

# World Journal of *Clinical Cases*

*World J Clin Cases* 2021 August 26; 9(24): 6964-7291



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**ABOUT COVER**

Editorial Board Member of *World Journal of Clinical Cases*, Vijaykumar Chava, MD, Professor, Department of Periodontology, Narayana Dental College and Hospital, Nellore 524003, Andhra Pradesh, India.  
chava7@hotmail.com

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The *WJCC* is now indexed in Science Citation Index Expanded (also known as SciSearch®), Journal Citation Reports/Science Edition, Scopus, PubMed, and PubMed Central. The 2021 Edition of Journal Citation Reports® cites the 2020 impact factor (IF) for *WJCC* as 1.337; IF without journal self cites: 1.301; 5-year IF: 1.742; Journal Citation Indicator: 0.33; Ranking: 119 among 169 journals in medicine, general and internal; and Quartile category: Q3. The *WJCC*'s CiteScore for 2020 is 0.8 and Scopus CiteScore rank 2020: General Medicine is 493/793.

**RESPONSIBLE EDITORS FOR THIS ISSUE**

Production Editor: *Ji-Hong Lin*, Production Department Director: *Yn-Jie Ma*, Editorial Office Director: *Jin-Lei Wang*.

**NAME OF JOURNAL**

*World Journal of Clinical Cases*

**ISSN**

ISSN 2307-8960 (online)

**LAUNCH DATE**

April 16, 2013

**FREQUENCY**

Thrice Monthly

**EDITORS-IN-CHIEF**

Dennis A Bloomfield, Sandro Vento, Bao-Gan Peng

**EDITORIAL BOARD MEMBERS**

<https://www.wjgnet.com/2307-8960/editorialboard.htm>

**PUBLICATION DATE**

August 26, 2021

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<https://www.wjgnet.com/bpg/gerinfo/208>

**ARTICLE PROCESSING CHARGE**

<https://www.wjgnet.com/bpg/gerinfo/242>

**STEPS FOR SUBMITTING MANUSCRIPTS**

<https://www.wjgnet.com/bpg/GerInfo/239>

**ONLINE SUBMISSION**

<https://www.f6publishing.com>

## Retroperitoneal bronchogenic cyst in suprarenal region treated by laparoscopic resection: A case report

Lei-Di Wu, Kan Wen, Zi-Rui Cheng, Osamah Alwalid, Ping Han

**ORCID number:** Lei-Di Wu 0000-0001-6445-1706; Kan Wen 0000-0002-6181-2171; Zi-Rui Cheng 0000-0001-8987-8052; Osamah Alwalid 0000-0001-9930-0932; Ping Han 0000-0003-0758-6668.

**Author contributions:** Wu LD acquired the data, performed the literature review, and drafted, reviewed, and edited the manuscript; Wen K took radiological images and made the radiological diagnosis; Cheng ZR performed the surgery and contributed to manuscript editing and revising; Alwalid O revised the grammar of manuscript; Han P revised the manuscript for the important content; all authors have read and approved the manuscript.

**Informed consent statement:** Informed written consent was provided by the patient for publication of this report and any accompanying images.

**Conflict-of-interest statement:** The authors declare that they have no conflict of interest to report.

**CARE Checklist (2016) statement:** The authors have read the CARE Checklist (2016), and the manuscript has been prepared and revised according to CARE Checklist (2016).

**Open-Access:** This article is an

**Lei-Di Wu, Kan Wen, Osamah Alwalid, Ping Han,** Department of Radiology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan 430000, Hubei Province, China

**Zi-Rui Cheng,** Department of Urology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, Wuhan 430000, Hubei Province, China

**Corresponding author:** Ping Han, MD, Full Professor, Department of Radiology, Union Hospital, Tongji Medical College, Huazhong University of Science and Technology, No. 1277 Jiefang Avenue, Wuhan 430000, Hubei Province, China. [cjr.hanping@vip.163.com](mailto:cjr.hanping@vip.163.com)

### Abstract

#### BACKGROUND

Bronchogenic cysts (BCs) are benign congenital foregut malformations that are mostly present in the mediastinum and pulmonary parenchyma but rarely seen in the retroperitoneum.

#### CASE SUMMARY

We report the case of 17-year-old girl who complained of epigastric pain. A cystic lesion was found in the left suprarenal region on spectral computed tomography. The ovoid, well-defined, and homogeneous cystic lesion revealed slightly enhancement on conventional imaging but no enhancement on 40 KeV virtual mono-energetic images. The iodine density value of the lesion was 0.001 mg/mL and the Z-effective value was 7.25, which were close to those of fluid material in *in vitro* experiments. Magnetic resonance imaging revealed a cystic mass of intermediate signal intensity on T1-weighted imaging and high signal intensity on T2-weighted imaging. A laparoscopic surgery was carried out. Intraoperatively, a cystic lesion with a smooth surface was found in the left retroperitoneum. And the cystic wall was completely resected after intracystic fluid was suctioned. The histopathological examination findings of the lesion were compatible with BC. The patient recovered uneventfully without signs of recurrence during a 10-mo follow-up period.

#### CONCLUSION

Radiological examinations play a significant role in the diagnosis of suprarenal BCs and spectral images offer additional spectral parameters. Accurate preoperative diagnoses of retroperitoneal BCs based on thorough imaging examinations are beneficial to the operation of laparoscopic resection.

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**Manuscript source:** Unsolicited manuscript

**Specialty type:** Oncology

**Country/Territory of origin:** China

**Peer-review report's scientific quality classification**

Grade A (Excellent): 0  
Grade B (Very good): 0  
Grade C (Good): 0  
Grade D (Fair): 0  
Grade E (Poor): 0

**Received:** April 20, 2021

**Peer-review started:** April 20, 2021

**First decision:** May 24, 2021

**Revised:** June 3, 2021

**Accepted:** July 2, 2021

**Article in press:** July 2, 2021

**Published online:** August 26, 2021

**P-Reviewer:** Cimen SG

**S-Editor:** Wu YXJ

**L-Editor:** Wang TQ

**P-Editor:** Zhang YL



**Key Words:** Bronchogenic cysts; Suprarenal region; Spectral computed tomography; Laparoscopic resection; Case report

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**Core Tip:** Bronchogenic cysts (BCs) are benign congenital foregut malformations that are mostly present in the mediastinum and intrapulmonary but rarely seen in the retroperitoneum. We report a case of BC in the left suprarenal region which was diagnosed based on spectral computed tomography imaging and magnetic resonance imaging. After a laparoscopic surgery, the patient recovered uneventfully without signs of recurrence during a 10-mo follow-up period.

**Citation:** Wu LD, Wen K, Cheng ZR, Alwalid O, Han P. Retroperitoneal bronchogenic cyst in suprarenal region treated by laparoscopic resection: A case report. *World J Clin Cases* 2021; 9(24): 7245-7250

**URL:** <https://www.wjgnet.com/2307-8960/full/v9/i24/7245.htm>

**DOI:** <https://dx.doi.org/10.12998/wjcc.v9.i24.7245>

## INTRODUCTION

Bronchogenic cysts (BCs) are congenital foregut-derived aberrations of the respiratory tract in the process of embryonic development[1]. They are usually located in the mediastinum and pulmonary parenchyma, and rarely occurred in the retroperitoneum. In most cases, retroperitoneal bronchogenic cysts (RBCs) occur near the left adrenal gland or pancreas[2].

In dual energy computed tomography (CT), material decomposition and quantification depend on photoelectric effect and Compton effect in different elemental compositions generated by X-ray[3]. Dual layer detector spectral CT (also known as spectral CT) is a new detector-based dual energy CT for clinical use, in which the top and bottom layers of detector absorb low- and high-energy photons, respectively. Owing to the technology of novel dual layer detector, both conventional images and energy images reach perfect temporal and spatial registration. Spectral images are comprised of virtual mono-energetic images (VMI), iodine density map, Z-effective (Z-eff) map, and so on[4]. Spectral imaging has widely used in the study of abdominal diseases. It was reported that the optimal energy level of VMI could decrease pseudo-enhancement effect of renal cysts in *in vivo* and *in vitro* experiments based on spectral imaging[5,6]. Other spectral parameters such as iodine density value and Z-effective value were also used to differentiate cystic lesions from solid lesions[7,8]. Currently, there is no report about imaging manifestations of suprarenal BCs based on spectral CT imaging. Herein, we report the first case of suprarenal BC found on the spectral CT and review cases of RBCs in English-language publications.

## CASE PRESENTATION

### Chief complaints

A 17-year-old girl was referred to Union Hospital (Huazhong University of Science and Technology, Wuhan, Hubei Province, China) with epigastric pain.

### History of present illness

The patient had epigastric pain for a week without back pain or other symptoms.

### History of past illness

The patient had no previous history of hypertension or endocrine-related disease.

### Personal and family history

The patient was in good health in the past and had no family history.

**Physical examination**

Routine and specialized physical examination of the patient showed no obvious abnormalities.

**Laboratory examinations**

Except erythrocyte count was slightly decreased, other laboratory results of the patient were within the normal limits.

**Imaging examinations**

Spectral images showed a 29 mm × 17 mm × 28 mm ovoid, well-defined, and homogeneous cystic lesion in the left adrenal area. The lesion was slightly enhanced on conventional images while no enhancement on VMI<sub>40KeV</sub>. In coronary plane of VMI<sub>40KeV</sub>, the lesion margin was clear with significant contrast. The iodine density value of the lesion was 0.001 mg/mL and the Z-eff value was 7.25. Magnetic resonance imaging revealed a cystic mass with intermediate signal intensity on T1-weighted imaging (T1WI) and high signal intensity on T2-weighted imaging (T2WI). Chemical shift imaging revealed no fat component within the lesion (Figure 1).

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**FINAL DIAGNOSIS**

The histopathological characteristics of the lesion are compatible with BC. On gross examination, the cystic lesion was about 30 mm in size. Upon sectioning, the lesion contained gray yellowish fluid. The cystic wall with a smooth inner surface measured about 1-2 mm in thickness. The cyst contained pseudostratified ciliated columnar epithelium and mature hyaline cartilage (Figure 2).

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**TREATMENT**

To confirm the diagnosis, a retroperitoneal laparoscopic resection was carried out. During the surgery, a 2.0 cm skin incision was made in the mid-axillary line 1.5 cm above the iliac crest and a laparoscope was placed through a 10 mm trocar in this site. Two additional trocars (5 mm and 10 mm) were placed in the anterior and posterior axillary line below the inferior margin of the twelfth rib, respectively. The patient was placed in the left lateral position with the kidney rest elevated. The angle between the long axis of the patient's upper trunk and long axis of the table was 20°-30°. And surgeons were in the dorsal side of the patient close to buttocks. Meticulous dissection and fine operation technique were applied to avoid the injury to the peritoneum and vessels. Intraoperatively, a cystic lesion with a smooth surface was found in the left retroperitoneum. The cystic wall was completely resected after intracystic fluid was suctioned.

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**OUTCOME AND FOLLOW-UP**

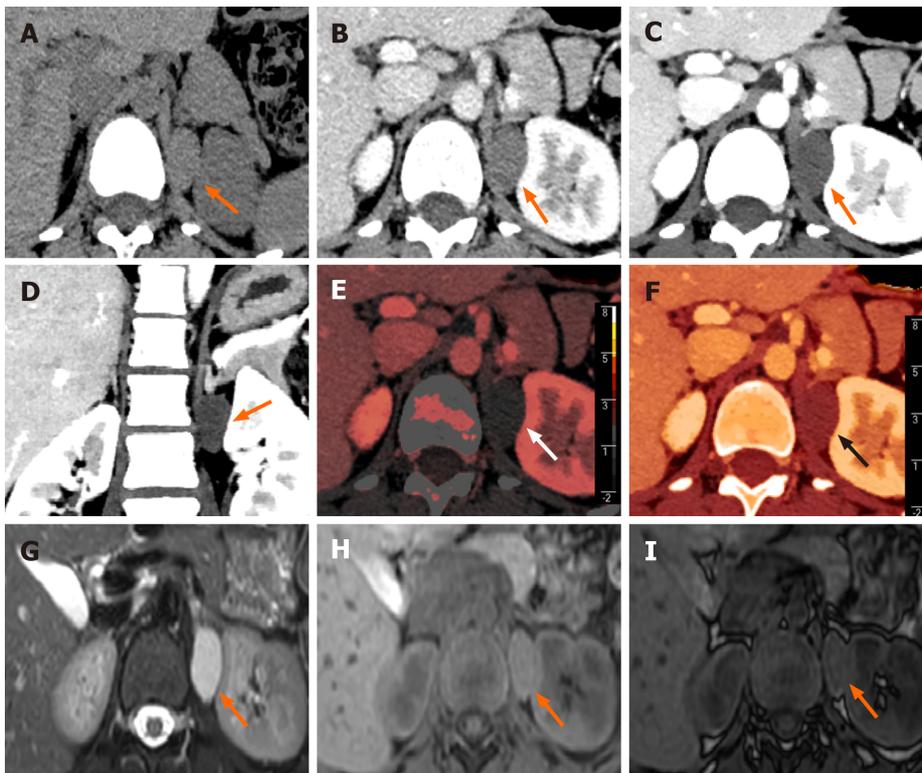
The patient recovered uneventfully without signs of recurrence during a 10-mo follow-up period.

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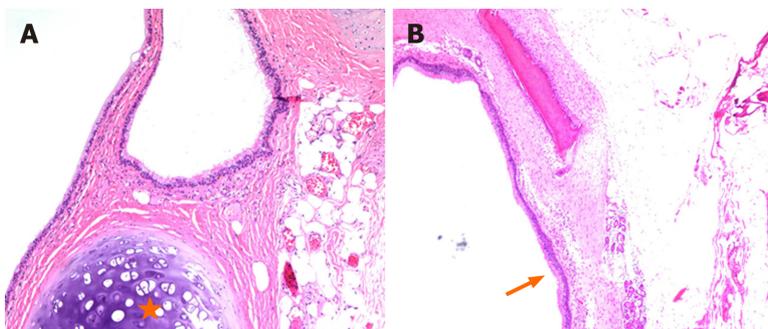
**DISCUSSION**

BCs are benign malformation as a result of the abnormal budding of the foregut at the 3<sup>rd</sup>-7<sup>th</sup> weeks of the embryogenesis. When the connection between bronchial buds and tracheobronchial tree completely separates, the buds may migrate to unusual locations such as neck, intraspinal, pericardiac, and subdiaphragm[9,10]. A search on PubMed database revealed 64 publications of RBCs published worldwide in the English literature. After screening the full texts and pathological results, 48 publications reporting on 50 cases of RBCs were eventually yielded.

RBCs tend to be found on the left side of the abdomen. About half of them were located in the suprarenal area. According to Rud *et al*[2], the left pericardioperitoneal canal shuts later and is larger than the right one, which provides an explanation for the predominantly left side location of RBCs. The mean age of these patients was 39.1 ±



**Figure 1 Spectral computed tomography imaging and magnetic resonance imaging in a patient with a bronchogenic cyst in the left suprarenal region.** A: Axial conventional plain image showing a 29 mm × 17 mm × 28 mm well-defined, ovoid, homogeneous lesion in left adrenal area with attenuation of 46 Hounsfield unites (Hu); B: Contrast-enhanced computed tomography (CT) image showing the lesion with slightly enhancement in the venous phase, and CT value was 58 Hu; C: Axial virtual mono-energetic (VMI<sub>40keV</sub>) image showing the lesion without enhancement in the venous phase, and CT value was 47 Hu; D: Coronal VMI<sub>40keV</sub> image showing an obvious lesion with a clear margin; E and F: Spectral images showing an ovoid lesion, in which iodine density value was less than 0.001 mg/mL and Z-effective value was 7.25; G and H: Axial magnetic resonance imaging images showing a lesion with intermediate signal intensity on T1-weighted imaging (T1WI) and hyperintense signal on T2-weighted imaging; I: Opposed-phase image showing the lesion without obvious signal loss compared to T1WI.



**Figure 2 Pathological appearance of a bronchogenic cyst in the left suprarenal region (hematoxylin and eosin).** A: Cystic wall parenchymal portion containing mature hyaline cartilage (asterisk, × 100); B: The cystic lining composing of a thin layer of pseudo-stratified, ciliated, and columnar epithelium (arrow, × 40).

15.2 years. The mean size of cysts was 5.9 ± 3.2 cm. Slightly over half of patients had been identified incidentally without related symptoms. Of symptomatic patients, the majority complained of nonspecific epigastric pain and a small number complained of thoracic pain and back pain. To relieve clinical manifestations and reduce the risk of complications and malignant change, surgical excision is recommended. And laparoscopic resection has been widely used to lessen economic burden as well as postoperative pain of patients[11]. More than half of patients with RBCs underwent laparoscopic resection and the majority were free of complications during the postoperative course.

BCs has ciliated, pseudo-stratified, columnar epithelium with parenchyma containing any one as follows: Seromucous glands, smooth muscle, or cartilage[12]. Ultrasonography generally shows a cystic hypo/isoechoic lesion. On conventional

images, RBCs are round or fusiform, well-circumscribed, and hypodense lesions with slight enhancement or without enhancement. RBCs usually manifest low or intermediate signal intensity on T1WI and high signal intensity on T2WI. In our case, increased attenuation on CT and increased signal intensity on T1WI suggested proteinaceous fluid. Some cases in the literature also reported this imaging appearance[13, 14]. Fat components were rarely seen in BCs. Most RBCs manifested as no signal loss on the opposed-phase imaging or no signal decrease on T1WI with fat suppression, which was similar to the appearance of our case[15].

We report the first case of RBC found on spectral CT. VMI are reconstructed by both high-energy and low-energy data sets of spectral imaging, which overcome pseudo-enhancement of cystic lesion caused by beam-hardening artifacts[5,6,16]. Also the image noise of whole spectrum is stable and relatively low. Therefore, the image quality of low-energy VMI is significantly improved. In our case, the lesion had slight enhancement on conventional images, while pseudo-enhancement of cystic lesion was eliminated and lesion conspicuity and margin delineation were more pronounced on VMI<sub>40KeV</sub>. On iodine density map, iodine remain or uptake in specific blood vessels, organs, or lesions could be quantified using region of interest method[4]. The extremely low iodine density of the lesion indicated a lack of cellular components, which excluded solid tumors with iodine uptake[7]. Z-eff values depend on Compton effect and photoelectric effect of the material in the process of spectral decomposition [4]. The Z-eff value of the lesion was 7.25, equal to that of fluid material reported on a phantom research of spectral imaging[8], which suggested that it was full of fluid contents.

The retroperitoneal laparoscopic resection was applied in our case. During the operation, the cystic wall was completely resected after intracystic fluid was suctioned. With thorough preoperative imaging and laboratory examinations, benign cystic lesions would be diagnosed confidently in the suprarenal region. Comparing to the laparoscopic surgery performed on other adrenal lesions, the surgery of RBCs has a clearer surgical field as well as shorter operative time.

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## CONCLUSION

BCs are benign congenital foregut malformations caused by abnormal migration of bronchial buds. We report the first case of suprarenal BCs found on spectral CT. Radiological examinations play a significant role in diagnosis of suprarenal BCs and spectral images offer additional spectral parameters. Accurate preoperative diagnosis of RBCs based on thorough imaging examinations is beneficial to the operation of laparoscopic resection.

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