

Coexistence of double gallbladder with cholangiocarcinoma

A case report

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Abstract

Rationale: Gallbladder duplication is a rare congenital disorder, which could cause an increasing risk of complications during surgery. The coexistence of cholangiocarcinoma with double gallbladder is extremely rare, which might lead to an even higher possibility of misdiagnosis and postsurgery complications.

Patient concerns: A 58-year-old female was presented with abdominal pain and jaundice. Abdominal ultrasonography showed duplication of gallbladder, one of which with a thickened wall and a rough surface. This was also confirmed by an abdominal computed tomography (CT), magnetic resonance imaging (MRI) and magnetic resonance cholangiopancreatography (MRCP) scan. During the surgery, we found a tumor inside one bile duct. The postsurgery pathology showed adenosquamous carcinoma.

Diagnoses: Gallbladder duplication, cholangiocarcinoma.

Interventions: The tumor was removed by surgery.

Outcomes: The patient died of tumor relapse six months after surgery.

Lessons: This is the first reported case with coexistence of gallbladder duplication and cholangiocarcinoma, which was diagnosed by abdominal ultrasound, CT and MRCP, as well as further confirmed in surgery and pathology. This case emphasized the importance of a thorough examination of gallbladder before surgery, especially in those cases with suspected double gallbladder, since each gallbladder could have the possibility of an independent cholangiocarcinoma.

Abbreviations: CT = computed tomography, D-BIL = direct bilirubin, IBIL = indirect bilirubin, MRCP = magnetic resonance cholangiopancreatography, MRI = magnetic resonance imaging, TBil = total bilirubin.

Keywords: cancer, duplication of gallbladder, ultrasound

1. Introduction

Duplication of gallbladder is an extremely rare congenital abnormality, which is easy to be ignored in ultrasonography, posing a great risk for surgical removal of gallbladder stones, which might lead to complications such as bile duct injury or leakage.^[1–3] There has been no report of the coexistence of double gallbladder and bile duct cancer till now, which make it easier for clinicians to neglect the possible carcinoma in each single gallbladder.

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Here, we report the first case with coexistence of duplication of gallbladder and cholangiocarcinoma, which was diagnosed by ultrasound, CT, MRCP, as well as surgery and pathology. These findings gave us a warning that in case of suspected double gallbladder, each gallbladder should be carefully examined.

2. Case report

A 58-year old Chinese woman with no medical history was admitted to the hospital because of 2 months' right upper quadrant pain and 1 week jaundice. Physical examination showed the vital signs were normal. The pulse was 70 beats/min, blood pressure was 116/69 mm Hg, temperature was 36.6°C, and respirations were 18 breaths/min. In general, she was weak.

Lab studies showed TBil (total bilirubin) was 97.8 μmol/L, D-BIL (direct bilirubin) was 79.7 μmol/L, IBIL (indirect bilirubin) was 18.06 μmol/L. Tenderness was found in the right upper quadrant. Murphy sign was negative.

Ultrasonography showed that the morphology and size of the liver is normal. Liver parenchymal echoed evenly. The common bile duct and intrahepatic bile duct were dilated. There is an irregularly hypoechoic mass with a size of 1.8 cm × 1.3 cm at the end of the common bile duct (Fig. 1). The hepatic portal veins are also in the normal range. Two separated ellipsoid-shaped cystic structures with well-defined wall, side by side in the gallbladder fossa were detected in the examination. The 2 gallbladders shared the same common bile duct. One of the necks of the gallbladder was unevenly thickened, with a hypoechoic mass in the cavity (the

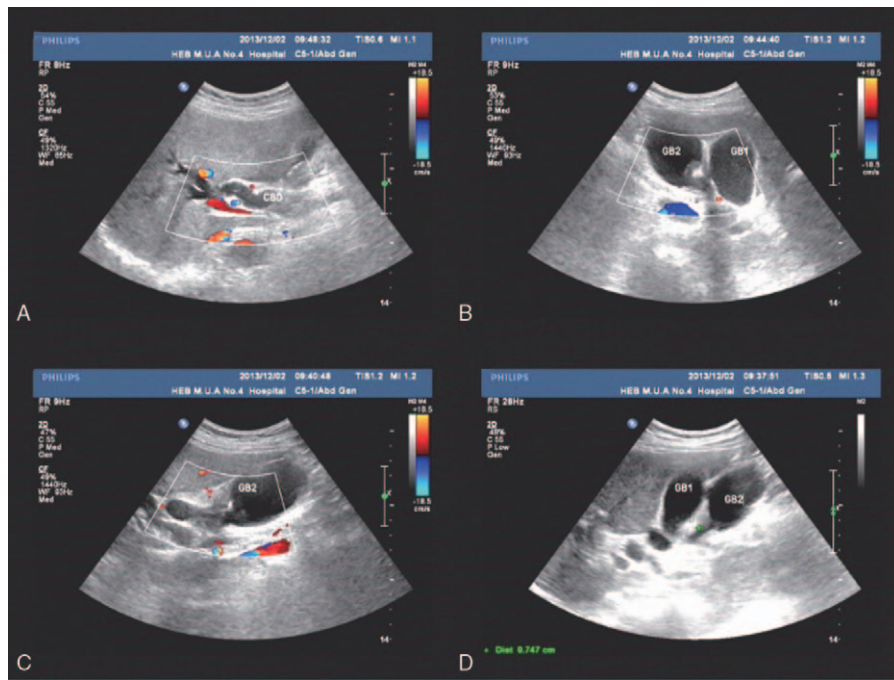


Figure 1. Ultrasound manifestation of the patient. (A–C) Ultrasound results showed that the morphology of the liver is normal. The common bile duct and intrahepatic bile duct are dilated. There is a low echo nodule with a lesion at the end of the common bile duct. (D) The “+” points out the location of the tumor.

size is 2.3 cm × 1.3 cm). The patient was diagnosed as duplication of gallbladder and cholangiocarcinoma. Abdominal computed tomography (CT) and magnetic resonance cholangiopancreatography (MRCP) also confirmed the true duplication of gallbladder and the tissue mass in the gallbladder.

During surgery, we could find double gallbladders. One tumor was located in the common bile ducts, with a diameter of 1.2 cm, which congested the bile duct. The other tumor was in the neck of 1 gallbladder, with a size of 2.5 cm × 2.0 cm × 1.0 cm. The pathology showed adenosquamous carcinoma (Fig. 2).

Patient informed consent was signed up for the publication of the case. The ethics committee of Hebei Medical University Fourth Affiliated Hospital and Hebei Provincial Tumor Hospital review board approved this study.

3. Discussion

To our knowledge, this is the first case report of the coexistence of double gallbladder and cholangiocarcinoma. The congenital abnormality of gallbladder is not uncommon; however, the

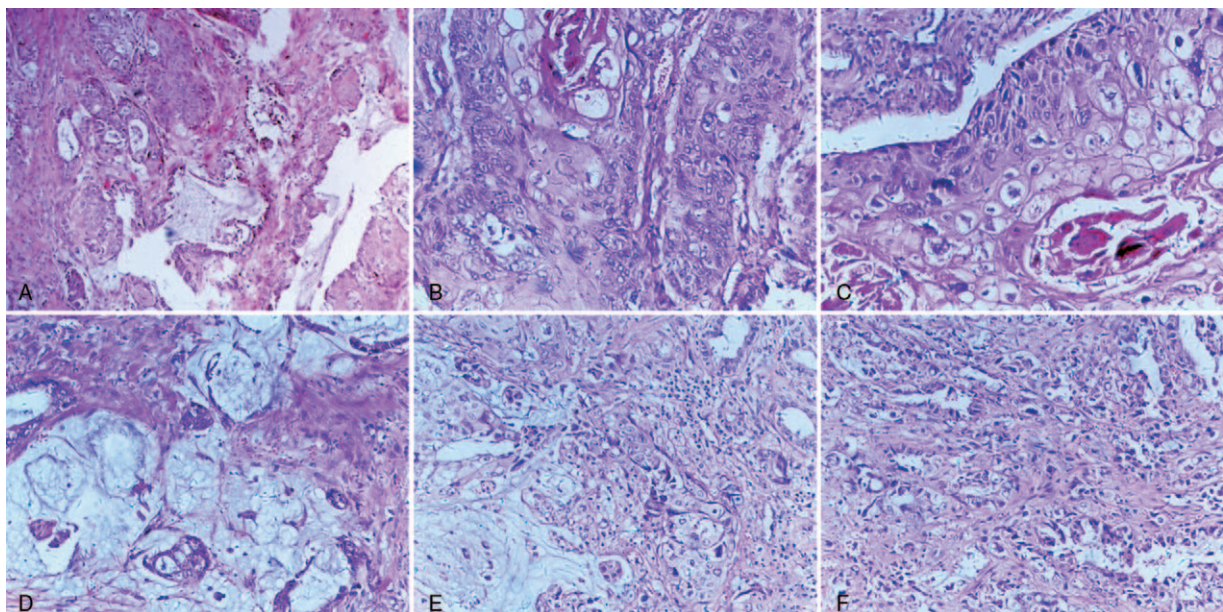


Figure 2. Pathology of the patient. (A, B, D) The pathology showed moderately differentiated adenocarcinoma. Granular glands distributed with a disorganized feature. The cavity was filled with mucus. (C, E, F) The pathology showed a squamous cell carcinoma, with evident keratin pearl.

number variation is rare, which were mostly found during health screening. Duplication of gallbladder is a rare congenital disorder, with an estimated incidence of 1/4000.^[4,5] It has not been reported that cholangiocarcinoma could coexist with duplication of gallbladder.

The anatomic variations of double gallbladder were classified according to the way that bile duct merged into the biliary system.^[6] Type I, or split primordial group, which means the 2 gallbladders shared the same bile duct, but divided by a septum in the bile duct. Type II indicates that each gallbladder arises from separate primordium from the biliary tree and has individual cystic duct for each gallbladder. However, in case that the septum is not consistent, or even missing from the neck of the gallbladder, is not considered as double gallbladder. Duplication of gallbladder is also different from diverticulum of gallbladder, which often showed a local outpouching of the entire gallbladder wall with a narrow neck.

Primary carcinoma of the gallbladder is the most common malignant tumor in biliary system, especially in women.^[7] The incidence of cholangiocarcinoma is increasing. Cholangiocarcinoma is asymptomatic at its early stage, which leads to its late diagnosis and poor prognosis. The tumor is often unresectable when diagnosed and rarely diagnosed preoperatively despite patients' symptoms. Early diagnosis can improve the clinical outcome and cure rate of gallbladder carcinoma. The most common pathological type of cholangiocarcinoma was adenocarcinoma (8%), followed by anaplastic carcinoma (4.6%), squamous cell carcinoma (4.4%), and acinar carcinoma (1.6%).^[8] Ultrasound is the most commonly used imaging modality for detecting early stage bile duct carcinoma, with a sensitivity of 80%. Signs suggesting the presence of bile duct carcinoma includes bile duct wall thickening, single or multiple

intraluminal mass, or polyps larger than 1 cm in diameter.^[8] Color Doppler often provide information on the position between tumor and the hepatic portal vein or hepatic artery, which makes it easier for assessing the potential value for surgeries.

Author contributions

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Data curation: Wei Chen.

Funding acquisition: Wei Chen.

Investigation: Ruoling Han.

Methodology: Wei Chen.

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