

# Retroperitoneal bronchogenic cyst resembling an adrenal tumor: two case reports and literature review

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Yang Wen<sup>1</sup>, Wanyuan Chen<sup>2</sup>, Junfa Chen<sup>1</sup> and Xiaodong He<sup>1</sup> 

## Abstract

Bronchogenic cysts are primitive, foregut-derived developmental anomalies with bronchial-type, pseudostratified cylindrical epithelium. They are usually discovered in the thorax. The occurrence of such cysts in the retroperitoneum is extremely rare. Imaging techniques are generally effective in the detection of retroperitoneal bronchogenic cyst. Here, we report two cases (a 27-year-old man and a 33-year-old man) who had no clinical symptoms and were found by chance to have masses in the adrenal gland area during routine physical examination. We found that they had some similar computed tomography imaging features, including complete adrenal structure, cystic fusiform mass in adrenal region, and inclusion of calcifications in the lesions. However, accurate preoperative diagnosis remains difficult and only histology can provide a definitive diagnosis. Surgery remains the treatment of choice.

## Keywords

Retroperitoneal, bronchogenic cyst, diagnostic imaging, adrenal, cystic fusiform mass, computed tomography

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## Introduction

Bronchogenic cysts are benign cystic congenital aberrations caused by abnormal budding of the tracheobronchial tree between 26 and 40 days of embryogenesis. They are usually found in the thorax, especially in the mediastinum. Rarely, they can occur below the diaphragm, and

<sup>1</sup>Department of Radiology, Zhejiang Provincial People's Hospital, People's Hospital of Hangzhou Medical College, Hangzhou, China

<sup>2</sup>Department of Pathology, Zhejiang Provincial People's Hospital, People's Hospital of Hangzhou Medical College, Hangzhou, China

### Corresponding author:

Xiaodong He, Zhejiang Provincial People's Hospital, People's Hospital of Hangzhou Medical College, No. 158, Shangtang Road, Hangzhou, 310014, Zhejiang, China.  
Email: [HXD820403@163.com](mailto:HXD820403@163.com)



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a retroperitoneal position is exceptionally unusual. In most cases, retroperitoneal bronchogenic cysts occur on the corpus of the pancreas or left adrenal gland region.<sup>1</sup>

Bronchogenic cysts are usually asymptomatic unless they are infected or enlarged enough to compress nearby organs. Retroperitoneal bronchogenic cysts are occasionally identified by imaging analyses but are often misdiagnosed. At present, only histopathological examination can yield a definitive diagnosis of retroperitoneal bronchogenic cyst. Therefore, surgical resection is the only therapeutic strategy to establish definitive histology and to provide symptom resolution.

## Case reports

Institutional review board approval was obtained from the Research Ethics Committee of Zhejiang Provincial People's Hospital. Both patients provided written informed consent.

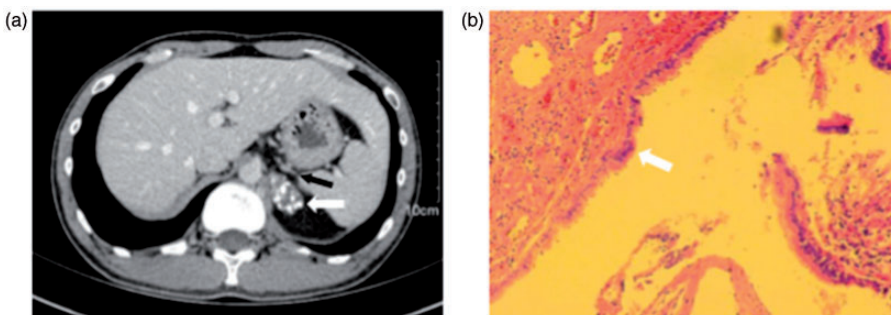
### Case 1

In June 2017, a 27-year-old man was referred to the Zhejiang Provincial

People's Hospital for examination of a left adrenal tumor that was incidentally discovered on physical examination. The patient's history was insignificant and systemic examination was normal. Complete blood counts and liver and kidney function tests were within normal limits. The secretory levels of all adrenal gland hormones were normal.

On abdominal computed tomography (CT) imaging, a 2.1-cm × 4.1-cm fusiform lesion with a clear boundary was seen in the left suprarenal area. The lesion was heterogeneous in density and had much calcification. After injection of contrast medium, the lesion showed no obvious enhancement. The left adrenal gland was located in front of the lesion, and the boundary between the lesion and the left adrenal gland was clear (Figure 1a).

We suspected a cystic adrenal tumor with calcification. To confirm the diagnosis and document the risk of malignant transformation, the cyst was removed laparoscopically. The lesion could not be discriminated from the left adrenal gland. The stomach, diaphragm, and aorta were adjacent to the cyst but not involved.



**Figure 1.** Mass in the left upper retroperitoneal space (case 1). (a) Axial abdominal computed tomography showed a fusiform soft tissue mass (white arrow) in the left adrenal area, with clear boundaries and heterogeneous in density. There was some calcification in the mass, and the mass was not contrasted after injection of contrast medium; the complete adrenal structure can be seen adjacent to the mass (black arrow). (b) Histologic section of the left retroperitoneal cyst (hematoxylin and eosin staining; 20× magnification) showed that the cyst was lined by pseudostratified ciliated columnar epithelium (white arrow), indicating the bronchogenic cyst.

In the pathological workup, the lesion had a smooth inner lining and contained a grey, viscous mass. Microscopically, it consisted of ciliated respiratory epithelium, seromucous glands, and fully developed cartilage. These findings were consistent with a bronchogenic cyst (Figure 1b). The patient was discharged on the fourth post-operative day, and no recurrence was found at follow-up.

## Case 2

A 33-year-old man was evaluated at the hospital in August 2018 for a right adrenal mass, which had been identified incidentally during a physical examination 2 months prior. Laboratory results, including serology, urea level, and liver and renal function tests, were within normal range. Ultrasonography revealed a fusiform mass in the right upper retroperitoneal space (Figure 2a). Abdominal CT showed a 3.1-cm × 5.9-cm fusiform heterogeneous lesion in the right upper retroperitoneal space. The right adrenal gland was visible at the lower edge of the lesion. Subsequently, enhanced CT showed no enhancement of the mass and no significant enlargement of lymph nodes in the retroperitoneal space. Calcification was observed in the lesion, and the complete adrenal structure could be seen beside the lesion (Figure 2b, c). Based on these findings, the radiologist suggested ganglioneuroma or tuberculosis as possible diagnoses.

Because the mass was close to important organs and had an untidy margin, surgical exploration was considered a reasonable treatment strategy. Laparoscopic surgery showed that the focus was located in the right retroperitoneal region, adjacent to the adrenal gland and adhered to the diaphragm. Pathological examination revealed a cystic mass filled with grey mucus (Figure 2d). Histopathological examination revealed that the cyst part

was lined with ciliated pseudolamellar epithelium (Figure 2e). Immunohistochemical staining showed that the epithelium surrounding the cyst was positive for cytokeratin 7 (CK7<sup>+</sup>), negative for cytokeratin 20 (CK20<sup>-</sup>), and positive for thyroid transcription factor-1 (TTF-1<sup>+</sup>) (Figure 2f), which supported its bronchogenic origin.

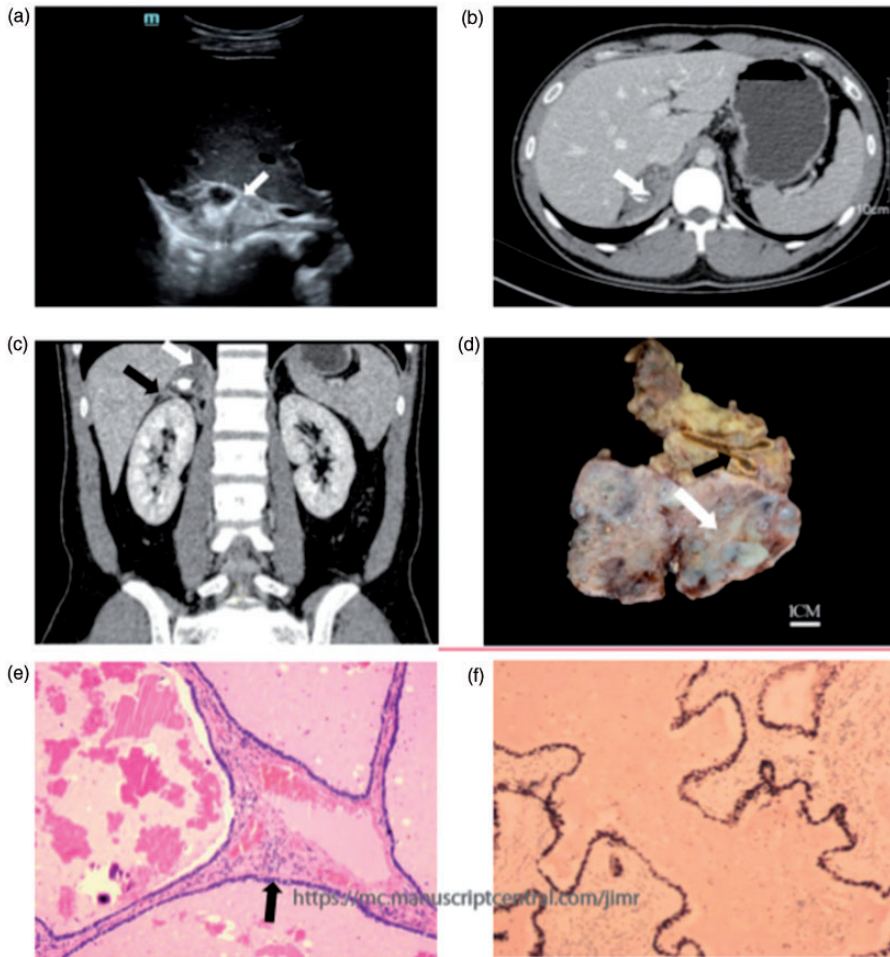
The patient recovered well and was discharged from the hospital on the fourth day after surgery. The patient did not receive further treatment because the lesion was benign, but regular follow-up was conducted.

## Discussion

Bronchial cyst is a benign congenital foregut malformation caused by abnormal budding of the tracheobronchial tree. Histologically, bronchial cysts are well defined. Microscopically, bronchogenic cysts are predominantly unilocular or oligolocular, lined by pseudostratified ciliated columnar epithelium with bronchial glands, cartilage, smooth muscle, and mucoid material.<sup>2</sup>

Bronchogenic cysts commonly occur in the mediastinal region of the thorax, usually located in the tracheobronchial tree, esophagus, or mediastinum.<sup>3</sup> Rarely, they develop below the diaphragm, and only a small number of bronchogenic cysts have been identified in the retroperitoneal space. Thus far, the exact mechanism underlying formation of retroperitoneal bronchogenic cyst is unclear. However, it is generally believed that the occurrence of bronchogenic cyst is related to the tracheobronchial tree and is caused by the separation of pulmonary buds.

If abnormal budding and extrusion of the tracheobronchial tree occur at approximately week 5 of gestation, the connection between lung bud and tracheobronchial tree is lost. In rare cases, the foregut and its derivatives of the trachea and bronchus may migrate to an atypical location,<sup>4</sup>



**Figure 2.** Mass in the right upper retroperitoneal space (case 2). (a) Ultrasound showed a mixed echo mass (white arrow) in the right adrenal gland, with a clear boundary, multiple strong echo spots in the lesion, and a less-obvious acoustic shadow behind the spots. (b, c) Post-contrast computed tomography scan revealed a fusiform-shaped, low-attenuation mass (white arrow) in the right adrenal gland area without enhancement. Calcification was seen in the lesion, and part of the lesion margin was indistinguishable from the right diaphragm and the mass (white arrow) abutting to right adrenal gland (black arrow). (d) The gross specimen showed a cystic solid mass (white arrow) with gelatinous changes; the adrenal gland is intact (black arrow). (e) High-power magnification of the cyst (hematoxylin and eosin staining; 20× magnification) revealed a lining of pseudostratified or tall columnar ciliated epithelium (black arrow). (f) Immunohistochemical staining (10× magnification) showed positive staining for thyroid transcription factor-1 (TTF-1<sup>+</sup>) in the epithelium surrounding the cyst.

including the neck, intraspinal locations, and below the diaphragm.<sup>5</sup> In the early stage of embryonic development, the thorax and the abdomen are a singular unit, and the pericardial–peritoneal canal

links the thoracic and abdominal cavities. When the canal is later divided by fusion of the pleuroperitoneal membranes, a portion of the tracheobronchial tree could be pinched off and migrate, and the migrated

lung buds may develop into retroperitoneal bronchogenic cysts.

Generally, the diameter of a bronchogenic cyst is less than 5 cm; peritoneal retroperitoneal bronchogenic cyst is often asymptomatic and often found incidentally. It can sometimes cause upper abdominal pain or back pain or result in secondary complications from infection, acute bleeding, perforation, or compression of other organs. Because the retroperitoneal bronchogenic cyst is caused by the extrusion and migration of pulmonary buds, it is not an abnormal metaplasia of the tissue at the site, and the adjacent structures are only pushed or adhered to.

In the two cases of adrenal area bronchial cysts reported here, the lesion was adherent to the adrenal gland, and the normal shape of the adrenal gland could be traced. The bronchogenic cysts had a fusiform appearance. Because of their small size, the stimulation of the adrenal gland was relatively minor, and endocrine examinations related to the adrenal gland were negative.

We believe that as the bronchial cyst occurs, growth of the ectopic lung bud occurs on both ends of the blind side. The wall of the cyst contains cartilage structure, and the space between the retroperitoneal adrenal region is spindle-shaped and relatively small; therefore, the early shape of the bronchial cyst in the adrenal region is likely to be fusiform. This appearance of the cyst has been shown in some cases in the literature.<sup>1,6-10</sup> This fusiform appearance or characteristic imaging appearance of the adrenal bronchial cyst may serve as a basis for the differential diagnosis of other adrenal disorders, but more cases are needed to support this. When secretion of mucus from the cyst increases or secondary infection and bleeding occur, the shape of the lesion will change to be round or irregular, making the cyst more difficult to distinguish from other adrenal diseases.

Because of an atypical clinical manifestation, retroperitoneal bronchogenic cysts are often incidentally detected and diagnosed by imaging modalities such as CT or magnetic resonance imaging (MRI). The fluid within a retroperitoneal bronchogenic cyst is typically a mixture of water and proteinaceous mucus. Retroperitoneal bronchogenic cysts frequently appear as heterogeneous or homogeneous hypoattenuating lesions that are not enhanced following intravenous administration of contrast agents.<sup>10</sup> However, in conditions of hemorrhage, thick mucinous or proteinaceous secretions of the lesions often show a high attenuation signal.<sup>11</sup> In addition, calcification is more common in bronchial cysts, and calcium is thought to be the predominant contributor to high attenuation on CT scans.<sup>12</sup> Considering that CT may be unable to definitively diagnose specific lesions as cysts due to a lack of mural enhancement or internal heterogeneity, MRI may be useful for correctly detecting the true cystic nature of a lesion.<sup>13</sup> Ultrasound is also an effective method to detect retroperitoneal bronchial cysts if the cysts are well transduced or accompanied by calcification. It is helpful to differentiate the nature of the cysts. However, the value of ultrasound in the diagnosis of retroperitoneal bronchial cysts is limited by the interference of gases in the gastrointestinal tract.

The two cases presented here indicate that, although extremely rare, bronchogenic cysts must be considered in the differential diagnosis of retroperitoneal tumors. We believe that complete adrenal structure, cystic mass in the adrenal region, fusiform mass, and calcification inclusions in the mass should suggest a diagnosis of bronchogenic cysts in the retroperitoneal adrenal region, but the specificity of these imaging features remains to be further verified. Considering that retroperitoneal bronchogenic cysts are frequently located

adjacent to or surrounding the adrenal gland, the condition is easy to misdiagnose as an adrenal tumor, an adrenal cyst, a cystic lymphangioma, a teratoma, or adrenal tuberculosis.

Because retroperitoneal bronchogenic cysts are benign and most patients are asymptomatic, we recommend that patients be closely followed if evidence suggests a diagnosis of bronchocyst in preoperative examination. However, preoperative diagnosis is sometimes difficult, and surgical diagnosis is recommended. Surgical excision may relieve symptoms, prevent infection or bleeding, and reduce the risk of malignant transformation of cysts.<sup>14,15</sup> The mainstay of treatment for retroperitoneal bronchogenic cysts is surgical removal, and laparoscopic resection has been widely used.<sup>16</sup> Laparoscopic surgery can reduce postoperative pain, shorten the hospital stay, and reduce the cost of surgery, among other advantages; moreover, postoperative outcomes are reliable and no complications have been reported. If the cyst is adhering to adjacent structures, the residual cyst wall should be cauterized by electrocoagulation to prevent recurrence. We performed laparoscopic excision successfully in our two patients and there have been no postoperative complications.

## Conclusions

Retroperitoneal bronchogenic cyst is rare, but when it occurs it is often seen in the adrenal gland and adjacent area. It has some characteristic imaging features but it can be easily misdiagnosed. When encountering a retroperitoneal mass, we should pay attention to the differential diagnosis to exclude other tumors. However, only a histological analysis can currently provide a definite diagnosis. Surgical resection is the most effective method to make a clear diagnosis, relieve the patient's discomfort, and

reduce the possibility of malignant transformation.

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## Declaration of conflicting interest

The authors declare that there is no conflict of interest.

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## ORCID iD

Xiaodong He  <https://orcid.org/0000-0003-3961-4328>

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