Bronchogenic cysts in rare sites (retroperitoneum, skin, spinal cord and pericardial cavity): A case series and characterization of epithelial phenotypes

YUSUKE ONISHI[1], SHIGERU KAWABATA[1], EMI YASUDA[1], NAOKAZU IBUKI[2,3], HARUHITO AZUMA[2], YUKA HIROTA[4], KOICHI UEDA[4], MASASHI NEO[5], MASAHIRO DAIMON[6], YOSHITAKA KURISU[1] and YOSHINOBU HIROSE[1]

Departments of [1]Pathology and [2]Urology, Faculty of Medicine, Osaka Medical and Pharmaceutical University, Takatsuki, Osaka 569-8686; [3]Department of Urology, Rakusai Newtown Hospital, Kyoto 610-1142; Departments of [4]Plastic and Reconstructive Surgery, [5]Orthopedic Surgery and [6]Thoracic and Cardiovascular Surgery, Faculty of Medicine, Osaka Medical and Pharmaceutical University, Takatsuki, Osaka 569-8686, Japan

Received August 20, 2023; Accepted November 22, 2023

DOI: 10.3892/br.2023.1709

Abstract. Bronchogenic cysts are congenital malformations of the bronchial tree, detected as a cystic and/or mass lesion in the thoracic cavity. Although it occurs in distant locations, such as skin and retroperitoneum, to the best of our knowledge, little is known about the components and phenotypes of the epithelium that line a bronchogenic cyst in rare sites. The present study reviewed 34 bronchogenic cysts that were surgically resected at Osaka Medical and Pharmaceutical University Hospital (Osaka, Japan) from January 1998 to December 2020. Bronchogenic cysts in rare sites were detected and diagnosis was confirmed based on the presence of pseudostratified, ciliated and/or columnar epithelium together with at least one of the following: Cartilage, smooth muscle or seromucous glands. The phenotypes of epithelium lining the cyst were characterized using immunohistochemical analysis. A total of six bronchogenic cysts in rare sites (two cases each in the retroperitoneum and skin and one case each in the cervical spinal cord and pericardial cavity) met the criteria for confirmation of the diagnoses. The epithelium lining the cyst stained positive for cytokeratin CK7 and thyroid transcription factor 1 (a marker expressed in thyroid follicles and bronchial epithelium) and negative for CK20, indicating that the phenotypes were similar to those of the respiratory epithelium. The present study demonstrated that a bronchogenic cyst can occur in rare sites, such as the retroperitoneum, skin, spinal cord and pericardial cavity, suggesting that it should be considered as a differential diagnosis before surgical approach to implement relevant management modalities such as follow-up, simple or radical resection.

Correspondence to: Dr Shigeru Kawabata, Department of Pathology, Faculty of Medicine, Osaka Medical and Pharmaceutical University; 2-7 Daigakumachi, Takatsuki, Osaka 569-8686, Japan
E-mail: shigeru.kawabata@ompu.ac.jp

Key words: bronchogenic cyst, retroperitoneum, skin, spinal cord, pericardial cavity

Introduction

Bronchogenic cysts are rare congenital malformations of the bronchial tree (a type of bronchopulmonary foregut malformation) with an incidence of ~1 in 42,000 admissions at St. Joseph Hospital between May 1975 and August 1986 (Houston, TX, USA) (1) and are detected as a cystic and/or mass lesion in the thoracic cavity, particularly in the mediastinum or pulmonary parenchyma (2,3). Bronchogenic cysts account for 6-15% of primary mediastinal masses (4), while pulmonary parenchymal bronchogenic cysts are ~20% of all bronchogenic cysts in the thoracic cavity (5,6). Clinical manifestations of bronchogenic cysts vary from asymptomatic incidental radiological findings to large mass lesions with severe symptoms. The most common symptom is chest pain, in particular, retrosternal or lateral chest pain, depending on the lesion location. Other symptoms include cough, dysphagia and fever, which are also dependent on compression to other organs, rupture or complicating infection (5-7). In pediatric patients, bronchogenic cysts often cause life-threatening emergencies with airway obstruction resulting in atelectasis, air trapping and respiratory distress as compressive symptoms. Conversely, in adult cases, although some symptoms are seen in 20-40% of patients, bronchogenic cysts often result in incidental radiological findings with computed tomography (CT) and/or magnetic resonance imaging (4,5). Surgical therapy should be considered for symptomatic patients with bronchogenic cysts as a primary treatment to prevent complications and histopathologically determine a final diagnosis (3).

The key histopathological characteristic of bronchogenic cysts is the pseudostratified ciliated columnar epithelium (3). Govaerts *et al* (8) defined the criteria for bronchogenic cysts as the presence of pseudostratified, ciliated, columnar epithelium in addition to the presence of at least one of cartilage, smooth

muscle or seromucous glands. Although bronchogenic cysts are primarily located in the thoracic cavity, however, they could ectopically occur anywhere along the developmental pathway of the foregut. Other distant locations have been reported, such as the skin, left ventricle and intraabdominal and retroperitoneal regions (9-11). However, it is unclear whether these distant lesions meet the aforementioned criteria and express the phenotypes of the respiratory epithelium. The present study retrospectively reviewed cases diagnosed as bronchogenic cysts that were surgically resected, including location of the bronchogenic cysts and how many lesions in rare sites there were. For bronchogenic cysts in rare sites, we reconfirmed the diagnoses based on the aforementioned criteria and characterized the epithelial phenotypes.

Materials and methods

Samples. The present study reviewed 34 available bronchogenic cysts from patients (age, 1 to 85-year-old, male/female: 22/12), that were surgically resected at the Osaka Medical and Pharmaceutical University Hospital (Takatsuki, Osaka, Japan) from January 1998 to December, 2020. Representative hematoxylin and eosin (H&E)-stained sections were selected and two pathologists reexamined the pathological features and diagnoses according to anatomopathological criteria (8).

Immunohistochemistry (IHC). The phenotypes of the epithelium lining the cyst were characterized using IHC. The tissue specimens were fixed in 10% neutral buffered formalin overnight at room temperature and embedded in paraffin wax for H&E staining. IHC analysis was performed using Vectastain Elite ABC kits (cat. no. PK-6100) from Vector Laboratories, Inc. (Maravai LifeSciences) according to the manufacturer's instructions for blocking, dilution of biotinylated secondary antibody and labeling. Briefly, 4-μm-thick sections were cut from the paraffin block. Following deparaffinization with xylene and hydration with 100% ethanol, endogenous peroxidase activity was quenched by incubation in 3% hydrogen peroxide solution for 10 min at room temperature. Antigen retrieval was performed using a preheated target retrieval solution (pH 6.0; Dako; Agilent Technologies, Inc.) for 30 min in a boiling rice cooker. The sections were incubated with primary antibodies and biotinylated secondary antibody at room temperature for 60 and 30 min, respectively. 3,3-diaminobenzidine was freshly prepared from tablets (Sigma-Aldrich; Merck KGaA) for chromogenic staining at 20˚C for 10 min. IHC-stained sections were evaluated using a light microscope at x20 to x400 magnification.

Primary antibodies against CK7 (cat. no. OV-TL 12/30; 1:200), CK20 (cat. no. Ks20.8; 1:200), thyroid transcription factor 1 (TTF-1, cat. no. 8G7G3/1; 1:50), synaptophysin (Syp, cat. no. DAK-SYNAP; 1:50), and surfactant protein A (SP-A, cat. no. PE10; 1:800) were obtained from Dako (Agilent Technologies). Anti-p40 (cat. no. BC28; 1:100) antibody for detecting basal cells was obtained from Nichirei Biosciences, Inc. Anti-α-smooth muscle actin (α-SMA, cat. no. 1A4; 1:800) and napsin A (cat. no. IP64; 1:400) antibodies were obtained from Sigma-Aldrich (Merck KGaA) and Leica Biosystems, respectively. Biotinylated secondary antibody (dilution, 1:200) was included in Vectastain Elite ABC kit.

Table I. Location of bronchogenic cysts.

Site	Number
Mediastinum	21
Intrapulmonary	6
Neck	1
Retroperitoneum	2
Skin	2
Spinal cord	1
Pericardial cavity	1

Results

Bronchogenic cysts are found in rare sites. The locations of the 34 bronchogenic cysts are listed in Table I; most were located in the mediastinum or intrapulmonary region. Rare sites included the retroperitoneum, skin, spinal cord and pericardial cavity.

Retroperitoneal bronchogenic cysts. A total of two cases of retroperitoneal bronchogenic cyst were identified. One was a mass and cystic lesion on the left side of the retroperitoneum in a 40-year-old female, which was detected during a medical checkup (Fig. 1A). The patient was asymptomatic and medical and family histories were unremarkable. Laboratory data, including hormone levels (such as catecholamine), were within normal range. Based on CT, neoplastic disease in the adrenal gland was a differential diagnosis. Surgical excision of the lesion was performed to obtain a pathological diagnosis. The specimen was a brownish, soft mass lesion in the left adrenal gland and measured 5.5x5.0x3.5 cm (Fig. 1B). The divided surfaces after cross-sectioning revealed a multicystic lesion, including atmospheric gelatinous or light-yellow contents. Macroscopically, there was an intact adrenal gland (Fig. 1C), indicating that the lesion was not derived from it.

Microscopic examination revealed the cyst was lined with ciliated epithelium and separated from the intact adrenal gland (Fig. 1D and E). Furthermore, seromucous glands (Fig. 1F), cartilage (Fig. 1G) and smooth muscle stained with anti-αSMA antibody (Fig. 1H and I) were detected near the cystic lesion and met the criteria for bronchogenic cysts. Considering these results, the diagnosis of retroperitoneal bronchogenic cyst was confirmed.

Epithelial phenotype was characterized by IHC. CK7 (a cytokeratin marker), TTF-1, p40, Syp (a neuroendocrine marker), napsin A, which is preferentially expressed in bronchial and alveolar epithelium, and SP-A (secreted by alveolar type II cells) (12) showed positive staining in the ciliated epithelium (Fig. 2A, B and D-H), whereas, CK20 was negative (Fig. 2C). These IHC features were similar to those of respiratory epithelium (CK7-positive and CK20-negative; data not shown).

Another case involved a cystic lesion in the retroperitoneum of a 65-year-old male (Fig. S1). On microscopic examination, the cyst was lined with ciliated epithelium, and smooth muscle (Fig. S1A), which met the criteria for bronchogenic cyst. However, seromucous glands and cartilage were not

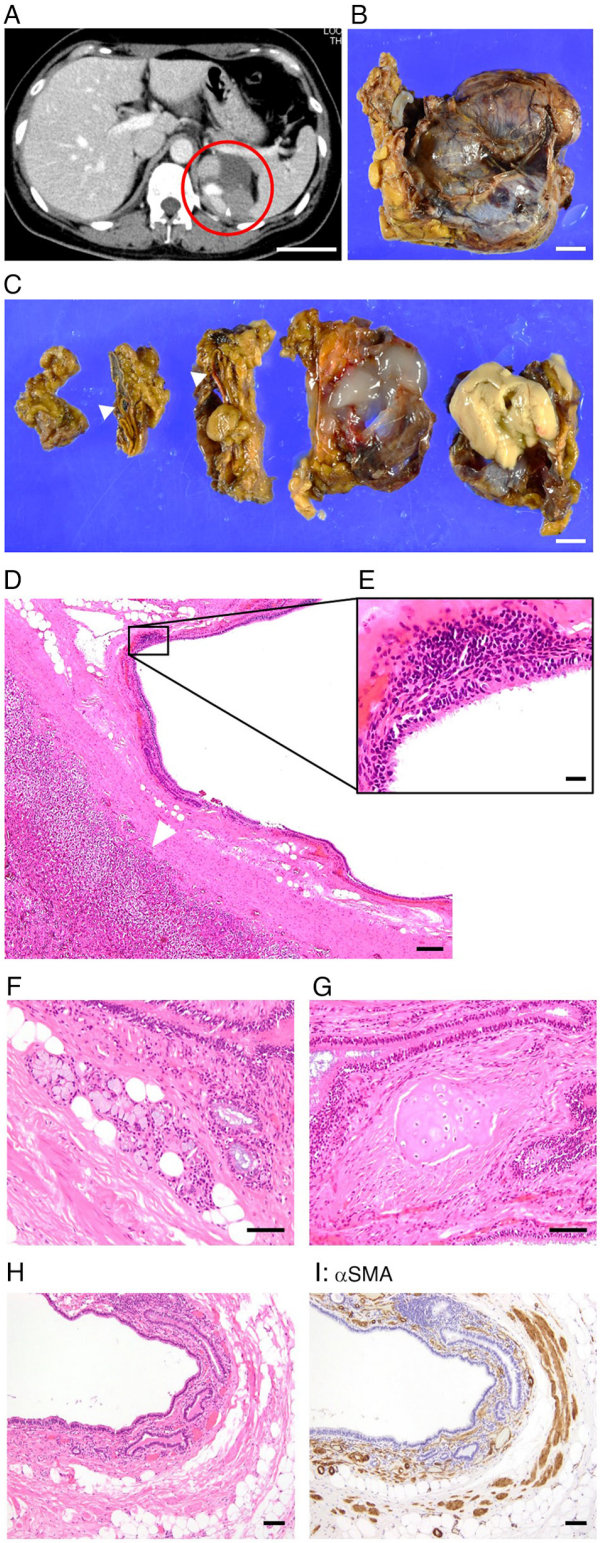

Figure 1. Retroperitoneal bronchogenic cyst. (A) Computed tomography (transverse section). Red circle, mass and cystic lesion. Scale bar, 5 cm. (B) Whole mass lesion. (C) Divided surfaces. Scale bar, 1 cm. H&E staining of ciliated epithelium. Scale bar, (D) 200 and (E) 20 μm. Arrowhead, left adrenal gland. Anatomopathological features for the diagnosis of bronchogenic cyst. (F) Seromucous glands. (G) Cartilage. (H) H&E and (I) α-smooth muscle actin staining of smooth muscle near ciliated epithelium. Scale bar, 100 μm. H&E, hematoxylin and eosin.

detected. IHC features were similar to those aforementioned, with positive staining for CK7, TTF-1, p40, napsin A and SP-A (Fig. S1B and D-G) and negative for CK20 (Fig. S1C) in the ciliated epithelium. Retroperitoneal bronchogenic cysts showed phenotypes similar to those of respiratory epithelium.

Bronchogenic cyst in the skin. There were two cases of bronchogenic cysts in the skin. A 59-year-old male had a polyp-like lesion on the medial anterior chest since childhood, which had grown with age (Fig. 3A). The patient did not complain of any pain around the lesion; physical examination showed skin flare and a fixed lesion on the sternal area. During radiological examination, chest CT showed a cystic lesion measuring 14.7x12.0x8.0 cm (Fig. 3B), thus, a median cyst was considered as a differential diagnosis. Surgical excision of the lesion was performed to obtain a pathological diagnosis. The cyst wall was lined with ciliated epithelium (blue), which was located in the deep dermal tissue (Fig. 3C). Seromucous glands were detected near the cyst wall (Fig. 3D). Goblet cells were also found among the cyst-lining cells (Fig. 3E). Epithelial phenotypes of ciliated epithelium were positive for CK7, TTF-1 and p40 (Fig. 3F-H) and negative for CK20 (Fig. 3J). Smooth muscle was found under the cyst, confirmed by αSMA-positive staining (Fig. 3I). Although cartilage was not found, a diagnosis of bronchogenic cyst was made based on the presence of ciliated epithelium, seromucous glands and smooth muscle. However, there was no evidence of atypical epithelium that indicated malignant transformation. There has been no recurrence for four years since the resection. For the giant skin bronchogenic cyst in Fig. 3, we previously described the treatment procedure (13).

Another case was a small polyp-like lesion on the chest of a 1-year-old male (Fig. S2A). The lesion was detected at birth and the differential diagnosis was soft fibroma. The lesion did not show rapid growth or infectious features and the diameter was ~10 mm, therefore, surgical excision of the lesion was performed. Microscopic examination revealed a multicystic lesion in the dermis (Fig. S2B). Ciliated epithelium with goblet cells lined the cyst, in addition to seromucous glands and cartilage (Fig. S2C and D) and showed positive staining for CK7, TTF-1, and p40 (Fig. S2E-G) and negative staining for CK20 (Fig. S2H). In this case, smooth muscle was not detected. Based on these findings, the diagnosis of bronchogenic cyst in the skin was confirmed.

Bronchogenic cyst in the cervical spinal cord and pericardial cavity. A nodular lesion (1.5x1.3x0.8 cm) was detected on magnetic resonance imaging in the cervical spinal cord (C4-5) of a 12-year-old male (Fig. 4A). The patient had pain and numbness in the left shoulder and upper arm, which had persisted for 1 month before he visited an orthopedist. Upon medical examination, he displayed loss of muscle strength in the left upper and lower extremities. Laboratory, respiratory, and cardiology tests showed no abnormalities. Therefore, bronchogenic cyst or spinal cord neoplasm such as schwannoma was the differential diagnosis and surgical excision of the lesion was performed. H&E staining showed a multicystic lesion (Fig. 4B) lined with ciliated epithelium (Fig. 4C). Epithelial cells were positive for CK7 and p40 (Fig. 4D and G) and negative for CK20 and TTF-1 (Fig. 4E and F). Smooth muscle was found under the cyst, which was confirmed by αSMA staining

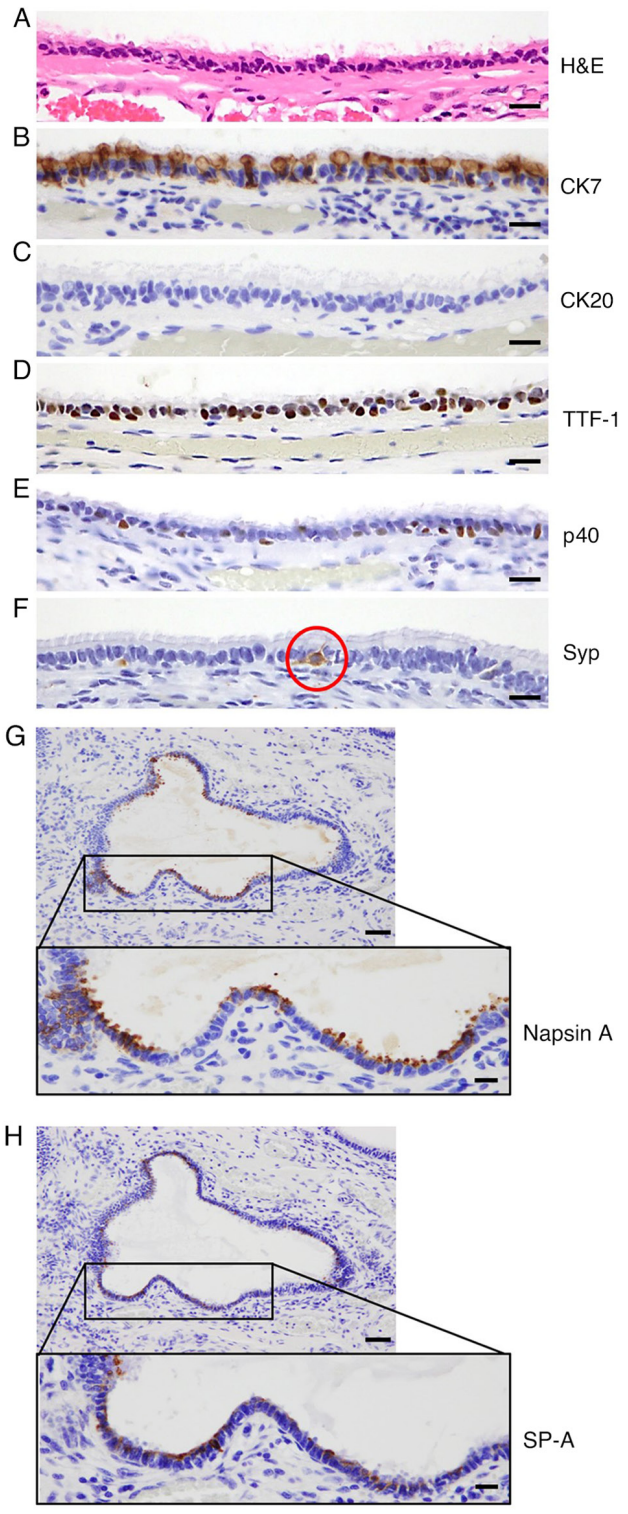

Figure 2. Characterization of epithelial phenotypes of retroperitoneal bronchogenic cysts. (A) Ciliated epithelium was stained for (B) CK7, (C) CK20, (D) TTF-1, (E) p40, (F) synaptophysin (circle). Scale bar, 20 μm. (G) napsin A and (H) surfactant protein A. Scale bar, 50 μm in lower magnification and 20 μm in black lined-higher magnification.

(Fig. 4H), resulting in the diagnosis of a bronchogenic cyst. Seromucous glands and cartilage were not detected.

A 56-year-old male underwent surgery for mitral regurgitation that was detected during a medical checkup. He did not experience discomfort. Physical examination revealed cardiac murmur. However, changes in the electrocardiogram such as arrhythmia were not observed. During surgery for the mitral regurgitation, a cystic lesion was incidentally detected in the pericardial cavity. On microscopic examination, it was lined with ciliated epithelium, seromucous glands and cartilage (Fig. 5A and B). Epithelial phenotypes of ciliated epithelium were positive for CK7, TTF-1, p40 and Syp (Fig. 5C and E-G) but were negative for CK20 (Fig. 5D). Smooth muscle was found under the cyst, which was confirmed by αSMA-positive staining (Fig. 5H), resulting in diagnosis of a bronchogenic cyst.

Discussion

The present study confirmed the diagnoses of bronchogenic cysts in rare sites, such as the retroperitoneum, skin, spinal cord or pericardial cavity according to the diagnostic criteria (8). Two cases each in the retroperitoneum and pericardial cavity showed bronchogenic cysts lined with ciliated epithelium with cartilage, smooth muscle and seromucous glands. All cases in rare sites except spinal cord bronchogenic cysts showed CK7-, TTF-1- and p40-positive and CK20-negative cyst-lining epithelium. These findings indicated that the aforementioned criteria were useful for diagnosis of remotely located bronchogenic cysts and that the epithelial phenotypes of bronchogenic cysts were similar to those of the respiratory epithelium.

Bronchogenic cysts of the central nervous system are also called neurenteric, enterogenous, or endodermal cysts, account for 0.3-1.3% of all spinal canal tumors and are more common in the lower cervical and upper thoracic spine (14). A total of ~90% of neurenteric cysts are located in the intradural/extramedullary compartment (15). Embryologically, the cyst is considered to be derived from the congenital maldevelopment of endodermal tissue displaced into the spinal canal through the interposed mesodermal layer that forms vertebral bodies (16,17). The histological classification of endodermal cysts of the central nervous system is as follows: Type A cysts contain columnar or cuboidal cells, with ciliated and non-ciliated components mimicking the respiratory or gastrointestinal epithelium; type B, type A features in addition to any of glands producing mucinous or serous fluid, smooth muscle, striated muscle, fat, cartilage, bone, elastic fibers, lymphoid tissue, nerve fiber or ganglion cells and type C, type B features in addition to ependymal or glial tissue (14,15). The treatment for cysts is total surgical resection; however, recurrence rates as of 37% have been reported with incomplete resection (15). The present spinal cord bronchogenic cyst was located in the lower cervical spinal cord (C4-5) and intradural/extramedullary compartment. Histologically, the cyst contained ciliated epithelium with smooth muscle but no other components such as seromucous glands and cartilage, indicative of type B cysts.

Recently, Kalfas and Scudieri (14) reported a case of intracranial supratentorial endodermal cysts and showed that the cells were positive for CK7. Here, epithelial cells of the spinal cord bronchogenic cyst were positive for CK7 and p40, consistent with the aforementioned study. Generally, the intestinal epithelium is CK7-negative and CK20-positive (18), whereas the respiratory epithelium is CK7-positive and CK20-negative. Although TTF-1 was negative in the present case of spinal cord bronchogenic cyst, based on the staining pattern of CK7

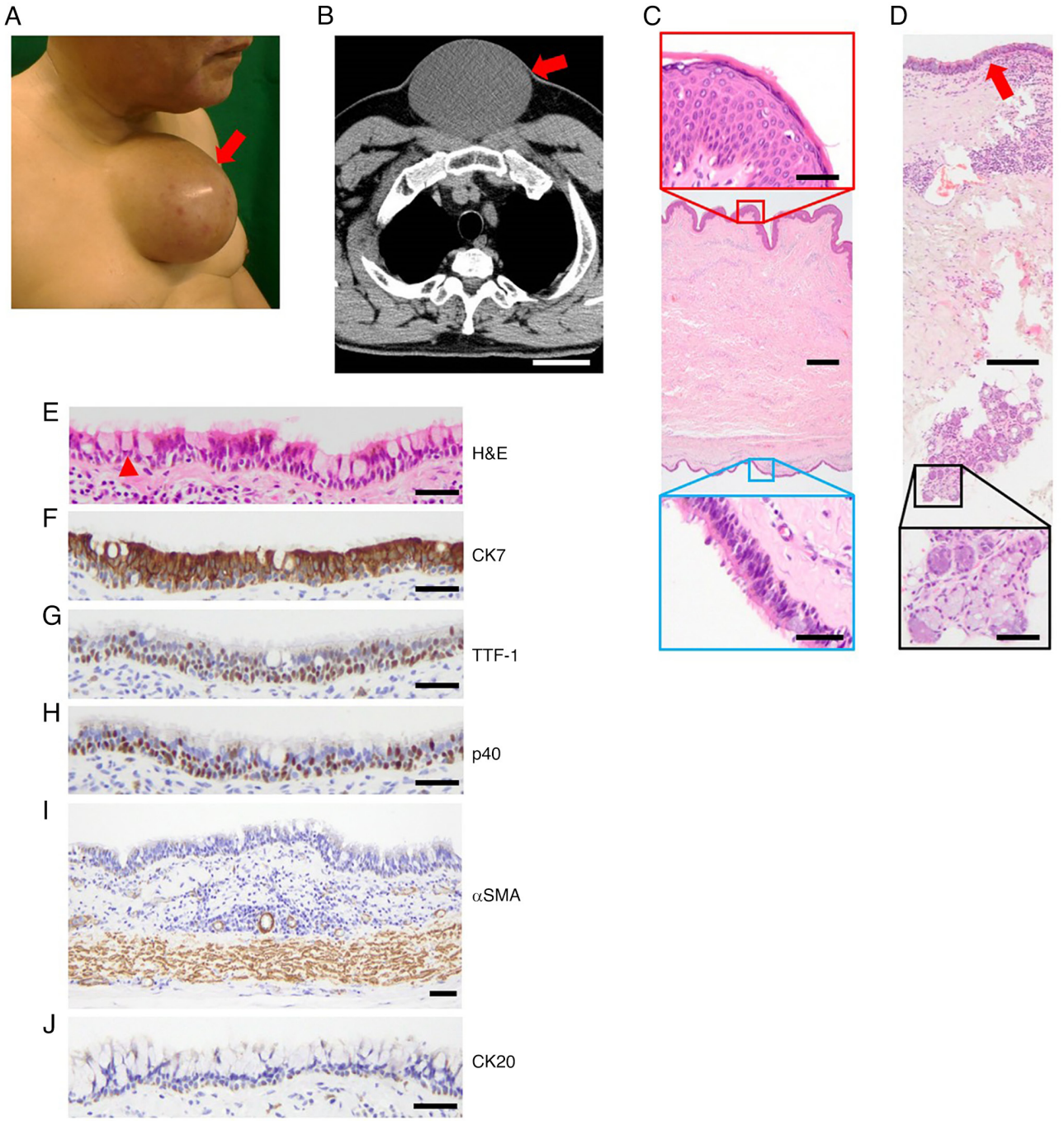

Figure 3. Bronchogenic cyst in the skin. (A) Polyp-like lesion (arrow) on the chest. (B) Chest CT (transverse section) showing a cystic lesion (arrow). Scale bar, 5 cm. (C) H&E staining of the wall of the cyst. Scale bar, 500 μm. Red, epidermis, scale bar, 50 μm; blue, ciliated epithelium, scale bar, 50 μm. (D) Seromucous glands. Arrow, ciliated epithelium. Scale bar, 200 μm in lower magnification and 50 μm in black lined-higher magnification. (E) Epithelial phenotypes of the ciliated epithelium. Arrowhead, goblet cell. Immunohistochemical staining for (F) CK7, (G) TTF-1, (H) p40, (I) αSMA and (J) CK20. Scale bar, 50 μm. CT, computed tomography; H&E, hematoxylin and eosin; TTF-1, thyroid transcription factor 1; αSMA, α-smooth muscle actin.

and CK20, an endodermal cyst in the central nervous system may have phenotypes of respiratory rather than intestinal epithelium.

To the best of our knowledge, the components and phenotypes in the epithelium of a bronchogenic cyst have been rarely evaluated (14). The present cases had two major components: Cyst-lining cells positive for CK7, such as ciliated epithelium and basal cells, as shown by p40-positive staining in the basal layer. Although there are some reports that show TTF-1-positive staining in the epithelium of extrapulmonary bronchogenic cysts in rare sites, such as the retroperitoneum, skin and spinal cord (8-11,17), few studies (10,14) have assessed other phenotypes for bronchial and alveolar epithelium. The present study confirmed the expression of napsin A and SP-A, which are primarily expressed in bronchial and alveolar epithelium. TTF-1, napsin A and SP-A staining demonstrated that the epithelium of bronchogenic cysts had the same structure as respiratory epithelium. A bronchogenic cyst of the retroperitoneum or pericardial cavity results from the abnormal budding of

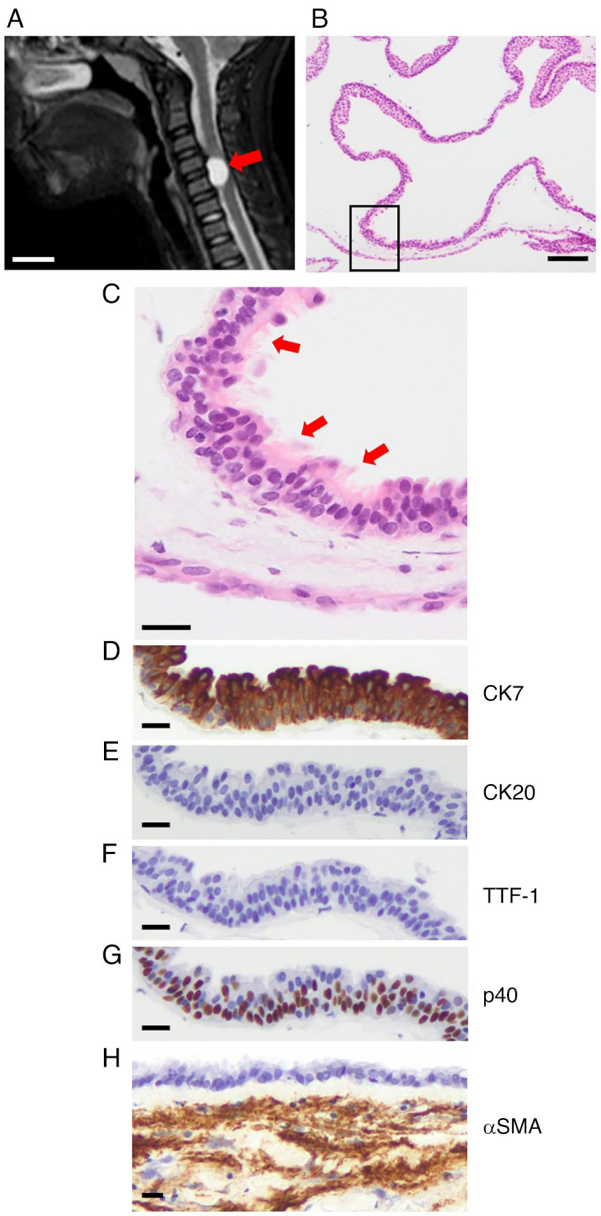

Figure 4. Bronchogenic cyst in the cervical spinal cord. (A) Magnetic resonance imaging (T1 image, sagittal section) showed a high-density nodule (red arrow). Scale bar, 2 cm. (B) Hematoxylin and eosin staining of the wall of the cystic lesion. Scale bar, 100 μm. (C) Ciliated epithelium (red arrow). Magnified inset of the black lined-area in B. Immunohistochemical analysis of (D) CK7, (E) CK20, (F) TTF-1, (G) p40 and (H) αSMA. Scale bar, 20 μm. TTF-1, thyroid transcription factor 1; αSMA, α-smooth muscle actin.

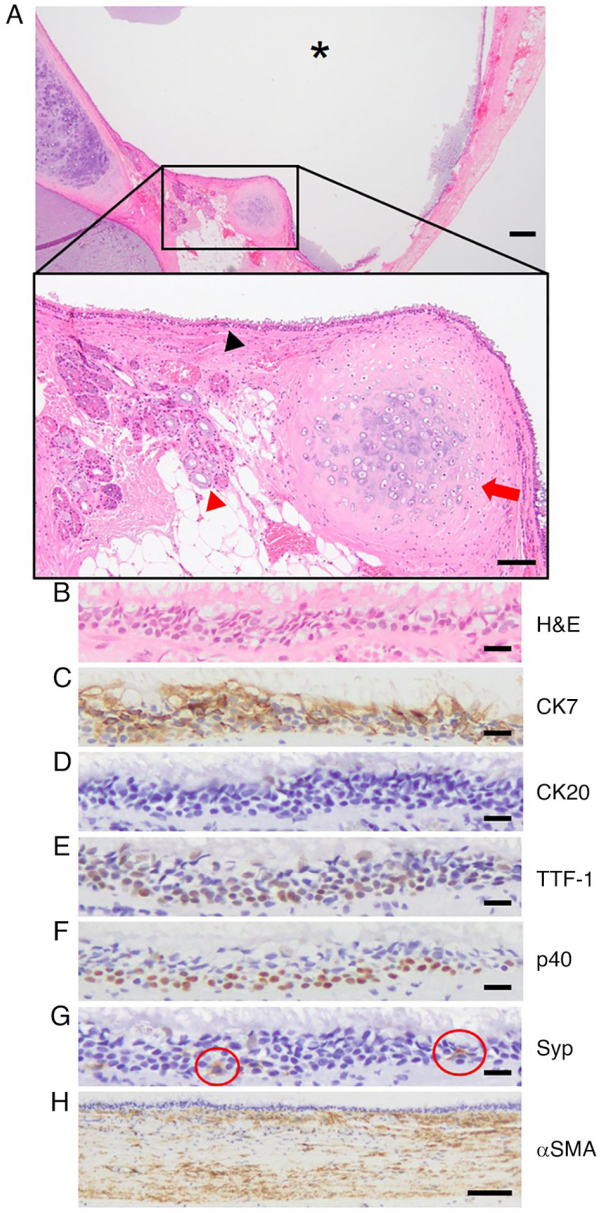

Figure 5. Bronchogenic cyst in the pericardial cavity. (A) H&E staining of a cystic lesion (asterisk) with ciliated epithelium (black arrowhead), seromucous glands (red arrowhead) and cartilage (red arrow). Scale bar, 200 μm in lower magnification and 100 μm in black lined-higher magnification. (B) Ciliated epithelium was assessed for (C) CK7, (D) CK20, (E) TTF-1, (F) p40 and (G) Syp (a positive cell in red circle), scale bar, 20 μm. (H) αSMA. Scale bar, 100 μm. H&E, hematoxylin and eosin; TTF-1, thyroid transcription factor 1; Syp, synaptophysin; αSMA, α-smooth muscle actin.

the respiratory diverticulum (8). However, the mechanism by which a bronchogenic cyst of the skin occurs is unclear. Moreover, a diagnosis of teratoma should be excluded before a diagnosis of skin bronchogenic cyst as teratoma is valid only when a tridermic lineage is present (9). Here, macroscopic and microscopic features representing germinal layers were not found, which ruled out teratoma. Additionally, goblet cells were observed among cyst lining-cells, which supported the diagnosis of skin bronchogenic cysts because these cells are components of the respiratory epithelium. Although the presence of goblet cells is not included in the Govaerts *et al* (8) anatomopathological criteria, detection of these cells is helpful for the diagnosis of bronchogenic cyst.

To the best of our knowledge, few studies have reported genetic analysis of bronchogenic cysts: Shiferaw *et al* reported an incidental intramyocardial bronchogenic cyst with p.H558R variant (a common polymorphism) in sodium voltage-gated channel alpha subunit 5 (*SCN5A*) gene, which is a human cardiac sodium channel gene (19). Mutations in the *SCN5A* gene are involved in pathophysiology of cardiac arrhythmia and cardiomyopathy. Moreover, the p.H558R variant in the *SCN5A* gene variant may result in ion channel mutation, leading to arrhythmias (20,21). The present case of the bronchogenic cyst in the pericardial cavity was incidentally found during surgery for mitral regurgitation and was

not located in any intramyocardial site. The patient did not show any change in electrocardiogram to suggest arrhythmia. Therefore, genetic analyses for *SCN5A* variants were not performed. Murakami *et al* (22) reported an abdominal bronchogenic cyst with a low-grade mucinous neoplasm harboring a *GNAS* mutation; *GNAS* mutation (p.R201C) in the atypical epithelium of the bronchogenic cyst indicated potential mechanism for tumorigenesis and malignant transformation. However, the present study did not find any atypical epithelium that suggested a neoplasm or malignant transformation. Nonetheless, genetic analyses may be warranted in some cases of symptomatic and/or malignant transformed bronchogenic cysts.

In conclusion, the present study evaluated bronchogenic cysts in rare sites, including the retroperitoneum, skin, spinal cord and pericardial cavity, and confirmed the diagnoses, epithelial components and phenotypes, which histopathologically suggested that the epithelium of bronchogenic cysts had the same structure as respiratory epithelium. None of the cases had recurrence since complete resection. Although most cases of bronchogenic cyst are asymptomatic and found incidentally during medical checkups or detailed examinations of other diseases, surgical treatment should be considered due to a risk of malignant transformation (2,22,23). The present study demonstrated that bronchogenic cysts can occur in rare sites and should be considered as a differential diagnosis before and after surgical resection to recognize a risk of malignant transformation and implement relevant management modalities such as treatment procedure and follow-up time.

Acknowledgements

The authors would like to thank Ms Megumi Miyauchi (Department of Pathology, Faculty of Medicine, Osaka Medical and Pharmaceutical University; Takatsuki, Osaka, Japan) for technical support with the IHC analyses, and Mr Kentaro Morimoto (Faculty of Medicine, Osaka Medical and Pharmaceutical University; Takatsuki, Osaka, Japan) for comments about pathological diagnostic approaches in this study.

Funding

No funding was received.

Availability of data and materials

The datasets used and/or analyzed during the present study are available from the corresponding author on reasonable request.

Authors' contributions

YO collected and analyzed data. SK and EY designed the study and evaluated the pathological findings. NI, HA, YuH, KU, MN and MD contributed to clinical data acquisition and interpretation. YK and YoH performed pathological diagnosis. SK wrote the manuscript. All authors have read and approved the final manuscript. SK and EY confirm the authenticity of all the raw data.

Ethics approval and consent to participate

The present study was approved by the Institutional Review Board of Osaka Medical and Pharmaceutical University (approval no. 2020-124; Takatsuki, Osaka, Japan). A total of four patients provided written informed consent to participate; two patients participated through an opt-out approach to them and their guardians.

Patient consent for publication

Written informed consent was obtained from four patients for publication of this case report. For the other two patients, the consent was obtained through the opt-out approach to them and their guardians.

Competing interests

The authors declare that they have no competing interests.

References

1. Coselli MP, de Ipolyi P, Bloss RS, Diaz RF and Fitzgerald JB: Bronchogenic cysts above and below the diaphragm: Report of eight aases. Ann Thorac Surg 44: 491-494, 1987.
2. Cuypers P, De Leyn P, Cappelle L, Verougstraete L, Demedts M and Deneffe G: Bronchogenic cysts: A review of 20 cases. Eur J Cardiothorac Surg 10: 393-396, 1996.
3. Gross DJ, Briski LM, Wherley EM and Nguyen DM: Bronchogenic cysts: A narrative review. Mediastinum 7: 26, 2023.
4. Ribet ME, Copin MC and Gosselin B: Bronchogenic cysts of the mediastinum. J Thorac Cardiovasc Surg 109: 1003-1010, 1995.
5. St-Georges R, Deslauriers J, Duranceau A, Vaillancourt R, Deschamps C, Beauchamp G, Pagé A and Brisson J: Clinical spectrum of bronchogenic cysts of the mediastinum and lung in the adult. Ann Thorac Surg 52: 6-13, 1991.
6. Suen HC, Mathisen DJ, Grillo HC, LeBlanc J, McLoud TC, Moncure AC and Hilgenberg AD: Surgical management and radiological characteristics of bronchogenic cysts. Ann Thorac Surg 55: 476-481, 1993.
7. Lee DH, Park CK, Kum DY, Kim JB and Hwang I: Clinical characteristics and management of intrathoracic bronchogenic cysts: A single center experience. Korean J Thorac Cardiovasc Surg 44: 279-284, 2011.
8. Govaerts K, Van Eyken P, Verswijvel G and Van der Speeten K: A bronchogenic cyst, presenting as a retroperitoneal cystic mass. Rare Tumors 4: e13, 2012.
9. Sun J, Yuan T and Deng H: Cutaneous bronchogenic cyst in the left scapular region of a boy. World J Pediatr 10: 365-367, 2014.
10. Nakagawa A, Nagase K, Inoue T, Miura Y and Fujisaki A: Upper arm bronchogenic cyst: A rare case presentation. Eur J Dermatology 33: 321-322, 2023.
11. Wen Y, Chen W, Chen J and He X: Retroperitoneal bronchogenic cyst resembling an adrenal tumor: two case reports and literature review. J Int Med Res 48: 300060520925673, 2020.
12. Kalhor N and Moran CA: Immunohistology of Pulmonary and Pleural Neoplasms. In: Diagnostic Immunohistochemistry. 6th edition. Dabbs DJ (ed). Elsevier, Inc., Philadelphia, PH, pp434-497, 2021.
13. Nakao T, Katayama M, Kino H, Hanaoka N and Ueda K: A case of giant subcutaneous bronchogenic cyst. Arch Clin Med Case Rep 4: 590-595, 2020.
14. Kalfas F and Scudieri C: Endodermal cysts of the central nervous system: Review of the literature and a case report. Asian J Neurosurg 15: 989-996, 2020.
15. Savage JJ, Casey JN, McNeill IT and Sherman JH: Neurenteric cysts of the spine. J Craniovertebr Junction Spine 1: 58-63, 2010.
16. Chen IH, Kao KP, Penn IW and Ho DM: Double intraspinal enterogenous cysts. J Neurol Neurosurg Psychiatry 58: 110-111, 1995.
17. Baek WK, Lachkar S, Iwanaga J, Oskouian RJ, Loukas M, Oakes WJ and Tubbs RS: Comprehensive review of spinal neurenteric cysts with a focus on histopathological findings. Cureus 10: e3379, 2018.

18. Krasinskas AM and Zheng W: Immunohistology of the Gastrointestinal Tract. In: Diagnostic Immunohistochemistry, 6th edition. Dabbs DJ (ed). Elsevier, Inc., Philadelphia, PH, pp528-560, 2021.
19. Shiferaw K, Lobrinus AJ, Grabherr S, Michaud K, Mangin P and Schrag B: One case, 3 rare simultaneous findings: Intramyocardial bronchogenic cyst, P.H558R variant of SCN5A gene, and granular cell tumor of the esophagus. Am J Forensic Med Pathol 33: 335-338, 2012.
20. Gouas L, Nicaud V, Berthet M, Forhan A, Tiret L, Balkau B and Guicheney P; D.E.S.I.R. Study Group: Association of KCNQ1, KCNE1, KCNH2 and SCN5A polymorphisms with QTc interval length in a healthy population. Eur J Hum Genet 13: 1213-1222, 2005.
21. Shan L, Makita N, Xing Y, Watanabe S, Futatani T, Ye F, Saito K, Ibuki K, Watanabe K, Hirono K, *et al*: SCN5A variants in Japanese patients with left ventricular noncompaction and arrhythmia. Mol Genet Metab 93: 468-474, 2008.
22. Murakami T, Shimizu H, Yamazaki K, Nojima H, Usui A, Kosugi C, Shuto K, Obi S, Sato T, Yamazaki M and Koda K: Intra-abdominal ectopic bronchogenic cyst with a mucinous neoplasm harboring a GNAS mutation : A case report. World J Clin Cases 10: 8709-8717, 2022.
23. Taira N, Kawasaki H, Atsumi E, Ichi T, Kawabata T, Saio M and Yoshimi N: Mucoepidermoid carcinoma of arising from a bronchogenic cyst of the diaphragm. Ann Thorac Cardiovasc Surg 24: 247-250, 2018.