






# Retroperitoneal Bronchogenic Cyst Located in the Presacral Space: A Case Report

## 전천추 부위에 생긴 후복강 기관지 낭종: 증례 보고

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Bronchogenic cysts are rare congenital anomalies that are most frequently found in the mediastinum along the tracheobronchial tree, especially in the posterior aspect of the superior mediastinum. Bronchogenic cysts have also been reported in intrapulmonary, intrapericardial, abdominal, and retroperitoneal locations. Herein, we report a case of a retroperitoneal bronchogenic cyst in the presacral space. The patient was diagnosed based on a post-operative histopathological examination.

**Index terms** Bronchogenic Cyst; Retroperitoneal Space; Computed Tomography, X-Ray

## INTRODUCTION

Bronchogenic cysts originate from the tracheobronchial bud which arises from the embryonic foregut (1). These lesions can be found in or near any organ derived from this embryonic foregut (1). Bronchogenic cysts are usually located in the thorax, but a few cases have been reported as a retroperitoneal mass (2-4). Among them, there is only one report, represented as a retrorectal and lower sacral mass (4), however, to the best of our knowledge, there is no report of the bronchogenic cyst located in presacral space. The diagnosis of bronchogenic cyst was made on the basis of histopathological examination.

Herein, we present the clinical, radiologic, surgical, and pathologic findings of the patient with retroperitoneal bronchogenic cyst, located in presacral space.

## CASE REPORT

An asymptomatic 40-year-old man visited to the health screening center. The patient was previously healthy and did not have any record of operational or procedural history

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in abdomen. On screening ultrasound, about 8.0 cm × 9.3 cm × 8.7 cm large cystic mass was incidentally found at the posterior aspect of urinary bladder without vascular flow (Fig. 1A).

For further evaluation, multiphase contrast-enhanced CT scan was performed (Fig. 1B). On CT image, it showed the cystic mass with relatively homogeneous attenuation, less than 25 Hounsfield unit (HU). The mass did not have calcification or enhancing portion. The mass was located in presacral area with mass effect on the rectum to right lateral side. Before surgery, the mass was suspected to be a tailgut cyst, lymphangioma, rectal duplication cyst or rarely mucinous cystic tumor.

The laparoscopic surgery was performed for accurate diagnosis. The surgical finding was the large pure cystic mass at posterior aspect of left iliac vessels. The peritoneum on the left side of the rectum was opened, and the structure surrounding the tumor was removed. Due to the large mass, it was excised before it was removed from the abdominal cavity, and the yellowish, viscous cyst fluid was discharged (Fig. 1C).

Microscopic examination showed that the mass was a cystic tissue lined with pseudostratified ciliated columnar epithelium with submucosal seromucinous glands. Interspersed goblet cells were noted in high power insert (Fig. 1D).

Considering these morphologic and pathologic results, the tumor was compatible with presacral bronchogenic cyst. After 1 week later, the patient discharged without complications.

## DISCUSSION

The pathogenesis of bronchogenic cysts was not revealed clearly. However, a reasonable speculation is that bronchogenic cyst is related to embryonic abnormal development that the tumor arises from primitive foregut because of the lung bud development malformation (5). Therefore, (the) many bronchogenic cysts located in the lungs and heart have been reported, but little has been reported in the abdominal cavity, especially in retroperitoneum (5).

There are some possible hypothesis of bronchogenic cysts arising from retroperitoneum. First, it could be considered an abnormally differentiated foregut duplication cysts that occurs in the abdominal cavity or in the retroperitoneum. These can be found in or near any organ derived from this primitive foregut, which can explain the occurrence of bronchogenic cysts in or around pancreas, liver and stomach (2). Second, before closing the parts forming the future diaphragm, abnormal lung buds may pinch from the tracheobronchial tree. As a result, they may be trapped in abdominal cavity, before complete closure (2). Considering our case, since the bronchogenic cyst was located in lower level abdominal cavity, we support the latter hypothesis.

Clinical manifestation of retroperitoneal bronchogenic cyst is usually asymptomatic, and mostly benign nature. In rare cases, the literature said repetitive inflammatory condition, subsequently resulting scarring and may increase the risk for malignant degeneration (6).

This tumor needs histopathologic examination of the excised specimen for exact diagnosis (7). The pathology of bronchogenic cysts is presence of normal bronchial tissue in the cyst, including ciliated columnar epithelium, mucous gland, smooth muscle fiber, cartilage fibrous tissue, and elastic tissue (7). In our case, pseudostratified columnar epithelium, sub-

**Fig. 1.** Presacral bronchogenic cyst in a 40-year-old male, which was incidentally found in a screening ultrasound.

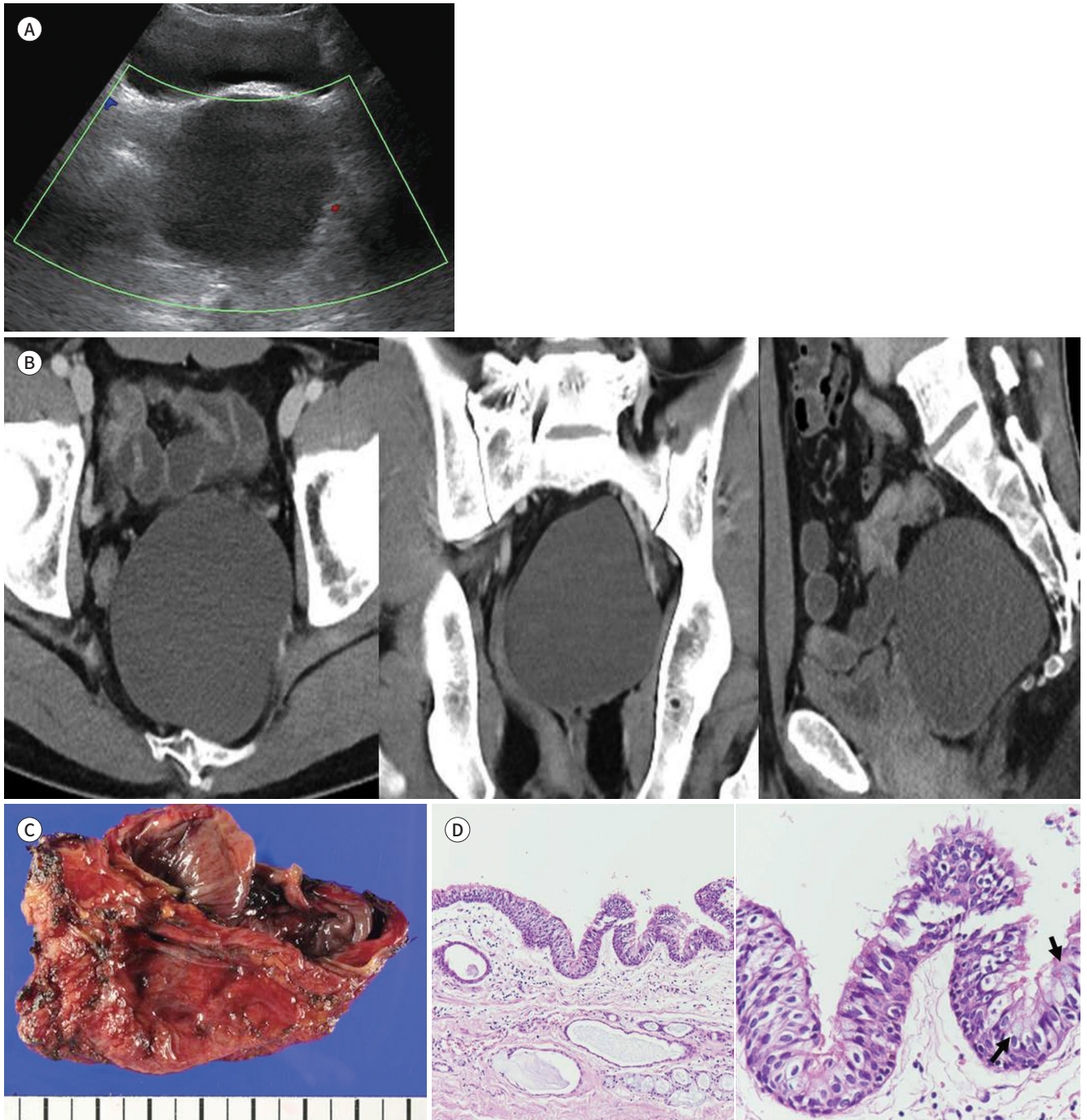
**A.** Trans-abdominal ultrasound reveals an 8 cm × 9.3 cm × 8.7 cm unilocular cystic mass located in the posterior aspect of the urinary bladder. There was no vascular flow in the mass.

**B.** Contrast-enhanced axial, coronal, and sagittal CT show a homogeneously attenuated, less than 25 Hounsfield unit, cystic mass without calcification or enhancing portion. The mass is located in the retroperitoneum, among the rectum, sacrum, and left iliac vessels, resulting in a right lateral deviation of the rectum.

**C.** The retroperitoneal cystic lesion was dissected and was found to contain a yellow colored fluid. Photograph of the gross specimen demonstrates the hemorrhagic and irregular surface of the tumor.

**D.** Microscopically, the cystic mass is lined with pseudostratified ciliated columnar epithelium with submucosal seromucinous glands (right, H&E stain, × 100). Interspersed goblet cells (arrows) are noted in the high power insert (left, H&E stain, × 400).

H&E = hematoxylin and eosin



mucosal gland, and goblet cell was shown which represents typical histologic finding of the bronchogenic tumor.

Radiologically, on CT image, the bronchogenic cyst have the typical appearance of a cyst. However, the fluid within the bronchogenic cyst usually contains protein-rich mucus (watery to a viscous fluid), it may show a relatively high HU (30 to 100 HU) (8). MRI findings of bronchogenic cysts are intermediate to high signal intensity on T1-weighted images, which also may be related to the presence of a high content of protein (9). In our case, the tumor was relatively pure cystic mass without enhancing solid portion. And the lesion made a space among the rectum, left iliac vessels and sacrum, that could be the clue for confirmation as retroperitoneal mass. Under the assumption that it is a lesion of the retroperitoneum, we discern presacral cystic tumor, such as foregut cyst, lymphangioma, and rectal duplication cyst, yet it was difficult to include retroperitoneal bronchogenic cyst in the differential diagnosis.

In general, the treatment of this tumor is not standardized and surgical excision for treatment is recommended to establish the diagnosis, treat any symptoms, and prevent complications. The complication(s) rate after surgery with infected cysts is high. In addition, future malignant degeneration or the presence of missed malignancies is also one of the possible late complications (2). After all, early surgical resection of asymptomatic retroperitoneal bronchogenic cyst is recommended for all great surgical procedures for these reasons (2).

Even though, a retroperitoneal bronchogenic cyst located in presacral space is unusual, this should offer the potential as differential diagnosis for presacral cystic tumor.

#### Author Contributions

Conceptualization, M.S.J.; investigation, K.A.Y.; M.S.J.; project administration, M.S.J.; supervision, M.S.J.; writing—original draft, all authors; and writing—review & editing, all authors.

#### Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

## REFERENCES

1. Cuypers P, De Leyn P, Cappelle L, Verougstraete L, Demedts M, Deneffe G. Bronchogenic cysts: a review of 20 cases. *Eur J Cardiothorac Surg* 1996;10:393-396
2. Govaerts K, Van Eyken P, Verswijvel G, Van der Speeten K. A bronchogenic cyst, presenting as a retroperitoneal cystic mass. *Rare Tumors* 2012;4:e13
3. Trehan M, Singla S, Singh J, Garg N, Mahajan A. A rare case of intra- abdominal bronchogenic cyst—a case report. *J Clin Diagn Res* 2015;9:PD03-PD04
4. Pasquer A, Djeudji F, Hervieu V, Rabeyrin M, Barth X. A rare retrorectal presentation of a bronchogenic cyst: a case report. *Int J Surg Case Rep* 2016;24:112-114
5. Chen HY, Fu LY, Wang ZJ. Ileal bronchogenic cyst: a case report and review of literature. *World J Clin Cases* 2018;6:807-810
6. Sullivan SM, Okada S, Kudo M, Ebihara Y. A retroperitoneal bronchogenic cyst with malignant change. *Pathol Int* 1999;49:338-341
7. Kurokawa T, Yamamoto M, Ueda T, Enomoto T, Inoue K, Uchida A, et al. Gastric bronchogenic cyst histologically diagnosed after laparoscopic excision: report of a case. *Int Surg* 2013;98:455-460
8. Sohn KM, Kim KJ, Maeng ES. Retroperitoneal bronchogenic cyst: a case report. *J Korean Radiol Soc* 2007; 57:451-453
9. Nakata H, Egashira K, Watanabe H, Nakamura K, Onitsuka H, Murayama S, et al. MRI of bronchogenic cysts. *J Comput Assist Tomogr* 1993;17:267-270

## 전천추 부위에 생긴 후복강 기관지 낭종: 증례 보고

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기관지 낭종은 기관이나 기관지를 따라 종격동에서 가장 흔하게 나타나는 드문 선천성 기형이며, 특히 뒤쪽 상부 종격동에 생긴다. 기관지 낭종은 다른 위치에서도 발생할 수 있으며 폐, 복강, 그리고 후복강에서 위치가 보고되었다. 저자들은 전천추부 공간에 생긴 후복강 기관지 낭종의 증례를 보고한다. 환자는 수술 후 병리 조직학적 검사에서 최종적으로 진단되었다.

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