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Case Report

Coexistence of Giant Hydatid Cyst and Hepatocellular Carcinoma: A Case Report and Literature Review

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Abstract

From a global perspective, hepatocellular carcinoma (HCC) and hydatid cyst disease are both common; however, the endemic and zoonotic nature of hydatid cysts (due to *Echinococcus* larvae) makes the simultaneous detection of the two conditions a rare occurrence. In this case report, in a 43-year-old male patient, we aim to draw attention to the potential coexistence of HCC and liver hydatid cysts by presenting a case in which HCC tissue was detected in the cyst wall—removed by emergency surgery due to cyst perforation. Hydatid lesions in the liver may exhibit tumor-like growth characteristics. Consequently, identifying a hydatid cyst concomitant with HCC can be challenging, particularly when HCC has developed within the cystic structure. Careful assessment of resected tissues and detailed diagnostic approaches can facilitate the identification of such cases, even if the risk of HCC in patients with hydatid cysts is marginal. It may be advisable to suggest periodic monitoring with HCC-related markers and liver imaging methods.

Introduction

Hepatocellular carcinoma (HCC) is the most common liver cancer and the most likely cause of cancer-related death in patients with cirrhosis (1,2). Hydatid cyst disease, also known as echinococcosis, is a zoonosis endemic in many countries including Turkey, and it is caused by the larvae of the Echinococcus tapeworm (3). Liver hydatid

cysts can resemble various liver pathologies (4,5), including cancer. Despite the high prevalences of HCC and hydatid cysts, it is rare for them to be seen simultaneously in the same patient, as demonstrated by the number of such cases reported in the literature (n = 27) (2,4,6-8).

In this case report, we aimed to draw attention to the coexistence of HCC and hydatid cyst by presenting a case in which HCC tissue was found in the cyst wall, detected pathologically following the emergency resection of the hydatid cyst due to perforation.

Case Presentation

A 43-year-old male patient was admitted to the emergency department of a tertiary hospital with complaints of severe abdominal pain, nausea, dizziness and skin rashes that had started approximately 12 hours prior. The patient had visited the General Surgery Outpatient Clinic 2 months ago with a complaint of abdominal discomfort that had persisted for a few months, and the radiological investigations revealed findings consistent with a liver hydatid cyst. Additionally, several hemangiomatous lesions were detected in different areas of the liver. Operative treatment had been recommended to the patient at that time, but the patient refused surgery.

Informed consent was taken from the patient before writing the paper.

Routine assessment revealed no known history of chronic liver disease or other comorbidities, severe pain upon palpation in all abdominal quadrants, and positive defense reflex. The patient had an icteric appearance with maculopapular skin rashes, which were more prominent in the abdomen and lower extremities. Abdominal ultrasonography was conducted, revealing irregular liver contours. The left liver lobe and spleen demonstrated increased size. Numerous nodular hyperechoic solid lesions were observed in both liver lobes. Furthermore, a cystic lesion, measuring 146×110 mm and exhibiting internal levels consistent with a CE-3 class hydatid cyst according to the WHO classification, was identified in the right lobe parenchyma adjacent to the diaphragm (9).

Abdominal computed tomography revealed an enlarged liver. Additionally, a 150-mm cystic lesion consistent with a hydatid cyst was observed in the right liver lobe. Numerous heterogeneous contrast-enhancing solid lesions were identified, with the largest measuring 9 cm. In contrast-enhanced upper abdominal magnetic resonance imaging, a 110 × 140 mm lesion compatible with a hydatid cyst was observed at the level of segments 6-7. Multiple lesions suggestive of hemangioma were also detected, the largest of which was 45 mm (in segment 3) (Fig. 1)

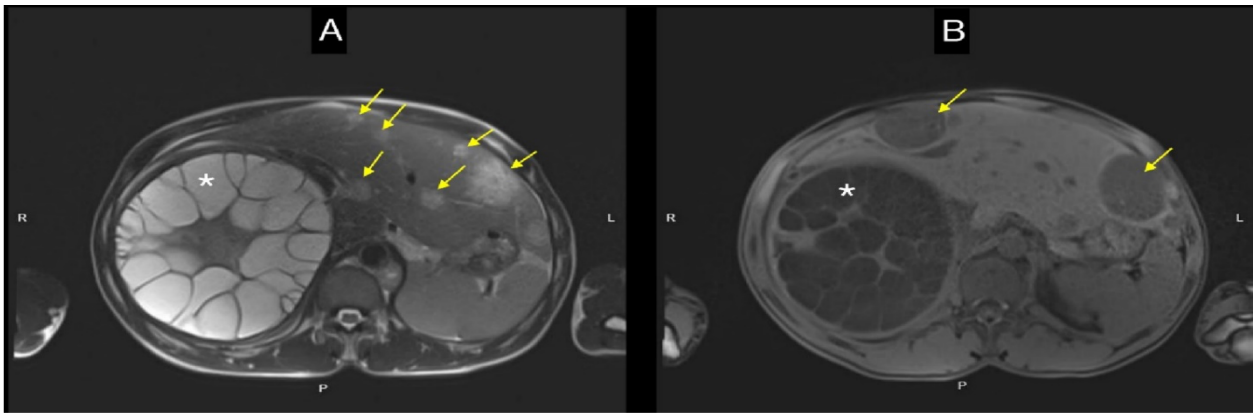

Fig. 1: The MRI examination shows a giant septate cystic lesion with thickened walls (asterisk) and multiple contrast-enhancing lesions (arrows)

The patient underwent emergency surgery. Approximately 300 ml of serous fluid was seen in the abdomen. A cyst was observed in the posterior right lobe of the liver, pushing the liver to the left. There were observable masses on the cyst wall that could not be clearly differentiated, and a 3-4 mm perforation was observed between these masses. Similar lesions were also present in other parts of the liver. Ultimately, a total cystectomy with right partial hepatectomy was performed. The pathological report of the material described moderately differentiated cirrhotomimetic HCC accompanied by echinococcal liver cysts. The Romic class of the lesion was Type 4b and the lesion was Grade 3 according to the modified Edmondson classification (Fig. 2). The patient was monitored in the intensive care unit during the postoperative period in poor general condition due to hepatic encephalopathy as a result of liver failure; unfortunately, the patient died on the 10th postoperative day.

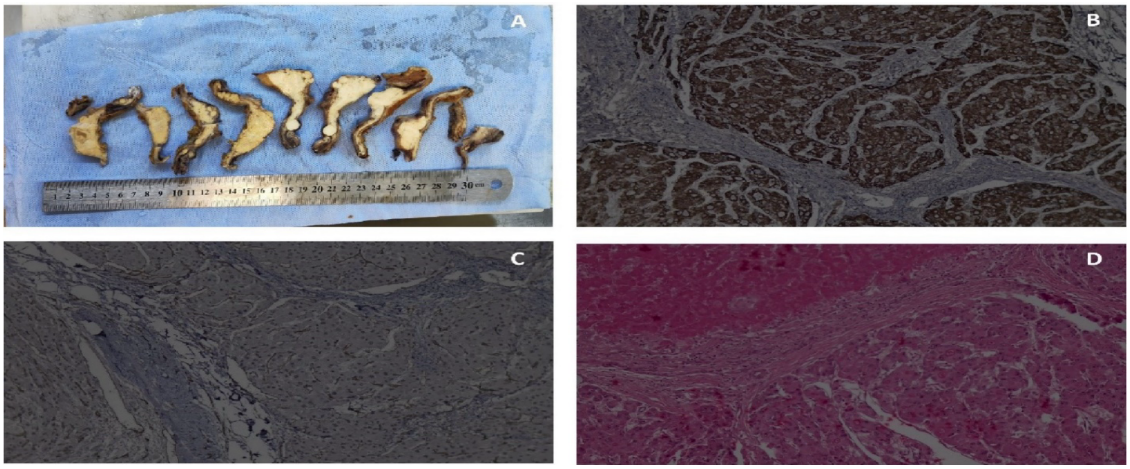

Fig. 2: (A) Cirrhotic macroscopic appearance of partial hepatectomy material. (B) Intense cytoplasmic staining with CK8/18 in hepatocellular carcinoma cells. (C) Abnormal sinusoidal pattern in hepatocellular carcinoma (capillaries appear to have diffuse CD34 positive staining). (D) Staining with hematoxylin and eosin shows areas of tumor necrosis and nodular carcinoma

Discussion

This case report highlights a case of the coexistence of a hydatid cyst and HCC. Despite the lack of a clear preoperative radiological diagnosis, HCC was identified through biopsy that was conducted on the mass connected to the cyst wall.

In the literature, case reports presenting the coexistence of HCC and hydatid cyst, though limited in number, are available. Kostov et al. presented a case of a single lesion involving HCC complicated by a hydatid cyst in the liver (9). Kalifu et al. identified a type II hydatid cyst in the left liver lobe, along with multiple small solid lesions surrounding it on preoperative imaging. The patient underwent radical left hemi-hepatectomy, and final pathological examination confirmed the coexistence of a hydatid cyst and well-differentiated HCC, consistent with the preoperative diagnosis (10). Kübeck et al. reported a case involving a 6 cm heterogeneous cystic lesion adjacent to a 3.7 cm solid mass in liver segments VI and VII, with a significant elevation of alpha-fetoprotein, leading to a diagnosis of HCC and hydatid cyst (8). Kovac et al. described a case initially suspected of HCC with extensive necrosis and peritumoral hematoma in the left liver lobe, which was confirmed as a coexisting hydatid cyst and HCC after histopathological examination following left hemihepatec-

tomy (11). Li et al. identified HCC during surgery in a patient who presented with a ruptured hepatic hydatid cyst (7). One of the most recent case reports suspected the coexistence of a malignant liver tumor and hydatid disease preoperatively, which was confirmed postoperatively (6). Wang et al. confirmed the coexistence of hepatic echinococcosis and HCC through postoperative pathological examination, even though preoperative imaging had not detected the presence of HCC in a patient initially diagnosed with liver cystic echinococcosis (12) (Table 1).

The concurrent occurrence and causal relationship between HCC and hydatid cyst are not well understood due to a scarcity of data in the literature. The prevalence of cancer has been observed to be notably lower in individuals with hydatid cysts, as reported in recent research (2,13-15). Bo et al. conducted a review of data from 3300 patients with hepatic hydatid disease, revealing that only 13 of these patients were diagnosed with concurrent HCC (0.39%) –only one of whom was definitively diagnosed through preoperative imaging (2). The researchers also compared the survival times between HCC patients with and without hydatid cysts, finding that those with hydatid cysts had significantly longer survival. They interpreted this observation as suggesting that *Echinococcus granulosus* might exert a protective effect against the development and progression of HCC (2).

Conversely, some recent studies have suggested that echinococcosis is closely related to the formation and progression of various cancers, including HCC (2,16). The background parenchyma is cirrhotic in most cases of HCC and hydatid cyst coexistence (2), suggesting that such cases may be due to sheer coincidence. However, instances of simultaneous HCC and hydatid cysts without underlying cirrhosis also exist (11), indicating the importance of understanding the potential relationships between these two conditions. These studies have proposed plausible pathophysiological mechanisms connecting hydatid cysts and HCC. Echinococcosis may have procarcinogenic effects by influencing the host immune response (8). *E. granulosus* might support the growth, migration, and invasion capabilities of HCC cells (17). The chronic inflammatory reaction triggered by this parasite in the surrounding parenchyma could lead to the development of HCC (7,8). Although our case involves HCC being part of the cyst wall, we believe that the conjecture put forth by Kovac et al. (11) may apply. That is, the echinococcosis may trigger carcinogenesis by causing mutations in hepatocyte DNA.

Table 1: The summary of case reports presenting the coexistence of hepatocellular carcinoma and hydatid cyst

References	*Summary of case report*
Li et al. (2015) (7)	A 27-year-old female patient presented to the clinic with sudden onset right upper abdominal pain. Ultrasound and PET/CT imaging revealed Echinococcus granulosus in the right lobe of the liver, and the parasite had spread to the abdominal cavity and bile ducts. During surgery, the hydatid cyst was removed, and a widespread cystic echinococcosis infection accompanied by hepatocellular carcinoma was discovered. The
Wang et al. (2022) (12)	A 69-year-old male patient presented with upper abdominal pain and discomfort that had worsened over the previous month. Preoperative imaging suggested hepatic echinococcosis; however, during surgery, nodules were found that were later confirmed to be moderately differentiated hepatocellular carcinoma and echinococcosis. The surgical procedure included hepatic segmental resection, portal vein thrombectomy, and removal of the echinococcal cysts. Postoperative pathology indicated a collision of two lesions, leading to the conclusion that the patient had multiple nodular hepatocellular carcinoma, which had invaded the portal vein, precluding the possibility of radical surgery. The patient was advised to seek further treatment in the medical oncology department after discharge.

Table 1: Continued …

Guo et al. (2022) (6)	A 54-year-old female patient presented with intermittent upper abdominal pain that worsened after consuming greasy foods. Imaging suggested hepatic hydatid disease and a malignant liver tumor, with laboratory tests confirming chronic HBV infection and elevated alpha-fetoprotein levels. She underwent surgery to remove the hepatic hydatid cysts and resect portions of the liver, with pathology revealing moderately differentiated hepatocellular carcinoma alongside hydatid infection. Postoperatively, she received antiviral and antiparasitic treatments, along with multiple sessions of transcatheter arterial chemoembolization due to rising alpha-fetoprotein levels.
Kovac et al. (2022) (11)	A 58-year-old female patient presented with persistent epigastric pain, weight loss, and nausea. Imaging studies revealed a large cystic lesion in the left liver lobe, which upon further MRI examination, indicated features of multinodular hepatocellular carcinoma with hemorrhagic transformation, alongside an echinococcal cyst. Following a left hemihepatectomy, the tumor was completely resected with negative margins. Histological analysis confirmed the coexistence of an echinococcal cyst and well-differentiated hepatocellular carcinoma. The patient had an uneventful postoperative recovery and showed no signs of disease recurrence during follow-up MRI examinations over the next year.
Kalifu et al. (2021)(10)	A 70-year-old man presented with upper abdominal pain lasting two weeks, with a physical examination revealing tenderness in the right upper quadrant. Laboratory tests showed normal liver function, but he tested positive for hepatitis B surface antigen. Imaging revealed a large polycystic lesion in the left liver lobe, consistent with a type II hydatid cyst, alongside multiple small solid lesions suggestive of malignancy. A multidisciplinary team diagnosed co-occurring hepatic polycystic echinococcosis and hepatocellular carcinoma, leading to a radical left hemi-hepatectomy. Postoperatively, the patient recovered well and showed no significant recurrence of the lesion at three-month follow-up MRI.
Kübeck et al. (2013) (8)	A 45-year-old man presented with upper abdominal pain and jaundice. Laboratory results showed elevated liver enzymes and bilirubin, along with a significantly increased alpha-fetoprotein level. Imaging revealed a liver lesion suspected to be an echinococcal cyst, with a co-occurring liver tumor. After partial liver resection, histology confirmed both echinococcal cyst and hepatocellular carcinom. Initially, alpha-fetoprotein levels decreased postoperatively, but a subsequent rise suggested recurrence. A multiregional hepatocellular carcinom was diagnosed, and treatment with Sorafenib was initiated.
Kostov et al. (2010) (9)	A 64-year-old woman presented with a history of weakness, weight loss, and abdominal discomfort. Imaging revealed a 10 cm cystic lesion and multiple nodules in the liver. During surgery, hepatocellular carcinoma complicated by an echinococcal cyst was confirmed. A left trisectionectomy was performed, and treatment with Mebendazole was initiated.

Conclusion

The challenge arises from the fact that hydatid lesions may exhibit growth characteristics resembling tumors in the liver, making the identification of hydatid cysts coexisting with HCC difficult. For patients who refuse surgery for hydatid cysts, it is essential to inform them about the risk of HCC and the potential adverse outcomes.

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Conflict of Interest

The authors declare that there is no conflict of interests.

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