

Bronchogenic Cyst of the Right Hemidiaphragm Presenting with Pleural Effusion

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Bronchogenic cysts are developmental foregut anomalies usually located within the mediastinum or lung parenchyma. An isolated bronchogenic cyst of the diaphragm is very rare. Our case was a 56-year-old female patient who presented with pleuritic chest pain in her right chest. Chest and abdominal computed tomography revealed a large lobulated cystic mass that was accompanied with pleural effusion in the right lower hemithorax. The tumor showed focally calcified areas in the wall and abutted against the diaphragm. We performed complete excision of the cyst including a portion of the diaphragm attached to it. The pathological diagnosis was established as the bronchogenic cyst originating from the diaphragm. We report this case with a review of the literature.

Key words: 1. Cyst
2. Bronchogenic cyst
3. Pleural effusion
4. Diaphragm

CASE REPORT

A 56-year-old female visited the hospital with the chief complaint of chest pain in the right chest for a week and occasional coughing. The patient had undergone a hysterectomy 11 years earlier due to uterine myoma, and had been told that a cyst was observed on the right lung by chest X-ray. However, there was no further follow-up or treatment. General blood tests produced normal findings; however, a great amount of pleural effusion was observed by chest X-ray. A huge calcified cystic tumor accompanied with pleural effusion was found on the right diaphragm by chest and abdominal CT (Fig. 1).

The thoracic cavity was observed by a video-assisted thor-

acoscope after intubation. Around 1,500 cc of brown pleural effusion was drained from the right thoracic cavity. A huge cystic tumor, which was the size of the head of a child, was observed on the diaphragm. Open heart surgery was performed on the right lateral side with the patient in the left lateral decubitus position to resect it effectively. The cystic tumor was excised and the fluid was aspirated to reduce the volume inside of the tumor. It was observed that the tumor was composed of calcified fibrous tissue after aspiration of 500 cc of brown fluid.

The 7×8 cm defect in the diaphragm (Fig. 2) was restored with a Gore-Tex 2 mm patch after removal of the tumor. The cystic tumor that was removed was diagnosed as a bronchogenic tumor by histopathological examination (Fig. 3). The

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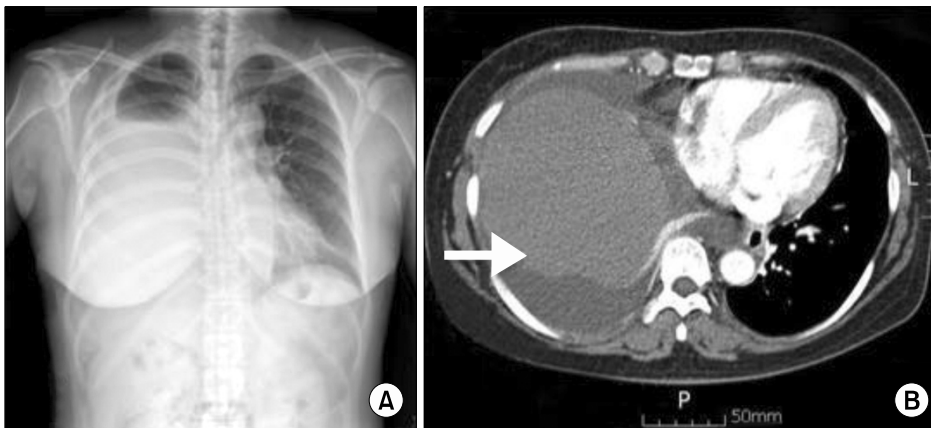


Fig. 1. (A) Preoperative chest X-ray shows pleural effusion in the right lung field. (B) Preoperative computed tomograph scan shows a huge lobulated mass in the sub-phrenic area (arrow).



Fig. 2. Gross finding of the specimen shows a unilocular cyst. The outer surface of the cyst is irregular and fibrotic. The inner surface of the cyst is white and relatively smooth.

chest tube was removed five days after surgery. The patient was discharged seven days after surgery, and has received follow-up care in the outpatient department for two months without major complications.

DISCUSSION

A bronchogenic cyst is formed of primitive foregut anomalies. It can be filled with fluid or air according to its connection with the bronchi and its excretion ability.

Bronchogenic cysts develop following tracheal branches in the mediastinum or pulmonary parenchyma. Otherwise, they are found in the pericardium, pleura, cervical area, or abdomen [1,2].

Bronchogenic cysts have round shapes with thin walls, and their sizes are average 2~10 cm in histopathological findings. A bronchogenic cyst has generally one cavity; however, it occasionally has multiple cavities. It has odorless, colorless mucinous fluid in most cases. A bronchogenic cyst is covered with pseudostratified ciliated columnar epithelium in most cases; however, it partially undergoes squamous metaplasia. Occasionally, a bronchogenic cyst in the esophagus is covered with squamous cells [1-3].

Bronchogenic cysts are detected during medical checkups because they rarely result in clinical symptoms. However, dyspnea by bronchial constriction can occur in infants and children. Surgical treatment is needed when it transforms from a benign cyst to a malignancy in adults [4].

Exact diagnosis is difficult with CT before surgery because its Hounsfield score ranges from 0 to 90 depending on the substances in the cysts. In fact, confirmation of a cystic mass inside the diaphragm is impossible [5]. Therefore, pathologic tests with surgical resection are needed to confirm diagnoses.

Surgical removal is a radical treatment. Although delicate surgical skill is essential in cases where cysts have adhered to important organs, the removal of a mediastinum tumor by video-assisted thoracoscope is popular because it can be dissected easily in case the mediastinum tumor is covered with pleurae [1,6,7].

In this study, a resection with video-assisted thoracoscope was attempted; however, open heart surgery in the right lateral side was performed due to the large size of the mass and restoration of the diaphragm after surgery.

Until now, only a few cases of bronchogenic cysts have

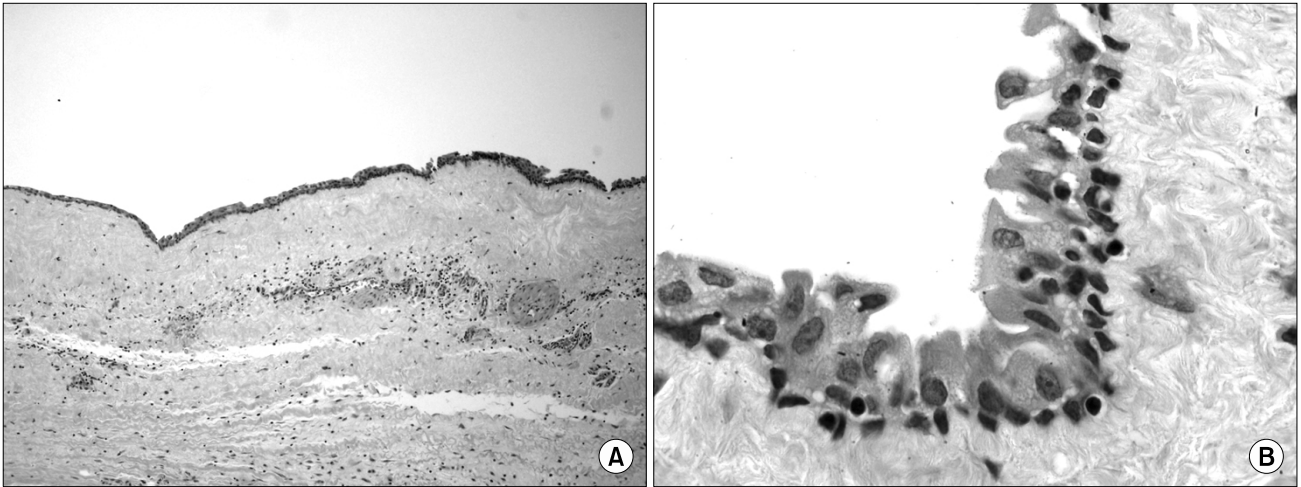


Fig. 3. (A) Microscopic findings of the bronchogenic cyst show a single layer of pseudostratified ciliated columnar cells. (B) Some of the cells show cilia at the apical border. Hematoxylin-Eosin stain, A: $\times 40$, B: $\times 1,000$.

been reported worldwide, and only one domestic case has been reported [8]. The authors experienced the rare case of a bronchogenic cyst that developed on the diaphragm, and they presented it here with a literature review.

REFERENCES

1. Chang YC, Chen JS, Chang YL, Lee YC. *Video-assisted thoroscopic excision of intradiaphragmatic bronchogenic cysts: two cases.* J Laparoendosc Adv Surg Tech A 2006;16: 489-92.
2. 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.
3. Sauvat F, Fusaro F, Jaubert F, Galifer B, Revillon Y. *Paraesophageal bronchogenic cyst: first case reports in pediatric.* Pediatr Surg Int 2006;22:849-51.
4. Endo C, Imai T, Nakagawa H, Ebina A, Kaimori M. *Bronchioloalveolar carcinoma arising in a bronchogenic cyst.* Ann Thorac Surg 2000;69:933-5.
5. Rozenblit A, Iqbal A, Kaley R, Rozenblit G. *Case report: intradiaphragmatic bronchogenic cyst.* Clin Radiol 1998;5: 918-20.
6. Suen HC, Mathisen DJ, Grillo HC, et al. *Surgical management and radiological characteristics of bronchogenic cyst.* Ann Thorac Surg 1993;55:476-81.
7. Kern JA, Daniel TM, Tribble CG, Silen ML, Rodgers BM. *Thoroscopic diagnosis and treatment of mediastinal masses.* Ann Thorac Surg 1993;56:92-6.
8. Park SC, Kuh JH. *A case of bronchogenic cyst in diaphragm.* Korean J Thorac Cardiovasc Surg 2000;33:847-50.