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Case Report

Bronchogenic cyst with atypical imaging findings and repeated ruptures in a short period of time: A case report[☆]

Mika Matsushita, MD^{a,*}, Osamu Honda, MD, PhD^a, Masasuke Kohzai, MD^a, Kotaro Minami, MD^a, Shintaro Yamamoto, MD^a, Kenichi Ueda, MD^a, Haruaki Hino, MD^b, Tomohiro Murakawa, MD^b, Asako Okabe, MD, PhD^c, Noboru Tanigawa, MD, PhD^a

^aDepartment of Radiology, Kansai Medical University, 2-5-1 Shin-machi, Hirakata, Osaka, 573-1010, Japan

^bDepartment of Thoracic Surgery, Kansai Medical University, 2-5-1 Shin-machi, Hirakata, Osaka, 573-1010, Japan

^cDepartment of Pathology, Kansai Medical University, 2-5-1 Shin-machi, Hirakata, Osaka, 573-1010, Japan

ARTICLE INFO

Article history:

Received 11 July 2022

Revised 24 July 2022

Accepted 28 July 2022

Keywords:

Bronchogenic cyst

Rupture

Mediastinal cyst

Back pain

Chest pain

ABSTRACT

Bronchogenic cysts are the most common primary cysts of the mediastinum. Although most are asymptomatic, some bronchogenic cysts cause symptoms such as chest pain and dyspnea. Here, we report a case of bronchogenic cyst that ruptured twice in a short period of time in a patient who presented with sudden back pain. The lesion was apparent on computed tomography (CT) as a mass lesion with heterogeneous and high attenuation in the posterior mediastinal region. CT-guided puncture performed for diagnostic purposes revealed the contents as bloody fluid. The patient suffered chest pain approximately 3 months after the first presentation, and re-growth and re-rupture of the mass was suspected. The lesion was surgically resected and pathologically diagnosed as a bronchogenic cyst. Spontaneous rupture is a very rare complication of bronchogenic cyst, usually into the trachea, pleural cavity, or pericardial cavity. However, there are no reports of multiple ruptures. This case highlights the importance of recognizing the atypical imaging findings of bronchogenic cyst and the rare complication of rupture.

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Introduction

Bronchogenic cysts are congenital malformations that arise from abnormal budding of the ventral diverticulum of the

foregut or the tracheobronchial tree during embryogenesis. They are the most common primary cyst of the mediastinum, with a prevalence of 6% [1,2]. Regarding location, approximately 79% occur in the middle mediastinum, 17% in the posterior mediastinum, and 3% in the anterior mediastinum [2]. They are found less frequently in the lung parenchyma, pleura, and diaphragm [3,4]. It is generally diagnosed by computed tomography (CT), although cysts with hemorrhage or infection may show atypical imaging findings, making diag-

[☆] Competing Interests: There is no conflict of interest to declare.

* Corresponding author.

E-mail address: matsumik@hirakata.kmu.ac.jp (M. Matsushita).

<https://doi.org/10.1016/j.radcr.2022.07.103>

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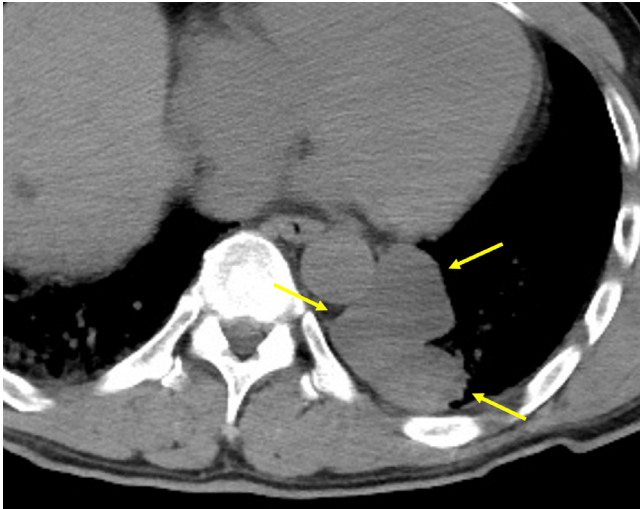


Fig. 1 – CT scan obtained at the time of presentation. Nonenhanced image in the axial plane shows a smooth, lobulated mass in the left paraspinal region adjacent to the aorta (arrow). The mass shows heterogeneous high attenuation.

nosis difficult [5,6]. For this reason, CT has limited usefulness in preoperative diagnosis, with a diagnostic rate of 60%-70% [6]. In contrast, magnetic resonance imaging (MRI) is highly sensitive and specific for bronchogenic cyst [2].

Bronchogenic cysts are usually asymptomatic, but sometimes enlarge and cause symptoms because of compression of surrounding organs [2,7]. Spontaneous rupture is a very rare complication, and has been occasionally reported to occur into the trachea, pleural cavity, or pericardial cavity [4,7,8]. Here, we report a case of mediastinal bronchogenic cyst that presented with severe back pain, and ruptured twice in a short period of time.

Case report

A 59-year-old man presented to the emergency department with dyspnea and sudden onset of back pain that moved to the left hypochondrium. Physical examination at the time of arrival revealed spontaneous pain and tenderness in the left anterior thoracic region. Chest radiograph and echocardiography showed no abnormal findings. Contrast-enhanced CT of the chest revealed a smooth-margined lobulated mass measuring $6.4 \times 4.7 \times 6.8$ cm, located on the diaphragm in the left paravertebral region adjacent to the aorta (Fig. 1). There was fluid retention in the left thoracic cavity. The mass was heterogeneous, with high attenuation values (20-40 HU). No contrast enhancement of the mass or extravasation of contrast media was seen.

As the imaging studies were inconclusive, CT-guided puncture was performed the next day for diagnostic purposes. The mass contained a dark red bloody component, and drainage was performed for treatment. The patient was discharged 7 days after onset with a provisional diagnosis of chronic ex-

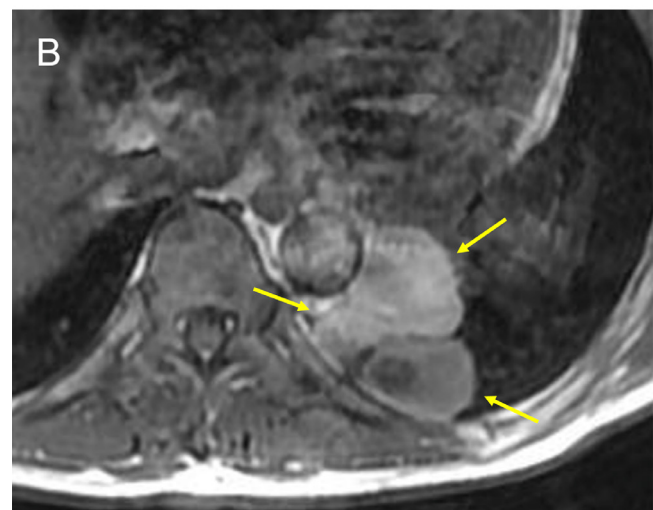
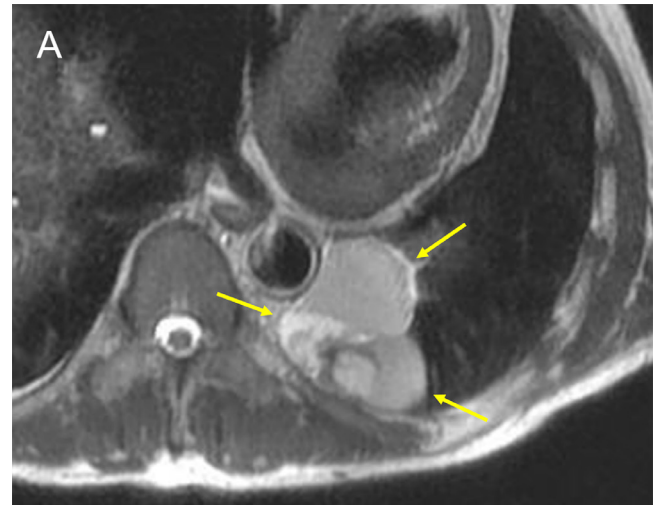


Fig. 2 – MRI of the pancreas. (A) T2-weighted image in the axial plane show a mass with heterogeneous high signal intensity and an internal septum (arrow). (B) T1-weighted image in the axial plane shows low to high signal intensity of the mass (arrow).

panding hematoma. In follow-up at the outpatient clinic approximately 3 weeks later, CT showed slight shrinkage of the mass on the left diaphragm to 6.1×3.6 cm in diameter and no fluid retention in the left thoracic cavity.

Due to a high CA19-9 level detected during hospitalization, MRI of the pancreas was performed for further investigation about 4 weeks after discharge. No abnormality was found in the pancreas, but the lesion identified on the left diaphragm on CT was visualized as a $6.2 \times 4.5 \times 6.5$ cm mass with smooth margins and divided by an internal septum (Fig. 2). The mass had low to high signal intensity on T1-weighted imaging and heterogeneous high signal intensity on T2-weighted imaging, suggestive of hemorrhage within a multilocular mass (Fig. 2).

As the mass did not appear to be increasing in size, the patient was scheduled for follow-up in 6 months. However, approximately 11 weeks after discharge from the hospital, the patient again presented to the emergency department with severe chest pain. Chest CT on arrival revealed increased size

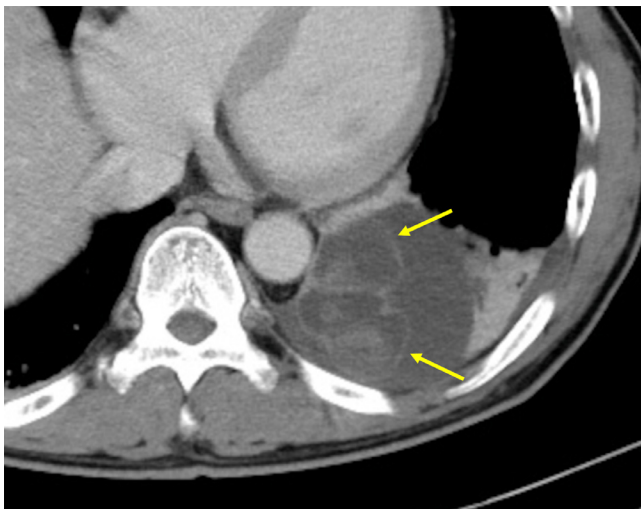


Fig. 3 – Contrast-enhanced CT of the chest at the second episode shows an enlarged mass (arrow) with fluid retention in the left thoracic cavity.

of the mass on the left diaphragm and fluid retention in the left thoracic cavity. The attenuation values of the fluid in the thoracic cavity were about 5-10 HU, and there was no extravasation or enhancing effect on contrast-enhanced CT (Fig. 3). A ruptured left posterior mediastinal cystic mass was suspected, and surgical resection of the mass was performed for diagnostic and therapeutic purposes. Upon thoracoscopic exploration, there were no adhesions or pleural effusions in the left thoracic cavity and fibrin clots were scattered. The mass was recognized as a cystic lesion on the left diaphragm in the posterior mediastinum, with partial adhesions to the lung. The cystic mass was completely removed through a small thoracotomy. The gross appearance of the resection specimen was of a multilocular mass with a reddish-brown and hemorrhagic

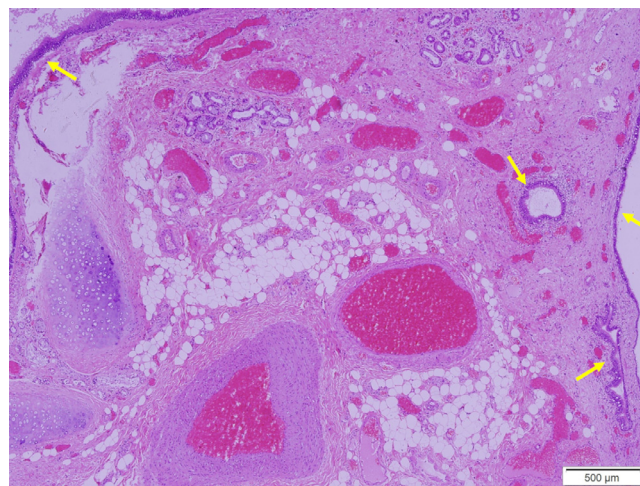


Fig. 5 – Pathological findings. The wall of the lesion is composed of bronchial epithelium (arrows), cartilage, blood vessels, adipose tissue, peripheral nerve tissue, and ganglion cells. (Hematoxylin & eosin [H&E] stain, 8 × 40.)

appearance inside the mass (Fig. 4). On pathological examination, the lesion contained bronchial epithelium, cartilage, blood vessels, adipose tissue, peripheral nerve tissue, and ganglion cells (Fig. 5). On this basis, the lesion was diagnosed as a bronchogenic cyst. The patient had an uncomplicated post-operative course and was discharged from the hospital after 6 days.

Discussion

Bronchogenic cyst is a congenital malformation originating from the primitive foregut. It is the most common primary cyst

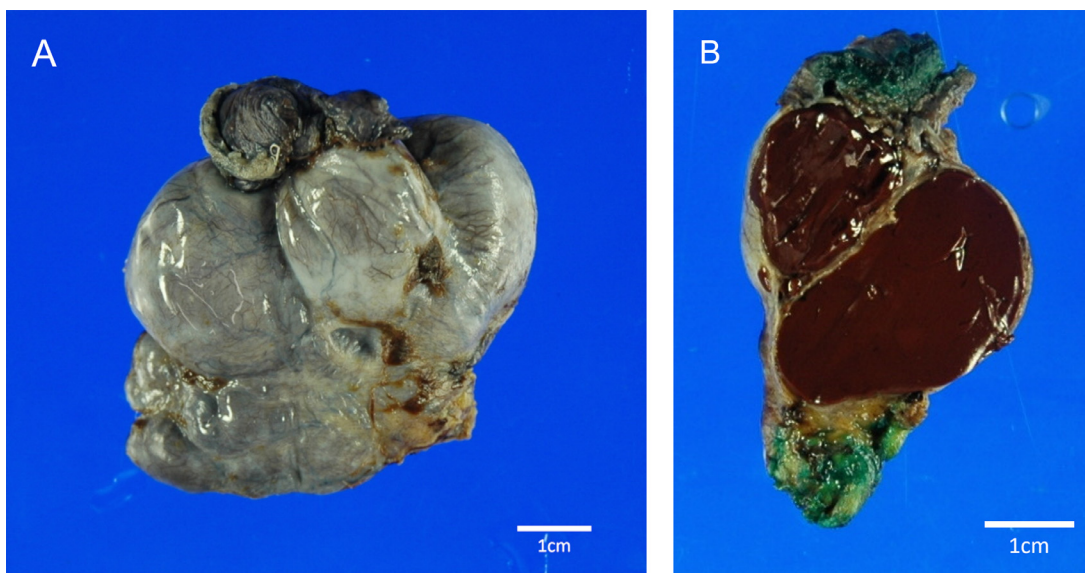


Fig. 4 – Photograph of the gross specimen. (A) The mass is multilocular with reddish-brown hemorrhagic components inside the mass (B).

of the mediastinum, occurring mostly in the mediastinum and lungs [2]. These cysts are usually diagnosed by chest radiographs or CT, and present as monolocular, smooth margined masses [3,5,8]. CT can clarify the relationship between the cyst location and the surrounding organs; however, cysts with hemorrhage or infection may show atypical imaging findings, which complicate the differential diagnosis [5,6]. The present patient presented with atypical imaging findings, of a mass with high attenuation values and fluid retention in the thoracic cavity on initial hospital admission, which made the diagnosis difficult. The feature of high attenuation values, which may sometimes lead to misdiagnosis as a solid tumor, can also be an important clue in distinguishing bronchogenic cysts from other cystic lesions [1,9]. Among cystic lesions recognized in the mediastinal region, bronchogenic cysts may be differentiated based on the feature of atypically high attenuation values, as in the present case. MRI has higher accuracy than CT for identifying bronchogenic cysts, which are characterized by high signal intensity on T2WI regardless of the cyst contents. In this case, the mass showed slightly higher attenuation values than water on CT, low to high signal intensity on T1-weighted imaging, and heterogeneous high signal intensity on T2-weighted imaging. With respect to the pathological findings, the imaging appearances were due to bleeding within the cyst.

The majority of bronchogenic cysts are asymptomatic and have a good clinical course. Symptomatic cysts are sometimes seen, with common symptoms of chest pain, dyspnea, cough, wheezing, and respiratory compromise due to tracheal/bronchial compression [2,7]. Most symptoms depend on the location of the cyst rather than its volume and result from compression of adjacent structures such as the esophagus, bronchi, and heart [2,7,10]. In contrast, pain is caused more by irritation of the pleura than by compression [7]. In the present case, the back pain was sudden rather than chronic, suggesting that the pain was not caused by compression of the pleura by the cystic mass but by pleural irritation from the sudden rupture of the cystic mass.

Spontaneous rupture of a bronchogenic cyst is a very rare complication and is reported to occur sometimes into the trachea, pleural cavity, or pericardial cavity [4,7,8]. In both of the present episodes, there was fluid retention in the thoracic cavity ipsilateral to the cyst, which was considered to have ruptured into the thoracic cavity. A literature search revealed only one such previously reported case of rupture into the thoracic cavity [11]. Rupture is caused by rapid enlargement of the cyst, most commonly due to infection [12]. Although cases of short-term enlargement without signs of infection have been reported, most cases of enlargement during the course of the disease are accompanied by inflammatory reactions such as leukocytosis or elevated C-reactive protein [12]. In our case, there was no sign of infection in either episode, suggesting a noninfectious mechanism of enlargement and rupture. One such mechanism is hypersecretion of the bronchial glands intrinsic to the cyst [12]. Bronchial gland secretions contain amylase, and abnormally high amylase levels have been reported in intracystic solutions in cases of symptomatic mediastinal occurrence [12]. Bronchogenic cysts can also enlarge and rupture as a result of increased fluid in the cyst due to bronchial gland hypersecretion and in-

creased intracystic pressure. However, because the cyst contents were bloody in the present case, it is reasonable to assume that the cause of the rupture was not bronchogenic gland hypersecretion, but rather rupture of blood vessels supplying the bronchogenic cyst, which caused hemorrhage into the cyst followed by rapid expansion and rupture. In the present case, the second rupture of the bronchogenic cyst may have been related to tissue fragility caused by the puncture.

The principal treatment for bronchogenic cysts is surgical resection [4]. In the present case, a puncture was performed for diagnostic purposes on initial admission, including differentiation between solid or cystic tumor, and the contents were diagnosed as cystic with bloody contents. Although drainage was performed for treatment and tumor shrinkage was confirmed, the lesion recurred about 3 weeks later and enlarged to the point of rupturing again in less than 3 months. This is consistent with the fact that transbronchial/percutaneous cyst aspiration, which has been proposed as an alternative to surgery in the treatment of bronchogenic cysts, has not been widely accepted because of the high risk of recurrence [4,5,8]. Therefore, in cases of suspected bronchogenic cyst, surgical resection is the treatment of choice whenever possible [4,5,6]. In addition to obtaining a histologic diagnosis, surgical resection also prevents complications and recurrence [5,7,8].

In the present case, if bronchogenic cyst had been included in the differential diagnosis at the time of the initial episode, surgical intervention at that time may have prevented recurrence. Accordingly, recognition of atypical imaging findings and the diverse symptoms of bronchogenic cyst would be helpful in diagnosis. Although cyst rupture is a very rare complication, it is important to recognize that cyst enlargement can be caused by mechanisms other than infection, such as hemorrhage or bronchial gland hypersecretion, and that even patients without signs of infection may have a risk of rupture. This case highlights the importance of recognizing the atypical imaging findings of bronchogenic cyst and the rare complication of rupture.

Conclusion

We experienced a case of mediastinal bronchogenic cyst that ruptured twice in a short period of time. When a mediastinal mass is present along with sudden chest pain, the differential diagnosis should include the possibility of rupture of a cystic mass, and it should be recognized that bronchogenic cysts, although very rare, can also cause rupture.

Patient consent

Written informed consent for the publication of this case report was obtained from the patient.

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