Original Article

Acute acalculous cholecystitis coexisting with spontaneous intra-hepatic bile duct rupture: a rare case

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Abstract: Rare cases of acute acalculous cholecystitis (AAC) coexisting with spontaneous rupture of intra-hepatic bile duct (IHBD) are available. In this case, a male patient aged 81 year-old with a history of diabetes and hypertension was admitted to our hospital due to pain in right upper abdomen. The patient was initially diagnosed with AAC with no signs of peritonitis. Two days after conservative therapy, sharp pain in abdomen was reported together with presence of peritonitis signs. Ultrasonic examination and abdominal paracentesis revealed bile peritonitis. Accumulation of bile and rupture of bile duct at the left liver were revealed after exploratory laparotomy. The patient was finally diagnosed with AAC coexisting with rupture of IHBD. For the treatment, removal of gallbladder and suturing of rupture was performed, followed by T-tube drainage. After discharge, the patient showed no complications or adverse events during the 12-month follow up. For AAC patients concurrent with bile peritonitis, special cares should be taken to identify the rupture of IHBD. Besides surgery, a systematic evaluation of the bile duct is necessary to exclude the possibilities of rupture of IHBD.

Keywords: Acute acalculous cholecystitis, intra-hepatic bile duct, bile peritonitis, simultaneous rupture

Introduction

Acute acalculous cholecystitis (AAC) is usually occurred in critically ill patients following cardiac surgery, abdominal vascular surgery, as well as severe trauma [1]. It is commonly reported in male patients and is associated with the stress, instability of haemodynamics and severe damages [2, 3]. Until now, the pathogenesis of AAC is still not well defined. To our knowledge, several factors have been reported to be related to the AAC, among which gallbladder ischemia and/or cholestasis are considered as the major causes [4, 5]. In clinical practice, the diagnosis of AAC is mainly based on the imaging techniques. Unfortunately, the prognosis of AAC is poor in patients, especially in children, due to the fact that the gastrointestinal function of critically ill patients is not adequate to enteral nutrition [6]. On this condition, early diagnosis and effective management are crucial for the prognosis of AAC.

Rupture of intrahepatic bile duct, also known as non-traumatic perforation of the bile duct, is un-frequently encountered entities that exteriorize the biliary content in the great peritoneal cavity. Patients with AAC usually showed concurrent diseases such as gangrene, perforation and empyema [7]. However, to our best knowledge, no patient with AAC coexisting with intrahepatic bile duct (IHBD) rupture is reported after literature research using the key words “acute acalculous cholecystitis” and “rupture of intrahepatic bile duct” or “intrahepatic bile duct rupture”. Thus, we present a patient with AAC concurrent with rupture of IHBD.

Case presentation

A male patient aged 81 year-old with a history of diabetes and hypertension was admitted to our hospital due to pain in right upper abdomen and chilly-sensation, together with continuous dull pain, vomiting and nausea. No rebound tenderness was noticed in right upper abdomen, and the Murphy’s sign was positive. The patient was diagnosed with AAC according to abdominal ultrasound examination (Figure 1). As no symptoms of peritonitis or other concurrent dis-
orders were observed, the patient received conservative therapy.

However, on the second day after conservative therapy, the patient reported unbearable pain in abdomen with fever, together with shortness of breath and peritonitis. Bed-side ultrasound examination indicated swelling in gallbladder together with edema and thickening in gallbladder wall and hydrops in right upper abdomen. About 5 ml liquid in a jasmine color was extracted through ultrasound-guided abdominal paracentesis. On this basis, AAC combined with bile peritonitis was suspected. Therefore, exploratory laparotomy was performed, during which bile-like ascites with a volume of about 50 mL was observed near the gallbladder. Meanwhile, thickening and edema was noticed in the gallbladder wall, combined with gangrene and condensed bile. A rupture with a size of about 3 mm was observed at the surface of the third segment of liver lobe, through which the bile was exteriorized. No significant dilatation was observed in the common bile duct. Choledochoscope indicated mucous hyperemia or significant swelling in common bile duct, while no calculus or neoplasm was observed within it. The rupture on left liver was confirmed through injecting 20 mL normal saline into common bile duct and observes the overflow of the fluid. On this basis, the gallbladder was removed, and the incision on the left liver was sutured using prolene suture. A T-tube was inserted in the common bile duct, followed by drainage via foramen of Winslow.

The surgery was successful, and the patient was transferred to the ICU department for additional caring. Postoperative pathological report revealed purulent cholecystitis (Figure 2). Upon no significant obstruction in the bile duct or bile leakage was observed, the T tube was removed. No adverse events or postoperative complications were reported by the patient during the 12 months follow up.

**Conclusion**

The exact pathogenesis of AAC combined with rupture of IHBD is still not well defined due to no cases available. In this case, we speculated the development of the AAC and rupture of IHBD may be related to the following aspects: (i). The terminal vessels of hepatic artery responsible for the nurturing of gallbladder were apt to subject to embolism and ischemia that finally resulted in ischemic injury and necrosis in gallbladder wall [4]. The small bile duct at the surface of the left liver was localized at the end of Glisson system, and the nurturing blood vessels shared similarities in the structure with those of the gallbladder. Therefore, rupture of IHBD may simultaneously occur in the presence of instability of haemodynamics with the AAC. (ii). In this case, the patient showed a history of coronary heart disease besides diabetes and hypertension, which were the risk factors for the peripheral angiopathy. In addition, these disorders may lead to coronary atherosclerosis and lesions of capillaries, which would attenuate the resistance of gallbladder and terminal segment of bile duct to the ischemic injuries [8, 9]. (iii). The patient showed significant congestive edema in bile duct wall, which may induce stenosis of bile duct cavity and finally resulted in elevation of the gallbladder wall, combined with thickening of gallbladder wall. The bile in the gallbladder was condensed. No significant calculus was observed.

![Figure 1. Ultrasonic examination revealed significant swelling of gallbladder combined with thickening of gallbladder wall. The bile in the gallbladder was condensed. No significant calculus was observed.](image-url)
biliary tract pressure. High pressure in biliary tract will impair the discharge of bile in the gallbladder, which finally lead to cholestasis. Meanwhile, infection of biliary tract was observed, which may lead to retrograde chemical and bacterial inflammation in the gallbladder. All these would responsible for the pathogenesis of AAC [5]. (iv). Significant increase was induced in the tension of intrahepatic bile duct with the elevation of bile duct pressure. Consequently, the terminal bile duct at the liver surface was dilated and extended. Meanwhile, the inflammation may result in inevitable edema at the bile duct surface, which would increase the fragility of bile duct surface in combination with the embolism and ischemic injury of the nurturing vessels. On this occasion, the sudden change of bile duct pressure may lead to rupture of bile duct surface and resulted in bile peritonitis [10]. Recently, extensive studies revealed AAC was a systemic critical illness other than acute calculous cholecystitis (ACC) which was merely a local disease of the gallbladder. Thus, the pathogenesis of AAC involved several organs, which would explain the simultaneous occurrence of AAC and rupture of IHBD.

Early diagnosis of AAC is of prime importance as the onset of AAC is comparatively faster. Ultrasonic technique and CT have been commonly used for the diagnosis of AAC [5]. In addition, cholescintigraphy plays an important role in the auxiliary examination of AAC [11]. The patient showed significant abdominal pain together with remarkable elevation of body temperature, white blood cells, peptase and bilirubin. Thus, bed-side ultrasound examination was performed, which revealed hydrops in the abdominal cavity. We speculate that it may be related to the perforation of intrahepatic bile duct. Rupture of IHBD is merely confirmed through surgery, and is commonly noticed in patients with bile peritonitis. In this case, the patient was initially diagnosed with AAC after admission, and rupture of IHBD was not confirmed prior to surgical exploration. We want to raise the attention of the surgeons about the fact that, for the patients diagnosed with AAC initially, the bile in the abdominal cavity may leak from the gallbladder induced by and/or the bile duct perforation. Therefore, in a previous study, a systematic evaluation of the bile duct is necessary for the patients with bile peritonitis [10].

To date, the treatment of AAC is still controversial. For the aged patients that were unfit for the surgery, cholecystostomy is preferred [12]. Nevertheless, cholecystostomy is considered to offer no survival benefit for AAC patients with sepsis and shock [13]. For the patients tolerated to surgery, removal of gallbladder and abdominal drainage are considered to be effective based on our experiences. For the patients with simultaneous rupture of IHBD, suturing and T-tube drainage were performed. In this case, we present an 81 year-old patient diagnosed with AAC and rupture of IHBD. The patient received removal of gallbladder, and the incision on the left liver was sutured using Prolene suture. Finally, the patient’s outcome was satisfactory and no complications were reported during the 12-month follow up.
In conclusion, AAC concurrent with rupture of IHBD may be induced by senior age, hypertension, DM, cardiovascular diseases, as well as bile duct infection and elevation of bile duct pressure. For the patients with AAC concurrent with bile peritonitis, special cares should be taken to the rupture of IHBD. Besides surgery, a systematic evaluation of the bile duct is necessary to exclude the possibilities of rupture of IHBD.

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Written informed consent was obtained from the patient for publication of this Case report and any accompanying images. A copy of the written consent is available for review by the Editor of this journal.

Disclosure of conflict of interest

None.

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