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# Adenomyoma of the distal common bile duct demonstrated by endoscopic ultrasound: A case report and review of the literature

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**Author contributions:** Xu LM was responsible for interpretation of the clinical data and drafting of the manuscript; Hu DM conceived the case report and revised the manuscript; Tang W revised the manuscript for important intellectual content; Wei SH and Chen W were in charge of the operation; Chen GQ provided the figures showing the computed tomography and magnetic resonance imaging findings.

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## Abstract

### BACKGROUND

Adenomyomatous hyperplasia of the distal common bile duct (CBD) is very rare, with only scarce case reports in the literature. Diagnosis is usually based on imaging findings, and endoscopic biopsy is very difficult before operation. It is believed that adenomyomatous hyperplasia has little or no risk of malignant transformation.

### CASE SUMMARY

A 68-year-old woman with abdominal pain in the right upper quadrant was referred to our hospital. Abdominal ultrasonography in the emergency ward revealed acute cholecystitis and dilated CBD. Laboratory findings showed elevated levels of transaminases, phosphatase, and  $\gamma$ -glutamyltranspeptidase. Pharmaceutical treatment for 3 d did not relieve the symptoms. Magnetic resonance cholangiopancreatography (MRCP) and computed tomography (CT) showed proximal bile duct dilatation but could not identify the cause. Endoscopic ultrasonography (EUS) demonstrated a mixed echogenic mass in the distal CBD. During surgery, a firm mass was found in the distal CBD and the Whipple procedure was performed with the initial concern of malignancy. Histology showed diffuse adenomyomatous hyperplasia.

### CONCLUSION

EUS may be a useful choice to diagnose adenomyoma of the distal CBD before operation, especially in patients with ambiguous MRCP/CT findings.



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**Core tip:** The distal common bile duct is an extremely rare site of adenomyomatous hyperplasia. Diagnosis is usually based on imaging findings, and endoscopic biopsy is difficult before operation. We present here a rare case of adenomyomatous hyperplasia of the distal common bile duct demonstrated by endoscopic ultrasound, which revealed a nodular change and bile duct wall thickening. We concluded that the mass was a benign, non-neoplastic lesion. This case highlights how endoscopic ultrasound may be a useful choice for the diagnosis of adenomyoma of the distal common bile duct, especially in patients with ambiguous magnetic resonance cholangiopancreatography/computed tomography findings.

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## INTRODUCTION

Most of adenomyomas are located in the gallbladder, stomach, duodenum, and jejunum<sup>[1-5]</sup>. The distal common bile duct (CBD) is an extremely rare site of adenomyomatous hyperplasia<sup>[1,5,6]</sup>, and here we report here our experience with such a case. For our case, histology demonstrated glandular structures that were surrounded by a fibroblastic or myofibroblastic proliferation. Reported symptoms for these rare cases are nonspecific and include jaundice, abdominal pain, nausea, vomiting, dysphagia, and unintentional weight loss<sup>[1,3,7]</sup>. A dilated CBD is common and sometimes presents intermittently in the adenomyoma of the Vaterian system<sup>[1,3]</sup>. It can be very difficult to distinguish an adenomyoma from a malignancy before operation; this is a valid concern as adenomyomas have little or no risk of malignant transformation<sup>[8-10]</sup>.

## CASE PRESENTATION

### Chief complaints

A 68-year-old woman with abdominal pain located in the right upper quadrant was referred to our hospital. Abdominal ultrasonography (US) performed in the emergency ward revealed stones in the gallbladder, with acute cholecystitis and dilated CBD.

### History of present illness

The patient's symptoms had begun 5 h prior to presentation at the hospital. The patient reported no vomiting or fever. Upon hospital admission, the initial treatment with antibiotics and anticholinergic did not relieve the symptoms.

### History of past illness

The patient had a history of hypertension and appendectomy. She was allergic to penicillin.

### Personal and family history

The patient had no habits of tobacco or alcohol intake. There were no risk factors for common diseases in the family history.

### Physical examination upon admission

On admission, the patient's temperature was 36.5 °C, heart rate was 85 beats per min, respiratory rate was 18 breaths per min, and blood pressure was 120/70 mmHg. Routine abdominal examination revealed tenderness and rebound tenderness in the

right upper quadrant. There was no shifting dullness. Normal active intestinal sounds were heard. There was no jaundice of the sclera or skin. There were no significant findings from palpation of the lymph nodes and no edema. Lung and heart auscultation was negative.

### Laboratory examination

Laboratory tests were conducted and the results were as follows: White blood cell count,  $5.7 \times 10^3/\mu\text{L}$ ; neutrophil count,  $4.7 \times 10^3/\mu\text{L}$ ; hemoglobin, 12.7 g/dL; platelet count,  $182 \times 10^3/\mu\text{L}$ ; total bilirubin/direct bilirubin, 18.7/9.5  $\mu\text{mol/L}$ ; aspartate aminotransferase/alanine aminotransferase, 540/482 U/L; alkaline phosphatase/ $\gamma$ -glutamyltranspeptidase, 111/175 U/L; amylase/lipase, 54/34 U/L; C-reactive protein 58.8 mg/L; carcinoembryonic antigen, 2.03 ng/mL; carbohydrate antigen 19-9, 76.11 U/mL; and carbohydrate antigen 50, 30.46 IU/mL. Hepatitis tests showed positivity for hepatitis B surface, e, and core antibodies. Symptoms were not relieved after 3 d of pharmaceutical treatments (reductive glutathione at 2.4 qdivgtt; ceftizoxime at 2.0 bid ivgtt). Laboratory findings showed decreased levels of transaminases (192/103 U/L) and elevated levels of phosphatase (203 U/L) and  $\gamma$ -glutamyltranspeptidase (496 U/L).

### Imaging examinations

Magnetic resonance cholangiopancreatography (MRCP) showed proximal bile duct dilatation, with the diameter being 17.5 mm (Figure 1A). 128-row multi-detector computed tomography (CT) and magnetic resonance imaging of the abdomen were consistent, showing diffused dilatation of the extra-hepatic bile duct and significantly enhanced bile duct wall (Figure 1B). Endoscopic US (EUS) was performed for the evaluation of distal CBD obstruction, which demonstrated a mixed echogenic structure (low-mild amplitude echoes, 7.1 mm  $\times$  6.6 mm) in the distal CBD (Figure 2).

## FINAL DIAGNOSIS

The final diagnosis prior to surgery was neoplasm of the distal CBD.

## TREATMENT

In accordance with the diagnosis, the patient agreed to undergo surgery. During the operation, a 7 mm  $\times$  7 mm sized, firm mass was found in the distal CBD near the ampulla of Vater. The surgeon performed radical pancreaticoduodenectomy by the Whipple procedure and cholecystectomy, having concern of a malignant tumor.

## OUTCOME AND FOLLOW-UP

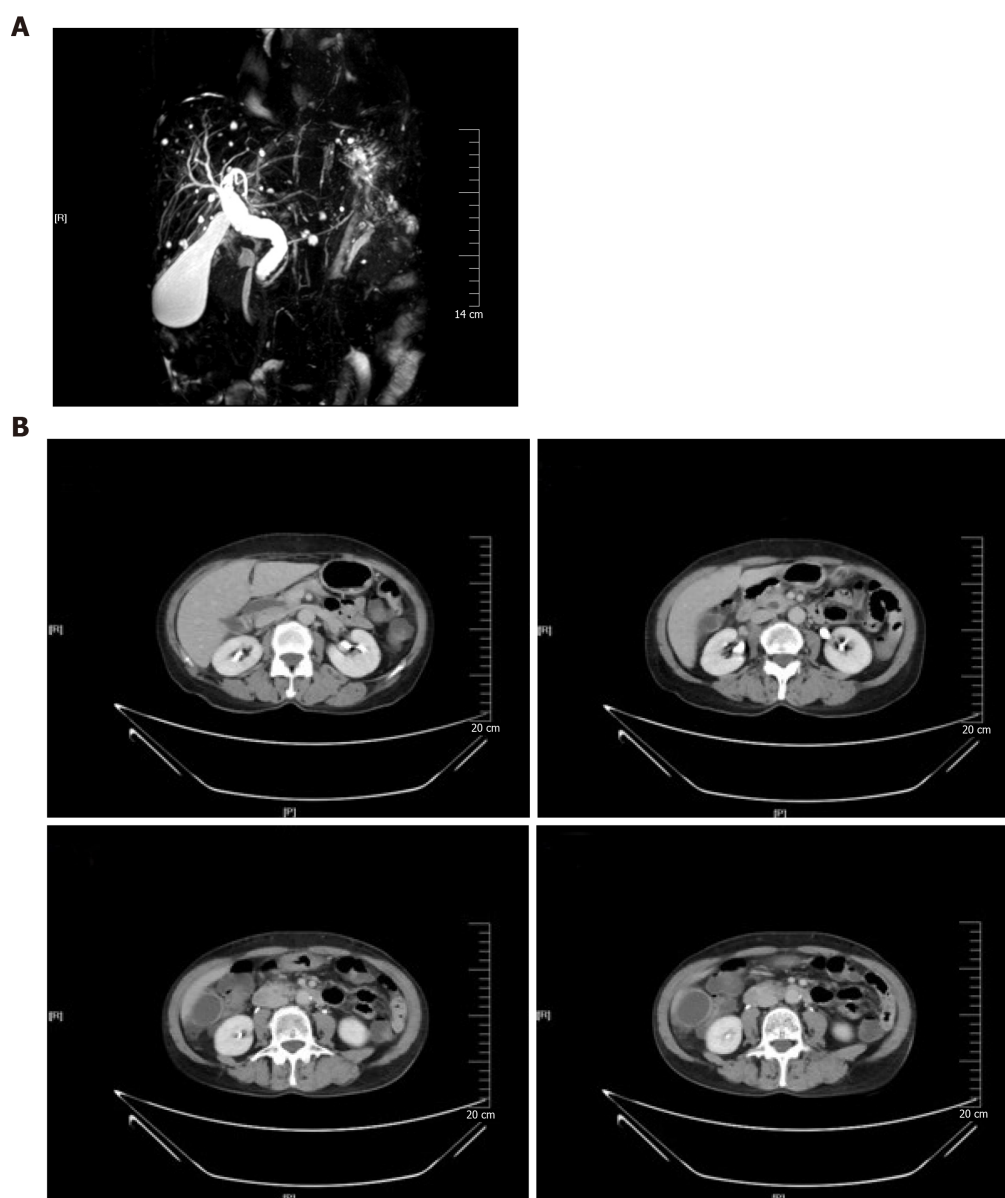
The gross specimen appeared as an irregular mass, measuring 1 cm and having an obscure boundary (Figure 3). For resection, the tumor was separated from the surrounding duodenum and pancreas. Histology of the specimen showed diffuse adenomyomatous hyperplasia of the distal CBD and acute cholecystitis. All the regional lymph nodes showed reactive hyperplasia. No evidence of vascular or perineural neoplastic invasion was observed (Figure 4). The diagnosis of adenomyoma was finally confirmed and no adjuvant therapy was needed.

## DISCUSSION

The overall accuracy for preoperative histopathological diagnosis is 62% for tumors in the papilla of Vater<sup>[11]</sup>. Diagnosis of adenomyomas of the CBD is usually based on imaging findings and endoscopic biopsy is difficult<sup>[5]</sup>. CBD dilation is demonstrated by US, CT, and MRCP in the cases of obstructive jaundice<sup>[12,13]</sup>. Although noninvasive and inexpensive, US-provided diagnosis can be operator-dependent (according to an operator's experience). CT has a lower sensitivity and accuracy in differentiating adenomyomatosis and gallbladder cancer<sup>[14]</sup>.

Endoscopic retrograde cholangiopancreatography (ERCP), another imaging option, is considered the gold standard for the diagnosis of distal CBD abnormalities<sup>[15-17]</sup>. However, it may induce severe complications such as post-ERCP pancreatitis<sup>[18]</sup>. MRCP and EUS are less invasive and useful in diagnosing malignancy and choledocholithiasis in the dilated biliary tree<sup>[19,20]</sup>. Studies have shown that MRCP and





**Figure 1** Magnetic resonance cholangiopancreatography and computed tomography images from our case. A: Magnetic resonance cholangiopancreatography image showing proximal bile duct dilatation but ambiguous findings for evaluation of the distal common bile duct; B: Computed tomography images showing diffused dilatation of the extra-hepatic bile duct and significantly enhanced bile duct wall.

EUS are comparable<sup>[20,21]</sup>. When MRCP findings are negative in the presence of dilated CBD, EUS provides better visualization of the biliary obstruction because its transducer is close enough to the CBD in the duodenum. Patel *et al*<sup>[23]</sup> preferred EUS rather than ERCP as the initial investigation in patients with a probability of choledocholithiasis. Endosonography may also be useful in the evaluation of cholangiocarcinoma<sup>[24]</sup>. Chon *et al*<sup>[25]</sup> considered EUS a key method for investigating biliary disorders of unknown reason.

In our patient, EUS revealed that the lesion was a nodular change with mixed echo, with the bile duct wall being thickened and enhanced. We concluded that it was a benign, non-neoplastic lesion but one not reported in the literature according to the best of our knowledge. There have been no long-term studies reported with large sample size for adenomyomatous hyperplasia of the CBD. The prognosis of the lesion is expected to be similar to that of the gallbladder, based upon the histological similarity<sup>[9]</sup>. EUS shows adenomyoma of the gallbladder with hyper and mixed echogenicity and the well-preserved multiple-layer pattern of the wall<sup>[26]</sup>, which is similar to the findings in our patient's mass. We regret that we did not perform fine needle aspiration (FNA) and then ERCP. The sensitivity of EUS-FNA for diagnosis of malignancy is 66%<sup>[27]</sup>. The sensitivity of ERCP with brush cytology and intra-ductal biopsy is 45% and 48.1%, respectively<sup>[27]</sup>. De Moura *et al*<sup>[28]</sup> reported that EUS-FNA was superior to ERCP with brush cytology for diagnosing malignant biliary strictures. If



**Figure 2** Endoscopic ultrasound image demonstrating a mixed echogenic structure in the distal common bile duct.

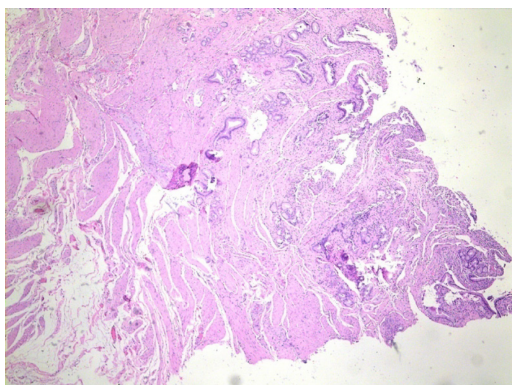
such was diagnosed definitively as adenomyoma, endoscopic treatments like drainage or local resection should be performed, as they carry less risk for complications than the radical surgical procedure<sup>[5,6]</sup>. Meanwhile, the method of EUS-guided biliary drainage might be a primary alternative in patients with malignant obstruction<sup>[29,30]</sup>.

## CONCLUSION

EUS is an important diagnostic modality that can help establish the diagnosis of adenomyomatous hyperplasia in patients with dilated CBD and ambiguous MRCP findings. We expect that the EUS image can be a useful choice to diagnose adenomyoma of the distal CBD. Moreover, EUS-FNA biopsy could be taken from the adenomyoma for further histopathological examination.



**Figure 3** The gross specimen appearance of an irregular mass of the distal common bile duct near the papilla.



**Figure 4** Histological findings from our case. The histological features of the resected mass are hyperplastic glandular lobules were surrounded by hyperplastic muscle fibers, fibroblasts, and myofibroblasts (Hematoxylin and eosin staining, ×200 magnification).

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