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CASE REPORT

A case of inferior vena cava thrombosis caused by compression due to growing giant liver cyst

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Abstract

We report a case of inferior vena cava (IVC) thrombosis caused by compression by a giant liver cyst. A 68-year-old man with a 1-day history of abdominal pain was referred to another hospital. Ultrasonography (US) and enhanced computed tomography (CT) showed a multilobular cyst on the right liver lobe that had increased to 300 mm in diameter from 90 mm 18 months earlier. Thrombosis was detected in the IVC, which was compressed by the cyst. Percutaneous transhepatic cyst drainage achieved no significant change in size. Cytological analysis from the percutaneous drainage tube fluid showed no evidence of malignancy. He was referred to our hospital for further assessment and treatment. Enhanced US using perfluorobutane, CT, and magnetic resonance imaging showed no tumorous lesions in the cyst. Thus, we diagnosed it as a multilobular cyst with no evidence of malignancy. A 3-week course of heparin resulted in the successful resolution of the thrombosis. Cystectomy was subsequently performed and pathological examination showed a multifocal cyst consisting of central suppurative inflammatory exudation and hemorrhagic material, with no malignancy. This case demonstrates that giant, expanding, non-tumorous cysts can cause IVC thrombosis. Careful treatment using heparin successfully resolved the thrombosis and allowed successful cystectomy.

Keywords Inferior vena cava · Thrombosis · Hepatic cyst · Anticoagulation

Introduction

Hepatobiliary cystic lesions can now be definitively diagnosed with advances in modern cross-sectional imaging. The prevalence of hepatobiliary cystic lesions has increased from the historical estimates of 2–18% [1]. Hepatobiliary cystic lesions can be classified into congenital or acquired. Congenital lesions include simple cysts, solitary or in the setting of polycystic liver disease (PCLD); ciliated hepatic foregut cysts; and bile duct cysts (Caroli disease). Acquired lesions include infectious cysts (abscesses and parasitic cysts) and neoplastic cysts, although some evidence suggests the latter may also be congenital. In many cases, hepatobiliary cysts are asymptomatic and are found incidentally on imaging performed for other reasons. Their management varies depending on the diagnosis, risks, and expected outcomes. Simple hepatic cysts should be treated only when they produce abdominal pain, vague discomfort or fullness, early satiety, palpable masses, or abdominal distention.

Occlusion of the inferior vena cava (IVC) is a well-recognized complication of malignant diseases, such as hepatocellular carcinoma. In the absence of malignancy, IVC obstruction is less frequent and may be due to external compression or internal occlusion. Although thrombosis of the IVC associated with PCLD has been reported previously [2], thrombosis in the absence of either PCLD or malignancy is rare. In this report, we present a rare case of thrombosis in the IVC due to a giant, growing, multilobular liver cyst.

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Case report

A 68-year-old man with a 1-day history of abdominal pain was referred to another hospital. He was born and residing in the Niigata prefecture in Japan. He had undergone rectal-sigmoid colon resection 2 years earlier due to perforation of the sigmoid colon. There was no other relevant medical history or medication. Follow-up computed tomography (CT) performed 18 months earlier showed a liver cyst 90 mm in diameter in liver segment 6 (Fig. 1a). CT was performed again at the same hospital; the cyst had grown to 300 mm in diameter and was compressing the IVC, which contained detectable thrombosis and no other findings of abnormalities beyond the compressing cyst (Fig. 1b). He was treated with percutaneous transhepatic cyst drainage (PTCD), but size of the liver cyst did not change. Thus, he was referred to our hospital for management of the giant cyst and IVC thrombosis. Magnetic resonance imaging (MRI) performed at our hospital showed a giant liver cyst and IVC thrombosis. The cyst was hypointense on T1-weighted imaging and hyperintense on T2-weighted imaging (Fig. 2a, b). He did not have a fever. His abdomen was distended, but there was no tenderness. Blood tests revealed liver dysfunction and an increase in inflammatory markers as follows: serum albumin, 2.6 g/dL [normal range (NR), 4.1–5.1 g/dL]; serum total bilirubin, 3.0 mg/dL (NR, 0.4–1.5 mg/dL),

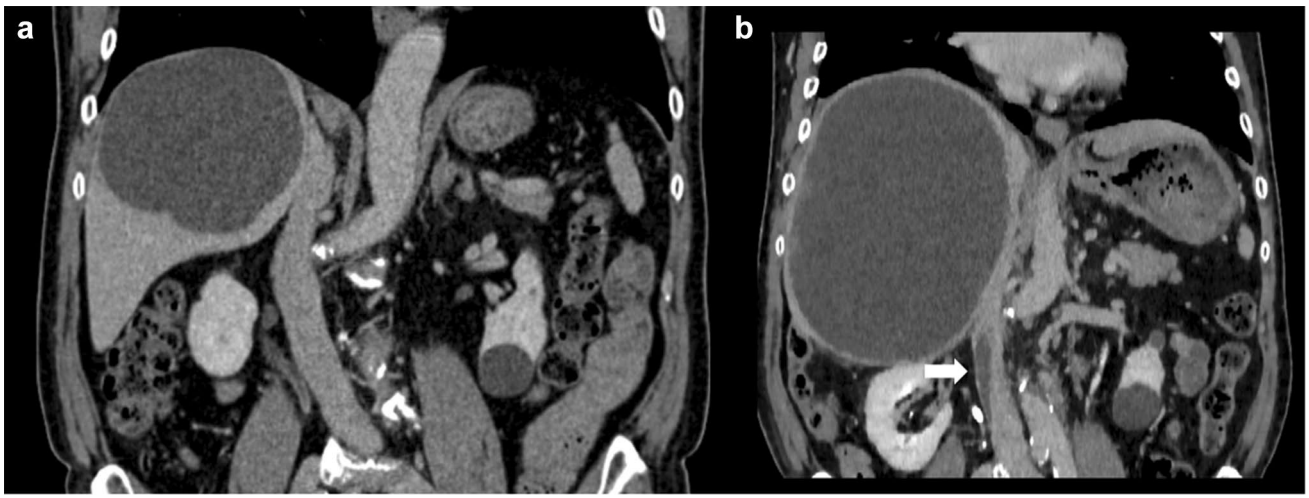

Fig. 1 Enhanced computed tomography. Enhanced computed tomography (CT) shows a cyst in the right liver lobe, 90 mm in diameter at 18 months before hospitalization (**a**). Enhanced CT shows a 300-mm liver cyst in the right lobe and thrombosis in the inferior vena cava (white allow), which was compressed by the liver cyst (**b**)

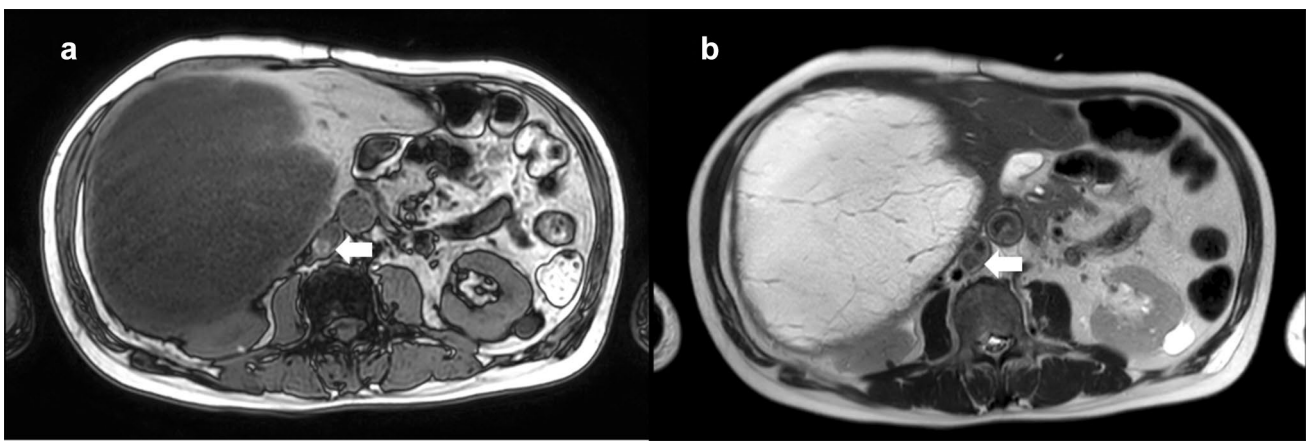

Fig. 2 Magnetic resonance imaging. Magnetic resonance imaging shows a multifocal 300-mm liver cyst in the right liver lobe. The cyst appears uniformly hypointense on T1-weighted imaging (**a**) and uniformly hyperintense on the T2-weighted image (**b**). Thrombosis is detected in the inferior vena cava (white allow), which was compressed by the liver cyst

prothrombin time, 51% (NR, 70–140%); international normalized ratio, 1.39 (NR; 0.80–1.20); aspartate aminotransferase, 101 U/L (NR, 13–30 U/L); alanine aminotransferase, 109 U/L (NR, 10–42 U/L); alkaline phosphatase, 1374 U/L (NR, 106–322 U/L); white blood cells, 14,710/μL (NR, 3300–8600/μL); and C-reactive protein, 16.10 mg/L (NR, < 0.14 mg/L). Levels of the tumor markers, carcinoembryonic antigen and carbohydrate antigen 19-9, were almost within normal range. Enhanced ultrasonography (US) showed a multilobular cyst where the contents of each lobe were different (Fig. 3a). There was no internal vascularity and the uptake of perfluorobutane was not detected (Fig. 3b). The giant cyst was severely compressing the IVC. Bacterial culture and cytological analysis from the PTCD fluid showed few blood components and no evidence of specific infection or malignancy. Thus, we determined that the cyst was multilobular, with no evidence of malignancy. There was no abnormality about portal vein, hepatic vein, and intrahepatic bile duct in the above imaging examinations. We suspected that the PTCD was not effective due to the multilobular nature of the cyst. Based on the physical findings, we considered the elevation in inflammatory markers to be unassociated with symptoms of infection; thus, we did not use antibiotics. To prevent pulmonary thrombosis, we considered placing an IVC filter. However, the short distance between the thrombosis and the right atrium precluded filter placement. We selected to continuously administer heparin (14,000 U/day) for 3 weeks, which resulted in the complete resolution of the thrombosis. During the treatment of the thrombosis, liver dysfunction and elevated inflammatory markers gradually improved, without the use of antibiotics. Subsequently, cystectomy was performed. Macroscopic and microscopic analyses showed a multifocal cyst consisting of central suppurative inflammatory exudation and hemorrhagic material without malignancy (Fig. 4a–c).

Discussion

In the present case, a multilobular liver cyst, which had grown from 90 to 300 mm in diameter in 18 months, compressed the IVC, resulting in thrombosis. Heparin treatment alleviated the thrombosis and the subsequent surgical treatment successfully excised the giant cyst without complications.

Simple cysts are the most common nonparasitic cystic lesions of the liver and are estimated to occur in approximately 5–18% of the population [1, 3]. They are usually asymptomatic, detected incidentally, and do not have malignant transformation potential. A liver cyst can cause extrinsic compression of the IVC, which predisposes it to thrombus formation by causing venous stasis [4]. To determine whether a cyst harbors malignant tissue can be challenging and cyst size is generally not helpful. During US examination, the presence of wall thickening or nodularity is suggestive of malignancy. Doppler sonography demonstrates a sensitivity and specificity in differentiating cystic malignancies from abscesses and simple cysts of 85 and 96%, respectively [5]. Enhanced US examination can also contribute to the diagnosis of intraductal papillary neoplasms [6]. Discordant findings on US and CT regarding nodularity or septation suggest hemorrhage and clotting, whereas concordance is suggestive of neoplasms [7, 8]. MRI can also be helpful in distinguishing simple hepatic cysts with intracystic hemorrhage from neoplastic cysts. In a recent review of cystic liver lesions, hemorrhagic simple cysts consistently had hyperintense elements on T1- and T2-weighted MRI, whereas neoplastic cysts were hypointense on both, reflecting their proteinaceous content [7]. In our case, enhanced US, CT, and MRI showed no evidence of either malignancy or active hemorrhage, but we suspected that the uneven content of the multiple lobes was a sign of previous bleeding and infection. IVC obstruction was inferred from the size of the giant hepatic cyst. It is reasonable to assume that the

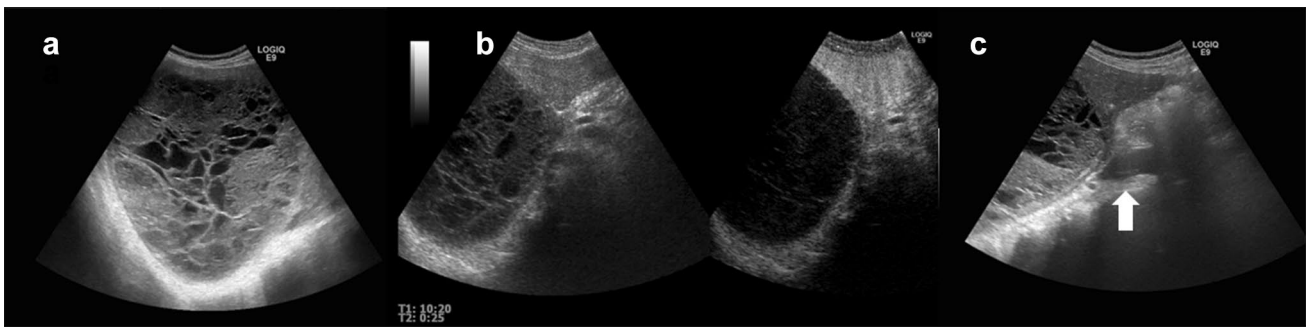

Fig. 3 Ultrasonography. Ultrasonography (US) shows a multilobular cyst in the right lobe. The contents of each lobe appear different in B mode (**a**) and enhanced US using perfluorobutane shows no uptake into the cystic lesion in the Kupffer phase (**b**). The cyst lesion is severely compressing the inferior vena cava (white arrow) (**c**)

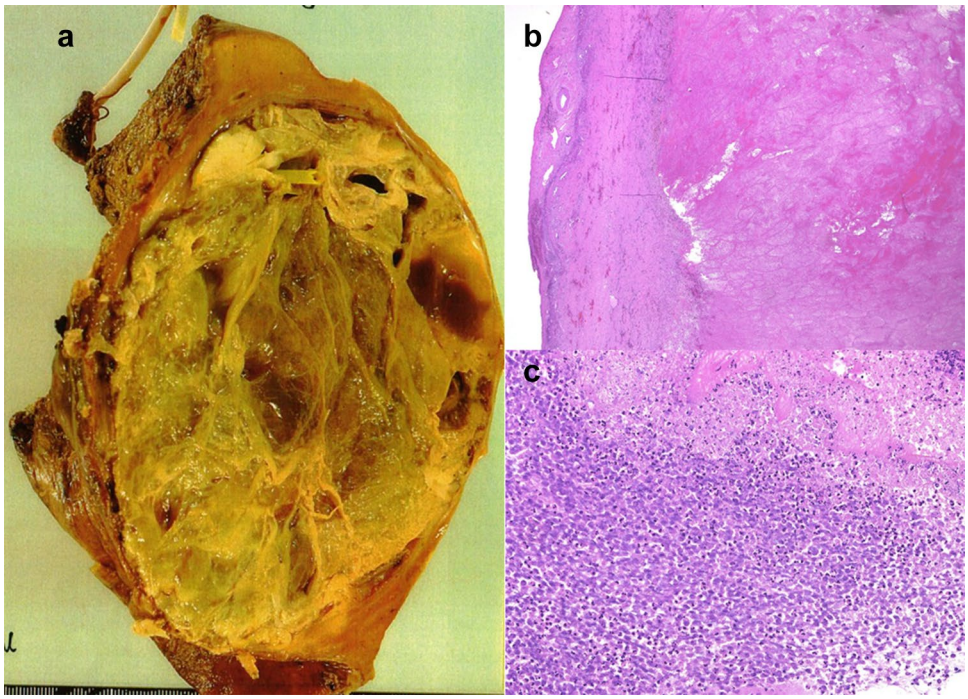

Fig. 4 Macroscopic and microscopic analyses of the cyst. Macroscopic analysis shows a multilobular cyst (**a**). Microscopic analysis shows that the cyst consists of central suppurative inflammatory exudation and hemorrhagic material without malignancy (**b** original magnification × 12.5 and **c** original magnification × 200)

marked narrowing of the IVC, and the resultant flow disturbance, was one factor which predisposed it to thrombosis. Although benign liver tumors or simple cysts can cause IVC thrombosis [3, 4], such extensive growth is rare in benign cysts. In our case, we suspected that the abdominal pain experienced by the patient was caused by hemorrhage of the cyst, and the increasing size leads to IVC obstruction and thrombosis.

To successfully treat the patient while avoiding adverse events, cystectomy was needed while preventing pulmonary embolism. Anticoagulation using heparin successfully treated the IVC thrombosis. Patients with acute (< 14 days) and subacute (15–28 days) presentations who are not at high risk of bleeding might benefit from catheter-directed thrombolysis [9]. Anticoagulation treatment with thrombolysis [10] and filter placement within the infrarenal IVC via a right-sided jugular approach above the distal thrombosis has also been reported [4]. The authors performed cyst aspiration after filtering, and venography showed a rapid increase in venous flow rate without pulmonary embolization.

In conclusion, we experienced a giant, growing, multilobular cyst that caused IVC thrombosis via compression. Prevention of pulmonary thrombosis by heparin followed by cystectomy successfully resolved the symptoms without adverse events.

Compliance with ethical standards

Conflict of interest Naruhiro Kimura, Atsunori Tsuchiya, Masahiro Ogawa, Yusuke Watanabe1 Kazunao Hayashi, Junji Yokoyama, Hajime Umezu, and Shuji Terai declare that they have no conflict of interest.

Human rights All procedures followed have been performed in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments.

Informed consent Informed consent was obtained from all patients for being included in the study.

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