A Rare Case of Retroperitoneal Bronchogenic Cyst: a case report

MEI-CHING HUANG¹ LIANG-KUANG CHEN¹,⁴,⁵ TAI-NEIN LU¹ THOMAS I. S. HWANG³ CHIN-CHENG LEE²

Department of Diagnostic Radiology¹, Department of Pathology², Department of Uro-surgery¹, Shin Kong Wu Ho-Su Memorial Hospital
School of Medicine², Fu Jen Catholic University
Department of Radio-Technologist², Yuan-Pei University

We reported a 54-year-old asymptomatic man who received PET-CT scan for physical health examination at our hospital. In PET-CT scan, we incidentally found a left suprarenal tumor located adjacent to diaphragmatic crus with almost no FDG activity. This tumor was cystic in nature and slightly enlarged in size on CT scan one year later. Laparoscopic adrenalectomy and resection of this suprarenal tumor were performed. Histopathologic examination revealed a thin-walled cystic mass lined by ciliated pseudostratiﬁed columnar epithelium and smooth muscle ﬁber which was diagnosed as bronchogenic cyst.

Bronchogenic cyst is regarded as a congenital abnormality of the primitive foregut. The most common location is in the mediastinum, particularly posterior to the carina; however, cervical, cutaneous, diaphragmatic, gastric, abdominal, and retroperitoneal bronchogenic cysts have all been described in English literatures [1].

Subdiaphragmatic bronchogenic cysts are unusual and that reported in English literatures have a tendency to locate on the left of midline. The most common location was within a triangle behind the stomach deﬁned by the midline, the splenic vein, and the spleen/diaphragm [1]. If a cystic tumor locates in this region, bronchogenic cyst should be listed in the differential diagnosis. Contrast-enhanced CT scan or MR image can be useful for differentiating hyperattenuating cysts from soft tissue masses [2].

CASE REPORT

A 54-year-old asymptomatic man underwent physical health examination at our hospital, during which the whole body PET-CT scan revealed a suprarenal incidentaloma. The patient appeared to be normal in physical examination. The blood pressure was 104/64 mmHg; the pulse rate was 68 beats per minute and regular. The laboratory data including complete blood count, electrolytes, blood chemistry and tumor marker were all within normal limits. The 24-h urinary VMA was also within normal range. PET-CT scan demonstrated an iso-dense mass at left suprarenal region, measuring around 2.9 cm in diameter, with almost no FDG activity detected (Fig. 1). Small calcification over the posterior border of this mass lesion was noted. Six months later, follow up abdominal CT scan revealed a 5.5 × 3.3 × 3.1 cm homogenous, iso-dense mass at left suprarenal region adjacent to crus of diaphragm. It was well-demarcated and slightly lobulated in border with focal marginal calcified spot. The average CT number was around 55 HU. No enhancement was noted after intravenous administration of contrast material. The surrounding fat was clear (Fig. 2). Keep following up one year later, sonography demonstrated an isoechoic mass with posterior enhancement at left suprarenal region, measuring around 2.9 cm in diameter, with almost no FDG activity detected (Fig. 1). Small calcification over the posterior border of this mass lesion was noted. Six months later, follow up abdominal CT scan revealed a 5.5 × 3.3 × 3.1 cm homogenous, iso-dense mass at left suprarenal region adjacent to crus of diaphragm. It was well-demarcated and slightly lobulated in border with focal marginal calcified spot. The average CT number was around 55 HU. No enhancement was noted after intravenous administration of contrast material. The surrounding fat was clear (Fig. 2). Keep following up one year later, sonography demonstrated an isoechoic mass with posterior enhancement at left suprarenal area, which was proposed to be a complex cystic lesion (Fig. 3). At the same time abdominal CT scan revealed that the tumor was still cystic in nature and slightly enlarged in size as compared with previous study. Due to progressive enlargement of the tumor, the patient underwent...
laparoscopic adrenalectomy for resection of the suprarenal tumor. Grossly, the tumor was a cystic lesion measuring $4 \times 3 \times 1.2$ cm in size and located superior to left adrenal gland. The tumor was separated from the adrenal gland and any other organs. Histologically, the cyst was found to be lined by ciliated pseudostratified columnar respiratory epithelium and smooth muscle fiber (Fig. 4). These findings were consistent with a bronchogenic cyst. There was no evidence of malignancy. Neither pulmonary parenchyma nor teratomatous components could be identified. The postoperative recovery was smooth and the patient was discharged 5 days after operation.

**DISCUSSION**

Bronchogenic cysts are regarded as congenital foregut malformations. Histopathologically, they are lined by ciliated pseudostratified columnar epithelium. The wall often contain cartilage and bronchial mucus glands [3]. The most common location is in the mediastinum, particularly posterior to the carina; however, cervical, cutaneous, diaphragmatic, gastric, abdominal, and retroperitoneal bronchogenic cysts have all been described in English literatures [1]. They originate from an abnormal budding of the tracheobronchial anlage of the primitive foregut during the 21st to 50th days of development. The cyst is usually associated with the tracheobronchial tree or the esophagus, if it attaches to the primitive foregut persistently. However, if complete separation occurs, the cyst may develop in other unusual locations mentioned above, presumably by migration [4]. People has assumed that a retroperitoneal bronchogenic cyst could result from the pinching off of an abnormal

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**Figure 1.** Positron emission tomography demonstrating almost no FDG activity of this suprarenal tumor.

**Figure 2.** A. Precontrast, and B. Postcontrast enhanced computed tomographic scan demonstrating a sharply marginated mass with focal marginal calcified spot (arrow) at left suprarenal region adjacent to left crus of diaphragm. No enhancement was obtained after intravenous administration of contrast material.
bud of the primitive foregut, with subsequent migration into the abdomen before fusion of the caudal pleuroperitoneal membranes that separate the pleural cavity from the peritoneal cavity. But if diaphragmatic closure has already happened, a bronchogenic cyst may become dumbbell shaped, straddling the diaphragm [5, 6].

Liang et al had reviewed the articles of the subdiaphragmatic bronchogenic cysts in 2005: there were 70 citations (48 English and 22 non-English) describing bronchogenic cysts below the diaphragm and only 38 cases reported in English literatures met the histopathologic criteria of bronchogenic cyst. These reported cases had a wide range in age from prenatally to 67 years-old with slightly male predominance. The most common presenting symptom was flank or abdominal pain (52%). It was not uncommon for these lesions to be found incidentally [1]. They are often asymptomatic, unless they become secondarily infected, perforated, or large enough to have mass effect to adjacent organ [7]. For example, if the tumor located in periadrenal region, it could be presented with mild elevations of metanephrine and catecholamine due to mass effect. Subdiaphragmatic bronchogenic cyst has a tendency to locate on the left of midline (82%). The most common location was within a triangle behind the stomach defined by the midline, the splenic vein, and the spleen/diaphragm. The malignant potential in subdiaphragmatic bronchogenic cyst is probably low [1].

Bronchogenic cysts typically manifest as well-demarcated masses of soft tissue or water attenuation on CT scan. It has variable CT density from +10 to +120 HU or even higher. The wall of cyst is thin and smooth. The cyst may contain milk of calcium or wall calcifications. The content within bronchogenic cyst is often mixed with water and proteinaceous mucus, which explains the variability of attenuation seen on CT scans and signal intensity characteristics on MR images. The cysts which presented soft-tissue attenuation on CT scans were hyperintense to CSF on T2-weighted MR images and isointense to skel-

Figure 3. Abdominal sonography showing an isoechoic mass with posterior enhancement at the left suprarenal region.

Figure 4. A. The cyst lining composed of ciliated pseudostratified columnar epithelium and smooth muscle fibers (H&E 40X). B. High power view of ciliated pseudostratified columnar epithelium (H&E 400X).
et al muscle on T1-weighted MR images. This finding suggests that mucus and proteinaceous debris, but not calcium, within the cyst is the most likely explanation for the increased attenuation seen on CT scans. McAdams et al believed that MR imaging can be useful for suggesting the true cystic nature of the lesion in difficult cases that could not be confidently diagnosed with CT scan, either because of internal heterogeneity, high attenuation numbers, streak artifact, lack of mural enhancement, or atypical location [2, 8].

The differential diagnosis of a retroperitoneal bronchogenic cyst can be classified as either neoplastic or nonneoplastic. Neoplastic lesions comprise cystic lymphangioma, mucinous cystadenoma, cystic teratoma, cystic mesothelioma, mullerian cyst, and cystic change in solid tumors. Nonneoplastic lesions comprise pancreatic pseudocyst, nonpancreatic pseudocyst, lymphocele, urinoma, and hematoma [8].

Surgical excision is recommended for retroperitoneal bronchogenic cysts to establish definite diagnosis, alleviate any symptoms, and prevent complications such as infection and risk of malignant change [9].

The retroperitoneal bronchogenic cysts are extremely rare and difficult to diagnosed preoperatively. Administration of intravenous contrast material at CT scan or MR image can be useful for differentiating hyperattenuating cysts from soft tissue masses. Although rare, these cysts should be considered in differential diagnosis of retroperitoneal tumors, especially for a cystic tumor in the region of the left adrenal gland.

**REFERENCES**

罕見之後腹腔支氣管原性囊腫：病例報告

黃眉菁 1 陳良光 1,4,5 盧大年 1 黃一勝 3 李進成 2 蘇誠道 1

新光吳火獅紀念醫院 放射診斷科 1 病理檢驗科 2 泌尿外科 3
輔仁大學 醫學系 4
元培科學技術學院 放射技術學系 5

支氣管原性囊腫（bronchogenic cyst）是原生於前腸的先天性異常，通常發生在縱隔腔，偶爾會有病例報告在頸部、皮下、橫膈膜、食道、胃、腹部及後腹腔。我們在此報告一位 54 歲沒有症狀的男性，在接受正子斷層造影的健康檢查時發現左腎上方緊貼橫膈腳有一腫瘤，經過手術切除及術後組織學檢查證實為支氣管原性囊腫。