Transplantation of splenic tissue after splenectomy: A case report

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Abstract. Transplantation of splenic tissue is a rare condition that usually occurs after splenic trauma and splenectomy. It usually requires surgery for diagnosis and treatment. A 38-year-old Asian male with familial hemolytic disease underwent laparoscopic splenectomy for a traumatic rupture of the spleen one year prior. The patient developed middle-upper abdominal pain without any obvious cause, radiating to the back and chest seven months prior to presentation. The condition improved with conservative treatment but the patient experienced recurrent episodes. Abdominal CT suggested multiple gallstones in the gallbladder that changed after splenectomy and multiple nodules in the original splenic area; thus, transplantation of splenic tissue was considered. MRI suggested thick gall bladder bile, multiple stones and cholecystitis, and the spleen was not observed (the patient underwent laparoscopic splenectomy at our hospital one year previously due to traumatic splenic rupture); furthermore, there were multiple abnormal signal foci in the splenic area, so the possibility of spleen implantation was considered. Considering the patient's family history of a hereditary hemolytic disease, laparoscopic cholecystectomy was performed simultaneously with laparoscopic accessory splenectomy. The final pathological report revealed chronic cholecystitis, mixed calculi, red pulp dilation, hyperemia and bleeding in round tissue with blood clot formation and acute and chronic inflammatory cell infiltration. Clinicians must bear in mind the possibility of splenosis after splenic trauma and its image variations.

Introduction

Transplantation of splenic tissue is caused by splenic trauma, including splenectomy and bodily injury, leading to partial rupture of the spleen and the fragments embedding in other parts of the body (1). Splenic hyperplasia may occur in implanted spleen tissue (2). Transplantation of splenic tissue is occasionally detected on imaging. Most cases have no clinical symptoms or signs, but certain cases of gastrointestinal splenomegaly may result in bleeding (3). The typical clinical manifestations of an implanted ectopic spleen are single or multiple, round or oval masses, that are sometimes misdiagnosed as malignant tumors, such as gastrointestinal stromal tumors (4), lymphomas (5), peritoneal mesotheliomas (6), renal cancer (7) or liver cancer (8).

It is generally thought that the best treatment may be planned only once a sample is obtained and the nature of the nodule is determined preoperatively. However, when the nature of the nodule cannot be determined preoperatively, the nodule size, the possible diagnosis, patient characteristics and clinical symptoms should be assessed prior to surgery (9).

The present study reported on a case of intraperitoneal transplantation of splenic tissue that was only able to be characterized after surgery.

Case report

A 38-year-old Asian male with a familial hemolytic disease had undergone laparoscopic splenectomy due to traumatic rupture of the spleen one year previously at our hospital (The Second Affiliated Hospital of Soochow University, Souzhou, China). At seven months previously, the patient developed middle-upper abdominal pain without any obvious cause that radiated to the chest and back. The condition improved with conservative treatment at our hospital, but the patient experienced recurrent episodes. The mass was discovered incidentally on a CT scan (May 2021; The Second Affiliated Hospital of Soochow University, Souzhou, China) performed to check for the cause of pain in the upper abdomen.

The patient had a white blood cell count of $12.6x10^9$/l (normal range, 4-$10x10^9$/l) and a platelet count of $458x10^9$/l (normal range, 100-$300x10^9$/l). Furthermore, the patient's blood test results indicated a slight increase in total bilirubin. However, the levels of other markers, including those of several serum tumor markers (carbohydrate antigen199, fetal protein, neuron-specific enolase, gastrin-releasing polypeptide and soluble interleukin-2 receptor) were normal.

Abdominal CT suggested multiple gallstones in the gallbladder, which changed after splenectomy, and multiple nodules in the original splenic area, suggesting transplantation of splenic tissue (Fig. 1). MRI revealed round T1-weighted

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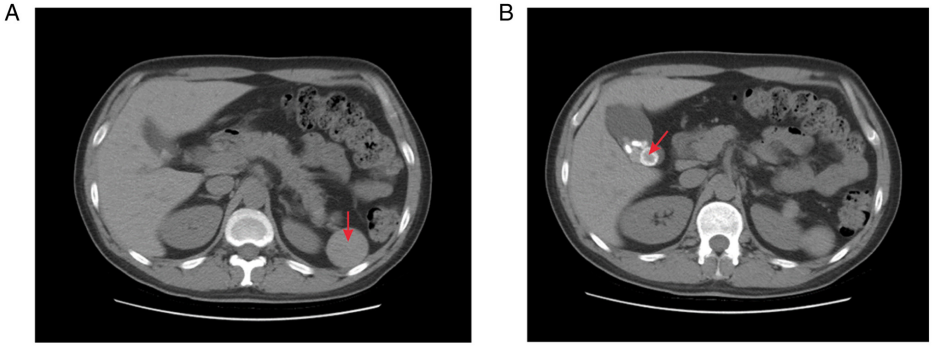

Figure 1. Abdominal CT images at presentation. (A) Multiple nodules in the original splenic area suggested transplantation of splenic tissue (indicated by arrow), which changed after splenectomy. (B) Multiple gallstones were observed in the gallbladder (gallstones indicated by arrows).

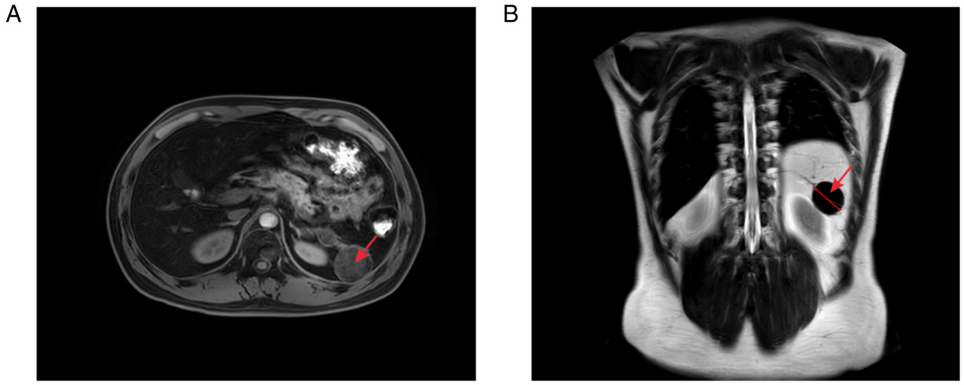

Figure 2. MRI revealed the following: Round T1WI/T2WI/T2W hyposignal foci in the splenic area (the larger foci were ~39 mm in length; the spleen is not shown). No retroperitoneal lymph nodes were observed. Gall bladder bile was thick, and multiple stones and cholecystitis were present, the spleen was not observed and there were multiple abnormal signal foci in the splenic area; therefore, spleen implantation was considered. (A) transverse axis; and (B) coronal axis. The lumps are indicated by arrows. T1WI, T1-weighted imaging.

imaging (T1WI)/T2WI/T2W hyposignal foci in the splenic area (the larger foci were ~39 mm in length). No retroperitoneal lymph nodes were observed. The gall bladder bile was thick. Multiple stones and cholecystitis were present. The spleen was not visualized (the patient underwent laparoscopic splenectomy at our hospital one year previously due to traumatic splenic rupture) and there were multiple abnormal signal foci in the splenic area, so the possibility of ectopic spleen implantation was considered. Due to the patient's history of a hereditary hemolytic disease, laparoscopic cholecystectomy and accessory splenectomy were performed simultaneously (Fig. 2). The results of abdominal ultrasonography indicated a stable strong echo sound group in the gallbladder cavity, suggesting that the patient had cholecystolithiasis and cholecystitis (Fig. 3).

The patient was subjected to laparoscopic cholecystectomy and a laparoscopic exploration of the abdomen was performed to remove the mass under laparoscopic guidance. Intraoperative exploration revealed that the gallbladder exhibited chronic inflammatory changes and it was resected successfully. A subphrenic broad-based protuberant lesion of ~40 mm in size was found and completely removed using an ultrasonic scalpel. The excised mass was ~40 mm in diameter and contained splenic parenchyma (Fig. 4).

The final pathological report (routine hematoxylin-eosin staining) revealed chronic cholecystitis, mixed calculi, red pulp dilation, hyperemia and bleeding in round tissue with blood clot formation and acute and chronic inflammatory cell infiltration (Fig. 5). Combined with the gross findings, the symptoms were consistent with the diagnosis of transplantation of splenic tissue.

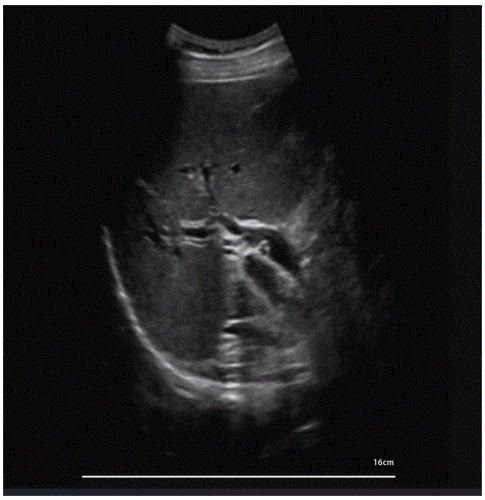

Figure 3. Abdominal ultrasonography revealed a stable strong echo sound group in the gallbladder cavity, suggesting that the patient had cholecystolithiasis and cholecystitis (scale bar, 16 cm).

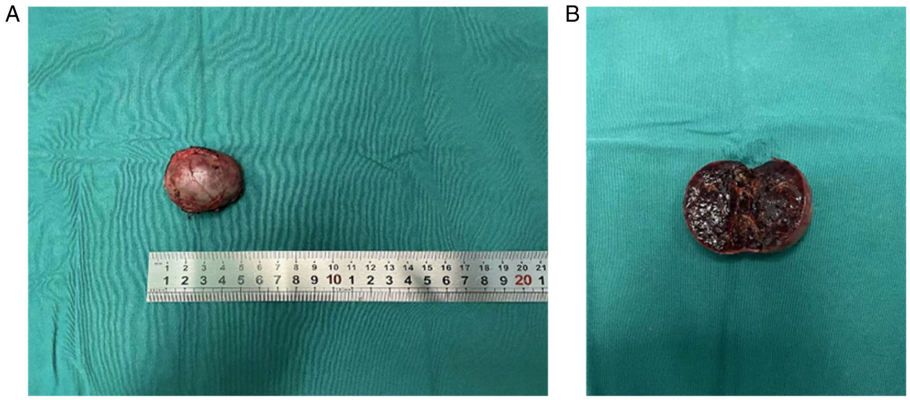

Figure 4. Intraoperative exploration revealed inflammatory changes in the gallbladder, which was successfully resected. A subphrenic broad-based protuberant lesion of ~40 mm was observed and completely removed with an ultrasonic scalpel. (A) The excised mass was ~40 mm in diameter (scale bar in cm) and (B) it exhibited splenic parenchyma (cross-section of the excised mass).

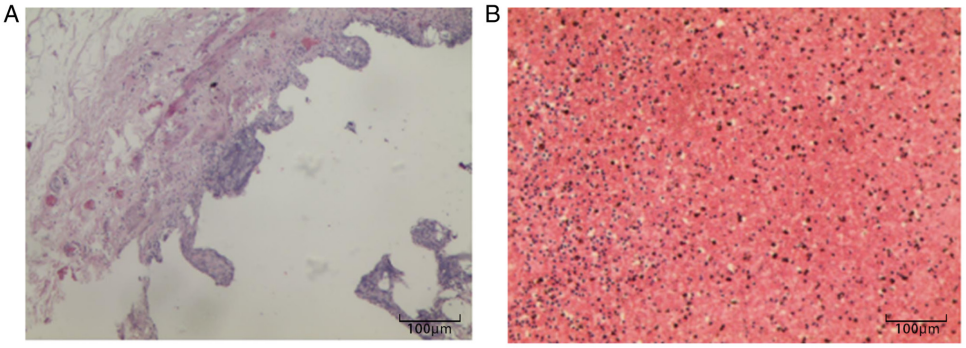

Figure 5. The final pathological report revealed the following: (A) Chronic cholecystitis, mixed calculi and red pulp dilation, as well as (B) hyperemia and bleeding in round tissue with blood clot formation and acute and chronic inflammatory cell infiltration (scale bars, 100 μm; H&E staining).

Discussion

Transplantation of splenic tissue refers to the regeneration of splenic tissue in other parts of the body through various means due to trauma or splenectomy (7). The regenerated tissue is composed only of undifferentiated reticular cells, which further differentiate into endothelial sinuses, capillaries and lymphocytes after forming a scaffold with fibrous tissue, and finally form splenic tissue (10). Splenic implanted nodules have no splenic hilum and only several small vessels from the penetrating nodular capsule maintain the blood supply to the ectopic splenic tissue. The splenic-implanted nodule's blood supply is derived from the arteries of the surrounding tissues rather than from the splenic artery (1).

Since most patients have a history of splenic trauma, a large amount of scar tissue remains after abdominal surgery (11). Asymptomatic spleen implantation is not a malignant disease and certain patients with clinical symptoms may survive for a long time without treatment (12). With the enlargement of the implanted spleen, abdominal pain, abdominal distension and other relevant clinical manifestations occur from occupation or compression. Intestinal obstruction or gastrointestinal bleeding are clear indications for surgery (13). The patient described in the present study was treated surgically due to symptoms of hypersplenism, the patient's medical history being suggestive of hypersplenism and the patient having a strong desire for surgical treatment.

The diagnosis of an implanted spleen may be challenging. An implanted spleen may resemble several abdominal malignancies. Several studies have reported cases of splenic masses mimicking pancreatic masses (14), lymphomas (5), neuroendocrine tumors (1), intramural colonic masses (15), liver masses (16) and gastrointestinal stromal tumors (17,18).

The imaging findings of the implanted spleen were also atypical. CT images indicated a slightly lower density, which was similar to that of normal splenic tissue and slightly higher than that of liver parenchyma. However, there were no signs of mosaic enhancement of normal spleen tissue in the arterial enhancement phase. Uniform enhancement was observed in the arterial enhancement phase and portal vein phase, with high density in the arterial phase and slightly high density in the portal vein and delayed phase, without any characteristic appearance (19). Most MR images revealed a slightly low signal on T1WI and a medium-high signal on T2WI, similar to signals from normal splenic tissue (20). It has been reported that ^{99m}Tc-DRBC has high specificity in spleen imaging and the concentration of radioactivity in the spleen is 2-4 times higher than that in the liver, which is of great significance when diagnosing an ectopic spleen (9). If the patient has no clinical symptoms, the mass may be diagnosed through needle biopsy and a pathological examination; however, the tissues may be difficult to distinguish from lymph nodes, so the puncture results must be scrutinized (21). However, it must be emphasized that the real key

to diagnosing splenopathy is to consider it in the context of a pertinent past medical history.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

WC conceived and designed this case report. XM, JG, XJ, WC and ZF participated in the planning and implementation of patient diagnosis and treatment, wrote the initial draft of the manuscript and collected data. JG and YL acquired the data in the diagnostic imaging. JX issued the pathological report and participated in the treatment. YL and ZF acquired staining and histological images. XM, WC and XJ confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent for surgery was obtained from the patient.

Patient consent for publication

Written informed consent for publication of the present report was obtained from the patient.

Competing interests

The authors declare that they have no competing interests.

References

1. Matsubayashi H, Bando E, Kagawa H, Sasaki K, Ishiwatari H and Ono H: A Multinodular mass of abdominal splenosis: Case report of uncommon images of a rare disease. Diagnostics (Basel) 9: 111, 2019.
2. Xiao SM, Xu R, Tang XL, Ding Z, Li JM and Zhou X: Splenosis with lower gastrointestinal bleeding mimicking colonical gastrointestinal stromal tumour. World J Surg Oncol 15: 78, 2017.
3. Reinglas J, Perdrizet K, Ryan SE and Patel RV: Splenosis involving the gastric fundus, a rare cause of massive upper gastrointestinal bleeding: A case report and review of the literature. Clin Exp Gastroenterol 9: 301-305, 2016.
4. Yang K, Chen XZ, Liu J, Wu B, Chen XL and Hu JK: Splenosis in gastric wall mimicking gastrointestinal stromal tumor. Endoscopy 45 (Suppl 2 UCTN): E82-E83, 2013.
5. Priola AM, Picciotto G and Priola SM: Diffuse abdominal splenosis: A condition mimicking abdominal lymphoma. Int J Hematol 90: 543-544, 2009.
6. Isopi C, Vitali G, Pieri F, Solaini L and Ercolani G: Gastric splenosis mimicking a gastrointestinal stromal tumor: A case report. World J Gastrointest Surg 12: 435-441, 2020.
7. Tandon YK, Coppa CP and Purysko AS: Splenosis: A great mimicker of neoplastic disease. Abdom Radiol (NY) 43: 3054-3059, 2018.
8. Falcão de Santana M, Menezes Marques L, Lopes Gibara V, E Magalhaes Neto G, Gustavo De Quadros L, Kaiser Júnior RL, Gouvêa Faria MA and Filho IZ: Ectopic spleen mimicking hepatocellular carcinoma in the late post-operative period of bariatric surgery. Cell Mol Biol (Noisy-le-grand) 64: 113-115, 2018.
9. Hiranyatheb P, Euanorasetr C, Suwanthanma W and Supsamutchai C: Upper gastrointestinal bleeding from gastric splenosis; A case report and literature review. J Med Assoc Thai 96: 749-755, 2013.
10. Carrara S, Rahal D and Repici A: A Case of gastric splenosis mimicking a stromal tumor. Clin Gastroenterol Hepatol 14: e50-e51, 2016.
11. Claassen B, Barneveld PC, Jager G and Rutten MJ: Abdominal splenosis. Ned Tijdschr Geneeskd 159: A8880, 2015 (In Dutch).
12. Fremont RD and Rice TW: Splenosis: A review. South Med J 100: 589-593, 2007.
13. Ksiadzyna D and Peña AS: Abdominal splenosis. Rev Esp Enferm Dig 103: 421-426, 2011 (In English, Spanish).
14. Mascioli F, Ossola P, Esposito L and Iascone C: A rare case of pancreatic splenosis and a literature review. Ann Ital Chir 9: S2239253X20032120, 2020.
15. Obokhare ID, Beckman E, Beck DE, Whitlow CB and Margolin DA: Intramural colonic splenosis: A rare case of lower gastrointestinal bleeding. J Gastrointest Surg 16: 1632-1634, 2012.
16. Kang KC, Cho GS, Chung GA, Kang GH, Kim YJ, Lee MS, Kim HK and Park SJ: Intrahepatic splenosis mimicking liver metastasis in a patient with gastric cancer. J Gastric Cancer 11: 64-68, 2011.
17. Luo X, Zeng J, Wang Y, Min Y, Shen A, Zhang Y, Deng H and Gong N: Hepatic splenosis: Rare yet important-A case report and literature review. J Int Med Res 47: 1793-1801, 2019.
18. Wang W, Li W, Sun Y, Zhao Y, Zhu R, Li J and Zhang H: Intra-gastric Ectopic Splenic Tissue. J Gastrointest Surg 20: 218-220, 2016.
19. Fishman EK, Urban BA and Hruban RH: CT of the stomach: Spectrum of disease. Radiographics 16: 1035-1054, 1996.
20. Park SH, Han JK, Kim TK, Lee JW, Kim SH, Kim YI, Choi BI, Yeon KM and Han MC: Unusual gastric tumors: Radiologic-pathologic correlation. Radiographics 19: 1435-1446, 1999.
21. Li B, Huang Y, Chao B, Zhao Q, Hao J, Qin C and Xu H: Splenosis in gastric fundus mimicking gastrointestinal stromal tumor: A report of two cases and review of the literature. Int J Clin Exp Pathol 8: 6566-6570, 2015.