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Imaging Findings of Mucinous Cystadenoma of Cystic Duct With Biliary Obstruction: A Case Report

Jian Liu and Zhiqun Mao*

Radiology Department,the First Affiliated Hospital of Hunan Normal University (Hunan Provincial People's Hospital) No 61, Jiefang West Road, Furong District, Changsha 410005, Hunan, China.

***Correspondence:**
Zhiqun Mao, Radiology Department, the First Affiliated Hospital of Hunan Normal University (Hunan Provincial People's Hospital) No 61, Jiefang West Road, Furong District, Changsha 410005, Hunan, China.

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ABSTRACT

Mucinous cystadenoma of the gallbladder account for 0.02% of the total number of cases in the hepatobiliary system. By searching the literature from 1900 to 2020, we found 16 cases of mucinous cystadenoma of the gallbladder. The first report of mucinous cystadenoma of the gallbladder was published in 1901 by Bishop. There are only 3 cases of bile duct obstruction caused by gallbladder mucinous cystadenoma extending into bile duct [1]. Meanwhile, the previous literature reports appear lack of complete imaging data. We report a rare case of mucinous cystadenoma of the cystic duct extending into the common bile duct causing biliary obstruction. It is the first case that describes the imaging findings of the disease in detail.

Keywords

Mucinous cystadenoma of cystic duct, Biliary obstruction, Ultrasound, Magnetic resonance imaging, Cystic lesions of the cystic duct.

Case Report

A 57-year-old woman with nausea and fatigue for more than one month was admitted to the hospital due to sclera and generalized jaundice. The patient had a history of hepatitis B virus infection. Physical examination revealed mild yellowing of skin and sclera throughout the body. Liver function tests were abnormal with a total bilirubin of 143.9umol/l (normal:5.1-20.0umol/l), a direct bilirubin of 109.6umol/l (normal: 0-6.1umol/l), a indirect bilirubin of 34.30umol/l (normal : 5.1-20.0umol/l), aspartate aminotransferase (ASAT) of 87.7U/L (normal: 7-40U/L), alanine aminotransferase (ALAT) of 78.25U/L (normal: 13-35U/L),a GGT of 682.0U/L (normal: 7-45U/L), and alkaline phosphatase of 201U/L (normal 35-100U/L). Virus serum examination showed increased hepatitis B virus surface antigen 1.45 (IU/ML), increased hepatitis B virus e antibody positive 0.02 (COI), positive hepatitis B virus core antibody 12.59 (COI). There were no positive results for tumor markers CA 19-9, CEA and AFP. After admission, the patient completed abdominal ultrasound, computed tomography (CT) scan and magnetic resonance imaging (MRI) /magnetic resonance cholangiopancreatography (MRCP) examinations. Then percutaneous transhepatic cholangial drainage (PTCD) was completed.

Abdominal ultrasound showed intrahepatic and extrahepatic bile duct dilation. A hypoechoic circular shape with a size of about 25mm * 15mm can be seen at the junction of the upper segment of the common bile duct(CBD) and the cystic duct, in which hyperechoic internal separation can be seen. Another hypoechoic circular shape lesion with a size of 26mm * 17mm is seen at the upper segment of the common bile duct. There is a clear margin with a hyperechoic capsule and gallstone (Figure 1). Contrast-enhanced ultrasonography demonstrated that the walls of means lesion showed the same enhancement as the adjacent bile duct walls in arterial phase, portal phase and delayed phase, and the contents were not enhanced.

Abdominal MRI and MRCP showed that intrahepatic and extrahepatic bile duct were significantly dilated, and there was a cystic tumor with a diameter of about 40 mm*20mm at the junction of the CBD and the cystic duct. The coronal T2 image showed that cystic tumors in the common bile duct was continuous with the

cystic ducts lesion. The cystic mass showed T1 hypointense and T2 hyperintense with thin hypointense capsule and hypointense intrinsic septas were visible. T1 postcontrast by Gd-EOB-DTPA showed a slight enhancement of the capsule. There was no contrast agent in the intrahepatic and extrahepatic bile ducts 20 minutes after contrast injection, and no contrast agent was shown in the intrahepatic and extrahepatic bile ducts until hepatobiliary phase after 40 minutes.

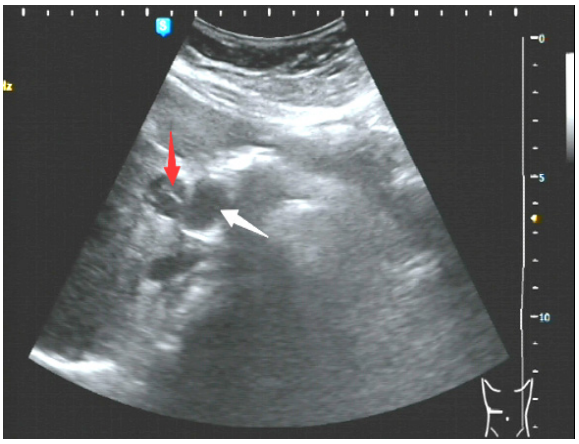

Figure 1: Ultrasound images. A hypoechoic circular shape of the cystic lesion can be seen at the junction of the CBD (white arrow) and the cystic duct with hyperechoic internal separation (red arrow).

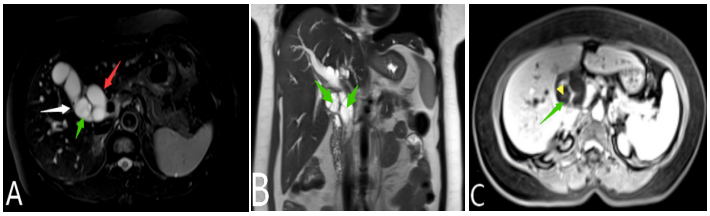

Figure 2: Magnetic resonance images. A, The axial T2 weighted imaging demonstrates the hyperintensity cystic lesion (white arrow) from the cystic duct extends into the CBD (red arrow). There is a thin hypointense capsule and intrinsic septas (green arrow) on T2WI. B. On the coronal T2 weighted imaging, the cystic tumor of the common bile duct is continuous with the lesions of the cystic duct. The capsule of the cystic mass is thin, and the low signal intensity can be seen in the internal septum (green arrow). C, The axial T1 weighted imaging postcontrast by Gd-EOB-DTPA shows the cystic duct is enlarged (green arrow), and the internal separation of the lesions in the cystic duct is slightly enhanced (yellow arrowhead).

Enhanced CT scan showed dilation of the bile ducts and significantly enlarged cystic duct that the diameter was 18 mm. There was almost no enhancement in the cystic duct area. There were many speckled hyperintense shadows at the bottom of the gallbladder, suggesting gallstones (not shown in the picture) (Figure 3a-b).

Cholangiogram (by PTCD) showed that there was a filling defect of 17mm * 30mm in size at the junction of the upper part of the common bile duct and the cystic duct. The intrahepatic and extrahepatic bile ducts were significantly dilated, and the gallbladder was not visualized (Figure 4).

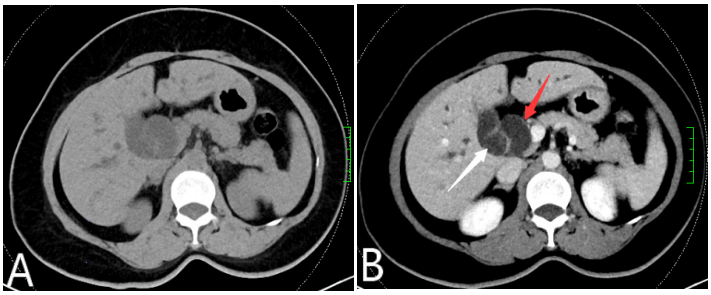

Figure 3: Computed tomography images. A, Non-enhanced axial abdominal CT showed low density fo ci in the cystic duct area. B, Contrast axial abdominal CT showed significant enlargement of the confluence of the cystic duct (white arrow), extended to the common bile duct (red arrow), There is minimum enhancement in the cystic duct area, and dilatation of the intrahepatic bile duct.

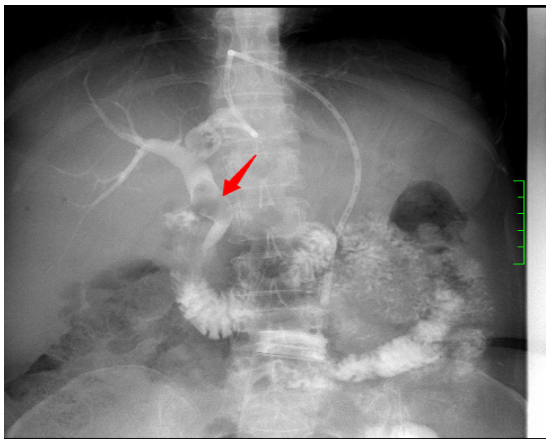

Figure 4: Cholangiogram.
On PTCD, a filling defect (arrow) is seen at the junction of the upper segment of the common bile duct and the cystic duct. The gallbladder cannot be shown due to biliary obstruction.

Later, the patient underwent surgical treatment. Cholecystectomy combined with cholangioplasty and T-tube drainage were performed by laparotomy. During the operation, a cystic hard mass was found at the junction of the cystic duct and the com) mon bile duct and the mass extended from the cystic duct into the common bile duct. The pedicle of the mass was at the cystic duct with a wide basal shape and the mass was about 4cm * 2cm * 2cm. The capsule of the mass is clear (Figure 5). The gross specimen showed that it was a multicystic mass and the cysts contained light yellow clear liquid.

Histological examination showed that the short fusiform cells in the lower layer of epithelial cells were ovarian-like mesenchymal stromal cells with mild atypical hyperplasia (Figure 6a). Immunohistochemistry showed CK7 (+), CK19 (Epithelial+), ER (+), PR (+), p53 (-), Ki67 (Scattered+) and was confirmed a cystic duct mucinous cystadenoma (Figure 6 b-c). The patient had fast recovery and was discharged from hospital on the 6th after the operation. As of this writing, three months after surgery, the patient showed no evidence of recurrence during follow-up.

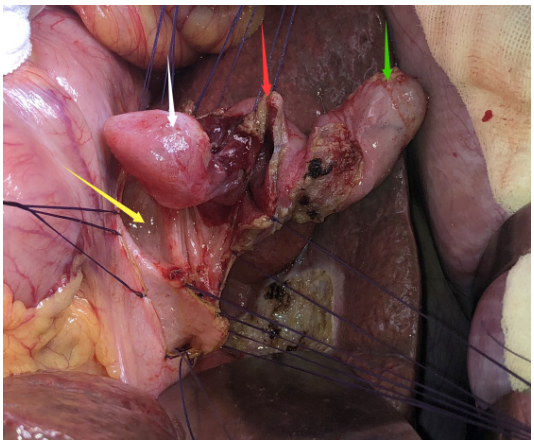

Figure 5: During the operation, a cystic hard mass (white arrow) could be found at the junction of the cystic duct (red arrow) and the common bile duct (yellow arrow) and the mass extends out from the cystic duct to the common bile duct. The gallbladder is seen (green arrow).

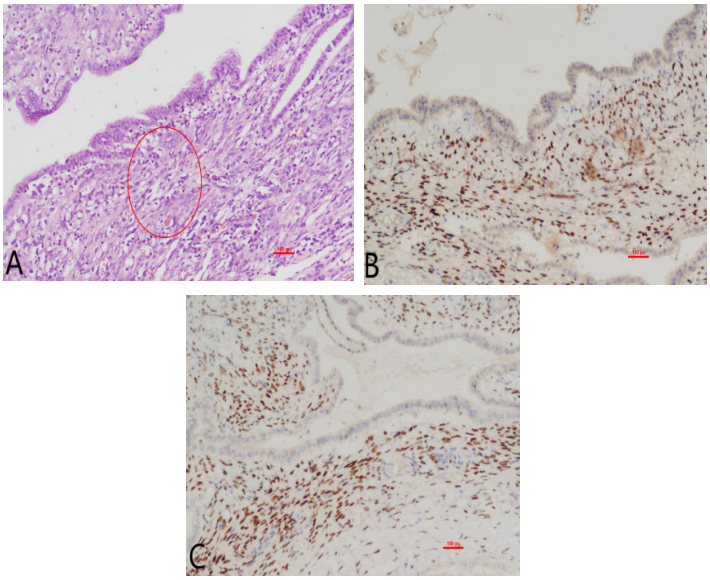

Figure 6: Ovarian-like mesenchymal stromal cells. A, Histology by spindle cells in the subepithelial layer(circle). B, Immunohistochemical images shows positive staining for PR (+). C, Immunohistochemical images shows positive staining for ER (+). PR and ER are specific biomarkers for diagnosing ovarian-like mesenchymal stromal cells.

Discussion

Hepatobiliary cystadenomas are most commonly seen in the liver (83%), followed by the extrahepatic biliary system (13%) and gallbladder (0.02%) [2]. It affects women more frequently, with a mean age at presentation of 45 years [3]. Symptoms vary depending on the location of the tumor, but it typically presents as acute or chronic right upper quadrant pain, epigastric pain, and nausea and vomiting [1]. Patients rarely present with jaundice; the size of cystadenoma lesions varies from 2 to 40 cm [4]. The mass in this case is 4*2*2cm, mainly manifested as whole body jaundice and nausea and fatigue. After reviewing literatures, we found that 16 cases of mucinous cystadenoma of gallbladder have been reported before, while only 3 cases of biliary obstruction caused by mucinous cystadenoma of gallbladder have been reported [1], and all of them lack detail radiological data. We report a case of mucinous cystadenoma of the cystic duct that extends into the CBD resulted in obstruction of bile duct. The patient has completed radiological examinations including ultrasound, contrast-enhanced ultrasonography, CT scan, MRI/MRCP, PTCD, which is of great value for clinicians to understand the radiological data of the disease and to make a correct diagnosis.

Mucinous cystadenomas are benign tumors with potential for malignant transformation [5,6]. The multilocular form is more common than unilocular. This case is a multilocular mass. The cystic lesions can be filled with serous, hemorrhagic, mucinous, or mixed fluids [1]. Histologically, there are three subtypes of mucinous cystadenoma: ovarian-like mesenchymal stroma occurring most often in middle-aged women, nonovarianlike mesenchymal stroma, which is most common in men and has a fibrous myxoid appearance, and eosinophilic subtype, a rare lesion lined with eosinophilic epithelial cells described as a premalignant variant [5,7]. This case belongs to ovarian-like mesenchymal stroma subtype.

Different radiological modalities reveal characteristic appearances of mucinous cystadenoma of cystic duct that is helpful to guide this diagnosis and treatment. Ultrasound is a useful diagnostic modality to demonstrate hepatic cystadenomas, particularly in definition of internal morphology [8,9]. Ultrasound appearances of biliary cystadenoma typically include well-demarcated, thick-walled, noncalcified, anechoic or hypoechoic, globular or ovoid, cystic mass with internal septations [10-12]. Meanwhile, ultrasound has been used to define the malignant potential of the lesion. When the lesion is composed of cysts with simply septation, the lesion is more likely to be cystadenoma, whereas septation together with mural nodules may indicate the presence of cystadenocarcinoma [13]. In this case, contrast-enhanced ultrasound was also completed. Combined with the results of contrast-enhanced ultrasound, the location and size of cystic lesions in the biliary tract can be preliminarily estimated. Therefore, ultrasound has certain advantages in the diagnosis of cystadenoma, but it is easy to be affected by subjective factors of personal operation.

It has been mentioned in the literature that contrast-enhanced CT with arterial and portal phase provides added surgical benefit in operative planning to determine size, morphology and anatomical relationship to adjacent structures, especially the blood vessels [14]. On CT scanning, biliary cystadenomas appear as a solitary cystic mass with a well-defined thick fibrous capsule, with mural nodules, internal septa causing multiloculation and, rarely, capsular calcification [15]. Common features of biliary cystadenomas on CT include hypointensity, well-defined, thick-walled and multilocular [16,17]. However, in this case, the CT scan showed a limited about the characteristics of cystic adenoma of cystic duct and almost no septum or cyst wall can be found. Only for the dilatation of cystic duct and CBD. Even it hardly enhanced, and it was difficult to distinguish the cystic mass by contrast CT. Therefore, when the capsule of the cystic mass is very thin, the CT

scan may not see the signs of the cystic mass. So the CT has certain limitations for the evaluation of cystic masses. MRCP is useful to establish the diagnosis and extent of hepatobiliary cystadenomas and cystadenocarcinomas [18]. Typically, on magnetic resonance imaging (MRI), biliary cystadenomas have a homogenous low signal on T1-weighted imaging and pronounce homogenous high signal on T2-weighted imaging [19]. In this case, MRI showed that the bile ducts and cystic duct were significantly dilated. On T1-weighted imaging, the tumor showed a uniform hypointensity and hyperintensity on T2-weighted imaging (T2WI). The capsule and septum of tumor could be easy seen on T2WI. Both the internal septum and capsule were hypointensity on T2WI and it can be diagnosed as cystadenoma; therefore, MRI is more advantageous for the diagnosis of cystic masses. However, MRI cannot distinguish mucinous cystic lesions from serous cystic lesions. Hepatobiliary specific contrast agent such as Gd-EOB-DTPA may show the hepatobiliary system very well, but for the patient with severe biliary obstruction, the application value is limited. Cholangiogram may shows filling defect of the biliary duct. In this case, the contrast agent could not enter the cystic duct and gallbladder because the cystic mass blocked the cystic duct. The mass of the cystic duct could not be displayed.

In summary, Mucinous cystadenoma of cystic duct is a rare disease, and rarely cause biliary obstruction. Ultrasound and magnetic resonance imaging have advantages in the diagnosis of the cystadenoma of bile duct. However, it is still difficult to distinguish between serous cystic lesions and mucinous cystic lesions. In view of the malignant potential and the possibility of recurrence of cystic duct mucinous cystadenoma, it is necessary to carry out a thorough surgical operation to remove the mass in time to prevent more invasive lesions and recurrence. In addition, close postoperative follow-up is also crucial.

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