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Ileal bronchogenic cyst: A case report and review of literature

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Abstract

We herein report a rare case of ileal bronchogenic cyst that was found in a 39-year-old Chinese man. He had no symptoms and the physical examination was normal. Tumor markers were within the normal range. Abdominopelvic enhanced computed tomography showed a mass in the lower abdominal cavity and the tumor had a complete capsule. Diagnostic laparoscopy was then performed, which showed that a spheroid mass with a complete capsule was located at the antimesenteric border of the distal ileum 20 cm from the ileocecal valve, measuring 6.0 cm × 6.0 cm × 5.0 cm. Considering that the malignancy of the tumor cannot be ruled out, and there is a risk of rupture during laparoscopic surgery, the patient was converted to an open surgery. Partial resection of the ileum with the tumor was performed, followed by a side-to-side anastomosis. The tumor was gray-red in color, filled with grayish yellow mucus and had no septum. The postoperative pathology revealed that the cystic wall was lined by pseudostratified ciliated columnar epithelium without cellular atypia. The wall consisted of bronchial mucous glands and smooth muscle fibers, and no abnormalities were found in adjacent ileum tissues. Thus, a diagnosis of bronchogenic cyst of the ileum was made.

Key words: Laparoscopy; Ileal neoplasms; Epithelium; Bronchogenic cyst; Abdominal cavity; Case report

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Core tip: Bronchogenic cyst is related to abnormal embryonic development. The most common site of bronchogenic cyst is the mediastinum. Subdiaphragmatic bronchogenic cysts are extremely rare and bronchogenic cyst of the ileum has not been reported in the literature. This case may help us better understand where bronchogenic cysts may occur. Bronchogenic cysts should be considered in the differential diagnosis of ileal masses.

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INTRODUCTION

Bronchogenic cyst is a rare disease, which is related to the abnormal embryonic development. It has been reported that bronchogenic cysts may occur in the mediastinum^[1], lung^[2], heart^[3], stomach^[4] and retroperitoneum^[5]. The diagnosis of bronchial cyst depends on pathological examination: The cystic wall is lined by pseudostratified ciliated columnar epithelium and consists of bronchial mucous glands and smooth muscle fibers. Here we report a rare case of ileal bronchogenic cyst and to the best of our knowledge, such case has not been previously reported in the literature.

CASE REPORT

A 39-year-old Chinese male was admitted for "finding an abdominal mass for 1 wk". The patient once accepted computed tomography (CT) examination because of low back pain and was diagnosed as having degenerative changes of the lumbar spine. There was no history of nausea, vomiting, hematemesis, melena, diarrhea or change of habit of discharge. Physical examination showed no obvious abnormality. Routine blood parameters and tumor markers were within the normal range. Abdominopelvic enhanced CT showed a mass of cystic density in the lower abdominal cavity with a complete capsule, which measured 5.2 cm × 4.1 cm. In the arterial phase, the CT value of the mass was 37 HU. The mass may arise from the ileum (Figure 1A). Based on these results, the preoperative diagnosis was tumor of the ileum. During a diagnostic laparoscopy, we found a spheroid mass with a complete capsule located at the antimesenteric border of the distal ileum 20 cm from the ileocecal valve, which measured 6.0 cm × 6.0 cm × 5.0 cm. Considering that the malignancy of the tumor cannot be ruled out, and there is a risk of rupture during laparoscopic surgery, the patient was converted to an open surgery. Partial resection of the ileum with the tumor was performed. The resection margin was 3 cm away from the tumor edge. Then, a side-to-side

anastomosis was performed. The patient recovered successfully after the surgery. The operative time was 135 min, and the volume of blood loss was 50 mL. The unilocular cyst was gray-red in color and filled with grayish yellow mucus. The capsule was smooth and about 0.4 cm in thickness. Microscopic examination of the tumor and partial ileum together with hematoxylin and eosin staining was conducted. The postoperative pathology revealed that the cystic wall was lined by pseudostratified ciliated columnar epithelium without cellular atypia. The wall consisted of bronchial mucous glands and smooth muscle fibers, and no abnormalities were found in adjacent ileum tissues (Figure 1B and 1C). Thus, a diagnosis of bronchogenic cyst of the ileum was made based on the histological appearance. The patient was discharged 8 d after the operation.

DISCUSSION

The pathogenesis of bronchogenic cysts remains unclear. A reasonable speculation is that the disease is related to abnormal embryonic development^[6]. Bronchogenic cysts are derived from the primitive foregut due to the lung bud development malformation during the third to seventh weeks of embryogenesis. The malformation occurs when the lung bud fails to attach to the trachea or esophagus and then migrates to the thoracic or abdominal cavity. The migration usually ends up in the posterior mediastinum. As the mucus in the ectopic lung bud fails to be discharged, the lung bud gets increasingly larger and eventually becomes a bronchogenic cyst. Bronchogenic cysts located in the mediastinum, lung and heart have been reported a lot, while subdiaphragmatic bronchogenic cysts such as gastric^[4] or retroperitoneal bronchogenic cysts^[5] are extremely rare. Among them, bronchogenic cysts in the abdominal cavity are mostly located in the left to the midline adjacent to the pancreas tail, spleen, and left adrenal gland^[7]. Only two cases of bronchogenic cyst of the ileal mesentery have been reported^[8,9]. However, ileal bronchogenic cysts have not been reported previously.

Most of the bronchogenic cysts were asymptomatic and found during thoracic or abdominal surgery^[9]. The patients with symptomatic bronchogenic cysts often complain of abdominal pain because of secondary infection or perforation. Generally, bronchogenic cysts appear as a well circumscribed cystic lesion accompanied with or without calcification and no significant contrast uptake on CT scan^[6,10,11]. On magnetic resonance images, those cysts were found to have low signal on T1WI but high signal on T2WI^[12]. As for the preoperative diagnosis, endoscopic ultrasound together with fine-needle aspiration may be the most effective way^[13].

Differential diagnosis of abdominal bronchogenic cysts includes gastrointestinal stromal tumor^[14], teratoma^[15], Meckel's diverticulum^[16], enteric duplication

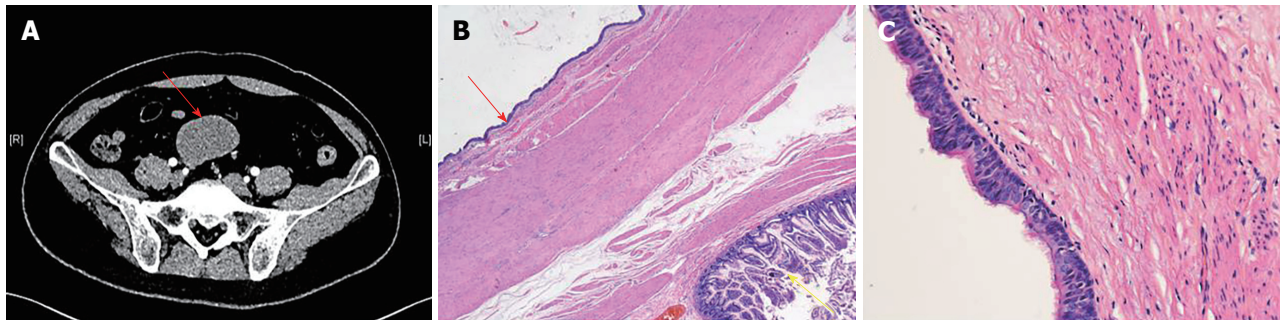


Figure 1 Imaging and pathological results. A: The abdominopelvic computed tomography scan reveals a cystic mass (red arrow); B: The microscopic findings showed the inner wall of the bronchogenic cyst (red arrow) and the ileal mucosa (yellow arrow) (HE staining; original magnification, $\times 20$); C: Pseudostratified ciliated columnar epithelium covering the inner wall of the cystic wall (HE staining; original magnification, $\times 200$).

cyst^[17], lymphangioma^[18] and echinococcosis^[19]. In this case, the pseudostratified ciliated columnar epithelium of the cystic wall had no cellular atypia and there was no sign of abnormality in the neighboring ileum tissue. Therefore, there is no possibility of malignancy.

Asymptomatic bronchogenic cysts in small size are hard to diagnose. However, as bronchogenic cysts enlarge, there is a risk of secondary infection, perforation or even malignant change^[5,20]. They may conceal a tumor and the malignant progression can occur both in adults and children^[20]. Thus, surgical resection is the most suitable choice recommended to treat bronchogenic cysts when identified^[21].

examination confirmed that the lesion was benign.

Related reports

Bronchogenic cysts are found mostly in the mediastinum, lung and heart, and occasionally in the stomach and retroperitoneal organs. Only two cases of bronchogenic cyst of the ileal mesentery have been reported. Ileal bronchogenic cyst has never been reported before.

Term explanation

Ileal bronchogenic cyst is congenital malformation which can occur at the antimesenteric border of the ileum.

Experiences and lessons

Surgical resection should be the most suitable choice to treat bronchogenic cysts. Clinicians should take bronchogenic cysts into consideration when the radiological examination shows a cystic lesion.

ARTICLE HIGHLIGHTS

Case characteristics

A 39-year-old man presented with an asymptomatic abdominal mass found by computed tomography (CT).

Clinical diagnosis

The diagnosis of an ileal bronchogenic cyst was made following radiological and pathological examinations.

Differential diagnosis

Differential diagnosis should include other abdominal space-occupying lesions, such as Meckel's diverticulum, gastrointestinal stromal tumor, teratoma, lymphangioma, enteric duplication cyst and echinococcosis.

Laboratory diagnosis

Serum total cholesterol and triglyceride were slightly elevated. Routine blood parameters and coagulation function indexes were in the normal range and the fecal occult blood was negative.

Imaging diagnosis

The abdominopelvic enhanced CT scan showed a cystic mass measuring 5.2 cm \times 4.1 cm that was located in the lower abdominal cavity. It may originate from the ileum. The imaging diagnosis was an ileal tumor.

Pathological diagnosis

The postoperative pathology revealed that the mass was an ileal bronchogenic cyst.

Treatment

The patient received partial ileal resection and anastomosis. No drugs or chemoradiotherapy were administered after the surgery as the pathological

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