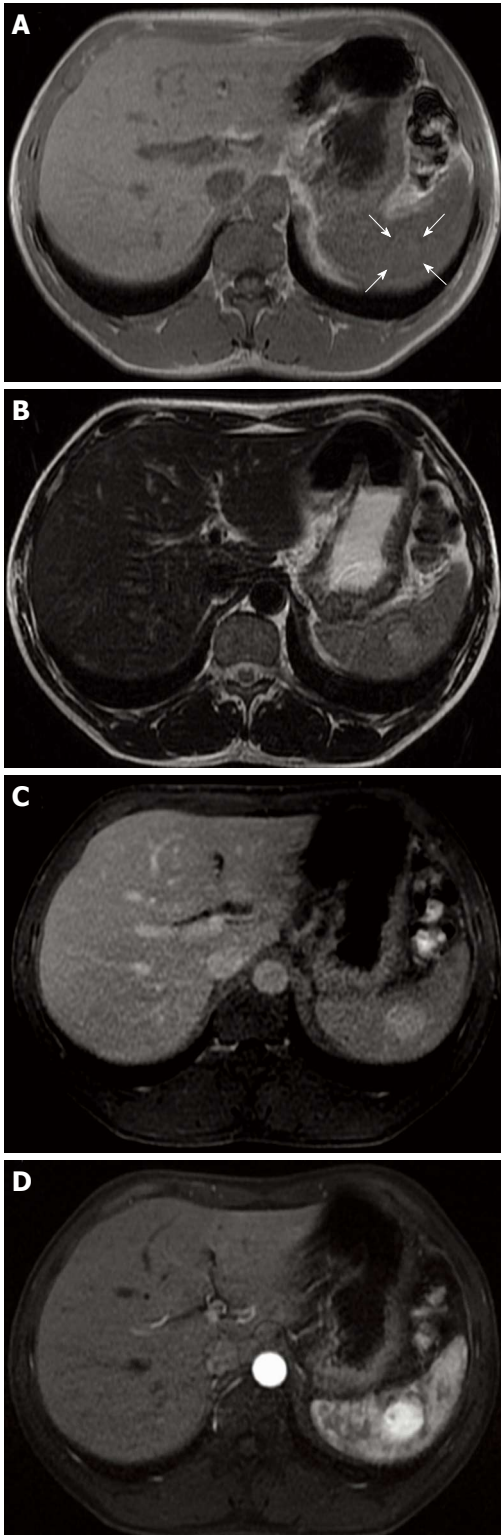
Abdominal US showed a distinct, round and hypoechoic mass measuring  $2.5 \text{ cm} \times 2.4 \text{ cm}$  in the spleen (Figure 1A). Color Doppler sonography showed blood flow signals along the edge of the mass and slight signals within the mass (Figure 1B). Computed tomography (CT) images before the administration of contrast material showed a slightly hypodense lesion relative to the normal splenic parenchyma, measuring  $2.4 \text{ cm} \times 2.3 \text{ cm}$  within the spleen (Figure 1C). Following a bolus injection of intravenous contrast material, the mass was strongly enhanced during the early and late hepatic artery phase (Figure 1D) and 3 min after the administration of contrast, the mass showed isodensity. The lesion was a homogenous mass with no calcification or cystic lesions observed. Magnetic resonance imaging (MRI) showed isointensity in the T1-weighted image and heterogeneous hyperintensity in the T2-weighted image (Figure 2). The lesion showed diffuse enhancement on gadolinium-enhanced MRI. Positron emission tomography (PET) using  $^{18}\text{F}$ -fluoro-2-deoxy-d-glucose (FDG) with CT showed no abnormal accumulation of FDG. Esophagogastroduodenoscopy and colonoscopy showed no significant lesion.

Although the diagnosis of a malignant tumor was not excluded completely, a hand-assisted laparoscopic



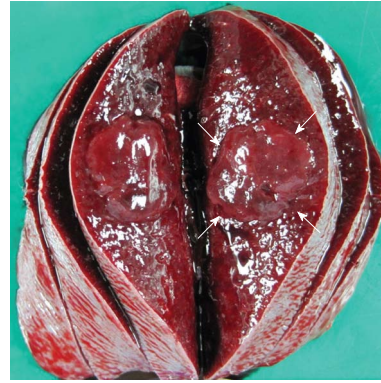
**Figure 1** Ultrasonography and computed tomography (CT) of the splenic lesion. A: Gray-scale ultrasonography shows a homogenous and hypoechoic mass at the mid-portion of the spleen; B: Color Doppler sonography shows multiple color circular flow signals within the mass; C: CT shows a slightly lower-density mass in the spleen (arrows); D: The mass demonstrated strong enhancement after the intravenous administration of contrast material.

splenectomy was performed because the splenic mass was limited in size and had not invaded adjacent organs. The patient was placed on the operating table in a right

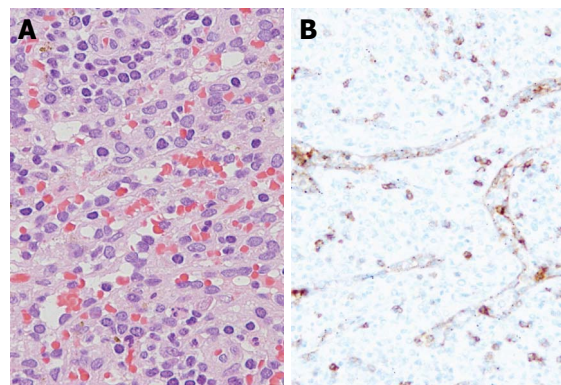


**Figure 2** Magnetic resonance imaging (MRI) of the splenic lesion. A: T1-weighted MRI shows the isointense mass (arrows); B: The mass is mildly hyperintense in the T2-weighted MRI; C: The early phase of dynamic MRI shows diffuse heterogeneous enhancement of the mass; D: The mass becomes more enhanced during the delayed phase.

semilateral decubitus position with the left arm tucked above the head. An incision measuring 7.5 cm, large enough for the surgeon's hand and forearm, was made



**Figure 3** Macroscopic appearance of the resected spleen showing a circumscribed mass measuring 2.5 cm × 2.4 cm in diameter (arrows).



**Figure 4** Pathology of the splenic lesion. A: Microscopically, the mass is composed of predominantly a red pulp splenic sinusoidal structure accompanied by lymphocytes and macrophages. Atypical cells or mitosis were not observed; B: The endothelial cells lining the sinusoidal structure are immunoreactive for CD8.

in the upper midline of the abdomen and three ports were used. The splenic artery at the tail of the pancreas was double ligated with absorbable suture. After mobilization of the spleen, the splenic vein at the splenic hilum was finally ligated and the resected spleen, contained in a plastic bag, was extracted through the upper midline incision. The total operation time and estimated intraoperative blood loss were 100 min and 20 mL respectively. The patient's postoperative course was uneventful and he was discharged on the seventh postoperative day.

The resected spleen weighed 110 g and measured 8.0 cm × 5.0 cm × 4.5 cm. The mass was noted deep inside the spleen near the hilum. Gross examination of the cut surface showed a dark-red, solid, well-circumscribed and firm lesion measuring 2.5 cm × 2.4 cm × 2.4 cm (Figure 3). There were no cystic lesions within the tumor.

Microscopically, the splenic tumor showed expansive growth, compressing the surrounding splenic tissue without a capsule. The tumor consisted of disorganized vascular channels lined by slightly plump endothelial cells without atypia, mixed with intervening splenic red pulp-like stroma without fibrous trabeculae and white pulp (Figure 4A). Immunohistochemical staining



Table 1 Imaging comparisons between splenic hamartoma and splenic hemangioma

Modality	Hemangioma	Hamartoma	Our case
US	Hyperechoic (capillary hemangioma) Hypoechoic (cavernous hemangioma) Doppler: Few flow signals	Hypo >> Hyper > Isoechoic Doppler: Multiple flow signals	Hypoechoic Doppler: Multiple flow signals
CT	Hypo-isodense Progressive centripetal prolonged enhancement	Hypo >> Iso > Hyperdense Diffuse heterogeneous enhancement	Hypodense Strong enhancement
MRI	T1: Hypo-isointense T2: Hyper >> Iso > Hypointense Progressive centripetal prolonged enhancement	T1: Iso > Hypointense T2: Hyper > Hypointense Diffuse heterogeneous enhancement	T1: Isointense T2: Hyperintense Diffuse heterogeneous enhancement

CT: Computed tomography; MRI: Magnetic resonance image; T1: T1-weighted image; T2: T2-weighted image; US: Ultrasonography.

was performed and the endothelial cells that lined the vascular channels stained positive for CD8 indicating splenic type endothelium, but negative for CD34 which detects the intervening cord capillaries (Figure 4B). The pathological diagnosis was splenic hamartoma.

## DISCUSSION

Splenic hamartoma is a rare benign tumor with vascular proliferation which is usually diagnosed pathologically after laparotomy, splenectomy or at autopsy<sup>[1,2]</sup>. The pathogenesis of splenic hamartoma varies and includes congenital malformation of the splenic red pulp, neoplasia or a reactive lesion to prior trauma<sup>[2,3]</sup>. Splenic hamartoma is often called splenoma or nodular hyperplasia because it is composed of an aberrant mixture of the normal tissue components of the spleen<sup>[4]</sup>. As the majority of patients are asymptomatic, most splenic hamartomas are found incidentally by modern imaging methods including US or CT scans<sup>[3,4]</sup>, as in this case.

Recent advances in imaging modalities have improved the ability to detect asymptomatic splenic masses<sup>[5,6]</sup>. However, the differential diagnosis at imaging in respect to more frequent focal lesions of the spleen is still not straightforward. Although splenic hamartoma should be considered as a differential diagnosis for all splenic masses, it is important to distinguish splenic hamartomas from splenic hemangiomas, the most common benign splenic tumor. Table 1 shows some of the differences observed in the US, CT and MRI findings of splenic hamartomas compared to splenic hemangiomas.

The macroscopic type of splenic hemangioma determines the US findings - a capillary hemangioma appears as a hyperechoic nodule whereas a cavernous hemangioma is seen as a heterogeneous hypoechoic mass, sometimes with calcification or multiple cystic areas<sup>[6]</sup>. Generally, splenic hamartomas have been described as solid, homogenous masses with various echogenic patterns relative to the normal splenic parenchyma, showing usually a hypoechoic, but occasionally hyperechoic lesion with or without cystic changes in the spleen<sup>[3,4,7]</sup>. The color Doppler sonograms of splenic hemangiomas show only a few color echoes which can appear in the tumor immediately after it is compressed by the probe<sup>[8]</sup>. On

the other hand, the color Doppler sonographic findings in splenic hamartomas are multiple radial blood-flow signals inside the mass<sup>[9]</sup>. However, masses inside the spleen are usually less vascular than the surrounding normal parenchyma because of the high vascularity of the spleen. Chou *et al*<sup>[10]</sup> demonstrated that the tumor could be markedly enhanced on color Doppler sonography by the administration of microbubble contrast agents. Thus, US may be an indispensable method for the diagnosis of splenic hamartoma<sup>[7]</sup>.

CT scans demonstrate that splenic hamartomas are isodense or hypodense masses and show a dense spreading and prolonged enhancement after intravenous administration of contrast material<sup>[11]</sup>. T2-weighted MRI shows most splenic hamartomas as heterogeneously hyperintense lesions relative to the spleen and demonstrates diffuse heterogeneous enhancement on early postcontrast images and more uniform enhancement on delayed images<sup>[5,6,12]</sup>. Similar findings were noted in our present case. In the case of splenic hemangiomas, dynamic enhanced imaging using either CT or MRI demonstrates a progressive centripetal pattern of enhancement with prolonged uniform enhancement on delayed images<sup>[6,12,13]</sup>. In addition, the fusion of images from FDG-PET with those of CT becomes an important tool in assessing patients with a malignant disease or suspected lymphoma to evaluate other malignant sites<sup>[14]</sup>. Avila *et al*<sup>[15]</sup> first reported the findings of PET-CT in splenic hamartomas which demonstrated a moderate FDG avidity, while our current case showed no abnormal accumulation of FDG.

Although there are some specific imaging features of splenic hamartomas, it is difficult to rule out the possibility of a malignant neoplasm based on such imaging studies. The diagnosis must be confirmed by pathological examination. Although there have been some studies about the efficacy and safety of fine needle aspiration biopsy of the spleen, the possibility of bleeding or tumor dissemination makes this technique problematic<sup>[16,17]</sup>. Therefore, splenectomy is still necessary for diagnostic and therapeutic purposes. Recently, less invasive treatments have been developed such as laparoscopy-assisted surgery.

The main pathological differential diagnosis for splenic hamartoma is a benign vascular tumor including hemangioma. They also have similar clinical and

Table 2 Characteristics of reported cases of splenic hamartoma treated by laparoscopic splenectomy

Author	Year	Age (yr)	Gender	Tumor size (cm)	Preoperative diagnosis	Number of ports	Operation time (min)	Estimated blood loss (mL)
Yoshizumi <i>et al</i> <sup>[25]</sup>	1997	45	Male	6.0 × 3.8	Benign tumor	4	305	450
Tatekawa <i>et al</i> <sup>[22]</sup>	2007	12	Female	5	Hamartoma	4	ND	ND
Our case	2009	37	Male	2.5 × 2.4	Benign tumor	3	100	20

ND: Not described.

radiological findings. Immunohistochemical staining can be used to distinguish a splenic hamartoma from a capillary hemangioma by their respective staining characteristics<sup>[18,19]</sup>. Endothelial cells which are positive for CD8 are a key feature that distinguishes a hamartoma from other vascular lesions of the spleen<sup>[3,19]</sup>. The endothelial cells of hemangiomas are CD8-negative and CD34-positive, in contrast to the CD8-positive and CD34-negative endothelial cells of splenic hamartomas<sup>[18,19]</sup>. The splenic hamartoma has been classified into two histological types: the white pulp type and the red pulp type, according to the tumor components<sup>[2]</sup>. The former is composed entirely of lymphoid tissue while the latter is composed of sinuses and is histologically similar to that of the normal red pulp of the spleen. Most reported cases are the red pulp type, including this present report. Multiple flow signals on color Doppler US and strongly enhanced findings on CT and MRI in our case are thought to be a reflection of the characteristic hypervascularity of the red pulp itself.

Laparoscopic splenectomy is the standard surgical procedure for the management of most cases of idiopathic thrombocytopenic purpura because of the lower operative and perioperative morbidity, compared to open splenectomy<sup>[20,21]</sup>. Moreover, laparoscopic splenectomy has become the surgical procedure of choice for not only the management of idiopathic thrombocytopenic purpura but also of some splenic tumors<sup>[14,21-24]</sup>. To the best of our knowledge, only three cases of laparoscopic splenectomy for splenic hamartoma, including our present case, have been reported in detail (Table 2)<sup>[22,25]</sup>. Hand-assisted laparoscopic surgery allows the surgeon to place one hand into the abdominal cavity, providing a tactile sense and improving the accuracy of manipulation while maintaining the pneumoperitoneum. This facilitates the surgical procedure by allowing identification of dissection planes or the hand to function as a retractor. Moreover, if bleeding occurs, it is easily controlled by compression of the hand on the splenic vascular pedicle or the injury site. In our case, the 7.5 cm incision made in the upper abdominal midline was large enough to allow entry of the surgeon's hand and forearm. The resected spleen can be easily extracted through this incision without the requirement for an additional incision.

In conclusion, although there are some imaging features that are specific to splenic hamartomas, the collection and analysis of more of these cases is necessary to improve the reliability of the diagnosis and management of this condition. Hand-assisted laparoscopic splenectomy may

play a pivotal role in the postoperative diagnosis and management of splenic hamartomas.

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