

## The First Case of Intraperitoneal Bronchogenic Cyst in Korea Mimicking a Gallbladder Tumor

We present a case of an intraperitoneal bronchogenic cyst located at inferior surface of the liver, next to the gallbladder which clinically mimicked a gallbladder tumor. This is the first case reported in Korea, and we offer reviews of the related literatures. A 48-yr-old woman was admitted to our hospital because of intermittent abdominal pain in right upper quadrant. Computed tomography showed a large mass alongside the gallbladder. During laparotomy, the mass showed an ovoid cystic nature, which was attached to the normal gallbladder and liver bed. Cyst excision with cholecystectomy was performed, and histopathological examination revealed a bronchogenic cyst. Most bronchogenic cysts have a benign nature, but malignant changes have also been reported. Therefore, if a cystic tumor in the abdomen is suspected during preoperative diagnosis, a bronchogenic cyst should be considered in the differential diagnosis.

Key Words : Bronchogenic Cyst; Gallbladder Neoplasms

Kee Hwan Kim, Ji Il Kim,  
Chang Hyeok Ahn, Jeong Soo Kim,  
Young Mi Ku\*, Ok Ran Shin†,  
Eun Jung Lee‡, Keun Woo Lim

Department of Surgery, \*Radiology and †Clinical Pathology, Uijeongbu St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Uijeongbu, Korea

Received : 27 March 2003  
Accepted : 18 August 2003

### Address for correspondence

Jeong Soo Kim, M.D.  
Department of Surgery, Uijeongbu St. Mary's Hospital,  
College of Medicine, The Catholic University of Korea,  
65-1 Gumo-dong, Uijeongbu 480-130, Korea  
Tel : +82.31-820-3048, Fax : +82.31-847-2717  
E-mail : drbreast@catholic.ac.kr

### INTRODUCTION

Bronchogenic cysts are derived from the embryologic bronchial cleft and are mainly of pulmonary origin. They are rarely located in an extrathoracic site, such as subdiaphragmatic retroperitoneal area (1-17). Only a few cases of intraperitoneal area (18-26) have been documented (Table 1). To the best of our knowledge, only 22 retroperitoneal cases have been reported in the world literature by the year of 2001, 17 of which are English language reports (17). Cases arising from an intraperitoneal position are more unusual. Only 8 cases have been reported by the year of 2001. We report upon the first isolated intraperitoneal bronchogenic cyst in a 48-yr-old woman, which was presented as a gallbladder mass in Korea.

### CASE REPORT

A 48-yr-old female was admitted to our hospital with one-year history of dyspepsia after meals and intermittent epigastric pain. A physical examination demonstrated no palpable mass in the abdominal region. White blood cell (WBC) count was at  $5.2 \times 10^9/L$ , and hemoglobin was at 11.9 g/dL. Blood chemistry results were normal and preoperative serum alpha-feto protein (AFP) was also within normal range (0.77 U/mL, normal 0-5 U/mL). Ultrasound sonography showed a cystic

mass adjacent to the gallbladder (Fig. 1). Abdominal CT showed a well defined and circumscribed, cystic mass  $3 \times 2.5$  cm in size at the inferomedial aspect of the gallbladder (Fig. 2). Radiological findings suggested a gallbladder tumor, a teratoma, bronchopulmonary sequestration, a complicated cyst or carcinoma, but the findings were insufficient for an accurate diagnosis to be made. Therefore a presumptive diagnosis of a gallbladder tumor was made. The lesion was explored because CT did not show a definite demarcation between the mass and the neighboring structures, nor did it confirm its isolation in the gallbladder area; moreover, the possibility of malignancy could not be ruled out. At laparotomy, a 3 cm-sized cystic mass was discovered adherent to the gallbladder (Fig. 3). The cyst was dissected from the liver bed, and the entire cyst and gallbladder were excised consequently. There was no connection between cyst and gallbladder. The gross appearance of the resected specimen seemed to be a benign cyst. On opening the specimen revealed one large cystic cavity, which contained thick brownish mucoid fluid (Fig. 4). Microscopically, the cyst is lined by a layer of pseudostratified ciliated columnar epithelial cells occasionally interspersed with goblet cells (Fig. 5). Thus, the cyst was histologically diagnosed as a bronchogenic cyst. The postoperative course was uneventful; the patient was discharged at 10th day post-operatively, and had remained asymptomatic through biweekly follow-ups for two months.

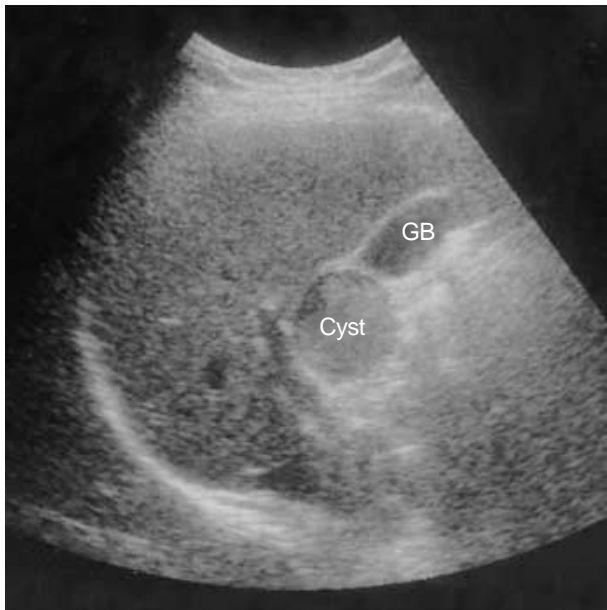


Fig. 1. Sonographic finding showing a well-defined round cystic mass adjacent to the gallbladder, the lesion is filled with echogenic materials.

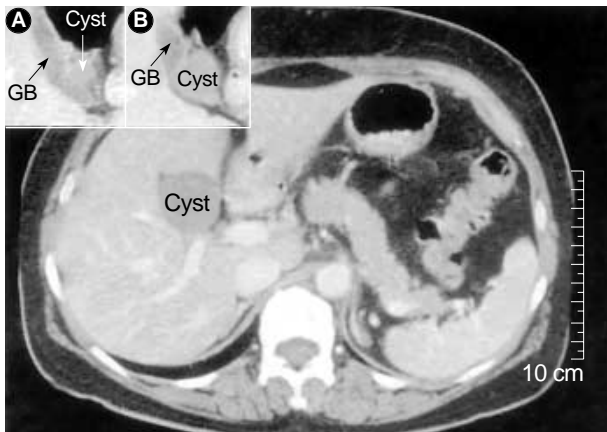


Fig. 2. Post-contrast sequential axial abdominal CT scan shows a well-defined round cystic mass at the inferomedial aspect of the gallbladder. The internal density of the cystic mass appears as a subtle increase than that of the gallbladder.

### DISCUSSION

Bronchogenic cysts are congenital abnormalities arising from the ventral foregut during the third to seventh week of fetal development. They are almost always lined, at least partially, by ciliated cuboidal to pseudostratified columnar epithelium and are often filled with mucus. Bronchial components such as cartilage, smooth muscle, elastic fibers, fibrous tissue and seromucinous glands may all be presented in the cyst wall (27). A retroperitoneal location is rarely reported. Although the exact mechanism is unknown, Sumiyoshi et al. (2) proposed the following theory. During early embryonic

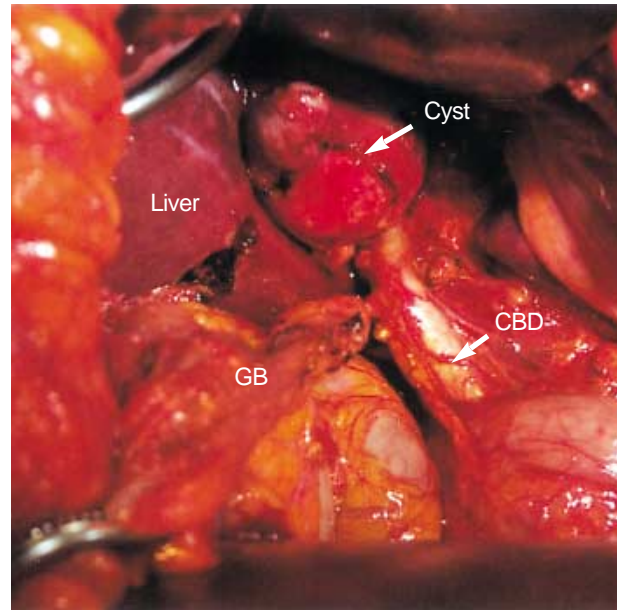


Fig. 3. On operation, the mass is ovoid and cystic and is attached to the normal gallbladder and liver bed.

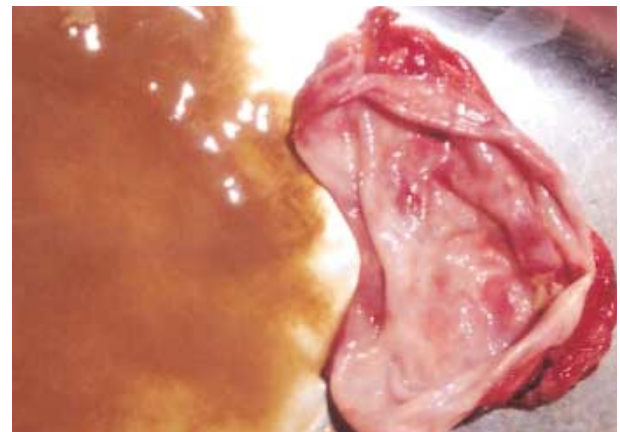


Fig. 4. The cut section of the specimen shows a single large cystic cavity, containing a thick brownish mucoid fluid.

life, the thoracic and abdominal cavities are linked via the pericardio-peritoneal canal. When the canal is later divided by the fusion of the pleuroperitoneal membranes (the future diaphragm), a portion of the tracheobronchial tree may be pinched off and migrate, resulting in a retroperitoneal bronchogenic cyst (2). However, subdiaphragmatic bronchogenic cysts, especially in the intraperitoneal region, are extremely rare. Only 8 cases have been reported in the world literature, and all had their locations adjacent to the stomach. Our case had an unique gallbladder location. To our knowledge, no intraperitoneal cyst arising near the gallbladder had been reported in either the Korean or the English literatures. Of these retroperitoneal bronchogenic cysts, nine cases occurred in

males and eight in females. The age of the patients varied because the cases of smaller cysts were asymptomatic and the masses were incidentally discovered. In the cases of larger cysts, the patients complained of various types of pains in the

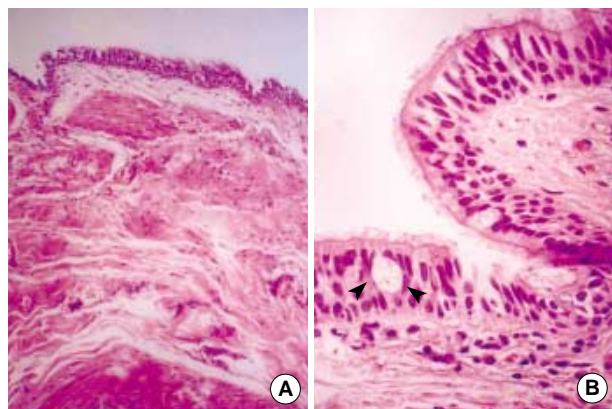


Fig. 5. Cyst lining is composed of respiratory type epithelium, underlying lamina propria, and smooth muscle (A, H&E,  $\times 40$ ). Pseudostratified ciliated columnar epithelial cells are interspersed occasionally with goblet cells (arrow head) (B, H&E,  $\times 200$ ).

suspected region. The size of the cyst showed an increasing tendency with ages of the patients. Table 1 summarizes the eight cases of isolated intraperitoneal bronchogenic cysts that have been reported by the year of 2001. Interestingly, eight cases were located adjacent to the stomach. All of the eight cases were considered to arise in the left side of stomach. Pre-operative clinical diagnosis included the followings; benign tumor (18), leiomyoma or lipoma (19), an intestinal obstruction (20), and a dermoid cyst (24). In our case, which was located beside the gallbladder, apart from stomach, and no connection to stomach and gallbladder wall.

Since there are no common symptoms and specific changes in laboratory findings, CT scan has an important role in making the diagnosis. CT scan is useful for evaluating cyst contents so it allows a further differential diagnosis of retroperitoneal cystic lymphangioma, hematoma, abscess, etc. (28). Bronchogenic cysts usually show high CT values ranging from 30 to 100 HU since the cysts are filled with protein-rich fluid (12).

For histological diagnosis, they should be differentiated from bronchopulmonary sequestration and cystic teratoma. Bronchopulmonary sequestration can be diagnosed by the fact

Table 1. Characteristics of the patients with subdiaphragmatic bronchogenic cysts reported in the English literature

| Author, year                              | Ref. no. | Age (yr)/sex | Size (cm)                      | Site  |
|---|----------|--------------|--------------------------------|---|
| <b>Retroperitoneal cysts</b>              |          |              |                                |   |
| Miller et al., 1953                       | 1        | 10 weeks/F   | 2                              | Anterior site of pancreas   |
| Sumiyoshi et al., 1985                    | 2        | 59/M         | 7                              | Superior body of pancreas   |
| Coselli et al., 1987                      | 3        | 35/F         | 5                              | Superior body of pancreas   |
| Foerster et al., 1991                     | 4        | 35/M         | 10.5 $\times$ 7.5 $\times$ 4.5 | Superior left adrenal gland   |
| Swanson et al., 1991                      | 5        | 4/F          | 2                              | Superior left adrenal gland   |
| Wirbel et al., 1993                       | 6        | 38/M         | 3                              | Superior left adrenal gland   |
| Fischbach et al., 1994                    | 7        | 12/M         | 1.5 $\times$ 1.3               | Right crus of the diaphragm   |
| Ojika et al., 1996                        | 8        | 62/M         | 2.2 $\times$ 1.5               | Right crus of the diaphragm   |
| Harvell et al., 1996                      | 9        | 57/F         | 2.2 $\times$ 1.7 $\times$ 1.5  | Superior body of the pancreas   |
| Resl et al., 1996                         | 10       | 21/M         | 4                              | Superior left adrenal gland   |
| Tokuda et al., 1997                       | 11       | 24/F         | 3                              | Superior left adrenal gland   |
| Menke et al., 1997                        | 12       | 35/M         | 8                              | Superior left adrenal gland   |
| Doggett et al., 1997                      | 13       | 44/M         | 10 $\times$ 10 $\times$ 6      | Adherent to left adrenal gland  |
| Cetinkursun et al., 1997                  | 14       | 20 months/F  | 5                              | Superior pancreatic tail  |
| Itoh et al., 1998                         | 15       | 46/F         | 8 $\times$ 8 $\times$ 7        | Superior left adrenal gland   |
| Sullivan et al., 1999                     | 16       | 55/F         | 10 $\times$ 8 $\times$ 4       | Inferior left adrenal gland   |
| Haddadin et al., 2001                     | 17       | 51/M         | 4.0 $\times$ 3.5               | Superior left adrenal gland I   |
| <b>Intraperitoneal cysts</b>              |          |              |                                |   |
| Dewing et al., 1956                       | 18       | 56/F         | 4 $\times$ 3 $\times$ 3        | Intramural in the posterior wall of the gastric cardia                              |
| Gensler et al., 1966                      | 19       | 46/F         | 6 $\times$ 8                   | Intramural in the gastric curvature of the stomach                                  |
| Pai et al., 1971                          | 20       | 67/M         | 9 $\times$ 2.5                 | Fused with posterior gastric wall proximally  |
| Tanenbaum et al., 1971                    | 21       | 60/M         | 10, 7.6                        | Between the spleen and stomach, intramural in the posterior gastric wall            |
| Benoit, 1972                              | 22       | 37/F         | Not stated                     | Juxtagastric  |
| Murley and Lenz, 1979                     | 23       | 17/M         | Not stated                     | Attached to distal esophagus and adherent to inferior surface of left hemidiaphragm |
| Shireman, 1987                            | 24       | 61/F         | 6                              | Intramural in gastric cardia  |
| Braffman et al., and Keohane et al., 1988 | 25, 26   | 64/F         | 15                             | Communicating with the posterior wall of the stomach                                |
| Present case                              |          | 48/F         | 3 $\times$ 2.5                 | Attached to gallbladder and adherent to inferior surface of liver                   |

that it contains lung parenchyma and pleural tissue. Cystic teratoma has endoderm-origin bronchial tissue and other structures from mesoderm and ectoderm. 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; and those with none of these distinguishing features are classified as foregut cysts (9). In contrast, the cysts of urogenital origin may rarely have pseudostratified ciliated epithelium, and submucosal seromucinous glands (4, 12). In our case, a teratoma was excluded by the absence of tissue, representing the three different germinal layers. In addition, bronchopulmonary sequestration can be diagnosed by the fact that it possesses lung parenchyma, pleural investment, and bronchial elements which were absent in our case.

Preferred treatment of intraperitoneal bronchogenic cyst is surgical removal. Although most are asymptomatic, excision is recommended to establish the diagnosis, alleviate symptoms, and to prevent complications, such as infections and the remote, but documented risk of malignant transformation (16).

Although the occurrence of bronchogenic cyst is rare, it should be considered in the differential diagnosis of an intra-abdominal mass, particularly in the case of a cystic tumor in the region adjacent to the gallbladder.

## REFERENCES

1. Miller RF, Fraub M, Pashuck ET. *Bronchogenic cysts: Anomalies resulting from maldevelopment of the primitive foregut and midgut. Am J Roentgenol Radium Ther Nucl Med* 1953; 70: 771-85.
2. 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.
3. Coselli MP, de Ipolyi P, Bloss RS, Diaz RF, Fitzgerald JB. *Bronchogenic cyst above and below the diaphragm: Report of eight cases. Ann Thorac Surg* 1987; 44: 491-4.
4. Foerster HM, Sengupta EE, Montag AG, Kaplan EL. *Retroperitoneal bronchogenic cyst presenting as an adrenal mass. Arch Pathol Lab Med* 1991; 115: 1057-9.
5. Swanson SJ III, Skoog SJ, Garcia V, Wahl RC. *Pseudoadrenal mass: Unusual presentation of bronchogenic cyst. J Pediatr Surg* 1991; 26: 1401-3.
6. Wirbel RJ, Uhlig U, Kiffner EM, Berger K. [Bronchogenic cyst as a rare differential diagnosis of retroperitoneal tumor.] *Chirurg* 1993; 64: 1056-9.
7. Fischbach R, Benz-Bohm G, Berthold F, Eidt S, Schmidt R. *Infra-diaphragmatic bronchogenic cyst with high CT numbers in a boy with primitive neuroectodermal tumor. Pediatr Radiol* 1994; 24: 504-5.
8. Ojika T, Mukouyama N, Tsuzuki T. [A case of bronchogenic cyst in the subdiaphragmatic region.] *Kyobu Geka* 1996; 49: 505-7.
9. Harvell JD, Macho JR, Klein HZ. *Isolated intra-abdominal esophageal cyst. Case report and review of the literature. Am J Surg Pathol* 1996; 20: 476-9.
10. Resl M, Navratil P, Krajina A. *Retroperitoneal bronchogenic cyst in a young adult. Respiration* 1996; 63: 387-9.
11. Tokuda N, Naito S, Uozumi J, Shimura H, Takayanagi R, Kumazawa J. *A retroperitoneal bronchogenic cyst treated with laparoscopic surgery. J Urol* 1997; 157: 619.
12. Menke H, Röher HD, Gabbert H, Schweden F. *Bronchogenic cyst: A rare cause of a retroperitoneal mass. Eur J Surg* 1997; 163: 311-4.
13. Doggett RS, Carty SE, Clarke MR. *Retroperitoneal bronchogenic cyst masquerading clinically and radiologically as a pheochromocytoma. Virchows Arch A Pathol Anat Histopathol* 1997; 431: 73-6.
14. Çetinkursun S, Öztürk H, Celasun B, Sakarya MT, Surer I. *Isolate abdominal bronchogenic cyst: A case report. Eur J Pediatr Surg* 1997; 7: 103-5.
15. Itoh H, Shitamura T, Kataoka H, Ide H, Akiyama Y, Hamasuna R, Hasui Y, Osada Y, Koono M. *Retroperitoneal bronchogenic cyst: Report of a case and literature review. Pathol Int* 1999; 49: 152-5.
16. Sullivan SM, Okada S, Kudo M, Ebihara Y. *A retroperitoneal bronchogenic cyst with malignant change. Pathol Int* 1999; 49: 338-41.
17. Haddadin WJ, Reid R, Jindal RM. *A retroperitoneal bronchogenic cyst: a rare cause of a mass in the adrenal region. J Clin Pathol* 2001; 54: 801-2.
18. Dewing SB, Roessel CW, Olmstead EV. *Enterogenous cyst of the stomach wall, a rare benign lesion: a case report. Ann Surg* 1956; 143: 131-5.
19. Gensler S, Seidenberg B, Rifkin H, Rubistein BM. *Ciliated lined intramural cyst of the stomach: case report and suggested embryogenesis. Ann Surg* 1966; 163: 954-6.
20. Pai SH, Cameron CT, Lev R. *Accessory lung presenting as a juxtagastric mass. Arch Pathol* 1971; 91: 569-72.
21. Tanenbaum B, Levowitz BS, Ponce M, Manubay S Jr. *Respiratory choristoma of stomach. N Y State Med J* 1971; 71: 373-5.
22. Benoit CG. *Compression de la grosse tuberosité de l'estomac par un kyste bronchogénique. Sem Hop Paris* 1972; 48: 2641-4.
23. Murley GD, Lenz TR. *Bronchogenic cyst, intra-abdominal. Rocky Mt Med J* 1979; 76: 243-4.
24. Shireman PK. *Intramural cyst of the stomach. Hum Pathol* 1987; 18: 857-8.
25. Braffman B, Keller R, Gendal ES, Finkel SI. *Subdiaphragmatic bronchogenic cyst with gastric communication. Gastrointest Radiol* 1988; 13: 309-11.
26. Keohane ME, Schwartz I, Freed J, Dische R. *Subdiaphragmatic bronchogenic cyst with communication to the stomach: a case report. Hum Pathol* 1988; 19: 868-71.
27. Rosai J. *Peritoneum, retroperitoneum, and related subjects. In: Rosai J, ed. Ackerman's surgical pathology. New York: Mosby-Year Book, 1996: 2135-72.*
28. Kajiya Y, Nakajo M, Ichinari N, Yamazumi K, Otuji T, Tanaka T. *Retroperitoneal foregut cyst. Abdom Imaging* 1997; 22: 111-3.