A retroperitoneal bronchogenic cyst clinically mimicking an adrenal mass: three case reports and a literature review

Bi-yue Hu¹, Hong Yu² and Jiang Shen¹

Abstract
Bronchogenic cysts are a congenital primitive foregut-derived developmental malformation, generally occurring in the posterior mediastinum. Their development in the retroperitoneum is extremely rare. Imaging techniques, such as multidetector computed tomography (MDCT), are typically effective in the detection of these lesions. Here, we describe three cases of a retroperitoneal cyst presenting as a para-adrenal mass. Only one boy presented with abdominal pain, and the other two showed no clinical symptoms. Endocrinological evaluation of all three cases was performed, and no adrenal hormone secretion was detected. All three cases were misdiagnosed preoperatively. Each patient underwent surgery, and one symptomatic patient became asymptomatic after surgery. Pathologic examination confirmed all three masses as bronchogenic cysts. The three cases showed some similar MDCT imaging features, including a complete adrenal structure, a cystic or solid mass in the adrenal region, and no obvious enhancement. Therefore, bronchogenic cysts should be considered in the differential diagnosis of retroperitoneal masses, even though accurate preoperative diagnosis remains difficult. A contrast-enhanced MDCT scan may be useful for differentiating hyper-attenuated cysts from other soft tissue masses.

Keywords
Retroperitoneal space, bronchogenic cyst, multidetector computed tomography, diagnostic imaging, developmental malformation, case report

Date received: 20 July 2021; accepted: 13 December 2021

¹Department of Radiology, West China School of Public Health and West China Fourth Hospital, Sichuan University, Chengdu, China
²Department of Radiology, Affiliated Hospital of Zunyi Medical University, Medical Imaging Center of Guizhou Province, Zunyi, China

Corresponding author:
Jiang Shen, Department of Radiology, West China School of Public Health and West China Fourth Hospital, Sichuan University, #18 South Renmin Road, Chengdu, Sichuan 610041, China.
Email: kfr111@163.com
Introduction

Bronchogenic cysts are rare congenital abnormalities of the tracheobronchial bud originating from the primitive foregut between the third and seventh weeks of embryonic life. They are typically found in the posterior mediastinum and are caused by anomalies in embryological budding of the bronchial tree. Their development in the abdomen or retroperitoneum is rare. Bronchogenic cysts are generally asymptomatic unless they are large enough to cause compression of nearby organs or infected. Symptoms are variable, depending on the diameter and location of the cyst. In most reports, retroperitoneal bronchogenic cysts occurred more often in women than in men. Here, we report three cases of bronchogenic cysts in male patients who presented with a para-adrenal mass. The aim of this case report was to increase awareness and share our experience with the imaging and diagnosis of this rare condition.

Case reports

The reporting of this study conforms to CARE guidelines. We de-identified all patient details. The need for ethical board approval was waived because this is a case report. Informed consent was not applicable because of the retrospective nature of this study. Three cases of retroperitoneal bronchogenic cysts were admitted to our hospital from October 2012 to October 2016 and retrospectively analysed. All three were male patients, ranging in age from 6 to 27 years, with a mean age of 17 years. Two cases with no clinical symptoms were diagnosed from physical examination (the patients in their late teens and late 20s), and one case complained of abdominal pain (the child with paroxysmal periumbilical abdominal pain that was aggravated at night for more than 2 months). They had no other gastrointestinal symptoms and no significant medical history (e.g. pancreatitis, hypertension or clinical signs of hypercortisolism). All physical examinations were unremarkable. Complete blood count, lipase, amylase, serum liver function and renal function tests were all within normal limits. The secretory levels of all adrenal gland hormones, plasma and urine catecholamine metanephrine concentrations and aldosterone/renin ratio were normal. Carcinoembryonic antigen and carbohydrate antigen 19-9 were not elevated.

All subjects underwent multidetector computed tomography (MDCT) examination. The cysts were located in the left adrenal region in two patients and the right adrenal region in the third patient. Abdominal dynamic contrast axial MDCT images revealed the characteristics of each lesion, which are summarised in Table 1. MDCT of the abdomen showed a hypodense mass and a hyper-dense mass adjacent to the left adrenal gland (Figure 1). The margins between the lesion and the left adrenal gland at the left para-adrenal level could not be clearly discerned. The abdominal MDCT images detected a hyper-dense mass adjacent to the right adrenal gland. There was some calcification in the margins of soft tissues, which had no septations. MDCT scans revealed cystic or solid masses and displayed no obvious enhancement following contrast medium injection. All three cases were misdiagnosed preoperatively. We obtained patient consent from our three subjects for treatment. To confirm the diagnosis, the mass was removed via retroperitoneal laparoscopic resection for two cases and a laparoscopic alternative to laparotomy for one case. The surgery details of the three cases are shown in Table 2. One symptomatic patient became asymptomatic after surgery.
All masses were pathologically diagnosed as bronchogenic cysts. Furthermore, the pathological examination revealed a mucous gland under the cyst wall and cartilaginous tissue (Figure 2).

**Table 1.** Baseline characteristics and CT findings of all subjects.

| Case | Sex | Age (years) | Clinical symptoms | Location | Number | Shape | Size (mm) | CT features |
|------|-----|-------------|-------------------|----------|--------|-------|-----------|-------------|
| 1    | male | 6           | abdominal pain    | left adrenal region | 1       | ovoid | 45 × 28 × 80 | well-defined, cystic, heterogeneous, no enhancement |
| 2    | male | 18          | none              | left adrenal region | 1       | fusiform | 71 × 36 × 70 | well-defined, soft tissue, heterogeneous, no enhancement |
| 3    | male | 27          | none              | right adrenal region | 1       | circular | 36 × 35 × 34 | well-defined, soft tissue, heterogeneous, calcification, no enhancement |

**Figure 1.** (a) Nonenhanced multidetector computed tomography (MDCT) scan showing a cystic mass in the left suprarenal region (arrow). (b, c) Contrast-enhanced MDCT scans demonstrating no enhancement of the lesion (arrow).

**Table 2** Surgical information of all cases.

| Case | Preoperative diagnosis | Surgical procedure                  | Position          | Diameter (mm) | Cystic fluid properties | Complications |
|------|------------------------|-------------------------------------|-------------------|---------------|------------------------|---------------|
| 1    | lymphocyst             | laparoscopic resection              | supine position   | 100 × 60 × 40 | gelatinous substance   | NA            |
| 2    | mucinous cystadenoma   | laparoscopic alternative to laparotomy | right lateral decubitus position | 100 × 80  | mucoid substance       | NA            |
| 3    | neurogenic tumour, heterotopic pheochromocytoma | laparoscopic resection | left lateral decubitus position | 40 × 35 | mucoid substance       | NA            |

**Discussion**

Bronchogenic cysts are rare congenital developmental foregut abnormalities resulting from aberrant budding of the
tracheobronchial anlage during the third to seventh weeks of development. They are typically found in the tracheobronchial tree, oesophagus or mediastinum when attachment to the primitive foregut persists. If complete separation occurs, the cyst may appear in other unusual locations presumably by migration but are especially rare in the retroperitoneum.

Retroperitoneal bronchogenic cysts were first reported by Miller et al. in 1953 and have been documented to occur almost equally in men and women and in a wide age range. Among all reported cases, the oldest patient was a 59-year-old man, and the youngest was diagnosed prenatally at 25 weeks of gestation during a routine scan. The majority of these cysts were found in the left adrenal gland or the superior body of the pancreas. Most cysts measured <5 cm in diameter, whereas two cysts in our study were >5 cm. The fusiform appearance of retroperitoneal bronchogenic cysts has been reported in some cases in the literature. In the three cases reported here, one cyst was spindle shaped, and the other two were round and oval, respectively. Clinical symptoms, including nausea, vomiting and abdominal pain, occur because of secondary bleeding, infection, perforation or compression of adjacent organs. Most cysts have no specific clinical manifestations and are found incidentally.

The differential diagnosis of retroperitoneal bronchogenic cysts includes several diseases, such as cystic lymphangioma, cystic mesothelioma, cystic teratoma, epidermoid cyst, tailgut cyst, bronchopulmonary sequestration, cysts of urothelial and mullerian origin, other foregut cysts and bronchogenic cyst. There are no specific imaging techniques for the preoperative diagnosis of retroperitoneal bronchogenic cysts. However, different imaging modalities can be used for their preoperative and differential diagnosis to distinguish them from other conditions, including a pleural fibroma, oesophageal leiomyoma, duplication cyst and lymphadenopathy. MDCT and magnetic resonance imaging (MRI) are useful diagnostic tools for retroperitoneal bronchogenic cysts that provide important information, including their location, shape, size, and wall thickness. Imaging may also reveal the involvement of adjacent structures and the presence of calcifications, septa or fat. On MDCT scans, retroperitoneal bronchogenic cysts commonly present as a homogeneous,
sharply defined, hypoattenuating mass with no obvious enhancement following intravenous administration of contrast agents. The thick mucinous, proteinaceous, calcium or haemorrhagic contents of bronchogenic cysts may demonstrate characteristics of solid or high attenuation masses. In most cases, the MRI finding of a long T2-weighted relaxation time is observed. MRI can be a good supplement to MDCT for detecting the true cystic nature of a lesion because MDCT might be unable to accurately diagnose specific lesions with a lack of internal heterogeneity or mural enhancement. Ultrasound is also a widely used and effective modality to detect retroperitoneal bronchial cysts, although its value in the diagnosis of lesions is limited by the interference of gases in the gastrointestinal tract.9,10 Our three cases support the previously published reports of retroperitoneal bronchogenic cysts and further emphasise the difficulty in preoperative diagnosis.1,3,6,7 MDCT images may suggest the presence of a solid retroperitoneal mass, as in our case, probably because of the thick proteinaceous secretions of the cyst. The relevant radiologic information in combination with clinical information allows adequate characterisation and diagnosis of the lesion. However, definitive diagnosis is difficult when the cysts are large and compressive or accompanied by infection.12 The signs of non-invasiveness are important, as they assist in differentiating the characteristics of retroperitoneal masses, guiding the therapeutic strategy for patients.

A precise diagnosis can be achieved by histopathological examination following surgical removal. Because of their mucinous ciliary glands, respiratory epithelium and well-differentiated cartilage, bronchogenic cysts are histologically well-defined.6 The treatment of a retroperitoneal bronchogenic cyst lies in its surgical excision. Although most cysts are asymptomatic, surgery (particularly laparoscopic resection) is recommended to establish a decisive diagnosis, prevent complications, alleviate any symptoms and assess the potential of malignant transformation.13,14 With no reports of recurrence, complete resection of retroperitoneal bronchogenic cysts yields a favourable prognosis.10

Conclusions

Although bronchogenic cysts are rare, they should be considered in the differential diagnosis of a retroperitoneal mass, especially a cystic lesion in the left adrenal gland.

Author Contributions

Conceived and designed the experiments: BH, JS and HY. Performed the experiments: BH and HY. Analysed the data: BH and HY. Contributed reagents/materials/analysis tools: BH, JS and HY. Wrote the paper: BH, JS and HY. All authors read and approved the final manuscript.

Declaration of conflicting interest

The authors declare that there is no conflict of interest.

Funding

This research received no specific grant from any funding agency in the public, commercial or not-for-profit sectors.

ORCID iD

Jiang Shen https://orcid.org/0000-0002-3825-5688

References

1. McAdams HP, Kirejczyk WM, Rosado-de-Christenson ML, et al. Bronchogenic cyst: imaging features with clinical and histopathologic correlation. Radiology 2000; 217: 441–446.
2. Cao DH, Zheng S, Lv X, et al. Multilocular bronchogenic cyst of the bilateral adrenal: report of a rare case and review of
3. Chung JM, Jung MJ, Lee W, et al. Retroperitoneal bronchogenic cyst presenting as adrenal tumor in adult successfully treated with retroperitoneal laparoscopic surgery. *Urology* 2009; 73: 442.e13–442.e15.

4. Huang H, Liu G, Li H, et al. Analysis of clinical features of retroperitoneal bronchogenic cyst. *Chin J Surg* 2015; 53: 856–859.

5. Gagnier JJ, Kienle G, Altman DG, et al. The CARE guidelines: consensus-based clinical case reporting guideline development. *Headache* 2013; 53: 1541–1547.

6. Haddadin WJ, Reid R and Jindal RM. A retroperitoneal bronchogenic cyst: a rare cause of a mass in the adrenal region. *J Clin Pathol* 2001; 54: 801–802.

7. Wang M, He X, Qiu X, et al. Retroperitoneal bronchogenic cyst resembling an adrenal tumor with high levels of serum carbohydrate antigen 19-9: A case report. *Medicine (Baltimore)* 2017; 96: e7678.

8. Miller RF, Graub M, Pashuck ET. Bronchogenic cysts; anomalies resulting from maldevelopment of the primitive foregut and midgut. *Am J Roentgenol Radium Ther Nucl Med* 1953; 70: 771–785.

9. Wen Y, Chen W, Chen J, et al. Retroperitoneal bronchogenic cyst resembling an adrenal tumor: two case reports and literature review. *J Int Med Res* 2020; 48: 30060520925673.

10. Cai Y, Guo Z, Cai Q, et al. Bronchogenic cysts in retroperitoneal region. *Abdom Imaging* 2013; 38: 211–214.

11. Yang DM, Jung DH, Kim H, et al. Retroperitoneal cystic masses: CT, clinical, and pathologic findings and literature review. *Radiographics* 2004; 24: 1353–1365.

12. Tong HX, Liu WS, Jiang Y, et al. Giant retroperitoneal bronchogenic cyst mimicking a cystic teratoma: A case report. *Oncol Lett* 2015; 9: 2701–2705.

13. Lim LL, Ho KY and Goh PM. Preoperative diagnosis of a paraesophageal bronchogenic cyst using endosonography. *Ann Thorac Surg* 2002; 73: 633–635.

14. Yoon YR, Choi J, Lee SM, et al. Retroperitoneal Bronchogenic Cyst Presenting Paraadrenal Tumor Incidentally Detected by (18)F-FDG PET/CT. *Nucl Med Mol Imaging* 2015; 49: 69–72.