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WJCC mainly publishes articles reporting research results and findings obtained in the field of clinical medicine and covering a wide range of topics, including case control studies, retrospective cohort studies, retrospective studies, clinical trials studies, observational studies, prospective studies, randomized controlled trials, randomized clinical trials, systematic reviews, meta-analysis, and case reports.

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Imaging presentation and postoperative recurrence of peliosis hepatitis: A case report

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Abstract

BACKGROUND

Peliosis hepatitis (PH) is a rare benign lesion of vascular origin with a pathological characteristic of multiple blood-filled cavities in the liver parenchyma. It is commonly misdiagnosed due to its lack of specificity in clinical presentation and laboratory test results. Herein, a case of a patient with PH who was misdiagnosed with hepatic echinococcosis before operation to remove the lesions was analyzed, with an emphasis on the computed tomography and magnetic resonance imaging characteristics of PH.

CASE SUMMARY

We outline the case of a 40-year-old Chinese female who was admitted with aggravated abdominal pain with fever for 1 wk. Ultrasound examination at the local hospital indicated hepatic echinococcosis. However, discordance between imaging diagnosis, clinical history and laboratory examinations in our hospital. Subsequently, the patient was pathologically confirmed as having PH-like changes, which recurred 1 year after operation removal of the lesion.

CONCLUSION

Our objective is to highlight the imaging diagnostic value of PH.

Key Words: Peliosis hepatitis; Computer tomography; Magnetic resonance imaging; Recurrence; Misdiagnosis; Case report

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Core Tip: The imaging presentation of peliosis hepatis is complex and diverse, which does not correspond with the clinical history and laboratory examination. In this case, computed tomography and magnetic resonance imaging characteristics of peliosis hepatis were valuable that had been proven.

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INTRODUCTION

In 1916, Schoenlank[1] described peliosis hepatis (PH) through histopathological biopsy: multiple blood-filled cavities of various sizes in the liver parenchyma. This is a rare disease and usually misdiagnosed due to clinical symptoms and laboratory findings are single with atypical symptoms and liver function abnormalities[2]. However, the imaging findings are diverse and complex with similar lesions, like liver hematoma[3], liver cyst[4], liver hemangioma[5], liver echinococcosis[6], liver metastasis[7], adenoma[8] and abscess. Therefore, imaging diagnosis will be an important challenge for the diagnosis of hepatic purpura. Here, we analyze the computed tomography (CT) and magnetic resonance imaging images in a case of a patient with PH who was misdiagnosed with echinococcosis before an operation to remove the lesion. The patient provided informed consent for publication of the case.

CASE PRESENTATION

Chief complaints

A 40-year-old woman reported persistent total abdominal distension and radiating back pain for 1 mo and aggravated abdominal pain with fever for 1 wk.

History of present illness

The patient had no other symptoms except for progressive abdominal pain with fever for 1 wk.

History of past illness

Her past medical history indicated antral gastritis for 10 years, unknown specific treatment, no underlying disease and no history of living in foci of infection or endemic areas.

Personal and family history

She had no personal or family history of other diseases.

Physical examination

On physical examination, the patient presented with abdominal distension and percussion pain in the liver area (+), and she had a blood pressure of 132/78 mmHg with pulse rate of 75 beats per minute.

Laboratory examinations

Laboratory examination indicated the following: total bilirubin, 41.9 mmol/L; direct bilirubin, 17.6 mmol/L; indirect bilirubin, 24.3 mmol/L; gamma-glutamyl transferase, 274 U/L; alkaline phosphatase, 309 U/L; lactate dehydrogenase, 327 U/L; serum 5'-nucleotidase, 28.8 U/L; and serum cholinesterase, 3542 U/L. Urinalysis, coagulation tests, complete blood count, infection marker tests and tumor marker tests did not reveal any obvious abnormalities.

Imaging examinations

Ultrasound examination at the local hospital indicated hepatic echinococcosis, and the patient was admitted to our hospital for further treatment. CT scanning of the right lobe of the liver revealed a cystic lesion approximately 11.7 cm × 15.2 cm in size and with clear margins. Multiple sacs of various sizes were present inside the cyst, with high-density blood accumulation opacities, and fluid–fluid levels in some of the sacs (Figure 1A). The margin of the lesion was slightly enhanced on enhanced scan (Figure 1B), and the inferior vena cava was compressed and unclear. Magnetic resonance imaging examination revealed a large mixed long T2 signal in the right lobe of the liver, the “capsule-in-capsule” sign, signs of intracapsular stratification (Figure 1C) and compression and stenosis of the inferior vena cava.

FINAL DIAGNOSIS

Pathology was consistent with PH-like changes (Figure 2). Microscopy showed multiple blood-filled cysts and liver parenchymal hemorrhage and necrosis in the lesion. The inner wall of the cyst cavity was not lined by endothelial cells.

TREATMENT

The lesion was removed by open surgery under general anesthesia.

OUTCOME AND FOLLOW-UP

The patient had no signs of recurrence at the 3 mo postoperative re-examination (Figure 3A). On abdominal CT re-examination 1 year after operation, multiple low or slightly low-density patches were detected in the liver parenchyma, and some lesions exhibited high-density opacities with blood-like density (Figure 3B). Recurrence of PH was considered.

DISCUSSION

The present case has several features. First, the patient presented with PH by operation and pathology. Second, imaging manifestations did not match symptoms and laboratory tests. Third, the lesions relapsed 1 year after the operation.

PH pathologically manifests as multiple blood-filled cysts of various sizes in the liver parenchyma[1,9]. PH mostly occurs in adults, and its incidence between males and females is the same[10]. The causes of PH are unknown, and its development may be associated with factors such as drugs, toxin exposure[5], cancer[11,12], infection, immunodeficiency[13] and organ transplantation[14]. In recent years, some researchers have reported cases of PH associated with centronuclear myopathy[15] and endocrine factors[16], but the etiology of PH in 20%-50% of patients is unknown[17]. A retrospective analysis of the treatment process and past medical history of the present case of PH did not reveal any of the abovementioned factors.

The results of the laboratory examination of patients with PH are generally nonspecific. The main manifestation is impaired liver function. Abnormal elevation of some biochemical indicators may indicate hepatocyte necrosis. The clinical manifestations also differ because of the different sizes of the lesions. When lesions are small, most patients do not present with any clinical symptoms[18]. But when lesions are large, some patients may present with abdominal distension, fatigue, loss of appetite and other symptoms. In addition, lesion rupture and bleeding may present as sudden severe abdominal pain and hemorrhagic shock[2,8,19].

The classic imaging features of PH are as follows: (1) the “capsule-in-capsule” sign, that is, multiple small sacs within a large lesion; and (2) fluid–fluid levels in the cyst (mixed old and new bleeding). The presentation of PH can be diverse depending on whether concomitant bleeding is present and the imaging characteristics of each period of bleeding[9]. Plain CT scan revealed the presence of equal- or high-density changes in low-density opacities. Enhanced scan can reveal the following[20-23]: (1) Persistent weak enhancement: lesions are relatively low- or equal-density in the

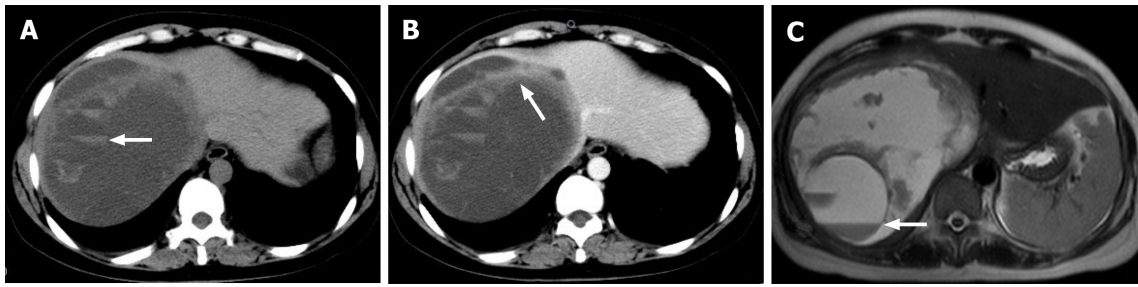


Figure 1 Imaging findings in a 40-year-old woman with peliosis hepatis. A: Plain computed tomography image showed multiple high- and low-density stratification signs (white arrows) within a massive cystic lesion in the right lobe; B: Enhanced computed tomography scan image showed slight enhancement in the margin of the lesion with density lower than that of the surrounding liver parenchyma (white arrow); C: Plain magnetic resonance imaging scan T2WI image showed massive "capsule-in-capsule"-like lesions in the liver parenchyma, mixed lesion signals, primarily high signal. Fluid-fluid (high-low signal) levels (white arrows) were visible in the capsule.

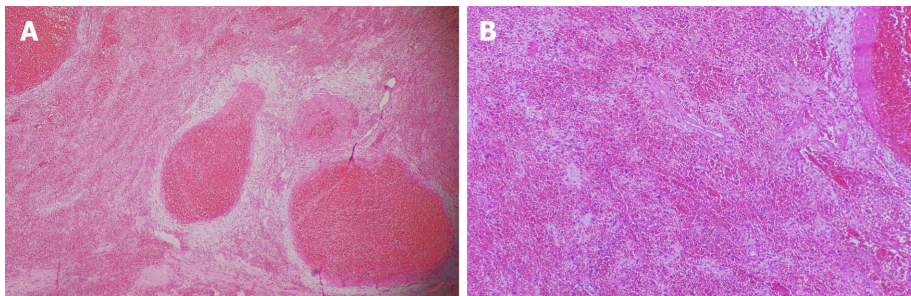


Figure 2 Postoperative pathology. Hematoxylin and eosin staining, 100× (A) and hematoxylin and eosin staining, 200× (B) microscopy showed multiple blood-filled cysts and liver parenchymal hemorrhage and necrosis in the lesion. The inner wall of the cyst cavity was not lined by endothelial cells. These results were consistent with the presentation of peliosis hepatis.

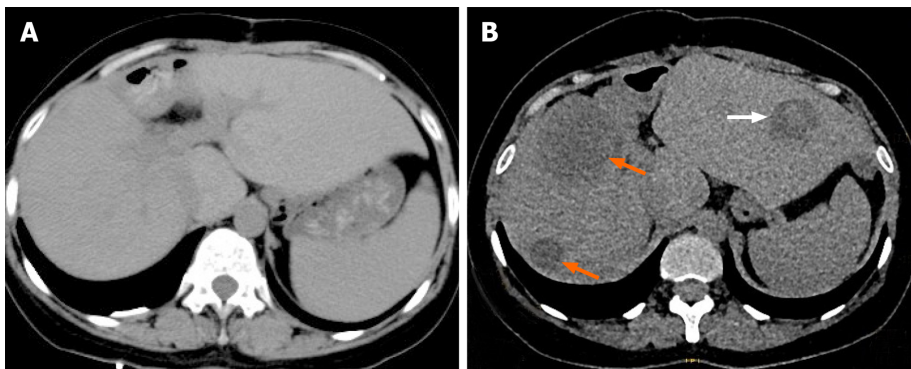


Figure 3 Postoperative re-examination images. A: Computed tomography re-examination at 3 mo postoperatively did not indicate obvious abnormal density opacities in the liver parenchyma; B: Computed tomography re-examination at 1 year postoperatively revealed multiple patches of low- or slightly low-density opacities in the liver parenchyma (orange arrow), with patchy high-density hemorrhage in the lesion (white arrow).

arterial phase, portal phase and delayed phase. When the lesions are large and combined with hemorrhage and necrosis, they may present with solid margins with partial weak enhancement similar to the present case; (2) Centripetal enhancement: the lesion margin exhibits annular or nodular enhancement in the arterial phase and gradually spread to the center of the lesion in the portal phase and delayed phase; and (3) Persistent and obvious enhancement: significant nodular or patchy enhancement of the lesion is present in the arterial phase; the lesion is relatively dense in the portal phase, and the density gradually decreases in the delayed phase. Plain magnetic resonance imaging scan T1WI showed mostly low signal and T2WI showed mostly high signal. T1WI can show equal- or high-density signal with concomitant bleeding. When the bleeding in the lesion is both new and old, the "liquid-liquid level" sign can be seen. The presentation on enhanced scan is consistent with that on enhanced CT[5, 20,21].

Asymptomatic patients with small PH lesions can be followed up for observation. For those with large lesions, lesion rupture and bleeding are usually prevented *via* surgical resection[5]. The most fundamental treatment for this disease involves eliminating or controlling its pathological factors. Thereafter, the prognosis is good, and recurrence is rare. But the present case experienced recurrence at 1 year after operation, strongly suggesting that hidden pathogenic factors may not have been addressed in this case.

CONCLUSION

The imaging presentation of PH is complex and diverse. When the imaging diagnosis does not correspond with the clinical history and laboratory examination, the possibility of this disease should be considered. However, pathological examination is required for a final diagnosis.

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