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Case Report

Misdiagnosis of a Patient with Hepatic Alveolar Echinococcosis with Hepatapostema: A Case Report

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Received 10 May 2023
Accepted 25 Aug 2023

Keywords:
Alveolar echinococcosis;
Misdiagnosis;
Ultrasound;
Case report

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Abstract

Alveolar echinococcosis (AE) is an important zoonotic tropical disease in China that affects people living in western endemic areas. The disease is prone to occur in the liver with a characteristic similar to slow-growing malignant tumors. We report a 31-year-old male patient with serious complication after hepatorrhaphy, who had presented with clinical manifestations of hepatapostema with infection. Ultrasound (US) and computer tomography (CT) are two important medical imaging modalities to diagnose hepatic AE. Based on the medical history, clinical findings, laboratorial and imaging results, the patient was misdiagnosed with hepatapostema. A series of subsequent treatments were ineffective. Finally, partial hepatectomy was performed, and postoperative pathological results confirmed hepatic AE. The patient has now recovered.

Introduction

Human alveolar echinococcosis (AE), caused by the metacestode of the fox tapeworm *Echinococcus multilocularis*, is a near-cosmopolitan zoonosis in the northern hemisphere (1, 2). Tibetan communities is the main epidemic area in China (3, 4). In the early stage, patients with AE have few clinical symptoms (5). Hepatic AE lesions were found accidentally in many patients (6). Here we report a 31-year-old male patient with AE misdiagnosed as hepatapostema.

Case description

The present study was performed in accordance with the Declaration of Helsinki and was approved by the Ethics Committee of West

China Hospital, Sichuan University, Chengdu, China.

In December 2021, a 31-year-old male patient was transferred to emergency department of our hospital (West China Hospital, Sichuan University, Chengdu, China), from Rikazhe People's Hospital in Tibet Autonomous Region because of drainage of purulent fluid for 4+ months after hepatorrhaphy in addition to cardiac fatigue and anhelation for 1 week. On June 24th, 2021, he underwent hepatorrhaphy in the local hospital because of a high falling with liver rupture. After the operation, the amount of purulent fluid was about 400 mL daily from the drainage tube with right upper abdominal pain. Two weeks ago, the patient developed edema of bilateral lower extremities without obvious incentives, without cardiac fatigue, anhelation, decreased urine output, pyrexia, cough or other discomforts. One week ago, the patient presented with cardiac fatigue and anhelation without significant pyrexia or chills. The daily urine output was about 800 mL. In the past 3 days, the patient had felt feverish, without chills, sweating or other discomforts. The patient was admitted to our hospital for further diagnosis and treatment.

Physical examination showed that the body temperature was 36.8 ℃, the regular pulse rate was 106 beats/min, the respiratory rate was 21 beats/min, and the blood pressure was 97/60 mmHg. The patient was conscious, with an acute appearance, and without yellow staining of the skin or sclera and cardiopulmonary abnormalities. The patient had tenderness in the right upper abdomen, and the drainage tube was draining freely. There was no edema in bilateral lower extremities.

Laboratory tests (normal values are given in parentheses) showed that leukocyte count ($4.0\text{-}10.0 \times 10^9$) was 3.47×10^9 cells/L, platelet count was ($100\text{-}300 \times 10^9$) 263×10^9/L, the percentage of neutrophils (50-70%) was 45.5% and monocytes (3-8%) was 11.2%. Clinical biochemical tests (normal values are given in parentheses) showed that hemoglobin (120-160) was 50 g/L, albumin (68-82) was 26.5 g/L, globulin (20-30) was 54.3 g/L, sodium (135-145) was 125.2 mmol/L, chloride (96-106) was 95.7 mmol/L, and calcium (2.25-2.75) was 2.02 mmol/L.

Imaging findings

Contrast-enhanced computed tomography (CECT) showed that the right lobe of the liver presented with an abnormal morphological mass characterized by a heterogeneous density shadow with areas of high-density shadow within it. Multiple areas of pneumatosis were also observed within the lesion, measuring approximately 15.1 × 9.4 cm in the largest cross-section (Fig. 1). Surrounding the lesion, there were multiple areas of slightly lower density shadow in a patchy pattern. The contrast-enhanced scan indicated peripheral enhancement of the lesion, with an increased number of small blood vessel shadows surrounding the lesion in the arterial phase, along with the presence of drainage tube shadow within it. Based on the above findings, infectious lesion was suspected in CECT.

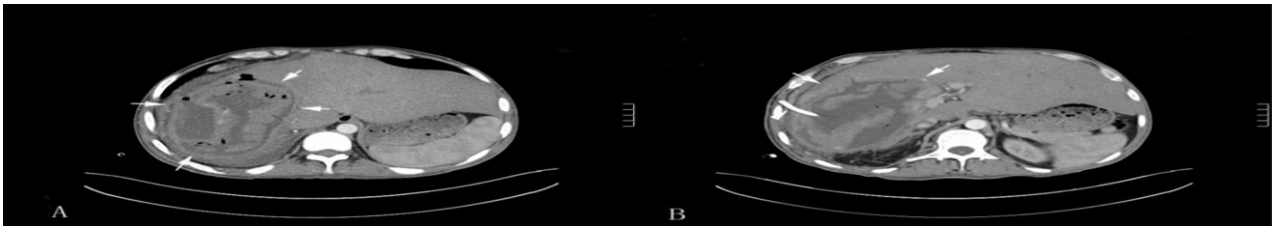

Fig. 1: CT findings. (A) A large heterogeneous mass (arrow) with high-density shadow and multiple pneumatosis. (B) Drainage tube (arrowhead) was found in the mass after percutaneous catheter drainage by US-guided

Ultrasound examination showed that a large hyperechoic heterogeneous lesion was about 13.5 × 11.0 × 15.0 cm in the right lobe of the liver with microcalcifications and irregular anechoic areas inside. Dopper signals few color in the lesion. Ultrasound indicated that the lesion might be a hepatapostema after hepatorrhaphy (Fig. 2).

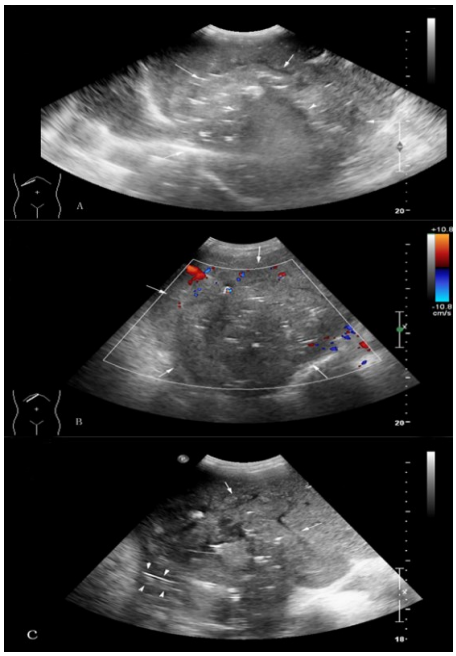

Fig. 2: US findings. (A) A large hyperechoic mass (arrow) with inhomogeneity was found in the right lobe of the liver with microcalcifications and irregular anechoic areas inside. (B) The lesion (arrow) was displayed by color Doppler flow imaging. (C) Drainage tube (arrowhead) was found in the mass (arrow) after percutaneous catheter drainage by US-guided

Subsequent treatments

The purulent fluid drained from the drainage tube exhibited abundant pyocytes (++++), and the bacterial culture test revealed the presence of predominantly G- bacilli. After admission, Piperacillin Sodium was administered to manage the infection, and blood transfusion was performed to address anemia, in addition to other symptomatic supportive treatments. However, the therapeutic response was unsatisfactory. The patient experienced persistent fever for three days, with a maximum body temperature of 39.2 ℃. On December 16th, 2021, the patient underwent US-guided percutaneous catheter drainage for the hepatapostema, resulting in the discharge of a significant amount of drainage fluid. The purulent fluid culture identified Methicillin-resistant *Staphylococcus aureus* (MRSA) and *Enterobacter cloacae*. Based on the results of bacterial culture and drug sensitivity testing, vancomycin and tienam were selected for infection control.

Despite three days of drainage and subsequent ultrasound reexamination, which showed no further fluid drainage from the drain, the patient continued to experience fever, and the size of the lesion did not decrease. As the infection was not fully controlled, the patient underwent hepatapostema resection surgery following completion of the necessary preoperative preparations. Postoperative pathological results confirmed the presence of hepatic AE (Fig. 3). Subsequently, the patient experienced a rapid recovery.

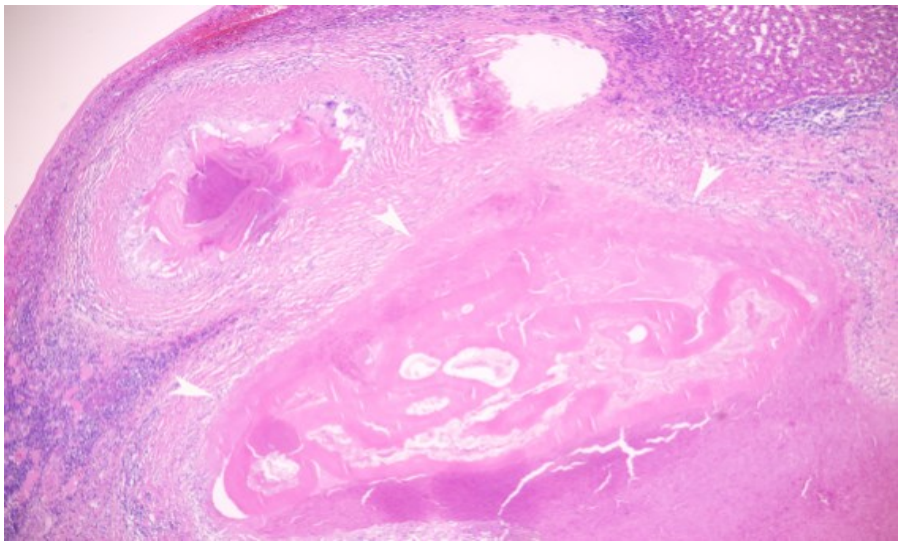

Fig. 3: Pathological findings. Wizened alveolar hydatid cysts (arrow head) were found microscopically, which were strongly positive for eosin (hematoxylin and eosin staining; magnification, x40)

Discussion

Hepatic alveolar echinococcosis (AE) infection can be fatal if left untreated. However, the initial symptoms of AE are often nonspecific (3, 6), and many patients are incidentally found to have AE lesions (7). In the early stages, AE may not present with clinical symptoms, leading to the diagnosis of large hepatic masses after infection (8-11). Surgery is the preferred choice of treatment for patients with hepatic AE, with radical resection of the entire parasitic lesions in the liver being the optimal treatment (12). The misdiagnosis of this patient primarily stemmed from the fact that the local hospital only performed hepatorrhaphy after liver rupture, without identifying the presence of intrahepatic hydatid lesions. As a result, there was a risk of removing the drainage tube, which could adversely affect the patient's quality of life and increase the risk of infection. The percutaneous catheter drainage performed under ultrasound guidance in this case was not as effective as expected for treating AE lesions. However, following the patient's radical resection of the AE lesion, a rapid recovery was observed (13).

The typical sonographic findings of hepatic AE include a mixed heterogeneous echogenic pattern with irregular contours, as well as the presence of cystic necrotic areas or multiple distributed calcified foci. Two studies, categorize the sonographic patterns observed in hepatic AE using ultrasound (14, 15). Accordingly, the present case should have been diagnosed as pseudocystic subtype. Unfortunately, it was misdiagnosed, likely due to the history of hepatorrhaphy, which served as the main interference factor. Despite the patient coming from an endemic area for AE and having no previous history of hydatid disease, the possibility of AE infection was not considered. Another significant factor contributing to the misdiagnosis was the lack of knowledge regarding hydatid disease or AE imaging, leading to the assumption that the lesion resulted from hepatic postoperative infection, particularly considering the atypical clinical manifestations compared to the traditional presentation.

Conclusion

Patients from endemic AE areas with hyperechogenicity, mixed echogenicity, or hypoechogenicity with dotted calcification presented in ultrasonographic images should be suspected of hepatic AE.

Conflict of Interest

The authors declare that there is no conflict of interests.

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