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Solitary necrotic nodules of the liver with "ring"-like calcification: A case report

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Abstract

BACKGROUND

Solitary necrotic nodule of the liver (SNNL) is a rare benign lesion with a complete necrotic core and a clear fibrous capsule containing elastic fibers. We present the case of a patient with a radiographic computed tomography (CT) finding of "ring"-like annular calcification within the lesion and postoperative pathologic diagnosis of necrotic nodules wrapped by dense fibers in liver tissue, as well as the patient's subsequent management and outcome.

CASE SUMMARY

A 38-year-old Chinese woman with a history of systemic lupus erythematosus treated with prednisone and hydroxychloroquine, without any symptoms, was found to have hepatic space-occupying lesions by imaging examination at a health examination. A subsequent CT scan suggested a space-occupying lesion of the liver with annular calcification, which was not defined to be benign or malignant. After that, a laparoscopic hepatic space-occupying resection was performed. The postoperative pathological diagnosis was necrotic nodules wrapped by dense fibers in the liver tissue, and the final diagnosis was SNNL. The patient had an uneventful postoperative recovery.

CONCLUSION

There is a "ring"-like calcification in SNNL. This patient had a history of systemic lupus erythematosus, without a history of parasite infection, trauma, or tumor. Therefore, whether the etiology and pathological changes of SNNL are related to

rheumatic immune diseases remains to be investigated.

Key Words: Solitary necrotic nodules of the liver; Annular calcification; Rheumatic immune disease; Laparoscopic hepatic space-occupying resection; Case report

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Core Tip: A 38-year-old woman with a history of systemic lupus erythematosus responded well to corticosteroids (prednisone) and hydroxychloroquine. A liver mass was found on examination, and subsequent computed tomography examination confirmed the liver mass with "ring"-like calcification, and thus malignancy was not ruled out. The patient received surgical treatment, and postoperative pathology showed necrotic nodules surrounded by dense fibers in the liver tissue. There were many lymphocytes and tissue cells around the nodules, which were consistent with isolated necrotic nodules in the liver. The patient is currently being followed.

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INTRODUCTION

Solitary necrotic nodule of the liver (SNNL) is a rare disease that is generally considered nonmalignant. It was first reported in 1983 by Shepard and Lee, who described four lesions with a completely necrotic core and a transparent fibrotic capsule containing elastic fibers. The pathogenesis of SNNL remains unknown. In their initial study, Shepard and Lee favored a traumatic or infectious etiology[1]. Sundaresan *et al*[2] found the presence of nourishing vessels within nodules, suggesting a hemangioma origin. They also described the central reticular fibers within the nodules, suggesting a sclerosing hemangioma origin. It has also been reported that SNNL may also demonstrate calcification[3]. Wang *et al*[4] studied 29 cases of SNNL and found that histopathological calcification was more likely to be used to confirm the diagnosis of SNNL. However, due to its non-specific features, SNNL is often mistaken for malignant lesions according to imaging[1,5]. A recent article questioned the traditional assumption that SNNLs are always benign, citing evidence of metastatic gastrointestinal carcinomas found in a small number of cases[6]. We report a patient with a previously indescribable imaging feature-annular calcification.

CASE PRESENTATION

Chief complaints

A 38-year-old Chinese woman was admitted to our Outpatient Department of Hepatobiliary Surgery for a space-occupying liver lesion.

History of present illness

No symptoms related to the hepatic mass were found, and no related treatment or management had been performed.

History of past illness

The patient had a history of systemic lupus erythematosus treated with prednisone and hydroxychloroquine. She had a history of cesarean section. She denied a history of other diseases, trauma or surgery. She had no abdominal tenderness or rebound pain and no palpable abdominal mass.

Personal and family history

This patient had no special personal or family history.

Physical examination

The physical examination of the patient's abdomen revealed no abnormalities. The patient had a flat abdomen with no pigmentation of the abdominal skin. The patient had no abdominal tenderness or

rebound pain, and no palpable abdominal mass.

Laboratory examinations

No elevated tumor markers such as carcinoembryonic antigen, alpha-fetoprotein, and CA-199 were found. Eosinophil counts were normal. Alanine aminotransferase and aspartate aminotransferase were normal before operation. Increased alanine aminotransferase and aspartate aminotransferase were found after the operation. The indexes of other assay items were normal.

Imaging examinations

Abdominal computed tomography (plain + enhanced scans) revealed an isolated, low-density, round lesion (3.4 cm × 2.7 cm in size) in the sixth segment of the liver with a "ring" calcification of 1.5 cm in diameter. There was no enhancement in the arterial phase, portal vein phase, or delayed phase (Figure 1). No radiographic evidence of other abnormalities was observed. No magnetic resonance imaging (MRI) examination was performed.

FINAL DIAGNOSIS

Considering the patient's history, laboratory examinations, and imaging examinations, she was diagnosed with a benign lesion of the liver, with the possibility of malignancy not being excluded.

TREATMENT

After undergoing laparoscopic hepatic space-occupying resection (Figure 2A and B), the patient recovered smoothly. Pathological examination after surgery revealed an irregular liver. A grayish yellow tough nodule measuring 4.2 cm × 3 cm × 2.9 cm was seen inside the lesion after incision (Figure 2C and D). It had a clear boundary with surrounding tissues. It invaded the liver capsule and was 0.4 cm near the resection surface. The remaining liver tissue was yellowish-brown and tough. Microscopically, there were necrotic nodules surrounded by dense fibers in the liver tissue. Many lymphocytes and histiocytes were surrounding the nodules. Immunohistochemical staining showed CD68 [tissue cell (+)], CK [liver tissue (+)], Hepatocyte [Hepatocyte (+)], CK19 [bile duct (+)], fungal immunofluorescence (-), and acid-fast mycobacterium fluorescence staining (-) (Figure 3). No granulomas, parasites, fungi, or atypical cells were found. The final diagnosis was SNNL.

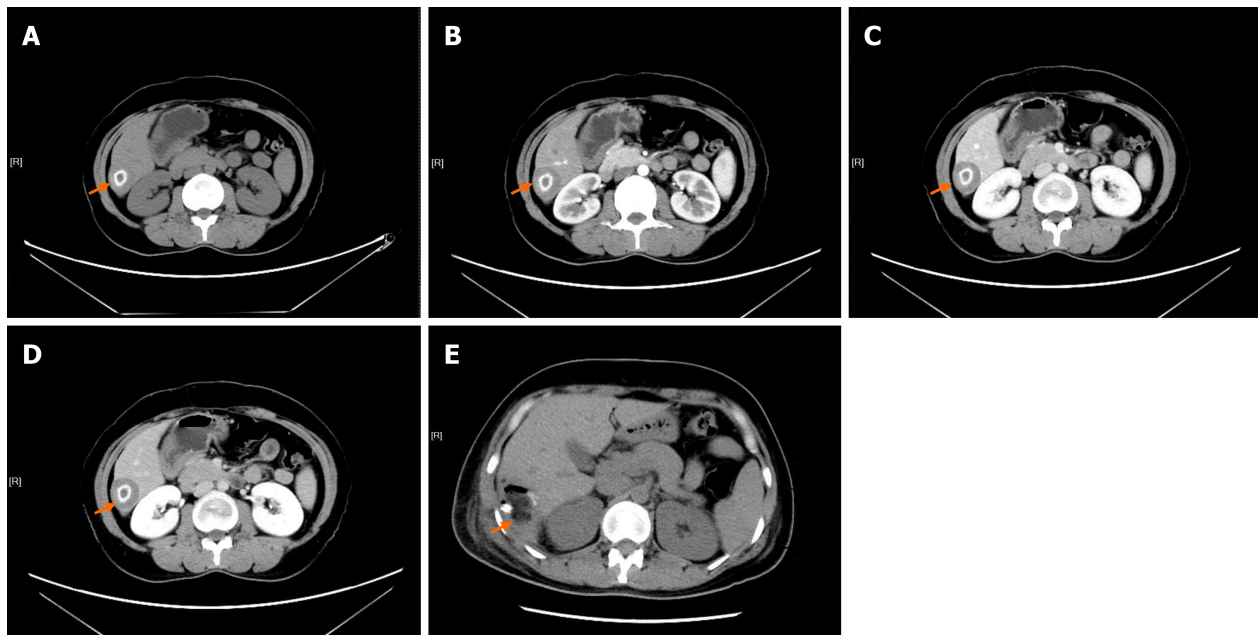
OUTCOME AND FOLLOW-UP

The patient had a smooth postoperative recovery and no adverse events occurred. The postoperative computed tomography (CT) scan showed no abnormal manifestations on the 4th day after liver lesion resection (Figure 1E). The patient is currently healthy and is being followed (lasting for 27 mo).

DISCUSSION

SNNL, a rare entity, was first described by Shepherd and Lee[1] as a rare benign lesion[5,7-10]. The origin of solitary hepatic necrotic nodules remains controversial. The type of cells in or around the nodules can be explained by the following hypotheses: The presence of eosinophilic multinucleated cells will lead to the origin of parasites, while the presence of epithelioid cells may lead to the origin of tuberculosis; however, these hypotheses have never been confirmed[11]. At present, there are two main theories about the pathogenesis of SNNL. One is that the lesion is a result of trauma or previous parasite infection. The other one is related to the sclerosis evolution of hemangioma[2]. The persistence of necrotic material in the center of the nodule may be ascribed to the formation of a dense fibrotic wall, in which blood vessels are destroyed, thereby preventing their reabsorption[7]. Patients with SNNL are often asymptomatic.

Preoperative examination to identify liver diseases such as SNNL, liver metastases, hepatic tuberculosis, inflammatory pseudotumor, *etc.* has an important influence on the treatment option. Hypoechoic nodules normally appear on abdominal ultrasound of SNNL[12], hepatic parenchymal tuberculosis[13], and liver metastatic tumors. Ultrasound has certain limitations in the differential diagnosis of solitary liver nodules. Some literature shows that simple coagulation necrosis type SNNL is manifested by low-density or iso-density nodules with clear borders during plain and enhanced CT scans. There tends to be no enhancement in the SNNL lesion, but thin edge enhancement could be seen in this case, which may result from calcification[3-4]. Liver metastases appear as low-density foci on the



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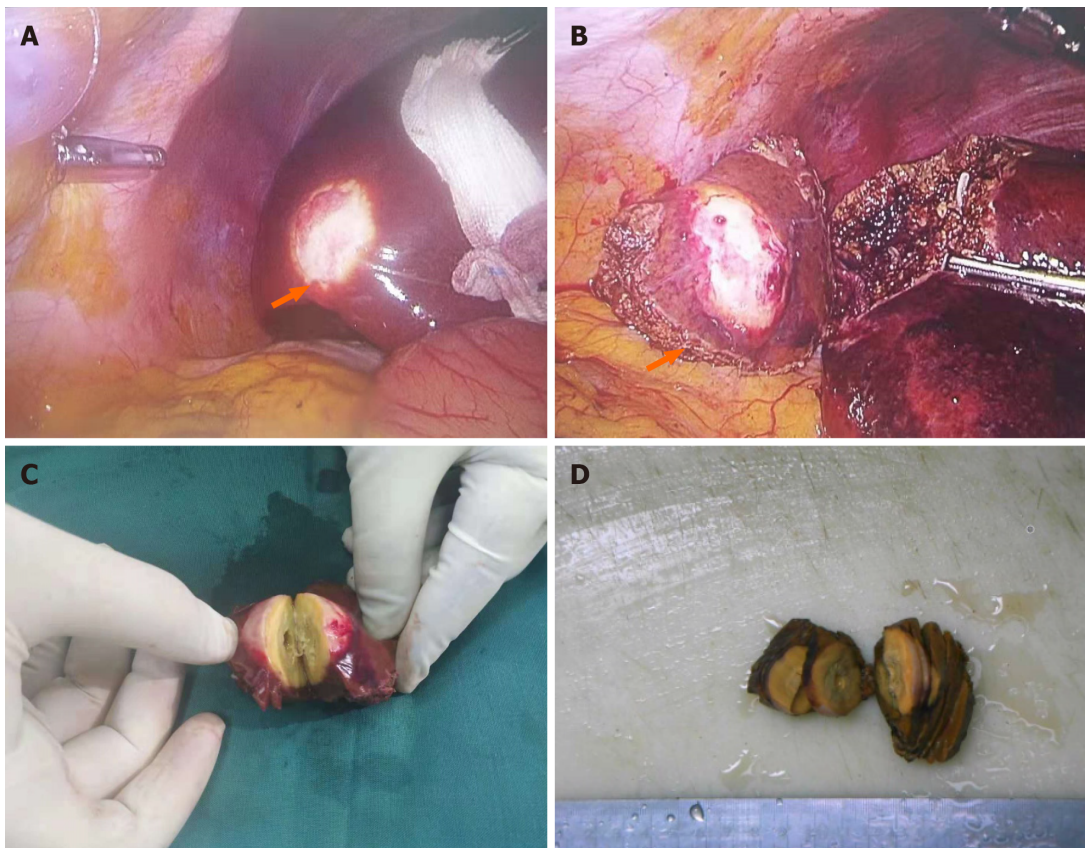
Figure 1 Computed tomography images of the lesion (orange arrow). A: Plain scan image showing a slightly low density, round lesion and "ring"-like calcification in the lesion, measuring 3.4 cm × 2.7 cm; B: Arterial phase; C: Venous phase; D: Delayed phase; E: After the operation. The high signal in the surgical area was the manifestation of drainage tube, and the lesion had no enhancement in the arterial, venous, and delayed phases.

plain CT scan, slightly lower density at the outer edge of the lesion during the enhancement phase and low density at the center of the lesion during the arterial phase. Liver inflammatory pseudotumor and liver parenchymal tuberculosis demonstrated low-density foci on CT, and there was calcification in the center of liver parenchymal tuberculosis. CT also has certain limitations in the differential diagnosis of solitary liver nodules. If SNNL is highly suspected, surgical resection should be avoided[8]. Because the appearance of SNNL overlaps with that of liver metastases, malignancy cannot be completely ruled out [4,14]. Therefore, liver biopsy cannot be performed on this lesion, and the pathological characteristics of the lesion should be determined after surgical resection. Some literature suggests that frequent calcification in the lesion is vital for the differential diagnosis of isolated necrotic nodules in the liver [15]. In this case, obvious "ring" circular calcification was found. Geng *et al*[16] reported that SNNL show a low signal or iso-signal with different degrees of necrosis changes on MRI, and T2WI can show variable signals. On enhanced MRI, there was no enhancement at all stages of SNNL. This patient did not undergo MRI examination.

The patient underwent laparoscopic hepatic space-occupying resection with wide margins. The histological feature of SNNL is the central necrotic core surrounded by a transparent fibrotic envelope. They are usually well-bounded from the surrounding liver and have a yellowish-white soft malleable solid center with hard white edge. Despite being often isolated, multiple necrotic nodules have been reported in the same patient. This case is consistent with related studies. The main pathological manifestation of SNNL is coagulative necrosis, with thin borders of surrounding collagen fibers, sparse monocytes, lymphocytes, plasma cells, inflammatory cells, and elastic fibers, and the central area is rough and patchy with cellular debris[5,9,14]. The pathological manifestation of this case was mainly transparent surrounding fibrotic sacs, lymphocytes, plasma cells, eosinophils, and a small amount of leukocyte infiltration. The calcification in the lesion plays an important role in the diagnosis of SNNL. SNNL is difficult to diagnose before surgery, especially in the case of tumors. Ultrasound-guided liver biopsy may be used to confirm the diagnosis, but it is less accurate than postoperative pathological diagnosis. Therefore, intraoperative and postoperative pathological examinations are the gold standards for the diagnosis of SNNL[10]. In the present case, the lesion was surgically resected and pathology revealed SNNL.

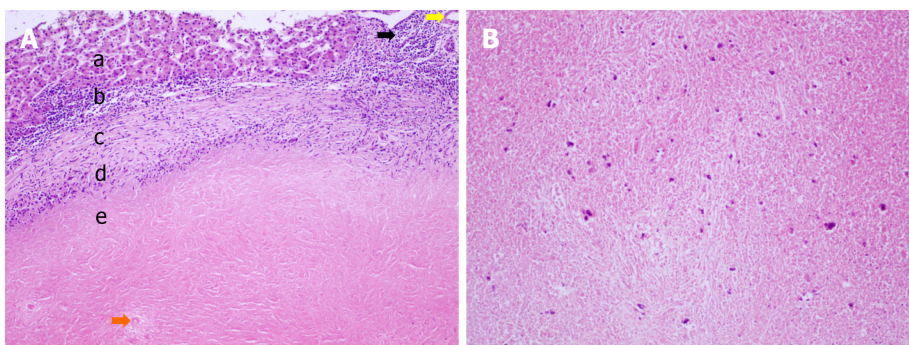
CONCLUSION

In conclusion, we report a case of SNNL with "ring"-like calcification. As SNNL is a rare lesion, there has been no previous similar literature or case reports. Since the manifestations of SNNL overlap with those of liver metastases, malignancy cannot be completely excluded[4,14], so the nature of the lesions needs to be determined through surgical treatment. The cause of SNNL remains unclear[1,2,11]. The patient described here has no history of parasitic infection, trauma, or tumor, yet has a history of



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Figure 2 Laparoscopic surgical resection of solitary necrotic nodules of the liver (orange arrow). A: The lesion was located in the liver and invaded the liver capsule; B: The lesion was completely excised; C and D: Necrotic tissue was seen inside the lesion after incision. An irregular liver tissue after surgical resection (7.6 cm × 5.2 cm × 2.5 cm) and a grayish-yellow nodule (4.2 cm × 3 cm × 2.9 cm) are shown.



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Figure 3 Pathological examination of the solitary necrotic nodule of the liver. A: Microscopic view of necrotic nodules wrapped by dense fibers in the liver tissue, surrounded by the infiltration of lymphocytes and histiocytes (HE staining, 40 × magnification). a: Normal liver tissue; b: Lymphocyte; c: Proliferation of fibrous tissue cells; d: Tissue cells; e: Necrotic tissue; Vascular necrosis (orange arrow); Lymphatic tissue (black arrow); Bile duct tissue (yellow arrow); B: Phagocytes are visible (HE staining, 100 × magnification).

systemic lupus erythematosus. Therefore, whether SNNL is related to rheumatic immune disease or pathological changes awaits further investigation.

FOOTNOTES

Author contributions: Tian H did the conception and design; Bao JP and Wang HC collected and assembly the data; Wang CC and Li B performed the patient follow-up; Bao JP, Wang HC, and Tian H analyzed and interpreted the data; Bao JP wrote the manuscript; all authors approved the final manuscript.

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