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Radical resection of hepatic polycystic echinococcosis complicated with hepatocellular carcinoma: A case report

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Abstract

BACKGROUND

Hepatic cystic echinococcosis (CE) is an infectious zoonotic parasitic disease, and the insidious onset and slow progression of hepatic CE usually contributes to delayed diagnosis and treatment. Hepatocellular carcinoma (HCC) is the fourth most common malignant tumor. Co-existence of CE and HCC is fairly rare in clinical settings and the association between the two is still not well recognized. We report a case of hepatic CE complicated with HCC which are radically resected and raise some questions worth thinking about.

CASE SUMMARY

A 70-year-old man presented with upper abdominal pain. On admission, laboratory data showed that, except for hepatitis B surface antigen positivity, other indicators were normal, including alpha-fetoprotein. Computed tomography of the abdomen revealed a huge polycystic lesion in left liver lobe, without reinforcement after enhanced scanning and sized about 16.9 cm × 12.2 cm, which was considered a type II hydatid cyst. Multiple small solid lesions were also found adjacent to it, and thus it was highly suspected as a malignant tumor. After a multidisciplinary team discussion, the diagnosis of co-occurrence of hepatic CE and HCC was made. According to Romic classification, the case belongs to type IIb, and radical left hemi-hepatectomy was performed. Postoperative pathological examination revealed CE co-existence with well-differentiated HCC, consistent with the preoperative diagnosis.

CONCLUSION

With the combination of hepatitis B and obvious extrusion by large hydatid, the HCC risk of a patient might be higher.

Key Words: Hepatocellular carcinoma; Cystic echinococcosis; Radical resection; Co-

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Core Tip: Hepatic cystic echinococcosis (CE) is an infectious zoonotic parasitic disease, hepatocellular carcinoma (HCC) is the fourth most common malignant tumor. Co-existence of CE and HCC is fairly rare in clinical settings and the association between the two is still not well recognized. We report a case of hepatic CE complicated with HCC which are radically resected, and present an opinion that with the combination of hepatitis B and obvious extrusion by large hydatid, the HCC risk of the patients might be higher.

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INTRODUCTION

Cystic echinococcosis (CE) is caused by *Echinococcus granulosus* (*E. granulosus*), an infectious parasitic disease. Liver is the most affected organ due to the special life cycle of *E. granulosus*^[1,2]. In recent years, with the development of tourism and the movement of populations, hepatic CE has become a serious public health problem around the world. In endemic areas, the incidence of cystic hydatid disease is 1-200/100000 per year, and fatality rate has been reported as 2%-4%^[3]. Hepatocellular carcinoma (HCC) is the most frequent primary liver cancer, accounting for more than 85%-90% of them, and is a leading cause of cancer-related mortality^[4]. Although both are hepatic space-occupying diseases, it is rare to co-occur, due to their biological characteristics. There have been few reports concerning hepatic CE accompanied by HCC^[5,6]. Whether hydatid infection impacts the occurrence and progression of HCC is still unclear.

Here, we share a case of huge hepatic CE lesion complicated with HCC, and present some controversial issues with detailed discussion.

CASE PRESENTATION

Chief complaints

A 70-year-old man presented to The Department of Hepatobiliary Surgery, People's Hospital of Xinjiang Uygur Autonomous Region, with complaint of upper abdominal pain for 2 wk.

History of present illness

The patient had upper abdominal pain for 2 wk, and no another special discomfort symptom.

History of past illness

The patient had no significant past illness.

Personal and family history

The patient had no significant personal and family history.

Physical examination

The physical examination was significant for pain in the right upper quadrant upon palpation.

Laboratory examinations

All laboratory testing, including glutamic-pyruvic transaminase, glutamic-oxalacetic transaminase, gamma-glutamyl transpeptidase, and alpha-fetoprotein, was normal, except for positivity for hepatitis B surface antigen.

Imaging examinations

Computed tomography of the abdomen revealed a huge polycystic lesion in left liver lobe, without reinforcement after enhanced scanning, sized about 16.9 cm × 12.2 cm (Figure 1), which was considered a type II hydatid cyst^[7]. Multiple small solid lesions were also found adjacent to it, and thus it was highly suspected as malignant tumor.

MULTIDISCIPLINARY EXPERT CONSULTATION

After a multidisciplinary team discussion, the preoperative diagnosis was made of co-occurrence of hepatic polycystic echinococcosis and HCC. According to Romic classification^[8], the case was type IIb. The postoperative pathological examination revealed CE co-existence with well-differentiated HCC (Figure 2), consistent with the preoperative diagnosis.

FINAL DIAGNOSIS

Co-occurrence of hepatic polycystic echinococcosis and HCC.

TREATMENT

Radical left hemi-hepatectomy was performed (Figure 3).

OUTCOME AND FOLLOW-UP

The patient recovered and was discharged on the eighth day after the operation and no treatment was received after discharge. The magnetic resonance imaging conducted at 3 mo after surgery showed no significant recurrence of the lesion (Figure 4).

DISCUSSION

Hepatic CE and HCC are both liver space-occupying diseases with high incidence, whereas the concomitant presence of hepatic CE and HCC is a fairly rare scenario, leaving evidence-based options of treatments unavailable worldwide. Based on the World Health Organization Informal Working Group on Echinococcosis (known as WHO-IWGE) international classification^[7], the current case belongs to the type II hydatid cysts, for which radical resection is recommended to be performed in the guideline. The BCLC staging system is the method currently used to stage HCC and widely accepted as a therapeutic guidance. For the current patient, there were more than three tumors with the size of 35 mm, and the case was considered as intermediate-staged HCC (stage B); furthermore, the lesions were located in the same lobe of the liver. Hemihepatectomy is advised by the European Association for the Study of the Liver clinical practice guideline^[9] and the American guideline (2018)^[10] for such cases. By applying the guidelines for both diseases, the radical left hemihepatectomy was performed and albendazole therapy for CE and HCC adjuvant therapy were not recommended after surgery. In addition, entecavir was administered since the hepatitis B surface antigen test was positive.

To our knowledge, there are few studies on co-infection of CE and HCC and the association between the two is still not well recognized. There are 25 cases reported concerning concomitant HCC and echinococcal cysts in the scientific literature worldwide and 20 of them have provided sufficient data for review analysis. The patients represent 12 males and 8 females, aged from 27 years to 82 years, with an average age of 67. Fifteen of them had underlying chronic liver diseases. It has been

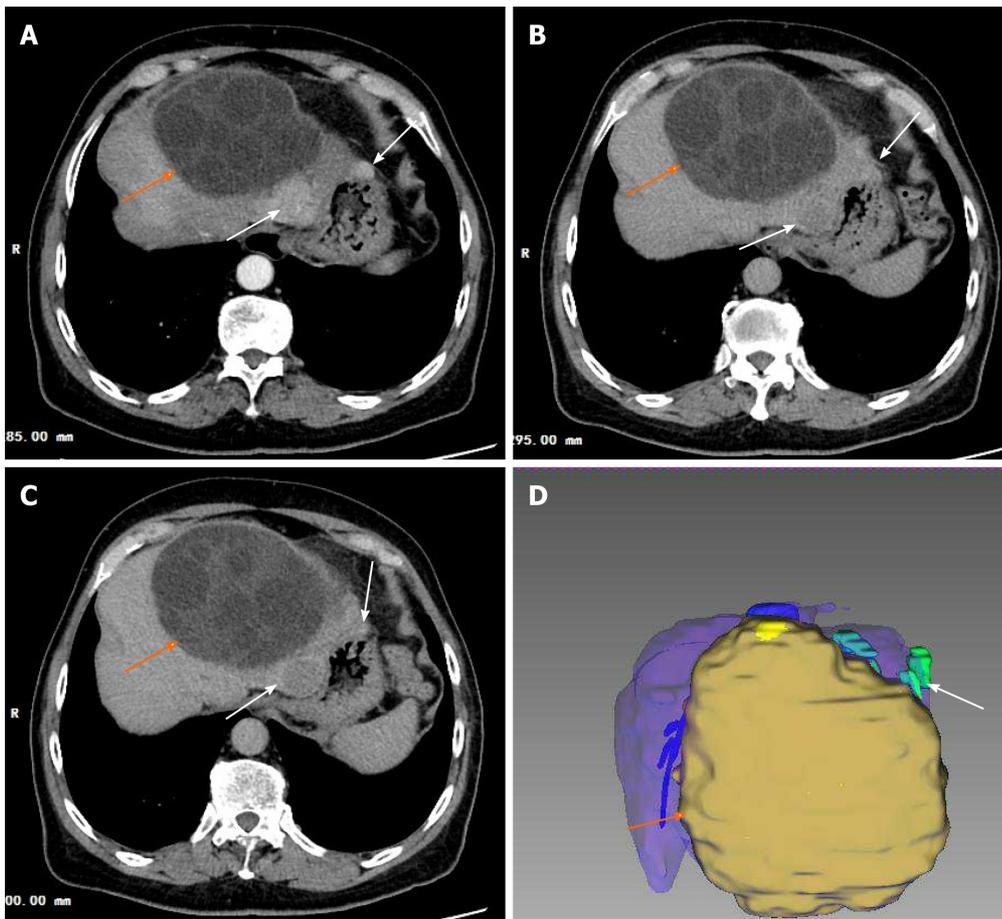


Figure 1 Preoperative images. A polycystic cystic echinococcosis (CE) lesion complicated with multiple hepatocellular carcinoma (HCC) nodules in the left lobe of liver (orange arrow represents hepatic CE lesion; white arrow represents HCC lesions). A: Computed tomography (CT) in the arterial phase showing obvious enhancement of the HCC lesions; B: The portal phase of CT revealed a washed-out pattern of the HCC lesions; C: A delayed CT scan showed HCC lesions without significantly retention; D: IQQA-Liver system three-dimensional reconstruction image.

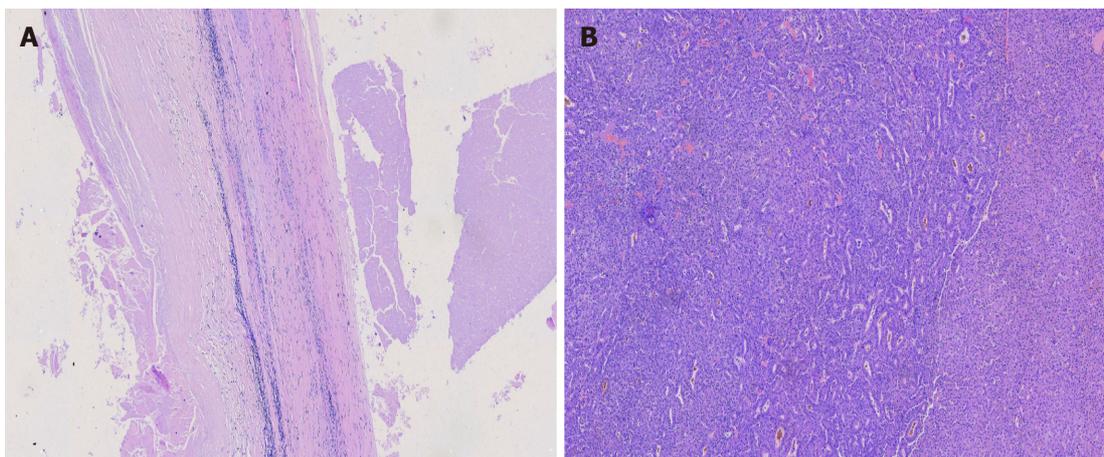


Figure 2 Microscopic histopathology images (× 100). A: Section of specimens showed complete cyst wall structure, consistent with histopathological characteristics of cystic echinococcosis; B: Multiple highly differentiated cancer cells are found in the section specimens.

reported that CE infection may lead to the development of liver carcinoma^[11,12]. By contrast, van Knapen^[13] put forward the hypothesis that echinococcus infection could suppress tumor growth. Recently, Bo *et al*^[14] reported 13 patients with co-existence of hepatic CE and HCC, being the report with the largest number of cases at present. Those authors consider that *E. granulosus* may elicit a protective effect against the development and progression of HCC.

The most common cause of HCC is liver cirrhosis caused by hepatitis and other

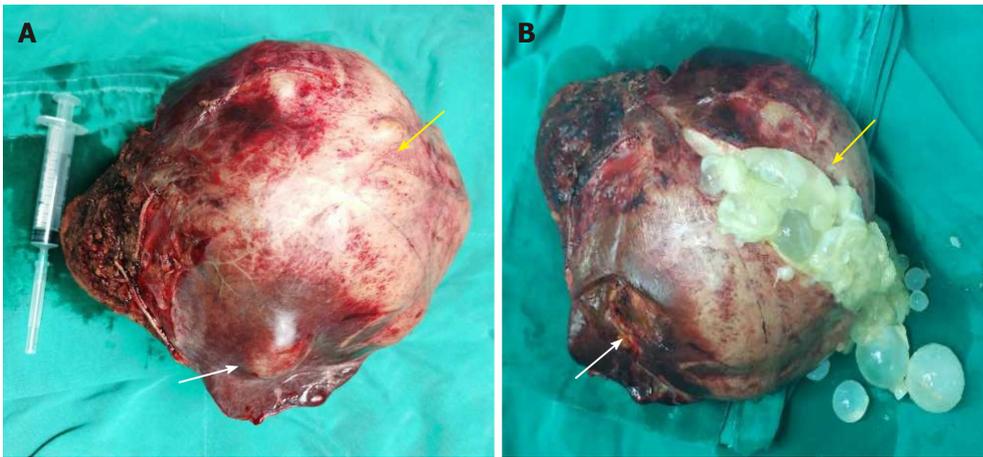


Figure 3 Intraoperative photos. A and B: The yellow arrows represent hepatic cystic echinococcosis lesions and the white arrows represent hepatocellular carcinoma lesions.

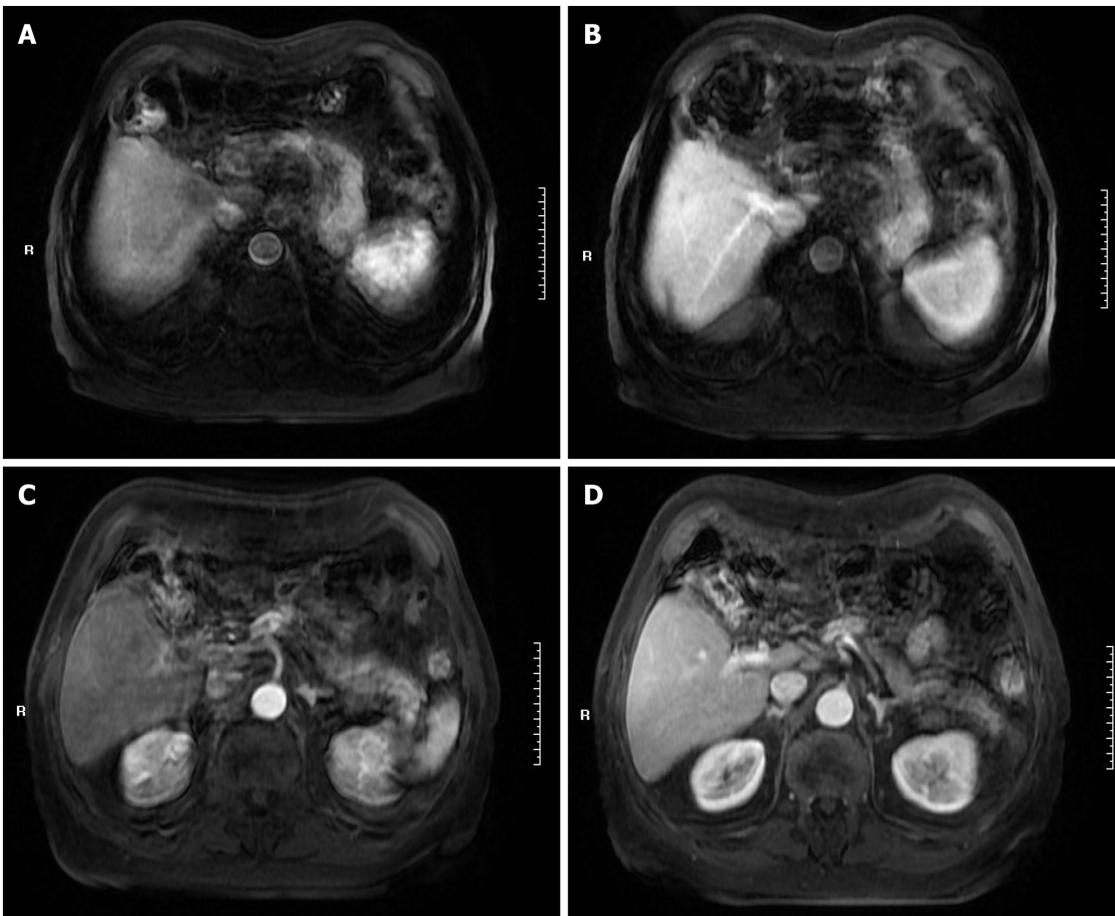


Figure 4 Magnetic resonance images 3 mo after surgery, showing no significant recurrence lesion (A-D).

causes^[15]. Although our case showed evidence of hepatitis B, there was no cirrhosis or elevation of alpha-fetoprotein. In addition, the tumors were located in the hydatid peripheral normal liver tissues, and there was no tumor in the right lobe of the liver. Therefore, we speculate that a chronic inflammatory reaction of liver tissue around the cyst, especially in the extruded area, may have led to increased risk of HCC development.

CONCLUSION

Based on the case and literature review described herein, the current knowledge may raise a hypothesis that human CE may induce the development of HCC, and increased cyst size and hepatitis B may increase the risk of such. When there is combination of hepatitis B and obvious extrusion by a large hydatid, the HCC risk of the patients might be higher. Nonetheless, further studies are needed to clarify the assumption.

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