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Ectopic bronchogenic cyst of liver misdiagnosed as gallbladder diverticulum: A case report

Cheng-Ji Dong, Ru-Ming Yang, Qi-Long Wang, Qing-Yuan Wu, Da-Ji Yang, De-Cai Kong, Ping Zhang

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Abstract

BACKGROUND

Ectopic bronchogenic cysts are a type of congenital cystic tumor that are extremely difficult to diagnose and can be ectopically located in various organs, with the possibility of malignant transformation. Here we report a case of an ectopic bronchogenic cyst in the liver initially misdiagnosed as a gallbladder diverticulum.

CASE SUMMARY

The patient was a middle-aged woman whose chief complaint was intermittent pain in the upper abdomen. Imaging examination revealed a cystic space in the left inner lobe of the liver. She was admitted to our hospital for treatment. Based on abdominal examination and imaging findings, the initial diagnosis was gallbladder diverticulum with cholestasis combined with chronic cholecystitis. However, following intraoperative observations and postoperative pathologic assessment, the diagnosis was revised to ectopic bronchogenic cyst of the liver.

CONCLUSION

Radiologists, hepatobiliary and pancreatic surgeons, gastrointestinal surgeons, urologists, and even neurosurgeons should be aware and consider a possible diagnosis of ectopic bronchogenic cysts, especially when other types of cyst, cystadenoma, and other diseases are excluded. The disease and its complications should be detected and correctly diagnosed and treated as early as possible in order to avoid adverse outcomes.

Key Words: Ectopic bronchogenic cyst; Misdiagnosis; Gallbladder diverticulum; Benign tumor; Cystadenoma; Case report

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Core Tip: Ectopic bronchogenic cysts are the rarest type of bronchogenic cyst and can be ectopically located in different organs. When various specialists diagnose cysts, cystadenomas, and other diseases, the possibility of ectopic bronchogenic cyst should not be discounted. We report a case of an ectopic bronchogenic cyst in the liver that was misdiagnosed as gallbladder diverticulum. Early surgical intervention is the most effective strategy to prevent disease transformation.

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INTRODUCTION

A bronchogenic cyst is a rare congenital cystic tumor that can generally be classified as one of three types – namely, intrapulmonary, mediastinal, and heterotopic (the most rare)[1-3]. Bronchogenic cysts often develop ectopically in the neck, retroperitoneum, and stomach wall; very few cases of ectopic cysts in the liver have been reported[4]. Ectopic bronchogenic cysts often have no specific clinical manifestations, imaging characteristics, or changes in laboratory indices, making preoperative diagnosis extremely difficult. The cysts are typically discovered during routine physical examinations or because of complications resulting from the compression of surrounding organs or tissues. Here we report a case of an ectopic bronchogenic cyst in the liver that was preoperatively diagnosed as cholecystic diverticulum with cholestasis.

CASE PRESENTATION

Chief complaints

A 40-year-old woman presented with intermittent upper abdominal pain.

History of present illness

Physical examination at the primary hospital revealed a space-occupying lesion above the gallbladder in the left inner lobe of the liver that was complicated by chronic cholecystitis. The patient was transferred to our hospital for further treatment.

History of past illness

The patient was physically fit.

Personal and family history

Her family history was noncontributory.

Physical examination

A physical examination of the abdomen showed no muscle tension, tenderness, or rebound pain. No abdominal mass was detected; the liver was not touched under the ribs, and the Murphy sign was positive.

Laboratory examinations

Laboratory examinations including routine blood and blood coagulation, liver function, biochemistry, tumor markers, and infection markers revealed no abnormalities.

Imaging examinations

Abdominal color Doppler ultrasound showed a cystic echo beside the gallbladder in the left inner lobe of the liver with a size of 3.2 cm × 1.6 cm. The boundary was clear, the ultrasound transmission was poor, a bile mud echo was observed, there was no blood flow signal, and there appeared to be a

connection with the neck of the gallbladder. Gallbladder diverticulum with cholestasis and chronic cholecystitis was suspected. Hepatobiliary, pancreatic, and splenic multi-slice plain computed tomography (CT) scanning and 3-phase enhanced scans showed an elliptical low-density shadow above the gallbladder in the left inner lobe of the liver, with a CT value of about 55 HU. Intrahepatic bile ducts were not dilated. The gallbladder was not enlarged but the wall was slightly thickened, and there was no abnormal density in the cavity. The cystic space-occupying lesion above the gallbladder in the left inner lobe of the liver strongly suggested gallbladder diverticulum with cholestasis. Magnetic resonance cholangiopancreatography (MRCP) was performed to determine the relationship between the lesion and the gallbladder. There was an oval abnormal signal in the gallbladder fossa area about 2.9 cm × 2.0 cm in size. T1-weighted imaging (T1WI) and T2-weighted imaging (T2WI) and diffusion-weighted imaging showed a high-signal shadow and the apparent diffusion coefficient was consistent with a slightly high-signal shadow; the edges of the shadow were thin, smooth, and clear, and there was no enhancement at the lesion center. The lesion appeared to be connected to the cystic duct given the limited fluid signal shadow in the gallbladder fossa area of the intrahepatic lobe (Figure 1).

FINAL DIAGNOSIS

Based on the patient's symptoms, abdominal examination, and imaging findings, the initial diagnosis was gallbladder diverticulum with cholestasis combined with chronic cholecystitis. However, based on intraoperative findings and postoperative pathologic results, the diagnosis was later confirmed as ectopic bronchogenic cyst in the liver.

TREATMENT

The patient underwent laparoscopic cholecystectomy and cystectomy of the left inner lobe of the liver. During the operation, it was observed that the cyst and gallbladder were connected to a pedicle but not to each other. Dark green bile was drained from the gallbladder, and light yellow turbid fluid was observed inside the cyst (Figure 2).

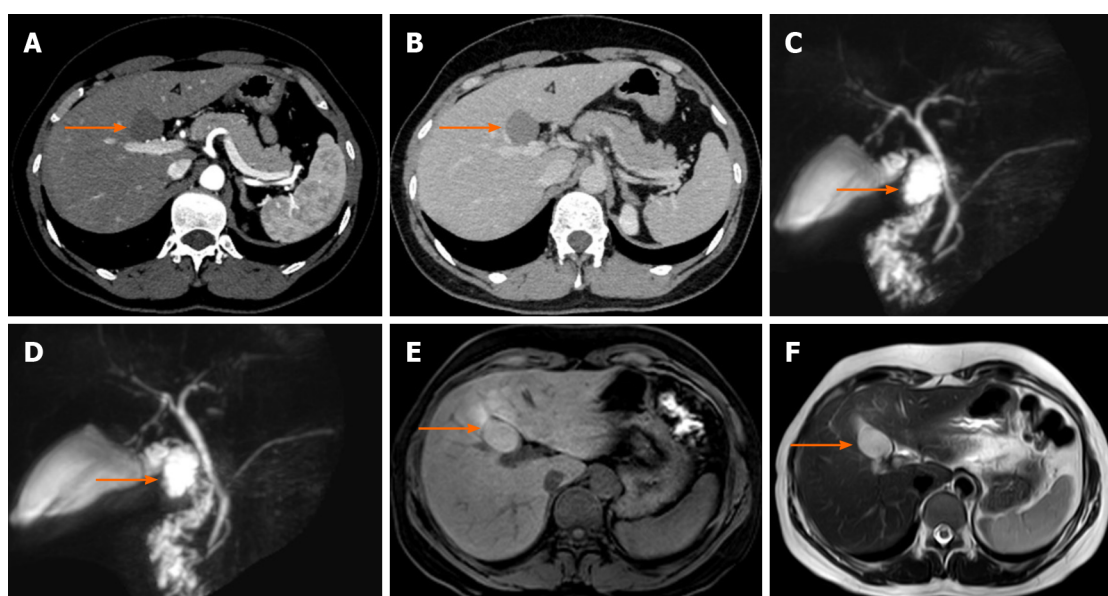
OUTCOME AND FOLLOW-UP

After the operation, we carried out a pathologic examination on the excised specimen. The outer part of the cyst was connected to the gallbladder; the volume was 2 cm × 2 cm × 2 cm, the wall thickness was 0.1–0.2 cm, and the inner wall was smooth and light brown. The cyst wall was fibrous under microscopic examination, with visible muscle tissue. Part of the mucosal epithelium on the inner surface was absent, and the remaining inner surface was partly lined with pseudostratified ciliated columnar epithelium and mucous epithelium. Serous and mucous glands were observed in the lamina propria, consistent with bronchogenic cysts. The intraoperative findings combined with histopathologic examination refuted the preoperative diagnosis of gallbladder diverticulum complicated by cholestasis (Figure 3). No recurrence was found in the color Doppler ultrasonography 6 mo after the operation.

DISCUSSION

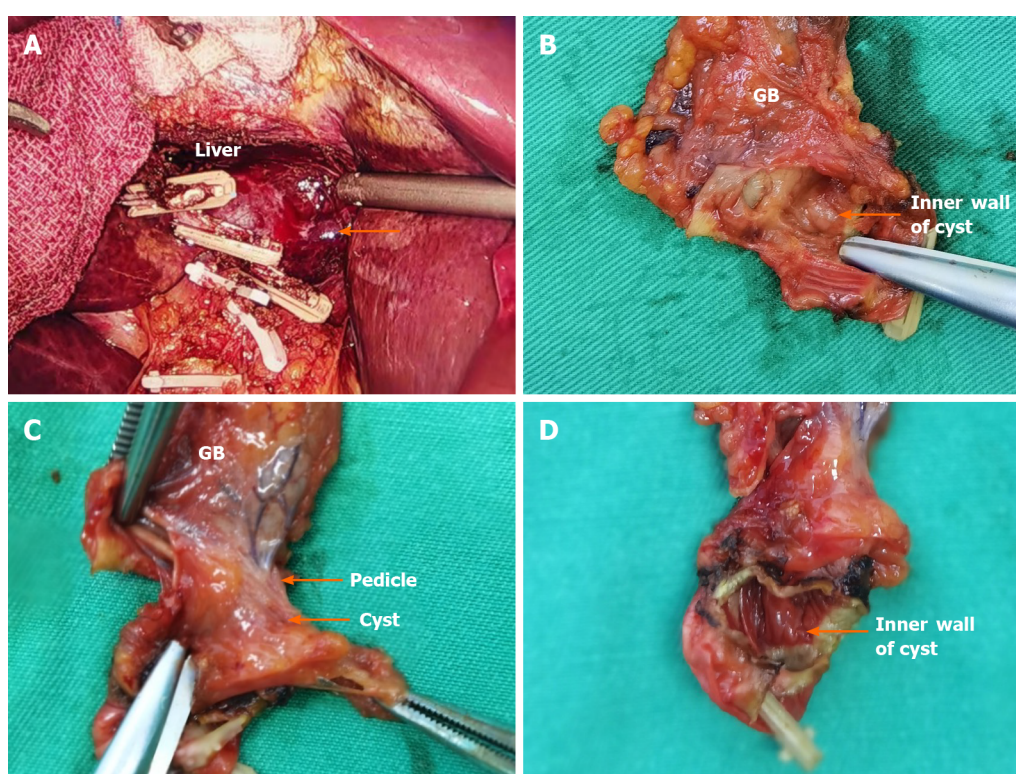
Ectopic bronchogenic cysts can occur at any age, with similar prevalence in men and women. There are few specific manifestations but clinical symptoms can arise when a cyst compresses nearby organs or tissues. For example, a patient developed new abdominal pain and gross hematuria that may have been caused by the aggravation of a lump originating from the left adrenal gland[5]; the pain was intermittent, which may have been related to chronic cholecystitis. Similarly, ectopic bronchogenic cysts have no special imaging findings. Magnetic resonance imaging (MRI) examinations often show isointense shadows on T1WI and high-intensity shadows on T2WI that are not enhanced by the contrast agent. However, in some patients, both T1WI and T2WI show significant enhancement[6]. In our patient, T1WI showed a slightly high-signal shadow and T2WI showed a high-signal shadow; a retrospective analysis of the MRI findings was more suggestive of ectopic bronchogenic cysts, which are associated with elevated levels of the tumor markers carcinoembryonic antigen, carbohydrate antigen (CA)19-9, and CA72-4[7,8].

In countries where bronchogenic cysts are endemic, they are often associated with hydatid disease [9]. The diagnosis and differential diagnosis of ectopic bronchogenic cysts is extremely difficult and they are often confused with other types of cyst and serous or mucinous cystadenoma. The pathogenesis of ectopic bronchogenic cysts in the abdominal cavity is complex and diverse. It has been proposed to



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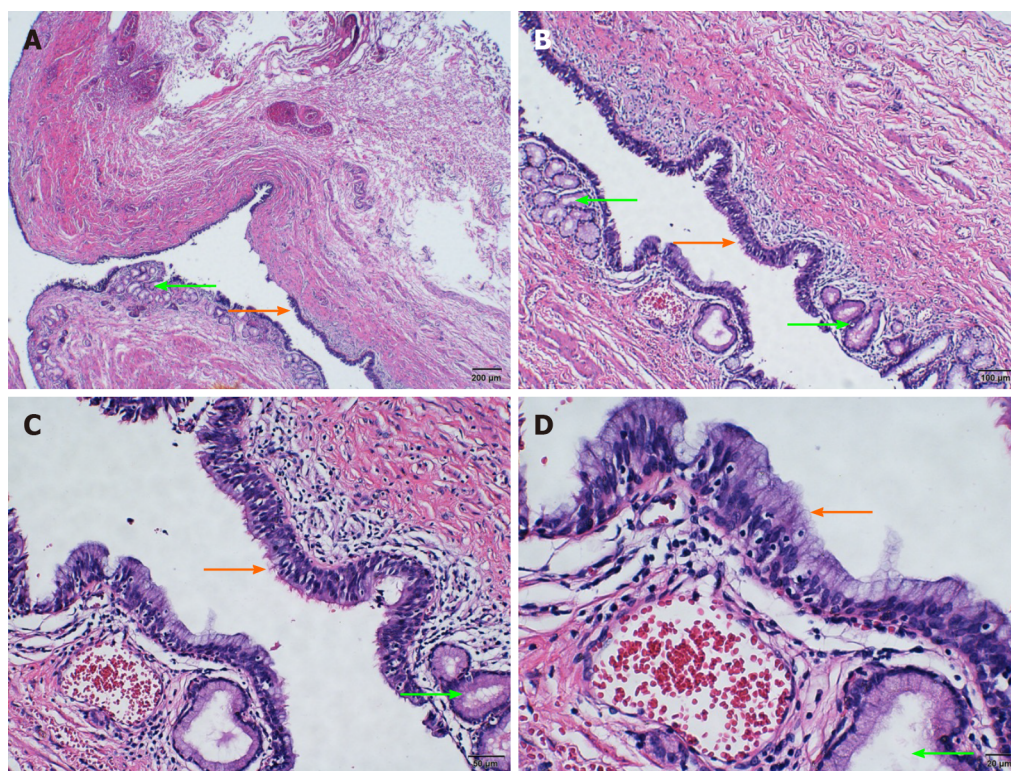
Figure 1 Ectopic bronchogenic cyst in a 40-year-old woman. The location of the cyst is indicated by an orange arrow. Magnetic resonance cholangiopancreatography (MRCP) showed that the cyst was locally connected to the cystic duct. A: Hepatobiliary and pancreatic enhanced computed tomography (CT) arterial phase; B: Hepatobiliary and pancreatic enhanced CT venous phase; C and D: MRCP; E: T1-weighted imaging showed a slightly high signal shadow; F: T2-weighted imaging showed a high signal shadow.



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Figure 2 Gross examination of the surgically excised ectopic bronchogenic cyst. A: The gallbladder and cyst were removed under laparoscopy, revealing the smooth wound surface (orange arrow) connecting the cyst and liver; B: The inner wall of the cyst. The cyst contained light yellow turbid liquid; C: The cyst was connected to the gallbladder via a pedicle; D: Inner wall of the cyst.

originate in the embryonic foregut, which develops from the primordial malformed trachea and bronchial tree in the early embryo; bacteria shed from this structure migrate to the caudal side of the foregut, which forms the abdominal organ or retroperitoneal bronchus cyst. The thoracic and abdominal cavities are thought to be connected in the early embryonic stage. At 6 wk of gestation, the pleura and



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Figure 3 Histopathologic examination by hematoxylin and eosin staining. The cyst wall was fibrous and muscle tissue was visible. The mucosal epithelium of the inner surface of the cyst wall was partially detached, and the remaining inner surface was partly lined with pseudostratified ciliated columnar epithelium (orange arrow) and mucous epithelium. Serous and mucous glands were observed in the lamina propria (green arrow). A: 4 ×; B: 10 ×; C: 20 ×; D: 40 ×.

peritoneum fuse and develop into a mature diaphragm. The body cavity is divided into the mature thoracic cavity and abdominal cavity. The abdominal buds of the tracheobronchial tree are squeezed and migrate into the abdominal cavity at the same time, leading to ectopic bronchogenic cysts in the abdominal cavity[10]. Clarifying the detailed mechanism underlying the development of bronchogenic cysts will facilitate their detection, diagnosis, and treatment.

Most ectopic bronchogenic cysts require surgical removal, and histopathologic examination of surgical specimens must be performed to confirm the diagnosis. Immunopositivity for cytokeratin (CK)7 and thyroid transcription factor 1 and immunonegativity for CK20 and calretinin in the epithelial lining also strongly support a diagnosis of bronchogenic cysts[7,11]. In terms of treatment, as ectopic bronchogenic cysts can be ectopic to different organs, the speed at which they develop and their biological behaviors vary[1,2]. Ectopic bronchogenic cysts are also easily confused with cystic diseases of adjacent organs, patients with bronchogenic cysts or suspected ectopic bronchogenic cysts should be examined by a surgeon, especially when the cyst is small as it may be missed. Thus, immunohistochemistry is useful when bronchial cysts cannot be differentially diagnosed based on histology and microscopic examination alone, and excision by using laparoscopy or robotic surgery and histopathology remain the first choices for treatment and definitive diagnosis[12].

CONCLUSION

Ectopic bronchogenic cysts are a congenital cystic tumor that are extremely difficult to diagnose and can be ectopically located in various organs, and have a risk of malignant transformation. Radiologists, hepatobiliary and pancreatic surgeons, gastrointestinal surgeons, urologists, and even neurosurgeons should be aware of these cysts. During diagnosis and treatment of a suspected cyst or cystadenoma, the possibility of ectopic bronchogenic cysts should be considered. The disease and its complications should be detected and diagnosed and treated as early as possible to avoid adverse consequences.

FOOTNOTES

Author contributions: Zhang P and Dong CJ performed the surgery, reviewed the literature, and contributed to manuscript drafting; Dong CJ, Yang RM, and Wang QL wrote the manuscript; Dong CJ, Wu QY, Yang DJ, and Kong

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