

Retroperitoneal bronchogenic cyst with fluid-fluid level: A case report and literature review

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Abstract. Bronchogenic cyst is a benign lesion with congenital dysplasia. Although the occurrence of this type of cyst is rare in the retroperitoneum, the presence of fluid-fluid levels is an even rarer phenomenon in bronchogenic cysts. Therefore, it can be easily misdiagnosed due to the lack of a universal guideline of specific imaging manifestations. The present report describes the case of a patient with a bronchogenic cyst with fluid-fluid levels whilst also performing a brief literature review to summarize the findings of previous reports on this condition. A 48-year-old male individual presented with severe lower back pain without any obvious causes. A CT scan revealed a low-density cystic mass of ~3x4x6 cm in the left front of the T12-L2 area, which originated from the left crus of the diaphragm. MRI revealed a fluid-fluid level in the cyst. Anterior thoracolumbar surgery was performed to completely resect the mass. During the surgery, it was confirmed that the cyst originated from the left crus of the diaphragm and the lesion was diagnosed to be a bronchogenic cyst by pathological analysis. The patient's symptoms improved after the surgery and no recurrence of the cyst was observed during the 3-year follow-up period. The presence of a fluid-fluid level in a retroperitoneal bronchogenic cyst is rare, particularly in the abdominal aorta and paravertebral regions, rendering it easily misdiagnosed. It may be associated with protein, hemorrhage and calcium-containing mucus deposition in the cysts. In the present study, a rare case of fluid-fluid level in bronchogenic cyst was reported and a literature review was provided.

Introduction

Bronchogenic cyst is a congenital, dysplastic disease derived from the foregut, with pseudostratified ciliated columnar epithelium (1). It most frequently occurs in the mediastinum and lung, whilst it can also occur in the diaphragm and retroperitoneum on rare occasions (2). The majority of bronchogenic cysts are benign lesions with a slow growth rate and an insidious onset (3). Retroperitoneal bronchogenic cysts are mostly located above the pancreas and the left adrenal gland, but rarely cause clinical symptoms. Therefore, they are usually discovered coincidentally during a physical examination for other conditions (4). It may occur at any age and may affect both males and females equally (5). In a small number of cases, clinical symptoms may occur when the cyst is infected or compresses adjacent structures (3).

The present report describes a case of bronchogenic cyst originating from the left diaphragmatic crus and extending into the retroperitoneal space. MRI revealed that there was a fluid-fluid level in the cyst. Due to the severe back pain and the inefficacy of conservative treatment, the cyst was completely resected with a favorable post-operative recovery. The presence of a fluid-fluid level in a bronchogenic cyst is rare. When the cyst contents have a high density and precipitate, a CT scan, MRI or B-ultrasound may reveal the density or signal stratification in the cyst. It may be related to protein, hemorrhage and calcium-containing mucus deposition in cysts. Fluid-fluid levels have also been reported in other mediastinal masses, including cystic teratoma and mediastinal lymphangioma, which may be misdiagnosed. In addition, the present report performed a literature review, summarizing the findings on bronchogenic cysts with fluid-fluid level reported in previous studies.

Case report

A 48-year-old male patient presented at Xiamen University Affiliated Southeast Hospital (Zhangzhou, China) in October 2018 complaining of 'back pain for 2 days'. The pain was relieved when bending over and the patient had no other symptoms/complaints, including fever, night sweats, abdominal pain or abdominal distension. The patient denied any history of trauma or surgery. The patient was in

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Abbreviations: L2, lumbar 2; T12, thoracic 12

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a forced position of bending and crouching due to extensive thoracic and lumbar spine tenderness and percussion pain. The visual analogue scale score of pain was 7 out of 10. There was no abdominal tenderness or rebound tenderness. Muscle strength, muscle tension, skin sensation and leg activity were all normal. Laboratory tests, such as white blood cell count, neutrophils, biochemistry, erythrocyte sedimentation rate, C-reactive protein and tumor markers all yielded normal results. A CT angiography of the abdominal aorta (Siemens Sensation 16; Siemens AG) revealed that there was a low-density cystic mass with a Hounsfield units value of 30 originating from the left diaphragm foot on the left front of the T12-L2, mildly compressing the aorta. The size of the cyst was $\sim 3 \times 4 \times 6$ cm (Fig. 1A). There was no change in the density of the cyst in all images. The upper segment of the cyst was in the retrocrural space, whereas the lower segment was in the retroperitoneal space (Fig. 1B-D). The CT scanning parameters were as follows: Slice thickness, 5 mm; tube voltage, 120 kV; tube current, 200 mA. An MRI scan (Siemens Magnetomveri 3.0T; Siemens AG) revealed that the cyst originated from the left diaphragmatic crus, close to the spine and abdominal aorta, with a fluid-fluid level in the cyst (Fig. 2A). An enhanced scan indicated that the fluid signal in the upper layer of the cyst was higher compared with that in the lower layer (Fig. 2B). The contents of the cyst had no enhancement, whereas the cyst wall was smooth with mild enhancement (Fig. 2B). The MRI scanning parameters were as follows: Repetition time/echo time, 2,000 msec/96 msec; matrix, 256x256; and section thickness, 5 mm. Celecoxib and tramadol were used to relieve pain after admission. However, their effectiveness was poor.

To confirm the diagnosis and relieve the symptoms, an anterior thoracolumbar surgery was performed. Following the surgical exposure, the size of the cyst was confirmed to be $\sim 3 \times 4 \times 6$ cm. The upper segment was located in the retrocrural space, closely connected to the left diaphragmatic crus, whereas the lower segment was located in the retroperitoneal space. A transparent fluid flowed out of the cyst after puncturing the cyst, following which a large amount of viscous white liquid was drawn out using a syringe. The cyst wall was then completely scraped off using a spatula. No bacterial or fungal growth was observed in the culture of the liquid. The cysts were analyzed pathologically by hematoxylin and eosin staining, the protocols as follows: The tissue of cyst was fixed in 4% paraformaldehyde for 24 h and then dehydrated and embedded in paraffin. The specimen was cut into 10- μ m slices, then dewaxed and immersed in hematoxylin solution for 5 min. After rinsing with water, the slices were stained with eosin Y solution for 2 min. After dehydration with alcohol and clearing with xylene, the slices were sealed with neutral resin and prepared for pathological analysis under the light microscope. The results revealed that the cyst wall was covered with pseudostratified columnar epithelium (Fig. 3A and B). In addition, there were chondrocytes and bronchial glands but no blood cells or calcium components in the cyst, which was suggestive of a bronchogenic cyst (Fig. 3C and D). After surgery, the pain symptoms of the patient markedly improved. Re-examination using MRI revealed that the

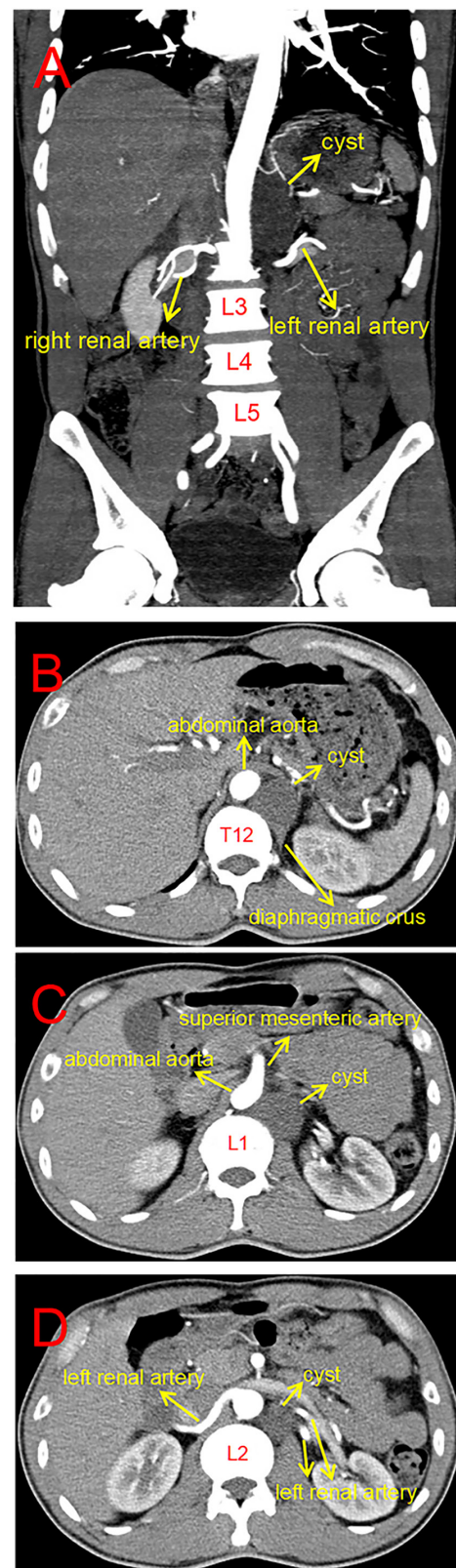


Figure 1. CT angiography of the abdominal aorta. (A) Coronal imaging revealed a low-density cystic mass with a Hounsfield units value of 30 on the left front of the T12-L2. The size of the cyst was determined to be $\sim 3 \times 4 \times 6$ cm. The lower part of the cyst is adjacent to the left renal artery and the medial part is adjacent to the abdominal aorta. (B) Upper cross-sectional imaging indicated that the cyst slightly compressed the abdominal aorta and originated from the left diaphragmatic crus, with the upper part located in the retrocrural space. (C) Middle cross-sectional imaging revealed that the lower part of the cyst extended into the retroperitoneal space. (D) Cross-sectional imaging below indicated that the lower edge of the mass was adjacent to the left renal artery.

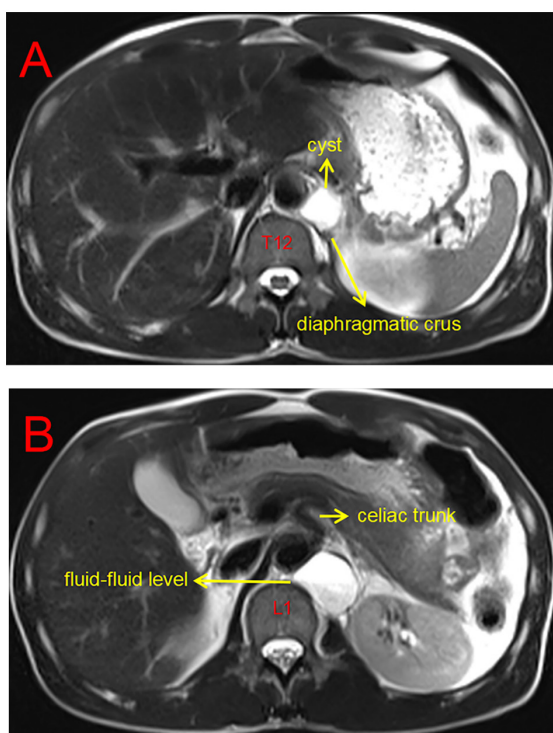


Figure 2. Enhanced MRI images. (A) A cyst located in the retroperitoneal area originating from the left diaphragmatic crus, close to the spine and the right side of the abdominal aorta. (B) The fluid-fluid level was visible in the cyst. The cyst compressed the right side of the abdominal aorta was mildly compressed, with higher fluid signal values in the upper layer and lower signal values in the lower layer. Enhanced scan revealed that the cyst wall was smooth with no enhancement of cyst contents, but there was mild enhancement of the cyst wall.

cyst had been completely removed. Of note, no recurrence of the cyst and any clinical symptom was observed at the immediate, 3 months, 1 year and 3 years postoperatively (Fig. 4A-E).

Discussion

Bronchogenic cyst (also known as bronchial cyst) is a rare, mostly benign, cystic mass caused by congenital dysplasia of the respiratory system (6). According to the location of the cyst, it may be classified as the intrapulmonary type, mediastinal type or ectopic type (7). The ectopic type may occur in various parts of the body, including the brain, spinal canal, diaphragm, retroperitoneum and neck (2,8-12). A systematic review in the PubMed database was conducted using the following keywords: [Bronchogenic cyst (title)] and [fluid level (title)]. Articles with a diagnosis of bronchogenic cysts without fluid level were excluded. A total of 31 articles were searched and 27 articles were excluded based on title screening and abstract screening. Only 4 relevant articles have been described in previous case reports (Table I) (13-16). In the present case, the fluid-fluid level in the cyst was considered to be caused by calcium or protein deposition and symptoms were usually related to compression of adjacent structures. Fluid-fluid level is mainly detected by CT scans, MRI and B-ultrasound, which may be related to the precipitation of protein, hemorrhage or calcium-containing mucus deposition

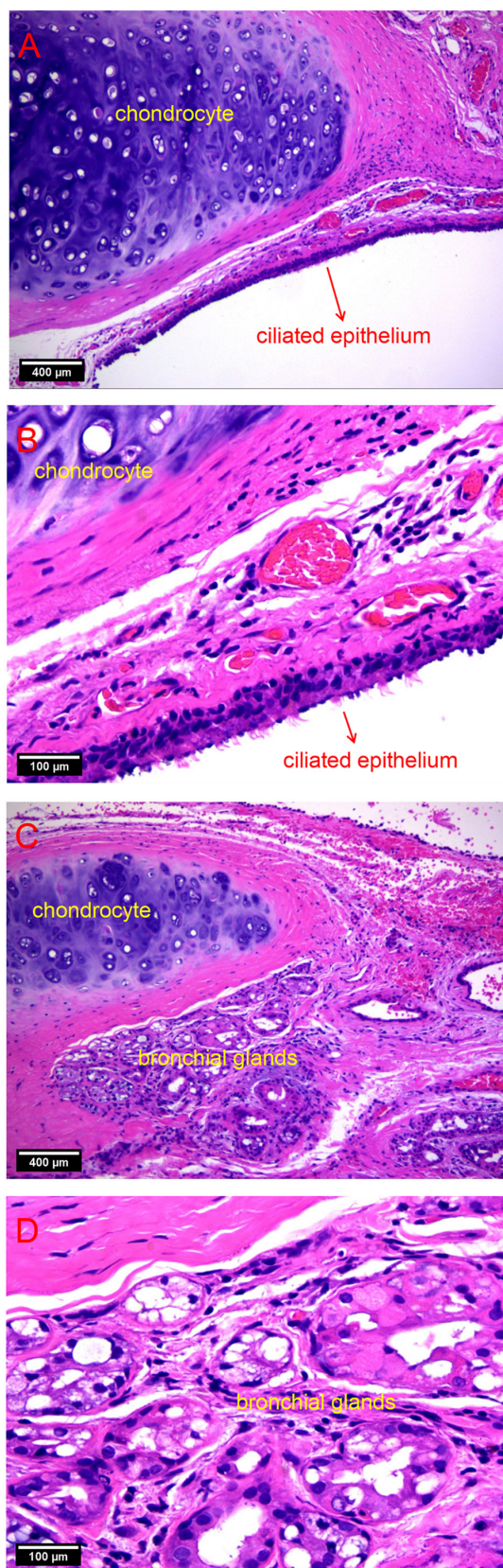


Figure 3. H&E staining histopathological examination. (A and B) The cyst wall was covered with well-differentiated, pseudostratified and ciliated columnar epithelial cells. There were numerous cartilages and blood vessels in it and inflammatory cells had infiltrated. (A) magnification, x100 and scale bar, 400 μ m. (B) Magnification, x400 of (A) and scale bar, 100 μ m. (C and D) Cartilages and bronchial glands were found under the epithelium but no blood cells or calcium components were observed within the cyst. (C) Magnification, x100 and scale bar, 400 μ m. (D) Magnification, x400 of (C) and scale bar, 100 μ m.

Table I. Summary of clinical characteristics of previously reported cases of fluid-fluid level in bronchogenic cyst and the present case.

Author, year	Sex	Age, years	Symptoms	Tumor size, cm	Location	Treatment	(Refs.)
Lyon, 1993	F	33	Dysphagia	Unknown	Middle mediastinum	Surgery	(13)
Bargalló, 1993	F	55	Asymptomatic	3	Right paraspinal region	Surgery	(14)
Aydingöz, 1997	M	38	Chest pain	7x6x5	Aortopulmonary window	Transbronchial needle aspiration	(15)
Han, 2017	F	31	Chest and back pain	2.2	Right lower posterior mediastinum	Surgery	(16)
Han, 2017	F	74	Asymptomatic	3	Right lower paratracheal region	Imaging follow-up	(16)
Present case	M	48	Acute back pain	6x4x3	Retroperitoneal space	Surgery	N/A

F, female; M, male.

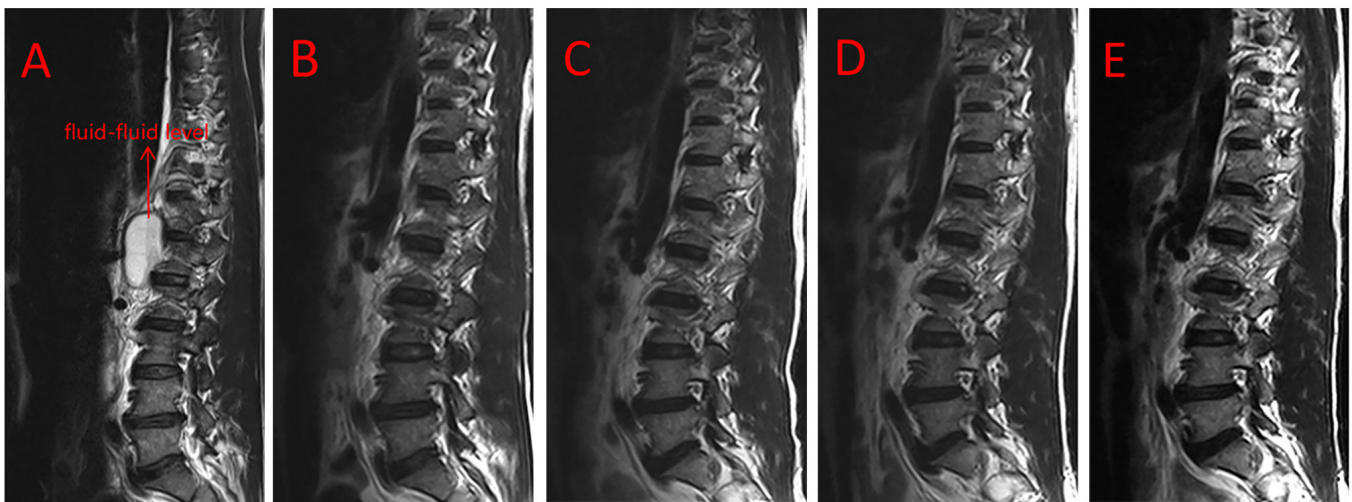


Figure 4. No recurrence of the cyst can be observed with a T2-weighted imaging scanning on the sagittal were conducted during a 3-year follow-up period. (A) Pre-operation (fluid-fluid level indicated by arrow). (B) Immediately after, (C) 3 months, (D) 1 year and (E) 3 years after the operation.

in the cyst (17). Retroperitoneal bronchogenic cysts are rare, the majority of which occur in the posterior triangle of the stomach composed of the midline of the abdomen, the splenic vein and the diaphragm (4). In the case described in the present report, the cyst was in the left front of T12-L2 and adjacent to the abdominal aorta in the posterior pararenal space. This cyst originated from the left diaphragmatic crus. The upper segment was in the retrocrural space, whereas the lower segment was in the retroperitoneal space. Since this presentation has been rarely reported in the literature, the pathogenesis of this disease remains elusive (18). At present, the hypothesis of a pinched and migratory bud is generally accepted (19). During embryonic development, certain abnormal buds of the tracheobronchial tree are 'pinched off'

and migrate to an aberrant location. In cases where the endocrine cells produced by the buds cannot be discharged, an ectopic bronchogenic cyst may form at the transfer site (20).

Bronchogenic cyst lacks a characteristic clinical presentation guideline and is frequently detected by accident upon physical examination (21). When the cyst becomes too large in size, compression may cause abdominal and back pain. In rare cases, symptoms similar to hypertension and hypokalemia may occur due to compression of the adrenal glands (22,23). In addition, if the cyst becomes infected, secondary infection manifestations may then occur, such as chills and fever (22,23). The severe back pain observed in the case of the present study may be caused by the cyst compressing the sympathetic nerves and the lateral

communicating branches in front of the spine. Bending over may reduce pain, as it may relieve compression by reducing the tension of the fascia around the cyst.

Pathological examination remains to be the gold standard for detecting bronchogenic cysts (21). The typical pathological finding is a cyst wall covered with pseudostratified ciliated columnar epithelial cells. Furthermore, this type of cyst contains cartilage or bronchial glands without any cell atypia (24). A respiratory epithelial cyst lacking cartilage or glands should be diagnosed as a foregut cyst (4). Due to the lack of specific imaging guidelines, auxiliary examinations unfortunately provides minimal assistance for the diagnosis of retroperitoneal bronchogenic cysts. A typical bronchogenic cyst exhibits as a well-circumscribed, round, low-density lesion on a CT scan. However, if the protein content in the cyst fluid is relatively high, the cyst may manifest as a soft tissue density shadow, which can easily be misdiagnosed as an adrenal tumor or neurogenic cyst (1). MRI is superior to CT scan with regard to diagnosis of cysts. Fat-suppressed images may be used to identify fat-containing masses, such as teratomas and chylocysts (7). The case of the present study exhibited a fluid-fluid level in the cyst, as revealed using MRI. This is a rare phenomenon, as suggested by previous literature reports (13). The lower layer was a denser, protein-containing precipitate. Due to the potential risk of malignant transformation, the majority of clinicians tend to select surgical treatment for retroperitoneal bronchial cysts, which is associated with a more favorable prognosis (25). In the case described in the present study, no blood cells or calcium components were found in the fluid in the cyst. All of the cases listed in Table I were found in mediastinal and retroperitoneal regions. The fluid-fluid level was frequently detected by CT or MRI, the formation of which was indicated to be related to calcium or protein deposition. Consistent with the clinical manifestations of bronchogenic cysts, those bronchogenic cysts with fluid-fluid level also tended to lack clinical symptoms, which only occur when the cyst became too large or infected. For patients able to endure surgical treatment according to their age and physical condition, surgical treatment may result in a favorable prognosis.

In conclusion, the presence of a fluid-fluid level in a retroperitoneal bronchogenic cyst is rare and can be easily misdiagnosed, particularly in the abdominal aorta and the paravertebral region. Surgical resection would be ideal in providing a definitive diagnosis and relieving symptoms, with favorable patient prognosis.

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Availability of data and materials

The datasets used and/or analyzed during the current study are available from the corresponding author on reasonable request.

Authors' contributions

WX and ZeH designed and performed the study, collected data and wrote the manuscript. ZiH, LX, ZC and BZ performed the literature search and pathological analysis. LZ and KL collected data and approved the manuscript. DL conceived and designed the study, collected data and wrote the manuscript. WX and DL checked and confirmed the authenticity of the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

The present report was approved by Xiamen University Ethics Committee (Zhangzhou, China) and was conducted in accordance with the Declaration of Helsinki.

Patient consent for publication

The patient provided written informed consent for the publication of his data and images.

Competing interests

The authors declare that they have no competing interests.

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