

# Retroperitoneal Multilocular Bronchogenic Cyst Adjacent to Adrenal Gland

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## Abstract

Bronchogenic cysts are generally found in the mediastinum, particularly posterior to the carina, but they rarely occur in such unusual sites as the skin, subcutaneous tissue, pericardium, and even the retroperitoneum. A 30-year-old Korean man underwent surgery to remove a cystic adrenal mass incidentally discovered during routine physical checkup. At surgery, it proved to be a multilocular cyst located in the retroperitoneum adjacent to the left adrenal gland. Microscopically, the cyst was lined by respiratory epithelium over connective tissue with submucous glands, cartilage and smooth muscle, thereby histologically confirming bronchogenic cyst. This is the first reported case of retroperitoneal bronchogenic cyst in an adult without other congenital anomalies in Korea.

**Key Words:** Bronchogenic cyst, multilocular, retroperitoneum, CT

## INTRODUCTION

Bronchogenic cyst is a developmental anomaly that results from abnormal budding of the tracheobronchial tree, the condition being closely related pathogenetically to aberrant pulmonary tissue, as well as to congenital intrapulmonary cysts which represent the persistence of bronchial tree continuity. If a mass of developing cells separates from the main tracheobronchial lumen, an extrapulmonary cyst develops and as its epithelial secretion continues, it enlarges by gradual distention.<sup>1</sup> Ninety percent of thoracic bronchogenic cysts are generally found in the posterior mediastinum or in the posterior portion of the superior mediastinum near the tracheal bifurcation.<sup>2</sup> Apart from the thorax, they rarely occur at sites such as skin, subcutaneous tissue and pericardium, but subdiaphragmatic bronchogenic cysts are particularly uncommon with ones in the retroperitoneum being distinctly rare. Bronchogenic cysts

may be spherical, elliptical, or lobulated and they are usually unilocular. A few cases of retroperitoneal bronchogenic cyst had been reported before the availability of computerized axial tomography without mentioning any connection to the adrenal area. Indeed some of the cases reported to occur in the retroperitoneum seemed to be actually intraabdominal, being loosely attached to the superior pancreatic body so that only a few cases presented as an adrenal mass.<sup>3</sup>

To our best knowledge, only 16 cases of subdiaphragmatic bronchogenic cyst have been reported in the literature in English.<sup>4,5</sup> We herein present a truly retroperitoneal multilocular bronchogenic cyst presenting as an adrenal mass.

## CASE REPORT

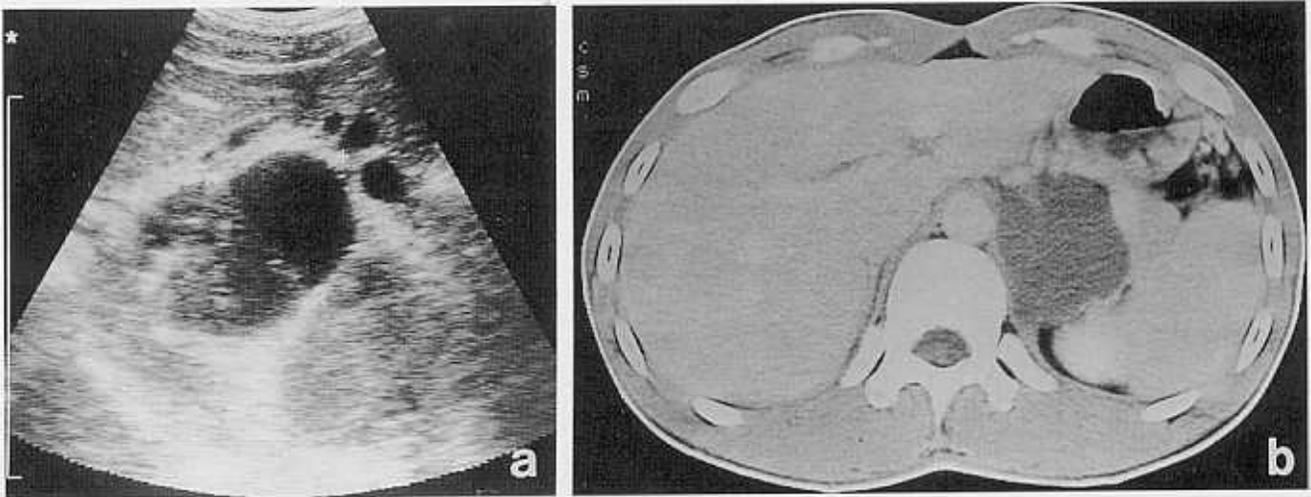
A 30-year-old Korean man was admitted for further evaluation of an adrenal mass which had been fortuitously discovered at roentgenologic examination during a routine physical checkup. He had been in good health without any evidence of chronic disease or anomaly. An abdominal ultrasonography and computed tomography (CT) scan of the abdomen revealed a cystic mass in the left suprarenal area, measuring 7 cm in diameter (Fig. 1a and 1b). Radiologically, it was considered to be a multilocular cystic mass

Received August 24, 1999

Accepted September 27, 1999

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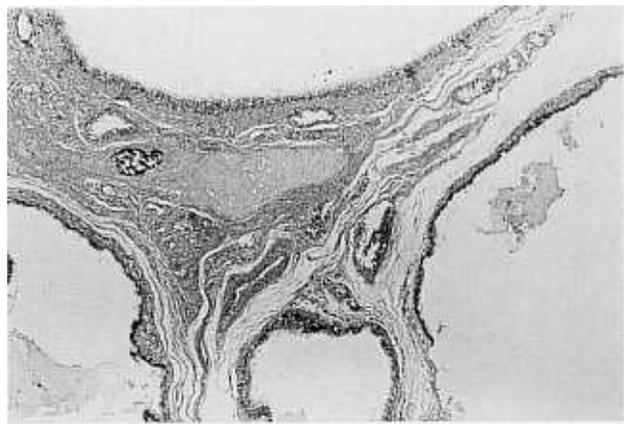


**Fig. 1a.** Longitudinal abdominal ultrasonography shows a  $6.7 \times 4.4$  cm-sized, well-defined, anechoic cystic mass at left suprarenal fossa. Several echogenic septa can be seen within the cystic mass. **b.** Contrast-enhanced CT demonstrates thin-walled cystic mass at left suprarenal fossa compressing adjacent structures without invasion. The enhanced septa are also noted.

probably arising in the left adrenal gland. The liver, spleen, pancreas, and kidney appeared normal. At operation, the cystic mass was separated from the left adrenal gland, left kidney, pancreas, and spleen. It was supplied by both the left renal artery and inferior phrenic arterial branches, suggesting that the mass had originated from the adrenal gland. During operation, it was inadvertently perforated, extruding a dark-green mucoid content, and so cytologic and biochemical analysis of the contents of the cyst was not possible. There was no evidence of communication with or previous rupture into adjacent structures. However the left adrenal gland was only closely apposed to the cystic mass. The mass in continuity with the left adrenal gland was removed. The patient made an uneventful recovery after surgery without any complication.

### Pathologic findings

The resected cystic mass in continuity with the left adrenal gland was dark-gray to brown and soft in consistency. For the most part, it was collapsed weighing 15 gm and measuring  $6 \times 1.5 \times 0.3$  cm. The left adrenal gland was easily detached from the cystic mass and was grossly intact. On section, the cystic mass had numerous small locules of varying sizes containing dark-green mucoid fluid. Each showed a thin membranous wall of even thickness, while



**Fig. 2.** Sections show cystic cavities lined by pseudostratified columnar epithelium resting on fibrovascular connective tissue containing cartilage and seromucinous glands.

the internal surfaces were uniformly smooth. Microscopically, locules were lined by respiratory-type pseudostratified ciliated columnar epithelium resting on fibrous connective tissue with seromucous glands and hyaline cartilages fulfilling the histologic criteria of multilocular bronchogenic cyst (Fig. 2). Neither pulmonary parenchyma nor teratomatous components were present. There was no cytologic atypia in the lining epithelium or stromal tissue components.

## DISCUSSION

Bronchogenic cyst represents one type of foregut cyst that forms from accessory buds that became sequestered from the foregut during embryonic development. Foregut cysts (bronchogenic, esophageal, or gastroenteric) may be located in or near the respiratory tract, GI tract, pancreas, and liver. The most common location of intrathoracic bronchogenic cyst is posterior to the carina.<sup>1</sup> Subdiaphragmatic bronchogenic cysts are rare, especially in the retroperitoneum. Most reported subdiaphragmatic bronchogenic cysts have been located in the peritoneal cavity and have demonstrated an attachment to or a direct intraluminal connection with a portion of the gastrointestinal tract.<sup>6</sup> An embryologic explanation for the occurrence of abdominal bronchogenic cyst has been previously outlined.<sup>3,7</sup> Early in embryonic life, the abdominal and thoracic cavities are connected by the pericardioperitoneal canal. With closure of the pleuroperitoneal membranes which form the future diaphragm, abnormal lung buds of the primitive foregut could be pinched off and trapped, with subsequent migration into the abdomen before fusion of the diaphragm components. Interestingly, about 75% of the reported cases were described to develop on the left side of the retroperitoneal region, as in this case.<sup>5</sup>

In clinical and radiologic views, the retroperitoneal bronchogenic cysts are easily misdiagnosed as adrenal tumor, pheochromocytoma, gastric tumor, enteric cyst, bronchopulmonary sequestration, cyst of urothelial origin, etc. However, a histological diagnosis of bronchogenic cyst from these masses can be easily made.<sup>8</sup>

The differential diagnosis of a retroperitoneal cyst lined by ciliated epithelium includes cystic teratoma, bronchopulmonary sequestration, cyst of urothelial origin and foregut origin.<sup>9</sup> A possible resemblance to extragonadal mature teratomas with lung differentiation must be appreciated, but the overall organization of constituent tissues in bronchogenic cysts resembling those of normal bronchi should suffice in distinguishing between the two lesions.<sup>6</sup> Bronchopulmonary sequestration, although believed to share a common embryologic origin with bronchogenic cyst, possesses lung parenchyma and a pleural investment along with the bronchial elements. Since the bronchus and esophagus originate from the same anlage, the histological difference occurs during the

budding period and developmental state. Among the cysts of foregut origin, those containing cartilage or seromucinous respiratory glands are designated as bronchogenic cysts; those containing two well-developed layers of smooth muscle without cartilage are designated as esophageal cysts; those with none of these distinguishing features are classified as foregut cysts. In contrast, retroperitoneal cysts of urogenital origin may rarely have pseudostratified, columnar ciliated epithelium; however, submucosal mixed serous and mucous glands are not seen.<sup>6,9</sup>

As mediastinal bronchogenic cysts are usually asymptomatic, so are retroperitoneal bronchogenic cysts, unless they are secondarily infected or large enough to cause compression of other vital structures. The cyst shows a tendency to be larger as the patient gets older.<sup>5</sup>

Since there is a common lack of symptoms and specific changes in laboratory findings, radiographic examinations, in particular computerized axial tomogram, have an important role in making the diagnosis. The computerized axial tomogram allows a further differential diagnosis.<sup>4</sup>

The additional differential diagnosis of retroperitoneal cystic mass includes retroperitoneal cystic lymphangiomas, cystic pancreatic tumors, pseudocysts, leiomyosarcomas with cystic degeneration, hematomas, and abscesses.<sup>8</sup>

More than two-thirds of all retroperitoneal tumors are malignant.<sup>9</sup> The congenital foregut cysts are nonneoplastic and malignant transformation is exceptional.<sup>10</sup> Excision is recommended to establish diagnosis and alleviate associated symptoms and for the prevention of further complication.

In summary, an isolated retroperitoneal bronchogenic cyst has been described in an asymptomatic 30-year-old Korean man. Review of the literature suggested that there is a tendency for retroperitoneal bronchogenic cysts to occur in the left side retroperitoneal region. Bronchogenic cyst should be considered in the differential diagnosis of retroperitoneal cystic mass.

## REFERENCES

1. Marks C, Marks P. The embryologic basis of tracheobroncho-pulmonary maldevelopment. *Int Surg* 1987;72:109-14.
2. Keohane ME, Schwartz IRA, Freed J, Dische R. Sub-

- diaphragmatic bronchogenic cyst with communication to the stomach: a case report. *Hum Pathol* 1988;19:868-71.
3. Sumiyoshi K, Shimizu S, Enjoji M, Iwashita A, Kawakami K. Bronchogenic cyst in the abdomen. *Virchows Arch A Pathol Anat Histopathol* 1985;408:93-8.
  4. Buckley JA, Siegelman ES, Birnbaum BA, Rosato EF. Bronchogenic cyst appearing as a retroperitoneal mass. *Am J Roentgenol* 1998;171:527-8.
  5. Itoh H, Shitamura T, Kataoka H, Ide H, Akiyama Y, Hamasuna R, et al. Retroperitoneal cyst; Report of a case and literature review. *Pathol Int* 1999;49:152-5.
  6. Foerster HM, Sengupta EE, Montag AG, Kaplan EL. Retroperitoneal bronchogenic cyst presenting as an adrenal mass. *Arch Pathol Lab Med* 1991;115:1057-9.
  7. Coselli MP, de Ipolyi P, Bloss RS, Diaz RF, Fitzgerald JB. Bronchogenic cysts above and below the diaphragm: report of eight cases. *Ann Thorac Surg* 1987;44:491-4.
  8. Kajiya Y, Nakajo M, Ichinari N, Yamazumi K, Otuji T, Tanaka T. Retroperitoneal foregut cyst. *Abdom Imaging* 1997;22:111-3.
  9. Menke H, Röher HD, Gabbert H, Schweden F. Bronchogenic cyst: A rare cause of a retroperitoneal mass. *Eur J Surg* 1997;163:311-4.
  10. Sullivan SM, Okada S, Kudo M, Ebihara Y. A retroperitoneal bronchogenic cyst with malignant change. *Pathol Int* 1999;49:338-41.
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