*Int Surg* 2011;**96:**310–315

Case Report

# Liver Hemorrhage Due to Idiopathic Peliosis Hepatis Successfully Treated With Hepatic Artery Embolization

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**Peliosis hepatis is an extremely rare condition that may cause fatal hepatic hemorrhage and liver failure. We report a case of liver hemorrhage due to idiopathic peliosis hepatis. A 60-year-old woman was admitted to our hospital with slight right hypochondriac pain. She went into hemorrhagic shock, and computed tomography (CT) showed multiple low-density areas in the right liver with massive subcapsular blood collection. Selective transfemoral arteriography of the celiac artery revealed no signs of vascular malformation or tumor stain, but showed signs of pooling in the right posterior segmental artery. The artery was embolized with particles of gelatin sponge, and hemostatic control was successful. Although peliosis hepatis is extremely rare, the diagnosis is significant because of its urgent clinical status, and transarterial embolization is a useful and minimally invasive procedure for liver hemorrhage due to peliosis hepatis.**

*Key words:* Peliosis hepatis – Liver hemorrhage – Transarterial embolization

Peliosis hepatis is a rare condition characterized by the presence of multiple blood-filled spaces of variable size and morphology in the liver. Autopsy has sometimes revealed this disease incidentally. It is usually related to wasting conditions, including tuberculosis, solid organ malignancies, renal and liver transplantation, and human immunodeficiency virus (HIV) infection. It also has been reported after the use of various pharmacologic agents, such as anabolic steroids and oral contraceptives. In this

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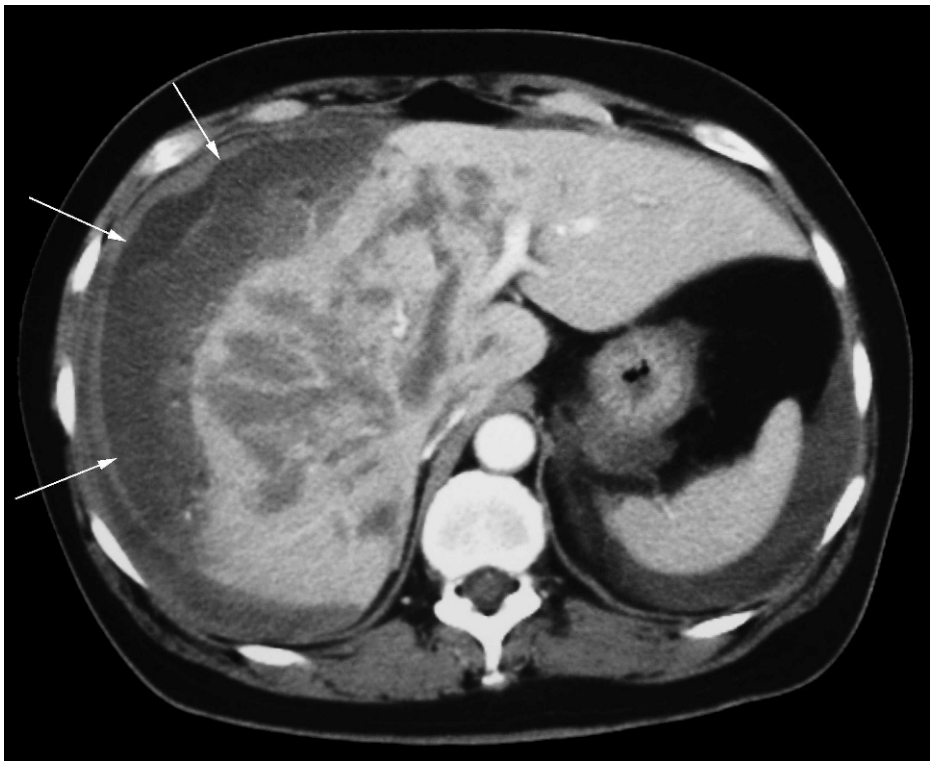

**Fig. 1** CT of the abdomen showed an irregular and low-density lesion in the right lobe of the liver and intra-abdominal hematoma (arrows).

report, we describe an extremely rare case of life-threatening intrahepatic hemorrhage due to idiopathic peliosis hepatis, successfully treated with embolization of the hepatic artery.

## Case Report

A 60-year-old woman presented to her local hospital with right hypochondriac pain. Computed tomography (CT) of the abdomen showed massive intrahepatic hematoma and cancer was suspected, so she was referred to our hospital. She was taking drugs for hypertension (amlodipine 5 mg/d) and allergic rhinitis (ramatroban 150 mg/d), but neither steroid-containing nor immunosuppressive drugs. Vital signs included pulse rate 67/min, body temperature 35.8°C, and blood pressure 109/70 mmHg.

Abdominal examination revealed right upper quadrant distention with mild tenderness. Complete blood count showed white blood cell count 10,120/μL, hemoglobin 10.8 mg/dL, and red blood cell count 338 × $10^4$/μL. Coagulation parameters showed prothrombin time 72% and an activated partial thromboplastin time of 26.1 seconds. Serum chemistry showed total protein 7.0 g/dL, total bilirubin 1.1 mg/dL, glutamic oxaloacetic transaminase 621 IU/L, glutamic pyruvic transaminase 765 IU/L, and C-reactive protein 3.8 mg/dL. Viral markers showed hepatitis B surface antigen [HBsAg(-)] and hepatitis C virus antibody [HCV Ab(-)]. Tumor markers showed carcinoembryonic antigen (CEA) 1.9 ng/dL and carbohydrate antigen (CA)19-9 25.1 U/mL.

On the following day, the blood cell count showed a fall in hemoglobin level of 8.3 g/dL, and blood pressure had dropped to 74/54 mmHg. Emergent CT showed an irregular and low-density lesion in the right lobe of the liver, revealing large subcapsular blood collection (Fig. 1). Under a diagnosis of massive bleeding from the hepatic artery and hemorrhagic shock, emergent angiography was performed. Selective transfemoral arteriography of the celiac artery revealed no signs of vascular malformation or tumor stain, but showed extravasation of the right posterior segmental artery (Fig. 2). The segmental artery was embolized with particles of gelatin sponge, and successful hemostasis was achieved.

A week later, the differential diagnosis was evaluated. T2- and T1-weighted magnetic resonance

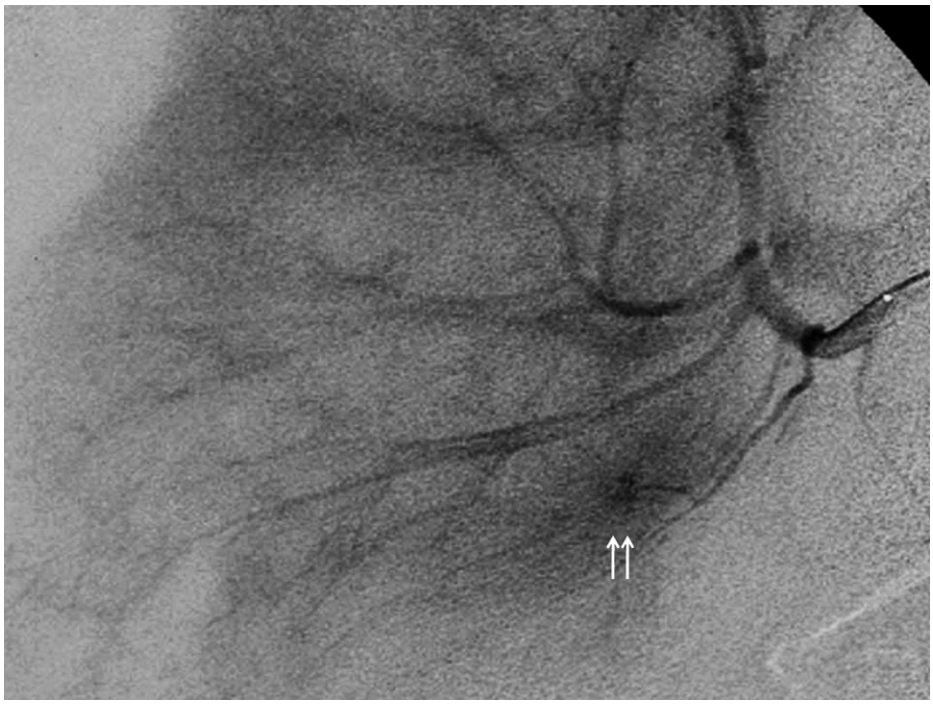

**Fig. 2** Selective angiography of the celiac artery showed signs of extravasation of the right posterior segmental artery with delayed washout (hemorrhagic spots, arrows).

imaging (MRI) showed subcapsular hemorrhage and intrahepatic hemorrhage, but not a tumor, which was the source of the bleeding (Fig. 3). Endoscopic retrograde cholangiopancreatography (ERCP) showed no biliary disorder. Fluorodeoxyglucose-positron emission tomography (FDG-PET) revealed little accumulation in the liver. At this point, we finally diagnosed peliosis hepatis based on these examination findings.

Ultrasound examination was performed repeatedly to monitor the size of the hematoma, which gradually decreased after embolization. We also confirmed the decrease in size of the hematoma with repeat CT scan (Fig. 4). Hepatic function had normalized by 2 months. The patient recovered well and was discharged from our department with no complaints.

## Discussion

Peliosis hepatis was initially recognized in the German literature in 1861 by Wagner, and was named by Schoenlank in 1916.[1] It is characterized by the presence of cystic, irregular, and multiple blood-filled spaces of variable size in the liver.[2] The cause of peliosis hepatis remains unknown, but various theories have been postulated. Blockage of liver blood outflow at the junction of the sinusoid and centrilobular veins, hepatocyte necrosis adjacent to the peliotic cavities,[3] or direct lesions of the sinusoidal barrier seem to be the most likely mechanism in adults.[4]

It has been reported mainly in adult patients in association with chronic wasting diseases, such as tuberculosis, solid organ malignancies, renal and liver transplantation, and HIV infection.[5] It has also been associated with long-term treatment with anabolic steroids and oral contraceptives.[6–8] However, in 20% to 50% of cases, no associated condition is identified.[9] The patient described had no clinical evidence of chronic wasting disease and was taking no drugs, except for hypertension and allergic rhinitis treatments, so we recognized this case as idiopathic peliosis hepatis.

Because the disease is very rare, data about its natural history are not well understood. The clinical findings of various patients have ranged from mild hepatic dysfunction to fatal outcomes, such as complications of liver cirrhosis or rupture in the peliosis; however, severe hemorrhage is controlled by angiographic management in most cases, and life-threatening liver hemorrhage is rare.[10,11] In our case, the patient temporarily went into shock but recovered to an almost normal condition after

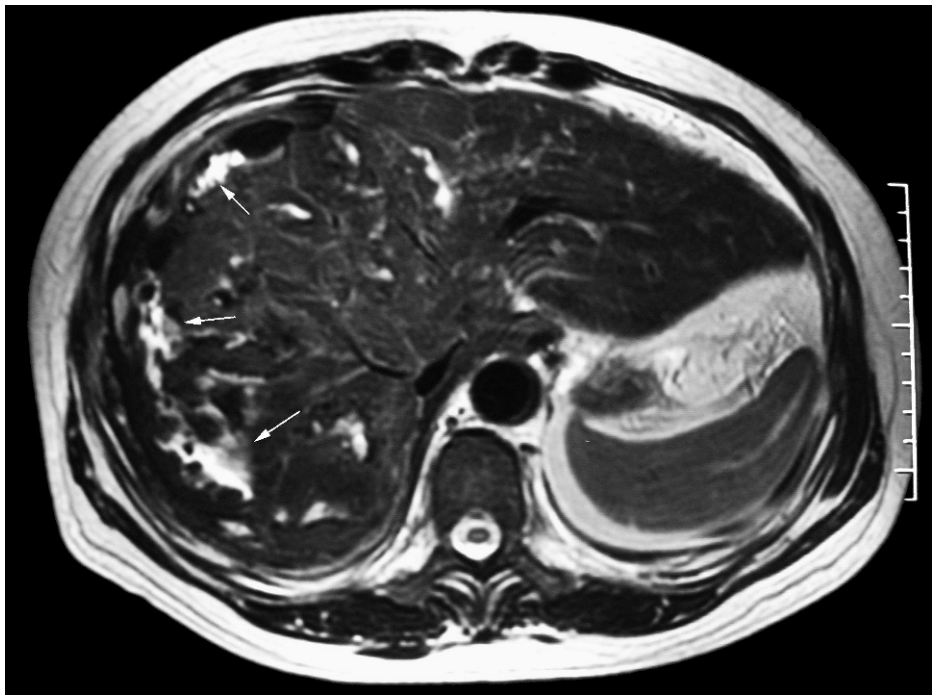

**Fig. 3** T2-weighted MRI showed subcapsular hemorrhage and intrahepatic hemorrhage and demonstrated a hyperintense hepatic lesion (arrows).

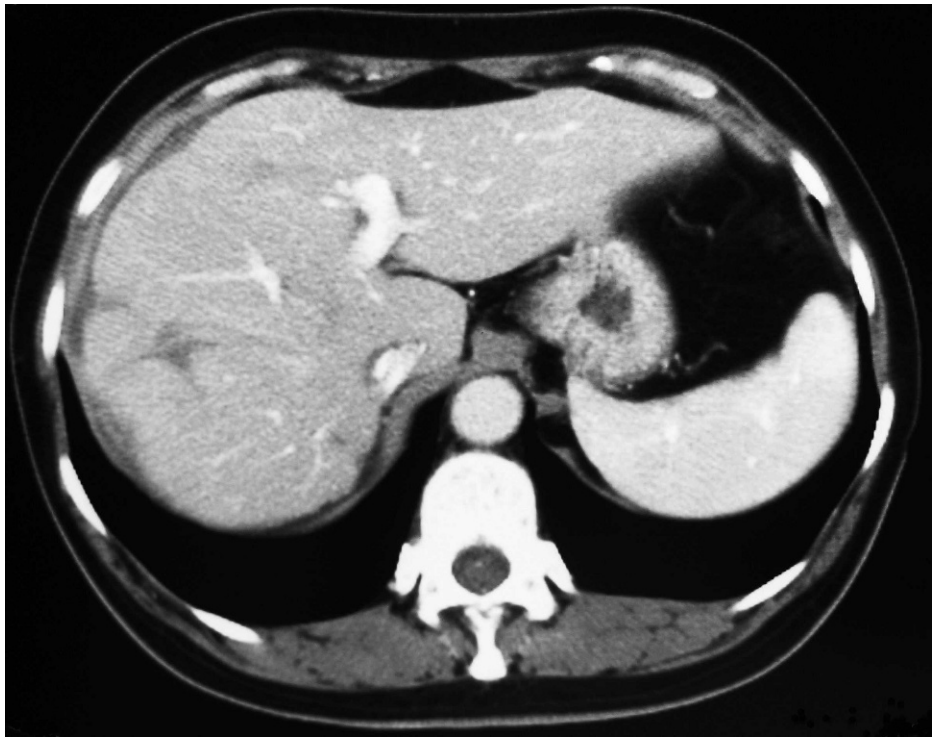

**Fig. 4** Follow-up CT scan 2 months after transarterial embolization revealed near-complete resolution of hematoma and a small low-density residual area.

transarterial embolization. This indicated that the disease was a reversible alteration, and controlling bleeding in the acute phase is the most important action to save a patient with peliosis hepatitis.

Diagnosis before treatment is difficult and sometimes is missed and delayed because of the rare clinical condition; its appearance on radiologic imaging is suggestive of a neoplasm or multiple abscesses.[12] Pathologic diagnosis is obviously ideal, such as an entity characterized by the gross appearance of multiple cyst-like, blood-filled cavities. In this case, because of rapid worsening of status, prompt medical care was necessary. Pathologic findings for this patient remain unclear, but no suspicion of malignancy was reported in any finding retrospectively. Sampling the liver parenchyma with blood-filled spaces as a tissue block is very difficult; even when ultrasound-guided, a percutaneous needle biopsy for diagnosis may promote bleeding and is a more dangerous procedure.[13] Comprehensive consideration, including the findings of clinical imaging, is necessary, and our diagnosis was considered reasonable and appropriate.

Few reports have described the radiologic findings of peliosis hepatis. CT findings frequently reveal patchy low-density areas that may or may not be enhanced with contrast medium[14]; however, the CT appearance of peliosis hepatis can be difficult to differentiate from multiple abscesses, hemangiomatosis, and metastases.[12] MRI findings are comparably specific. The signal intensities of lesions on MRI largely depend on the age and status of the blood component. T1-weighted images can demonstrate hypointense, isointense, or hyperintense foci. On T2-weighted images, the lesions are reported as hyperintense.[15] Our patient's MRI was typical of those described; an angiogram was also needed to arrive at the diagnosis. The diagnosis of peliosis hepatis is made on the basis of an angiography by visualizing small accumulations of contrast material in the late arterial phase, which persists into the venous phase; however, the decisive factor was lacking, so it was necessary to diagnose after considering various possibilities.

No specific treatment for peliosis hepatis is known. Several treatments for peliosis hepatis have been reported, such as partial hepatectomy,[16] liver transplantation,[17] and transhepatic artery embolization.[8] If a patient is taking drugs such as anabolic steroids and oral contraceptives, drugs should be halted immediately. In hepatic failure with cirrhosis, liver transplantation may be the only choice to save the patient's life. In our present case, because of worsening status, arrest of the hemorrhage was required and transarterial embolization, which is less invasive, was conducted. The best treatment for peliosis hepatis remains unknown, so an accumulation of cases is necessary; however, transarterial embolization is an alternative procedure for operative intervention in patients with hemorrhage.

Although peliosis hepatis is an uncommon disease, it should be considered in the differential diagnosis if images show atypical liver features caused by increasing immunosuppression drugs for liver or renal transplantation and HIV infection.

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