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Case Report

Multilocular bronchogenic cyst of the bilateral adrenal: report of a rare case and review of literature

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Abstract: Purpose: Bronchogenic cysts are rare benign congenital anomalies, originating from the embryonic foregut ventral segment. Adrenal bronchogenic cyst is a rare form of this anomaly. One extremely rare case of bilateral adrenal multilocular bronchogenic cyst in our hospital was reported and the relevant literatures were reviewed. Significant findings: A 51-year-old man suffered from an intermittent vague headache, fatigue and hypertension history for 2 years, which were gradually worsened in a week. Imaging tests showed bilateral suprarenal mass and left renal cysts. After underwent two retroperitoneal laparoscopic adrenal gland tumor separately, they were all proved to be both the multilocular bronchogenic cyst located in bilateral adrenal gland by histopathological examination. Conclusions: This report confirms the bronchogenic cyst that can be involved bilateral joint in the adrenal gland. And we demonstrated retroperitoneoscopic surgical management is effective in the treatment of the disease.

Keywords: Bronchogenic cyst, multilocular, bilateral, adrenal, retroperitoneum

Introduction

Bronchogenic cysts arise from developmental abnormal of the tracheobronchial tree in the early embryologic foregut period [1]. Most commonly they can be found in the posterior mediastinum, rarely, they located in the adrenal gland [2]. Additionally, this located bilateral adrenal is very exceptional. Searching relevant literatures was performed by using the following search strategy: (bronchogenic cyst) AND (adrenal or retroperitoneal). The 82 literatures of retroperitoneal bronchogenic cysts through our search of Medline were reviewed, of which none case about bilateral adrenal has been reported up to now. We describe an extraordinary case of bilateral adrenal multilocular bronchogenic cyst concurrent bilateral renal cysts and discuss the cases of reported in English.

Case report

This case was obtained from the hospital consultation files of the department of urology, West China Hospital of Sichuan University. This study was approved by the institutional ethics committee at the West China Hospital of Sichuan University, patient consent and conducted in accordance with the ethical guidelines of the Declaration of Helsinki. Clinical information and radiological details were obtained from the case files and electronic medical record files. Two senior pathologists reviewed all original slides of hematoxylin-eosin (HE) staining from the present case. And the relevant literatures were reviewed by authors.

We report a 51-year-old male patient with complaint of an intermittent vague headache, fatigue and hypertension history for 2 years and condition worsen for 1 week. No unusual family history was presented. The physical examination was unremarkable. Multiple test after admission included hypertension and a low serum potassium level (2.7-3.1 mmol/L). Endocrine tests for adrenal hypersecretion were negative. Chest radiography showed no abnormality. An abdominal ultrasonography from another hospital revealed bilateral suprarenal mass and the left renal cysts.

In order to understand his complaint more thoroughly, subsequently contrast enhanced computerized tomography (CT) of the abdomen was underwent. The examination revealed the fol-

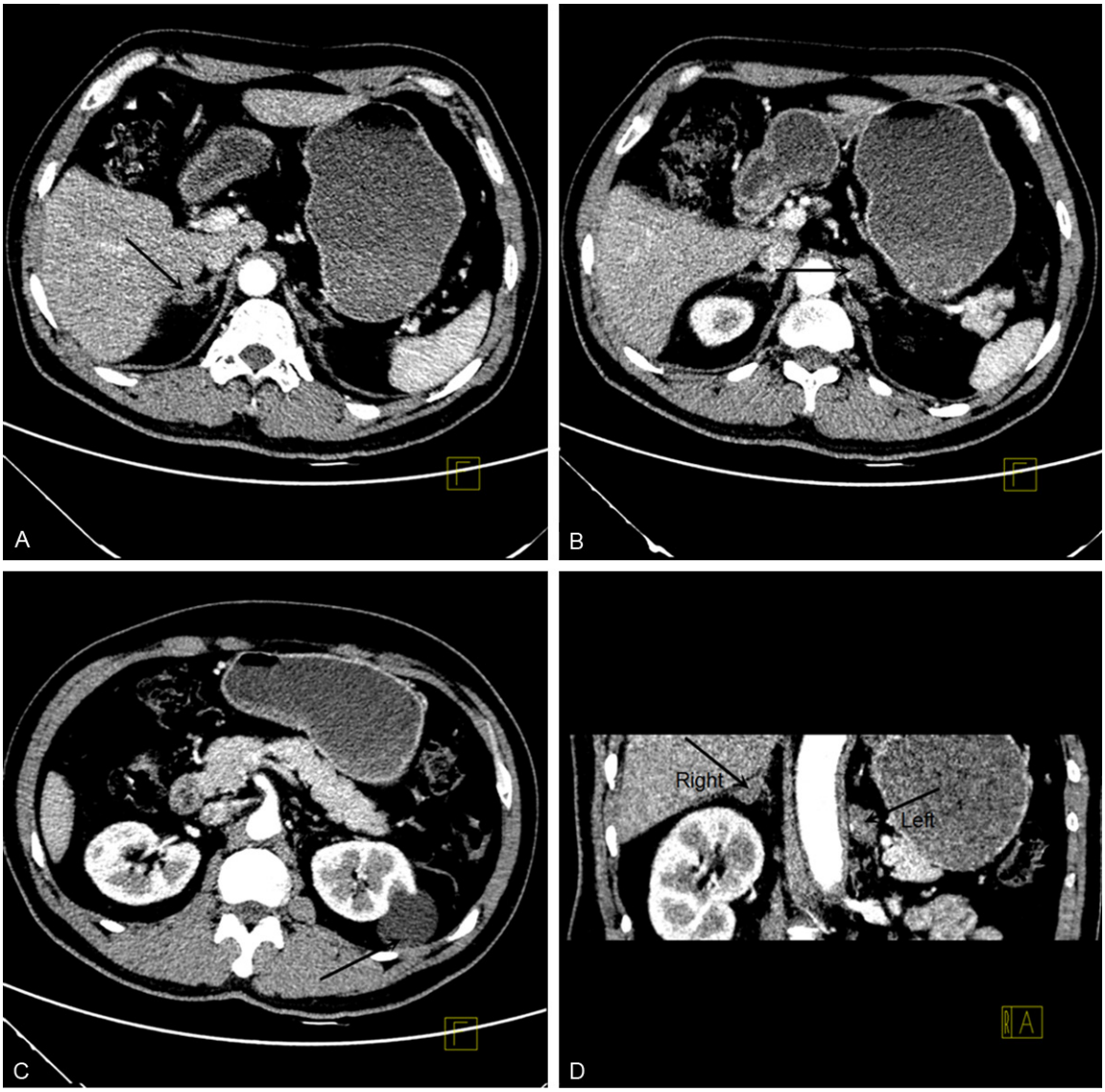

Figure 1. CT scan showing a right adrenal mass, about 1.5×0.9 in diameters (black arrow point to mass) (Panel A, Axial view). CT scan showing a left adrenal mass, about 1.7×2.1 cm in diameters (black arrow point to mass) (Panel B, Axial view). CT scan showing a left, rounded renal cyst, about 4.5 cm in diameters (black arrow point to renal cyst) (Panel C, Axial view). CT scan independently showing a left and right adrenal mass (black arrows point to the mass) (Panel D, Oblique coronal view).

lowings: 1. A soft tissue density nodular shadows are showed independently in the right and left adrenal glands, about 1.5×0.9 and 1.7×2.1 cm in diameters, respectively, which were highly suggestive of adenoma. 2. A rounded, low density area was seen within the left renal parenchyma. The area was about 4.5 cm in diameters without contrast enhancement, and the suggestion was renal cysts (**Figure 1A-D**).

Because of the occupied effect of uncertain entity and the renal cyst of the greater were also obvious in the left. After communicated with the patient and family, subsequently, the left side of adrenal tumor and the renal cyst unroofing were preferentially managed by retroperitoneal laparoscopic surgery as diagnostic treatment. However, the histopathology revealed adrenal multilocular bronchogenic cyst (**Figure 2A-C**). The operation was smooth. Unfortunately, the patient's symptoms showed no markedly improvement after surgery. The first 6 months after previous surgery, retroperitoneal laparoscopic right adrenal area mass

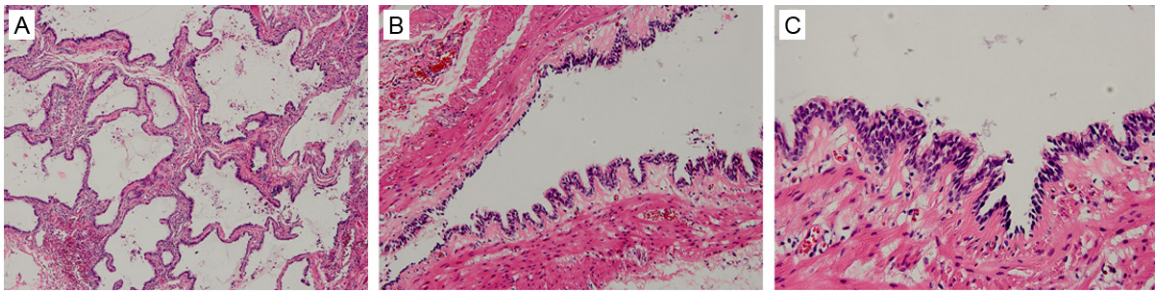

Figure 2. Histopathologic section in resection of the left adrenal revealed multilocular cystic lesions, and the cyst wall lined with ciliated columnar epithelium, smooth muscle bundles, seromucous glands. (Hematoxylin and eosin stain, original magnification, Panel A, ×100; Panel B, ×200 and Panel C, ×400).

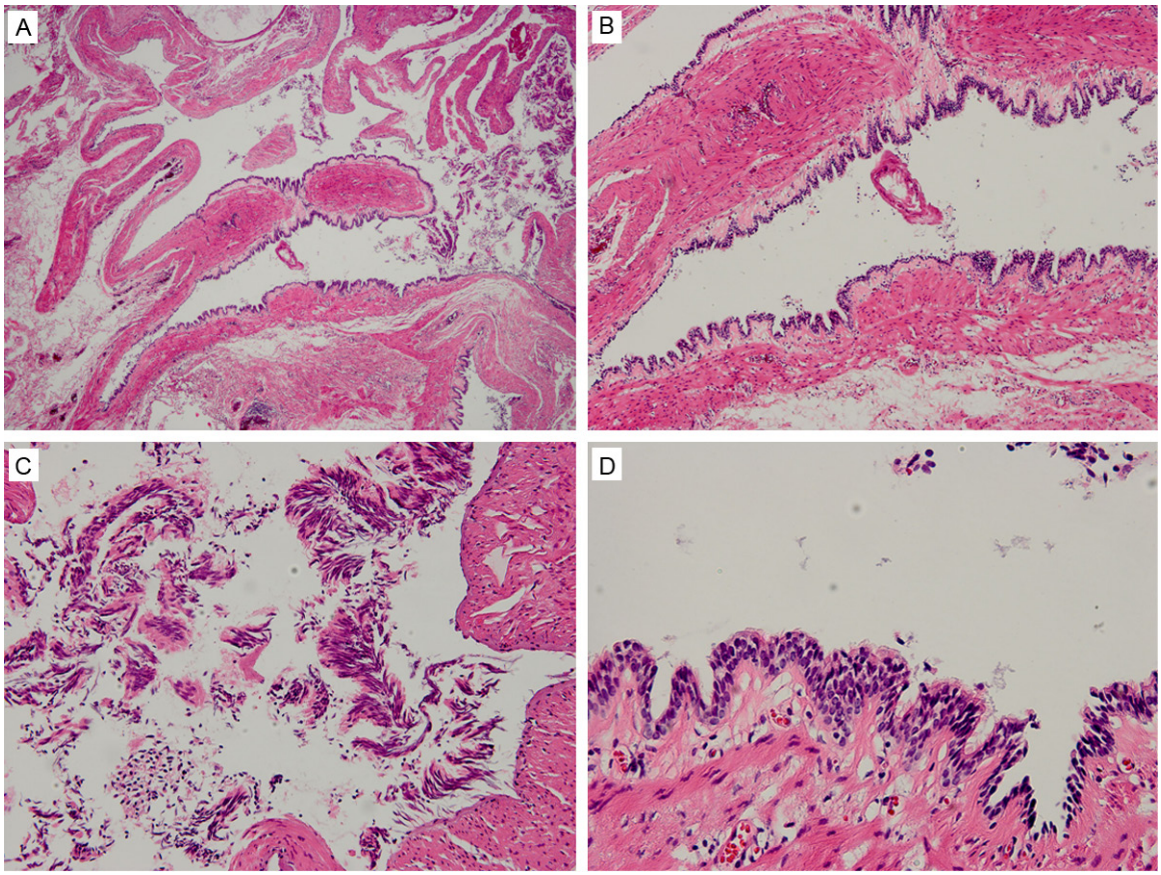

Figure 3. Histological section in resection of the right adrenal revealed ciliated columnar epithelium and part of the cyst wall containing smooth muscle bundles, seromucous glands. (Hematoxylin and eosin stain, original magnification, Panel A, ×40; Panel B, ×100; Panel C, ×200 and Panel D, ×400).

was performed again. To our surprise, the same result applied through histopathology (**Figure 3A-D**).

The operation was successful, and the patient was discharged a week later uneventfully and soon returned to work.

Discussion

Bronchogenic cysts are usually benign, can occur at any age, congenital anomalies that arising outside the thoracic cavity is rare [3-5]. Through reviewing the related literatures, most retroperitoneal bronchogenic cysts are found

near the left adrenal gland. Our findings agree with the results of previous reported in literature from Govaerts and colleagues [1-5]. In 2009, Liu and partners reported a case of bilateral pulmonary bronchogenic cysts [6]. This is the first reported case of bilateral adrenal multilocular bronchogenic cyst without other congenital anomalies. Nevertheless, the exact prevalence and pathogenesis of adrenal multilocular bronchogenic cysts is still unknown.

Clinical symptoms are due to the location and size of the cyst, and they mostly have no symptom [7]. When patients have symptomatic, it is commonly caused by secondary complications. As normal growth, acute hemorrhage, infection, or perforation, bronchogenic cysts could compress surrounding structures. In present case, perhaps this may cause of phaeochromocytoma-like symptoms due to pressure adrenal gland [8]. Also more subtle symptoms, such as abdominal discomfort and flank pain, could be explained by this mechanism.

As the adrenal space is not lightly accessible for abdominal ultrasonography, the imaging method is not the unique diagnostic test of preference. The best imaging modalities for the exploration of a bronchogenic cyst are CT scan and magnetic resonance imaging (MRI) [9]. The CT features the lesions are commonly mixed cystic-solid masses, sharply defined, homogenous hypoattenuating lesion without enhancement, and possibly with calcification of cyst wall. Additionally, hyperattenuation may suggest the present of thick mucinous, hemorrhage and protein mixture secreted in the lesion [2, 7]. These features can be misdiagnosed as solid masses, as in the present case. In T1-weighted and T2-weighted MRI the cyst usually shows a high-and middle-signal intensity and extremely bright signal, respectively [2, 7]. These features can be misdiagnosed as solid masses. Unfortunately, MRI was not performed in this case.

Reviewed related literatures, for cyst is located the adrenal gland, an endocrine differential tests is often considered. However, it does not help the diagnostic process, in that it is practically impossible to exclude the presence of a neo-plastic disease, surgery is recommended [2, 7]. In this case, and endocrine workup was performed, and no abnormality was indicated.

The preoperative diagnosis of adrenal bronchogenic cysts remains challenging. They are usually easily misdiagnosed as non-neoplastic such as adrenal adenoma or adrenal cyst and neoplastic such as pheochromocytoma or adrenal cortical carcinoma [2, 10]. Despite the rarity of this pathologic entity, bronchogenic cysts should be considered in the differential diagnosis of adrenal lesions. Unfortunately, only histology can provide the definitive diagnosis.

In our case, the cysts were diagnosed incidentally during clinical examination. The bronchogenic cysts were independently found in the left and suprarenal region, mimicking adrenal tumour. Additionally, renal cysts were detected by imaging tests. The hypertension and hypokalemia detected in this patient led to a possible diagnosis of pheochromocytoma; however, endocrinological evaluation revealed the cysts to be non-secreting. On the basis of clinical and radiological features, the adrenal bronchogenic cysts can easily be misdiagnosed as an adrenal cystic degenerated adenoma. Therefore, the histological diagnosis for bronchogenic cyst is usually essential.

Surgical resection is recommended treatment of a symptomatic or asymptomatic bronchogenic cyst to confirm the diagnosis, relieve symptoms and prevent further complications [11]. Chung et al. recommend retroperitoneal laparoscopic operation as an effective and safe procedure for the management of retroperitoneal bronchogenic cysts [12]. In our case, we successfully performed this method and our patient experienced a stable rehabilitation. We also reviewed the relevant literatures on the medical prognosis that the postoperative outcome is excellent with no report of recurrence. Latest follow-up results demonstrated no signs of relapse in this case.

In conclusion, although very rare, bronchogenic cysts should be considered in the differential diagnosis of an adrenal mass. Retroperitoneoscopic surgical removal is recommended to establish the diagnosis and treatment. The postoperative outcome is excellent, with no report of recurrence.

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Disclosure of conflict of interest

None.

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