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Mass-forming intrahepatic cholangiocarcinoma with portal vein tumor thrombus and bile duct tumor thrombus: A case report

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ABSTRACT

INTRODUCTION: We report the first case of mass-forming intrahepatic cholangiocarcinoma (ICC) with portal vein tumor thrombus (PVTT) and bile duct tumor thrombus (BDTT), where the extrahepatic bile duct was preserved with thrombectomy.

PRESENTATION OF CASE: A 70-year-old male. Magnetic resonance imaging (MRI) showed the tumor extending from the hepatic hilum to the left hepatic duct with complete obstruction of the left hepatic duct and a defect at the left portal vein. We planned to perform extended left lobectomy, lymph node dissection, extra hepatic bile duct resection and reconstruction based on the diagnosis of mass-forming ICC with left portal vein and left hepatic duct infiltration (cT3N0M0 Stage III). Intraoperative cholangiography revealed a crab claw-like filling defect at the left hepatic duct, which suggested tumor thrombus. Accordingly, we performed thrombectomy. The margin of the left hepatic duct was tumor negative, so we performed extended left lobectomy, lymph node dissection and thrombectomy. Pathologically, the tumor was diagnosed as ICC (pT4N0M0 Stage IVA, vp3, b3). Tumors in the left hepatic duct and left portal vein proved to be tumor thrombus. The postoperative course was uneventful. He is doing well without recurrence.

DISCUSSION: Thrombectomy is performed for hepatocellular carcinoma (HCC) with tumor thrombus. Furthermore, extrahepatic bile duct resection and reconstruction are recommended for ICC. In this case, intraoperative cholangiography was effective for precisely diagnosing. Thrombectomy could reduce surgical stress and prevent complications.

CONCLUSIONS: Thrombectomy can be a valid option for ICC with tumor thrombus, as well as for HCC.

1. Introduction

Portal vein tumor thrombus (PVTT) and bile duct tumor thrombus (BDTT) are pathognomonic for hepatocellular carcinoma (HCC) [1–4]. Intrahepatic cholangiocarcinoma (ICC) commonly infiltrates into the portal vein or bile duct [5,6]. There are a few reports of mass-forming ICC with PVTT or BDTT; however, a case of ICC with both PVTT and BDTT has not been reported to-date [7,8]. Here we report a case of mass-forming ICC with PVTT and BDTT, where the extrahepatic bile duct was successfully preserved with thrombectomy.

2. Presentation of case

A 70-year-old male was referred to our hospital who was noted to have a hepatic tumor. Blood biochemistry studies showed liver dysfunction (AST 52 IU/L, ALT 87 IU/L, ALP 593 IU/L, γ-GTP 894 IU/L), an increase in tumor markers (CEA 7.0 ng/ml, CA19-9 42.5 U/ml, AFP 8.7 ng/ml, PIVKA-II 37836 mAU/ml), and an absence of infection by hepatitis B or hepatitis C viruses. A computed tomography (CT) scan revealed a solitary hypovascular mass located at the hepatic hilum, being 4 cm in diameter and attached to the left portal vein. Magnetic resonance imaging (MRI) showed the tumor extending from the hepatic hilum to the left hepatic duct, with complete obstruction of the left hepatic duct and separation of B1, B2, B3 and B4 (Fig. 1). The MRI also showed a small defect at the left portal vein (Fig. 2). Endoscopic ultrasonography revealed bile duct wall thickening at

Abbreviations: BDTT, bile duct tumor thrombus; CT, computed tomography; ERCP, endoscopic retrograde cholangiopancreatography; HCC, hepatocellular carcinoma; ICC, intrahepatic cholangiocarcinoma; MRI, magnetic resonance imaging; PVTT, portal vein tumor thrombus.

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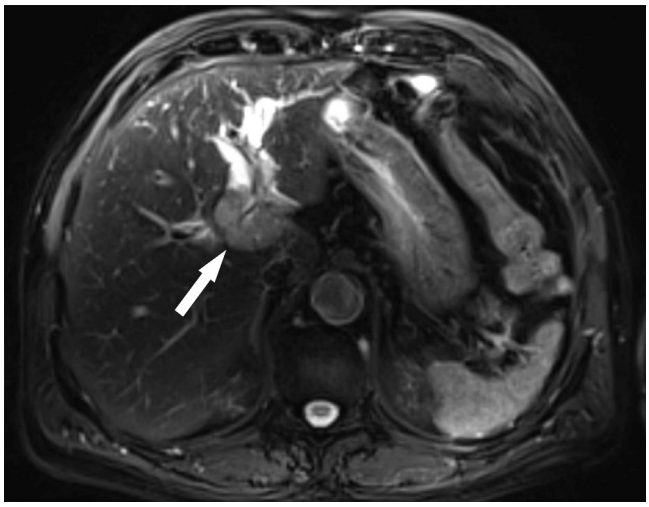

Fig. 1. MRI T2 weighted image. A tumor extending from the hepatic hilum to left hepatic duct (arrow) with a complete obstruction of the bile duct confluence, and separation of B1, B2, B3 and B4.

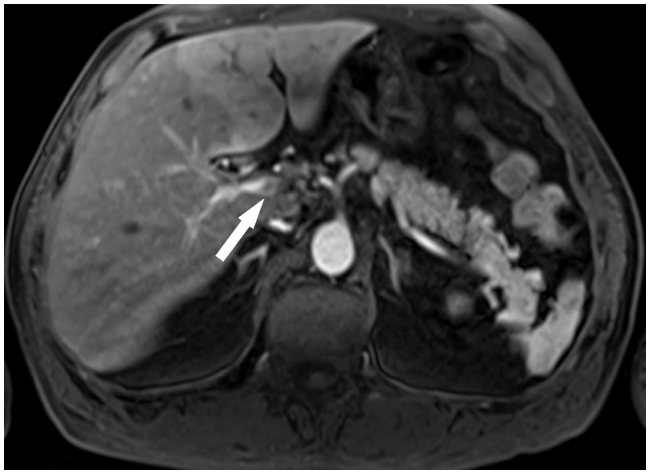

Fig. 2. Dynamic contrast enhanced MRI of the artery phase showed a small defect in the left portal vein (arrow).

the confluence of B2 and B3. Endoscopic retrograde cholangiopancreatography (ERCP) showed a small defect at the hepatic hilum and no enhancement of the left hepatic duct (Fig. 3). Pathological diagnosis of the defect during ERCP showed an adenocarcinoma. These findings led to a diagnosis of mass-forming ICC with infiltration into the left portal vein and left hepatic duct (cT3N0M0 Stage III, UICC7th). Based on the diagnosis, we initially planned to perform extended left lobectomy, lymph node dissection, extra hepatic bile duct resection and reconstruction.

After laparotomy, the left hepatic artery was ligated and divided. Next, the left portal vein was cut and closed after confirming that the PVTT had not extended over the confluence. Intraoperative cholangiography revealed a crab claw-like filling defect at the left hepatic duct (Fig. 4); this suggested that the tumor in the bile duct was tumor thrombus floating in the left hepatic duct, rather than infiltration into the bile duct. As a consequence, we decided to perform a thrombectomy and preserve the extrahepatic bile duct. We incised the duct after identifying the confluence of the hepatic duct. Thrombectomy was successfully performed because the BDTT was not adhering to the left hepatic duct wall. Intraoperative pathological diagnosis revealed that the margin of the left hepatic duct was tumor negative. Therefore, we performed extended left lobectomy, lymph node dissection and thrombectomy (without extrahepatic

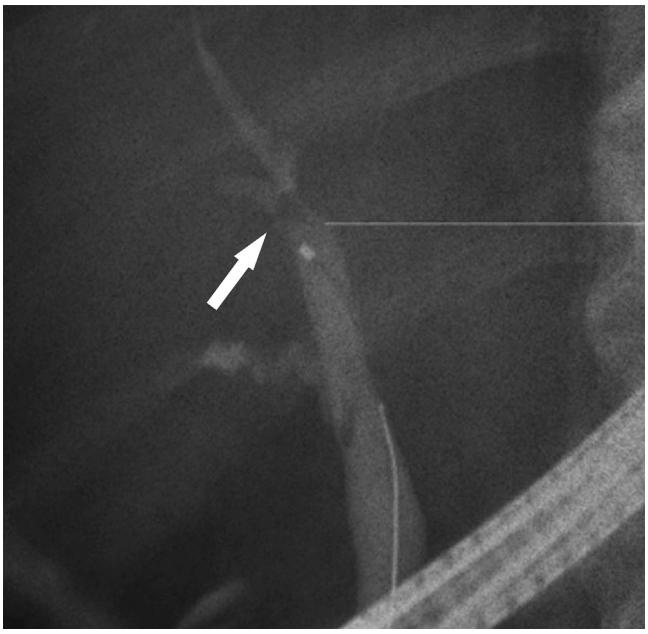

Fig. 3. Preoperative endoscopic retrograde cholangiopancreatography showed a small defect in the hepatic hilum (arrow) and no enhancement of the left hepatic duct.

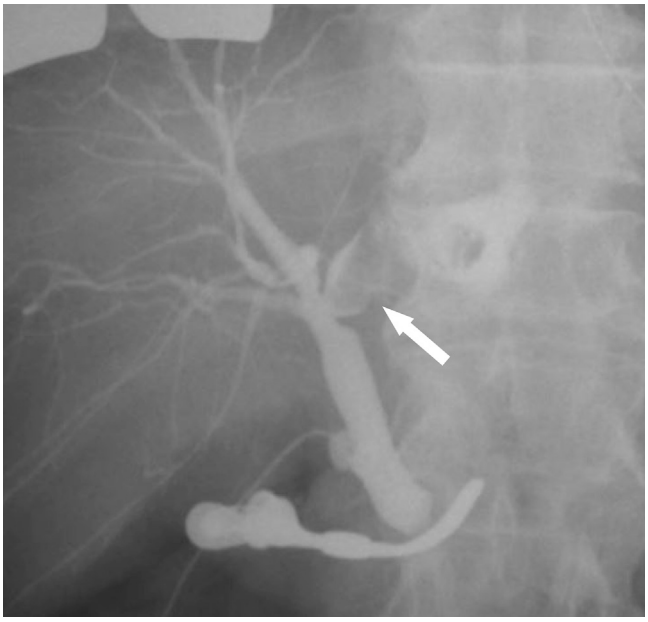

Fig. 4. Intraoperative cholangiography revealed a crab claw-like filling defect in the left hepatic duct (arrow).

bile duct resection and reconstruction). Pathologically, the tumor was 3.8 cm × 2.3 cm, and diagnosed as being moderately to poorly differentiated ICC (pT4N0M0 Stage IVA, vp3, b3, UICC7th). Tumors in the left hepatic duct and the left portal vein connected to the hepatic hilar mass. This finding led to a diagnosis of mass-forming ICC with PVTT and BDTT (Fig. 5A and B).

The postoperative course was uneventful. The patient was discharged on postoperative day 15. Postoperative adjuvant chemotherapy consisted of 8 courses of TS-1 (120 mg/day on days 1–14, every 21 days). Seven months after the operation, the patient is still doing well without any disease recurrence.

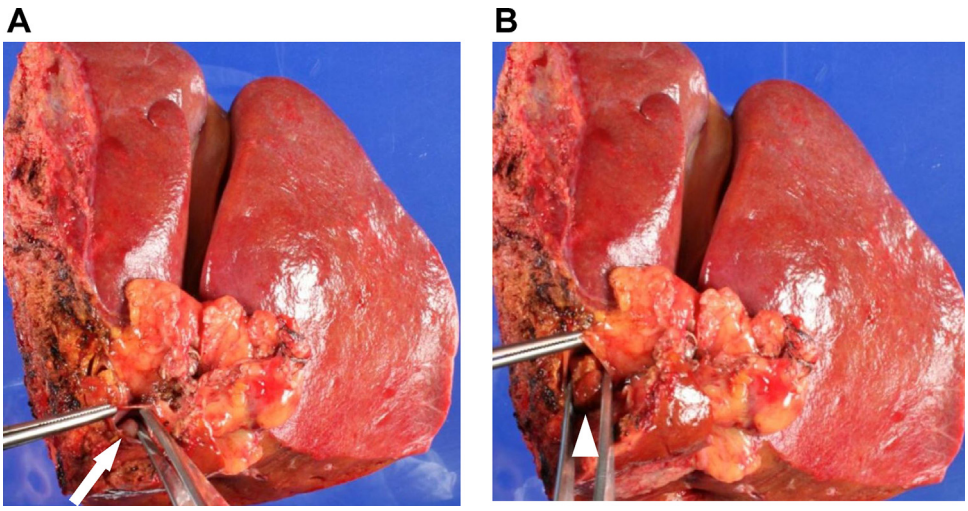

Fig. 5. The resected specimen. Portal vein tumor thrombus in the left portal vein (arrow) (A) and bile duct tumor thrombus in the left hepatic duct (arrowhead) (B).

3. Discussion

As far as we know, this is the first case of mass-forming ICC with PVTT and BDTT. PVTT and BDTT occur more frequently in HCC than ICC. Reports show that the incidence of HCC with PVTT ranges from 14 to 44% [4,9,10], and HCC with BDTT ranges from 2 to 9% [1–3,11]. Conversely, ICC with BDTT or PVTT is rare. Although a few cases of mass-forming ICC with BDTT or PVTT have been reported, a mass-forming ICC with BDTT and PVTT has not been reported to-date [7,8].

In HCC, BDTT loosely attaches to the bile duct epithelium. Because of this, systematic hepatectomy and thrombectomy through choledochotomy are often performed for HCC with BDTT, without resection of the bile duct [2,3,12]. Furthermore, extended lobectomy, lymph node dissection, extrahepatic bile duct resection, and reconstruction are recommended for ICC with BDTT; this is because ICC is a malignant tumor that originates from cholangiocytes and commonly infiltrates into the bile duct [5,13]. In this case, initially we planned to perform extended lobectomy, lymph node dissection, bile duct resection, and reconstruction. However, intraoperative cholangiography revealed that the tumor in the bile duct was tumor thrombus floating in the left hepatic duct. Because of this, we performed a thrombectomy and the BDTT was easily and successfully removed. Intraoperative pathological diagnosis revealed that the margin of the left hepatic duct was tumor negative. Consequently, we performed an extended left lobectomy only, lymph node dissection and thrombectomy, thus avoiding bile duct resection and reconstruction; this reduced surgical stress and prevented complications related to bile duct reconstruction, for example, cholangitis and bile leakage. Our literature search did not identify any reports describing the surgical operations for ICC with BDTT.

In this case, preoperative imaging analyses using MRI and ERCP were not useful for diagnosing BDTT. Intraoperative cholangiography led to a diagnosis of ICC with BDTT floating in the left hepatic duct, so we changed the procedural approach intraoperatively. Contrast medium was able to be injected with high pressure during the operation. Consequently, intraoperative cholangiography was effective for precisely diagnosing the BDTT in this case.

4. Conclusions

This case describes the first case of mass-forming ICC with PVTT and BDTT which was successfully removed without extrahepatic bile duct resection. Thrombectomy could be a valid option for ICC with BDTT, as well as for HCC.

Conflict of interest

The authors declare that they have no conflicts of interest.

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Ethical approval

Research ethical approval is not needed, since this paper is a case report.

Consent

Research ethical approval is not needed, since this paper is a case report. Written informed consent was obtained from the patient for publication of this case report and accompanying images.

Author contribution

Kentaro Iwaki and Toshimi Kaido collected the data and wrote this article. Kentaro Iwaki, Toshimi Kaido, Gen Yamamoto, Naoko Kamo, Shintaro Yagi and Kojiro Taura performed the operation and perioperative management. Shinji Uemoto gave guidance and final approval of the article. All the authors have reviewed this manuscript and approved it for submission.

Guarantor

Dr. Toshimi Kaido.

Materials and methods

The work has been reported in line with the SCARE criteria.

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