International Cancer Conference Journal (2019) 8:43–45
https://doi.org/10.1007/s13691-018-0352-z

CASE REPORT

A case of acute perforated cholecystitis with intracystic tumor thrombus of hepatocellular carcinoma

Yosuke Nakao[1] · Hirohisa Okabe[1] · Yo-ichi Yamashita[1] · Rumi Itoyama[1] · Toshihiko Yusa[1] · Yuki Kitano[1] · Kota Arima[1] · Tatsunori Miyata[1] · Shigeki Nakagawa[1] · Katsunori Imai[1] · Akira Chikamoto[1] · Hideo Baba[1]

Received: 20 August 2018 / Accepted: 21 November 2018 / Published online: 24 November 2018

Abstract
A 72-year-old man with hepatocellular carcinoma (HCC) was diagnosed with acute perforated cholecystitis requiring emergency surgery. Unexpectedly, a bile duct tumor thrombus was found to have spread into the intracystic space, mimicking a gall stone and presumably causing the severe acute cholecystitis. We herein report an extremely rare subtype of bile duct tumor thrombus of HCC.

Keywords Bile duct tumor thrombus · Hepatocellular carcinoma · Gall bladder

Introduction

Hepatocellular carcinoma (HCC), the most common malignancy of the liver, is the sixth most common cause of cancer-related mortality worldwide [1]. Advanced HCC sometimes produces tumor thrombi, such as a portal vein thrombus, venous tumor thrombus, or bile duct tumor thrombus (BDTT). Reportedly, a BDTT of HCC was found microscopically in 1.0–9.2% of resected specimens [2]. Furthermore, BDTT of HCC in gall bladder is extremely rare. We herein report a very rare case of acute perforated cholecystitis with an intracystic BDTT mimicking a gall stone.

Case presentation

A 72-year-old man with HCC was admitted to our hospital for abdominal pain. He exhibited upper abdominal tenderness without signs of peritonitis. His body temperature was 37.5 °C, his pulse rate was 88 beats per minute, his blood pressure was 88/53 mmHg, and his oxygen saturation was 97% with room air. He had liver cirrhosis caused by hepatitis C viral infection, and he had undergone partial hepatectomy of S4 2 years ago. Pathological finding shows the tumor size of 19 mm, moderately differentiated tumor, none of micro invasion (vp0, vv0, va0 and b0), and pathological stage I. Tumor markers were all within normal ranges at that time. However, he had multiple recurrences in both lobes 9 months after the hepatectomy. His serum AFP level was elevated at 42.5 ng/mL, and his AFP-L3% was 75.3%. He had multimodal treatments with repeated transarterial chemoembolizations, and chemotherapy using sorafenib. In this time, his serum AFP level was elevated at 29.7 ng/mL and his AFP-L3% was 60.9%. He also had high levels of white blood cell count (13,700/μL), C-reactive protein (18.7 mg/dL), alkaline phosphatase (409 U/L), γ-glutamyl transpeptidase (67 U/L), and total bilirubin (1.6 mg/dL). With an abdominal ultrasound investigation, debris was abundant in gall bladder, but an apparent gallstone with an acoustic shadow was not appreciated. Magnetic resonance imaging showed a dilated gall bladder with wall thickening, a gall stone-like mass in the gall bladder, and an extracystic fluid collection indicating the perforation of gall bladder (Fig. 1). Because the diagnostic investigation revealed the acute perforated cholecystitis, emergency surgery was performed. The transverse colon was tightly adhered to the gall bladder and liver edge, resulting in localized peritonitis. Cholecystectomy with almost full-thickness dissection was performed. The postoperative course was uneventful. The patient was discharged on postoperative day 8.

The resected specimen contained a large, soft mass with a size of 48 × 29 mm (Fig. 2a), which was pathologically

✉ Hideo Baba
hdobaba@kumamoto-u.ac.jp

[1] Department of Gastroenterological Surgery, Graduate School of Medical Sciences, Kumamoto University, 1-1-1 Honjo, Chuo-ku, Kumamoto 860-8556, Japan

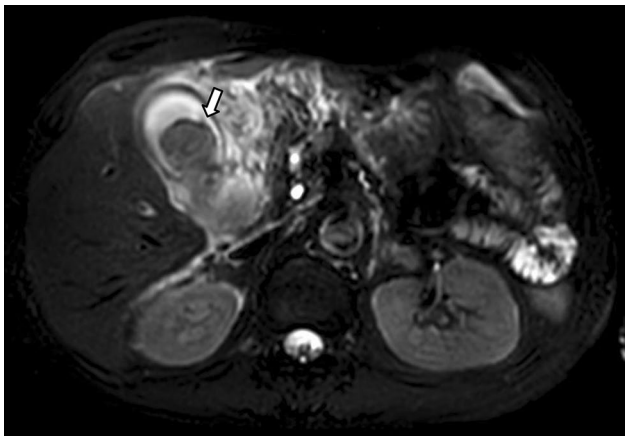

Fig. 1 Preoperative MRI imaging of the gall bladder. A mass mimicking a gall stone (open arrow) was present in the gall bladder on magnetic resonance imaging

found to be viable HCC (Fig. 2b). Pathological examination also revealed viable HCC cells in the center of the gall bladder wall (Fig. 2c). The mass in the gall bladder was finally diagnosed as a BDTT of HCC.

Five months after cholecystectomy, he restarted the treatment with sorafenib. Because of the increase of tumor marker level, the treatment was changed to Lenvatinib 3 months later. Chronological levels of the tumor markers are shown in Fig. 3. He has been alive without relapse of disease for 11 months after cholecystectomy.

Discussion

To the best of our knowledge, this is the first report of an intracystic tumor thrombus that resulted in acute perforated cholecystitis requiring emergency surgery. Compered to portal vein thrombus and venous vein tumor thrombus, BDTT of HCC is extremely rare. Several reports have described patients with BDTTs who exhibited obstructive jaundice because of the tumor thrombus in the common bile duct [3]. In contrast, the current patient did not show jaundice, because the tumor thrombus was intracystic.

In preoperative ultrasonography, it was difficult to find the gall stone of which we incorrectly obtained the information by MRI imaging. We considered that intracystic debris might make it difficult to obtain the image of gall stone by US. If we could confirm that an intracystic tumor never showed the acoustic shadow which is a golden-standard finding of the gall stone, we might be able to suspect the intracystic tumor preoperatively. However, emergent surgery was inevitable this time.

When the thick gall bladder wall was opened during surgery, the intracystic tumor had already been taken off from the necrotic mucosa. We presume that spreading of the BDTT into the intracystic space incidentally impacted the neck of the gall bladder, leading to an increase in intracystic pressure and acute perforated cholecystitis. Patients with BDTT are reportedly characterized to show more advanced tumor stage, portal invasion, and poor prognosis [4]. Given the clinical nature of BDTT based on such a

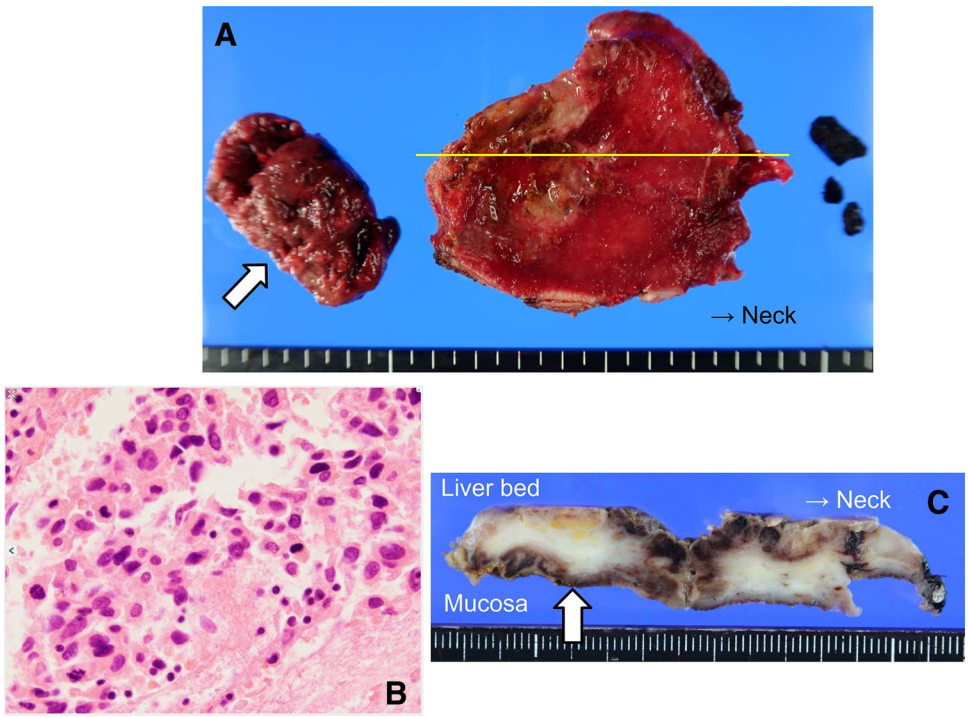

Fig. 2 Macroscopic and microscopic findings of the gall bladder. **a** The resected specimen is shown. The open arrow indicates the tumor thrombus. The membranous side of the gall bladder is shown in the center of the picture. Gall stones are shown in the right of the gall bladder. **b** Hematoxylin and eosin staining of tumor thrombus revealed the existence of viable hepatocellular carcinoma cells. **c** Cut surface of yellow line in **a** is shown. The open arrow indicates the location at which the hepatocellular carcinoma cells pathologically existed in the wall of the gall bladder

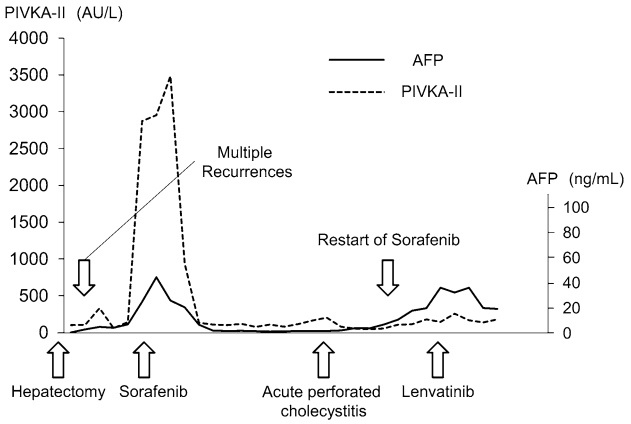

Fig. 3 Changes of tumor markers. The chronological changes of AFP and PIVKA-II levels are shown

report and the current case, BDTT might occur in the late stage of HCC.

Conclusions

Although an intracystic tumor thrombus in HCC is extremely rare, the current case suggested that we have to take the possibility of such disease into account during the follow-up of HCC patients. Unfortunately, CT was not performed in the current case. The ultrasonography might help us to suspect the possibility of intracystic tumor thrombus if the other modality strongly suggests that solid occupied lesion exists in the gallbladder. Furthermore, a BDTT in the gall bladder mimics a gall stone in terms of its clinical course, necessitating consideration of surgical treatment.

Compliance with ethical standards

Conflict of interest The authors have no conflicts of interest.

Ethical approval It was deemed to be unnecessary for this report.

Informed consent It was obtained from the patient and family according to the institutional review board protocols.

References

1. Ferlay J, Soerjomataram I, Dikshit R, Eser S, Mathers C, Rebelo M et al (2015) Cancer incidence and mortality worldwide: sources, methods and major patterns in GLOBOCAN 2012. Int J Cancer 136:E359–E386
2. Peng BG, Liang LJ, Li SQ, Zhou F, Hua YP, Luo SM (2005) Surgical treatment of hepatocellular carcinoma with bile duct tumor thrombi. World J Gastroenterol 11:3966–3969
3. Qin LX, Tang ZY (2003) Hepatocellular carcinoma with obstructive jaundice: diagnosis, treatment and prognosis. World J Gastroenterol 9:385–391
4. Orimo T, Kamiyama T, Yokoo H, Wakayama K, Shimada S, Tsuruga Y et al (2016) Hepatectomy for hepatocellular carcinoma with bile duct tumor thrombus, including cases with obstructive jaundice. Ann Surg Oncol 23:2627–2634