Case Report

ANNALS OF MEDICINE & SURGERY

OPEN

Retroperitoneal bronchogenic cyst resembling an adrenal tumor in adult: Three case reports and literature review

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Introduction and importance: Bronchogenic cyst is a rare congenital malformation of the tracheobronchial bud originating from the primitive foregut, especially in the retroperitoneal region. Retroperitoneal bronchogenic cysts in adults are difficult to make an accurate diagnosis preoperatively.

Case presentation: We present three cases of retroperitoneal bronchogenic cysts resembling adrenal tumors in adults. Three cases were asymptomatic, and all were located on the left side. There was no significant enhancement of the cyst walls on contrast-enhanced computed tomography. Two cases presented with typical multilocular sacs and scattered calcification on radiology, whereas the other one showed unilocular sacs, without calcification, and elevation of serum carbohydrate antigen (CA) 19-9 and CA 24-2. Three cases underwent retroperitoneal laparoscopic surgeries. Histopathologic examination confirmed the diagnosis of retroperitoneal bronchogenic cysts. There was no recurrence of the three cases during follow-up.

Clinical Discussion: A retroperitoneal bronchogenic cyst is mostly asymptomatic. It can be found in adults with variable findings in computed tomography. It can be likely ignored and misdiagnosed as an adrenal tumor.

Conclusion: The tests of CA 19-9 and CA 24-2 could help diagnose retroperitoneal bronchogenic cysts. Retroperitoneal laparoscopic surgery is recommended for the treatment of retroperitoneal bronchogenic cysts with a favorable prognosis.

Keywords: Bronchogenic cyst, Case report, Diagnosis or surgical intervention, Retroperitoneal

Introduction

The bronchogenic cyst is a rare congenital malformation of the tracheobronchial bud originating from the primitive foregut in the early embryonic stage[1]. It mostly occurs in the lung and posterior mediastinum, but rarely in the retroperitoneal region. A retroperitoneal bronchogenic cyst is usually asymptomatic and detected incidentally unless it is large enough to compress the surrounding organs, or undergoing infection, hemorrhage, or perforation. A retroperitoneal bronchogenic cyst resembling an adrenal tumor was typically misdiagnosed preoperatively in previous reports[2–4]. The retroperitoneal bronchogenic cyst should be considered in the differential diagnosis of retroperitoneal mass, but the preoperative diagnosis remains challenging. Herein, we reported three cases of retroperitoneal bronchogenic cysts successfully managed with retroperitoneal laparoscopic surgeries. In addition, a literature review was made to present a comprehensive description of retroperitoneal bronchogenic cysts. This case report has been reported in line with the SCARE 2020 Criteria[5].

HIGHLIGHTS

- Retroperitoneal bronchogenic cysts can be found in adults, most are asymptomatic and with multiple calcifications.
- The tests of carbohydrate antigen (CA) 19-9 and CA 24-2 could help diagnose retroperitoneal bronchogenic cysts.
- Retroperitoneal laparoscopic surgery is recommended for the treatment of retroperitoneal bronchogenic cysts with a favorable prognosis.

Case presentation

Case 1

A 24-year-old female presented to the outpatient department of our hospital in June 2020 for the incidental detection of a left retroperitoneal cystic mass in a routine ultrasound examination. She did not have a significant complaint and denied a drug history or a family history of inherited disorders. Physical examination was unremarkable. In laboratory tests, the endocrine evaluation did not reveal an overproduction of adrenal hormones. Serum levels of CA 19-9 and CA 24-2 were 58 U/ml (0–39 U/ml) and 54 U/ml (0–20 U/ml), respectively (Table 1). Other tumor markers and serum biochemical indices were within normal ranges.

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Sponsorships or competing interests that may be relevant to content are disclosed at the end of this article.

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Annals of Medicine & Surgery (2023) 85:473–476

Received 29 October 2022; Accepted 24 December 2022

Published online 9 March 2023

http://dx.doi.org/10.1097/MS9.0000000000000182

Table 1

Patient characteristics and perioperative outcomes

	Patient 1	Patient 2	Patient 3
Sex	Female	Male	Female
Age (years)	24	33	26
Clinical symptoms	None	None	None
CA19-9 (U/ml)	58 (0–39)	—	—
CA24-2 (U/ml)	54 (0–20)	—	—
Location	Left retroperitoneal	Left retroperitoneal	Left retroperitoneal
Shape	Ovoid	Fusiform	Ovoid
Size (cm^3)	$4.1\times3.1\times3.6$	$5.4\times2.2\times3.4$	$4.2\times3.4\times2.1$
Ultrasound features	Cystic	Solid	Solid
CT features	Unilocular sacs, no calcification, no enhancement	Multilocular sacs, massive calcification, no enhancement	Multilocular sacs, punctate calcification, no enhancement
Preoperative diagnosis	Adrenal cyst	Mature cystic teratoma	Bronchogenic cyst
Surgical procedure	Retroperitoneal laparoscopic resection	Retroperitoneal laparoscopic resection	Retroperitoneal laparoscopic resection
Complication	Acute pancreatitis	Diaphragm rupture	Acute pancreatitis
Postoperative hospitalization (days)	9	3	4

CA indicates carbohydrate antigen; CT, computed tomography.

Abdominal contrast-enhanced computed tomography (CT) scans revealed an oval-shaped hypodense mass, measuring $4.1\times3.1\times3.6$ cm^3 (Figs. 1A and B). There was no septation or calcification in the mass, which showed no significant enhancement following contrast medium injection. The ^{18}F-FDG PET/CT scan revealed a well-defined mass in the left retroperitoneal region without significant FDG uptake (maximum standardized uptake value = 1.6). It was preoperatively diagnosed as a benign cyst likely derived from the adrenal gland.

A retroperitoneal laparoscopic surgery was performed by a chief physician to remove the retroperitoneal mass. It was hard to demarcate from the adrenal gland and pancreas, but no invasion was observed during the operation. The mass was successfully removed by retroperitoneal laparoscopic surgery, and the adrenal gland was preserved. The operation time was 140 min, and the estimated blood loss was 20 ml. The mass was full of brown, thick secretions in gross pathology. Histopathological examination revealed that the cyst wall was lined by pseudostratified epithelium with patchy hemorrhage in the stroma of the cyst wall (Fig. 2A). The histopathological findings confirmed that the mass was a retroperitoneal bronchogenic cyst accompanied by hemosiderin deposition and remote hemorrhage in the cyst wall. The pathological diagnosis was unexpected to the patient and the doctors. The patient developed a postoperative complication of acute pancreatitis and was discharged on the ninth day after surgery. At the 1-month postoperative follow-up, CA 19-9 and CA 24-2 had declined to the normal range. The patient was subsequently followed up regularly in the outpatient department for 24 months without recurrence.

Case 2

A 33-year-old male presented to our hospital in August 2021 for a left retroperitoneal mass. It was found in a regular physical checkup with no symptoms. The patient had no remarkable drug history or family history. Physical examination and laboratory tests were normal. Ultrasound examination revealed a hypoechoic solid mass in the left retroperitoneal region adjacent to the diaphragm. Abdominal contrast-enhanced CT revealed a fusiform-shaped mixed density mass $5.4\times2.2\times3.4$ cm^3 in size, without significant enhancement on the enhanced scan (Figs. 1C and D). There were septation and scattered massive calcifications in the mass. It was preoperatively diagnosed as a mature cystic teratoma. A routine retroperitoneal laparoscopic procedure was performed by a chief physician. The operation time was 89 min, and the estimated blood loss was 10 ml. Intraoperatively, the mass was adjacent to the diaphragm and spine, but not closely related to the adrenal gland. Complications of diaphragm rupture and secondary pneumothorax occurred in the patient (Table 1). Diaphragmatic sutures and pleural punctures were performed with the help of a thoracic surgeon in the procedure. The patient recovered smoothly and was successfully discharged 3 days after

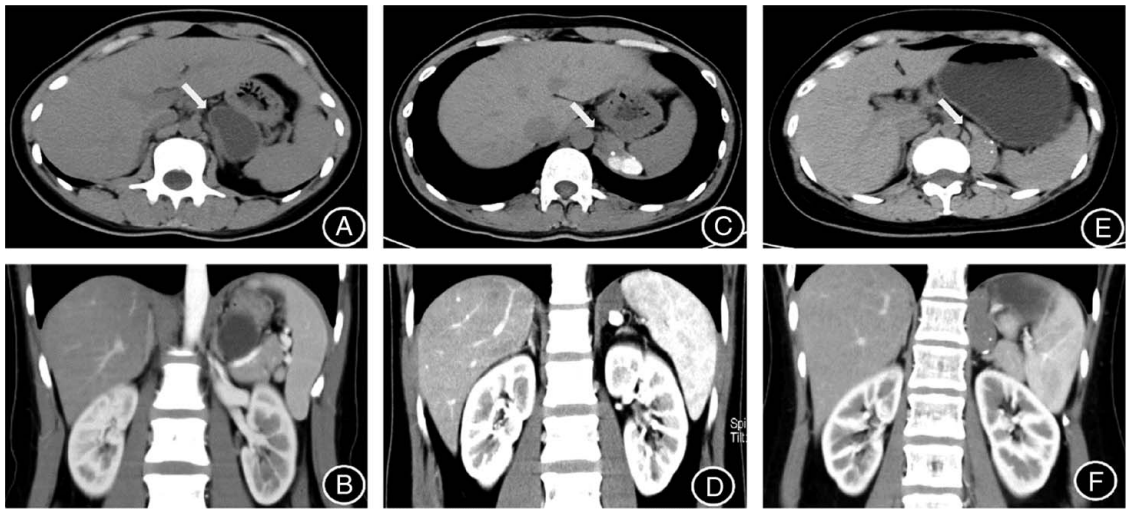

Figure 1. Abdominal contrast-enhanced computed tomography scans of the lesions in the left retroperitoneal region (white arrows). (A and B) An ovoid thick-walled cystic lesion without calcification and enhancement in case 1. (C and D) A fusiform solid lesion with massive calcification in case 2. (E and F) An ovoid solid lesion with punctate calcification in case 3.

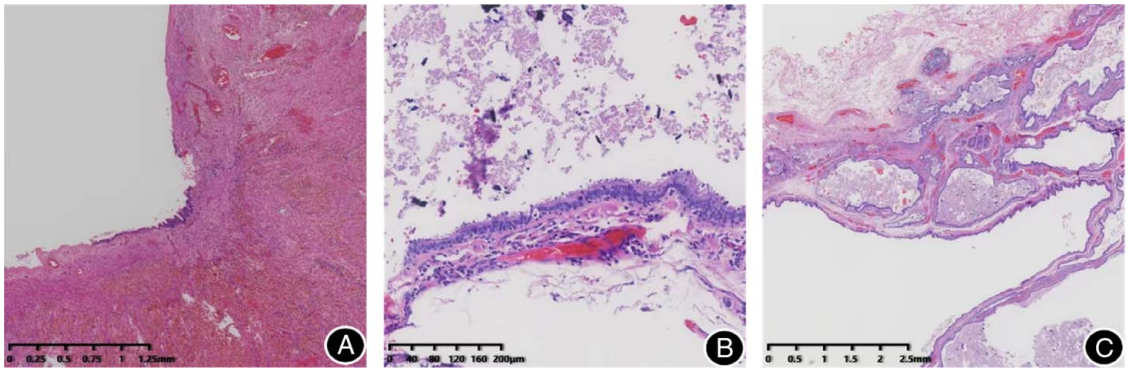

Figure 2. Microscopic appearances of retroperitoneal bronchogenic cysts with hematoxylin and eosin staining. (A) Low-power magnification of the cyst wall revealed a lining of pseudostratified epithelium with hemorrhage in the stroma in case 1. (B) High-power magnification of the cyst revealed a lining of pseudostratified columnar epithelium in case 2. (C) Low-power magnification of the cyst wall revealed a multilocular cyst-like structure lined with pseudostratified columnar epithelium in case 3.

surgery. Grossly, there were plenty of sediment-like calcifications in the cavity. The mass turned out to be a bronchogenic cyst after the histopathological examination (Fig. 2B). There was no recurrence within 10 months of follow-up in the outpatient department.

Case 3

In September 2021, a 26-year-old female without complaints presented to our hospital for a left retroperitoneal mass. The physical examination did not reveal any remarkable signs. There was no medical history, and the adrenal hormone screening was normal. Ultrasound examination revealed a hypoechoic solid mass in the left retroperitoneal region adjacent to the adrenal gland. Abdominal contrast-enhanced CT revealed an oval-shaped mixed-density mass, measuring $4.2 \times 3.4 \times 2.1$ cm^3 in size (Table 1). The mass in the left retroperitoneal region was characterized by multiple scattered punctate calcifications on radiology and no significant enhancement on the enhanced scan (Figs. 1E and F). Location and radiological characteristics were similar to those of case 2, leading to a preoperative diagnosis of the retroperitoneal bronchial cyst. Subsequently, a retroperitoneal laparoscopic procedure was performed by a chief physician. The operation time was 100 min, and the estimated blood loss was 15 ml. The patient did not complain of discomfort after surgery. Biochemical tests found that the amylase in plasma increased to 901 U/l (30–110 U/l), and the amylase in drainage fluid increased to 805 U/l. Considering a diagnosis of acute pancreatitis, fasting and being deprived of water for 2 days were performed. The drainage tube was removed, and the patient was successfully discharged 4 days after surgery. The histopathological examination revealed a cyst-like structure lined with ciliated columnar epithelium with surrounding cartilage, confirming the diagnosis of a retroperitoneal bronchogenic cyst (Fig. 2C). The patient was followed up for 11 months without recurrence.

Discussion

The retroperitoneal bronchogenic cyst resulted from the dysplastic tracheobronchial bud that was not completely separated from the primitive foregut and subsequently migrated into the peritoneal cavity during the third to seventh weeks of development[4,6]. There was no significant gender difference in the incidence of retroperitoneal bronchial cysts. West China Hospital of Sichuan University reported an extremely rare case of bilateral multilocular retroperitoneal bronchogenic cysts[7]. However, more than 80% of the cases were located in the left retroperitoneal region[8], because of the larger and later closing of the left pericardioperitoneal canal[9]. Most retroperitoneal bronchogenic cysts were asymptomatic and discovered incidentally during a routine checkup. A few patients complained of upper abdominal discomfort or back pain when the retroperitoneal bronchogenic cysts compressed the surrounding organs or developed secondary complications[10]. The average onset age was reported to be over 40 years old with an average diameter of 6.4 cm[9]. Mirsadeghi *et al.*[11] reported the largest retroperitoneal bronchogenic cyst as we know, which was up to 20 cm in diameter. The three cases we presented were all in the left retroperitoneal region but had younger onset ages and smaller tumor sizes.

Although the retroperitoneal bronchogenic cyst is rare, it should be considered in the differential diagnosis of retroperitoneal tumors. Elevated serum CA 19-9 were reported in two cases[12,13], prompting the serum CA 19-9 may be helpful in the diagnosis of retroperitoneal bronchogenic cysts. Preoperative serum CA 19-9 and CA 24-2 were all elevated in one of the presented cases, which returned to normal after resection of the cyst. To the best of our knowledge, retroperitoneal bronchogenic cysts with elevated serum CA24-2 have not been reported. CA 19-9 and CA 24-2 are tumor markers with important value for the diagnosis and prognosis of pancreatic cancer. The association between the retroperitoneal bronchogenic cyst and the two tumor markers is unclear. The fluid in the retroperitoneal bronchogenic cyst is a mixture of water and mucinous proteins and occasionally has intracapsular bleeding, leading to a solid-cystic heterogeneous ultrasonic echo. Furthermore, gastrointestinal gas affects the accuracy of ultrasound examination in retroperitoneal bronchial cysts. Contrast-enhanced CT is an effective method for preoperatively diagnosing retroperitoneal bronchogenic cysts, and the three-dimensional reconstruction could help with cyst localization and selection of surgical approach[14]. Most cases shared similar radiological features, including completed adrenal structure, multiple intracapsular calcifications, and no enhancement of the capsule wall[4,9,15]. MRI can accurately distinguish the cyst from the soft tissue mass. Retroperitoneal bronchogenic cyst presented as a benign mass without significant FDG uptake in the ^{18}F-FDG PET/CT scan[16], as same with case 1.

However, accurate preoperative diagnosis of retroperitoneal bronchogenic cyst remains challenging, only histopathological

examination could provide an accurate diagnosis. Endoscopic ultrasound can clearly distinguish the retroperitoneal bronchogenic cysts from the surrounding organs, playing a role in the differential diagnosis of adrenal tumors, esophageal duplication cysts, pancreatic cysts, and pulmonary sequestration. Above all, endoscopic ultrasound-fine needle aspiration is useful for the preoperative diagnosis of retroperitoneal bronchogenic cysts. It can identify cysts with malignant degeneration or benign complications[12,17,18]. Bronchogenic cysts in microscopical are lined by pseudostratified ciliated columnar epithelium with bronchial glands, mucinous proteins, cartilage, and smooth muscle[19]. Surgical resection is recommended for a symptomatic or asymptomatic retroperitoneal bronchogenic cyst to confirm the diagnosis, relieve compression symptoms, and prevention of the complications, such as infection, hemorrhage, and malignant degeneration[9,10]. Retroperitoneal laparoscopic surgery is a safe and effective intervention for retroperitoneal bronchogenic cysts. The complications in our cases suggested that precautions should be taken to avoid intraoperative pancreatic and diaphragmatic damage. Complete resection of the retroperitoneal bronchogenic cysts leads to a favorable prognosis, and to the best of our knowledge there is no report of recurrence.

Conclusion

Retroperitoneal bronchogenic cysts can be found in adults, most are asymptomatic and with multiple calcifications. The tests of CA 19-9 and CA 24-2 could help diagnose retroperitoneal bronchogenic cysts. Retroperitoneal bronchogenic cysts should be considered in the differential diagnosis of retroperitoneal tumors, especially mass with calcification. Retroperitoneal laparoscopic surgery is recommended for the treatment of retroperitoneal bronchogenic cysts with a favorable prognosis.

Provenance and peer review

Not commissioned, externally peer reviewed.

Ethical approval

This study is a case report and is not considered to require an ethics application.

Sources of funding

None.

Authors' contribution

L.M. and L.L. proposed the protocol and critically revised the manuscript. B.Y., X.T., X.H., and M.L. involved in data collection and management. B.Y. contributed to manuscript writing.

Conflicts of interest disclosure

The authors declare that they have no financial conflict of interest with regard to the content of this report.

Research registration unique identifying number (UIN)

This study is a case report and is not considered a research study registration.

Guarantor

Bin Yang.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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