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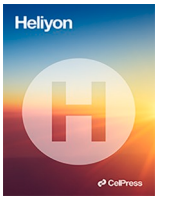

Case report: Two rare new cases of diaphragmatic bronchogenic cysts

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ABSTRACT

The bronchogenic cyst (BC) is a type of congenital anomaly that is most usually found in the mediastinum and lung, and rarely in the diaphragm. We report two cases of bronchogenic cysts originating from the left diaphragm. Case 1 involved a 50-year-old man who underwent computed tomography (CT) of the adrenal glands for hypertension, showing left adrenal changes. An adrenal CT at our hospital showed a space-occupying lesion above the left diaphragm. We performed a left-sided thoracotomy in the seventh intercostal space and found that the mass was entirely located in the diaphragm. Case 2 was a 58-year-old woman who underwent chest CT under a standard health check-up, and a subpleural lesion of approximately 4 cm was found in the posterior basal segment of the left inferior lung lobe. During thoracoscopic surgery, the mass was found to originate from the surface of the diaphragm, having no correlation with the lung tissue. The two cases of diaphragmatic bronchogenic cysts were confirmed by surgical pathology. Ectopic bronchogenic cysts arising from the diaphragm are very uncommon, and diaphragmatic bronchogenic cysts typically occur on the left side, in women, and in the angle between the vertebral column and the diaphragm. As far as we know, there are no relevant reports of intra-diaphragmatic BCs similar to case 1, which was entirely located within the diaphragm. Most diaphragmatic BCs are located on the surface of the diaphragm and project toward the thoracic or abdominal cavities. Moreover, bilocular diaphragmatic bronchogenic cysts, similar to case 2, have not yet been reported.

1. Introduction

The bronchogenic cyst (BC) is a type of congenital cyst that occurs as a result of abnormal differentiation of the ventral foregut outgrowth. BC is usually located in the mediastinum, lungs, and rarely in other locations, depending on the degree of abnormal germination during development [1]. However, few cases of diaphragmatic BCs have been published in English literature. Kara et al. [2] reported the 32nd case of diaphragmatic BCs. We identified four additional case reports published up until May 2022 [1,3–5]. Herein, we present two cases of surgically resected BC arising from the left diaphragm.

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2. Case presentation

2.1. Case 1

A 50-year-old Chinese man experienced a sudden increase in blood pressure without apparent cause or other symptoms. Clinical history and family history were uneventful. Subsequently, he underwent computed tomography (CT) of the adrenal glands, which suggested changes in the left adrenal gland. The patient visited the urology department for another CT scan, which showed a space-occupying lesion above the left diaphragm. A chest scan and enhanced CT showed a mass-like high-density shadow above the left diaphragm with poor demarcation between the lesion and the diaphragm, as well as multiple nodular calcified shadows measuring approximately 7.0 cm × 3.8 cm × 3.9 cm and with a CT value of approximately 65 HU (Fig. 1: A, B, C, D, arrows). A left-sided thoracotomy was performed through the seventh intercostal space, and the diaphragm surface was intact. The diaphragm was cut between the liver and spleen, followed by the opening of the abdominal cavity to expose the base of the diaphragm, which was also intact. A cystic occupancy was palpated within the diaphragm, and upon opening it, a dark yellow turbid fluid was observed flowing out (Fig. 2A). The cyst fluid was suctioned, and the cyst and its wall were removed intact. The wound was covered with anhydrous alcohol, and the diaphragm was sutured. The postoperative course went well, and the patient was discharged on the fifth postoperative day. Histopathological findings showed that the cyst was made up of smooth muscle, lax connective tissue, and pseudostratified ciliated columnar epithelium (Fig. 3: A, B). Due to the impact of the COVID-19 pandemic, the patient was not able to undergo a chest CT review in our hospital; however, three months after the operation, he underwent a chest CT in the outpatient clinic of the local hospital, and we learnt through telephone follow-up that the patient did not show any signs of recurrence.

2.2. Case 2

A 58-year-old Chinese woman underwent a chest CT as part of a health check-up, and the scan revealed a subpleural lesion in the posterior basal segment of the left inferior lung lobe, with a diameter of approximately 4 cm (Fig. 4: A, B, C, D, arrows). There were no clinical symptoms, surgical history, disease history, or family history. Under general anesthesia, the patient underwent exploratory video-assisted thoracic surgery (VATS) through the seventh intercostal space. A bilocular cyst (Fig. 2: B, C, D) was observed on the left

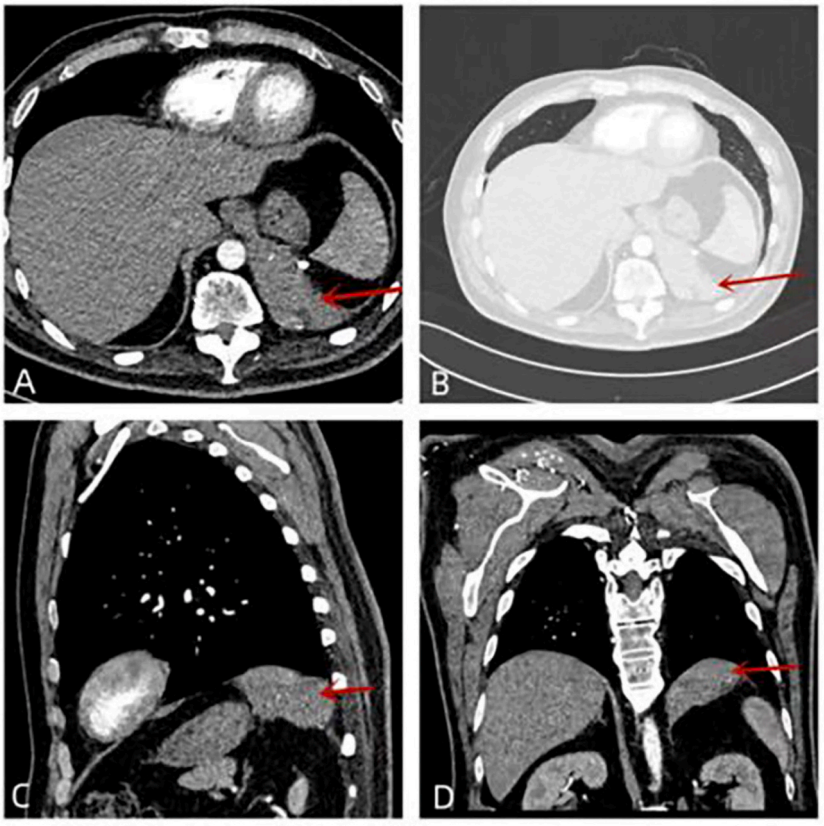

Fig. 1. (A–D) Preoperative computed tomography imaging showing a mass-like high-density shadow above the left diaphragm, measuring approximately 7.0 cm × 3.9 cm × 3.8 cm (red arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

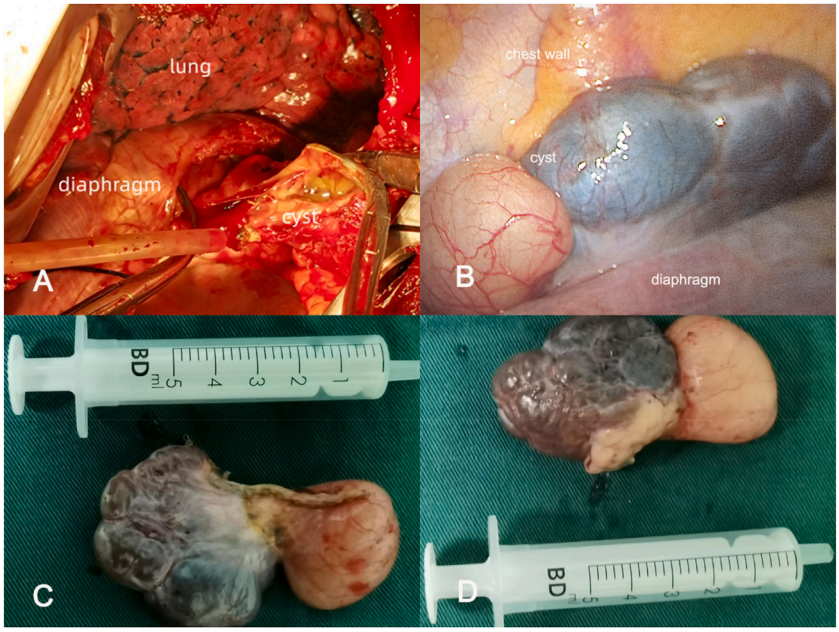

Fig. 2. (A) Case 1: Intraoperatively, the cyst was located within the diaphragm and perforated during dissection, resulting in the discharge of a thick, yellowish mucoid secretion from the cyst. (B) Case 2: During the operation, a bilocular cyst, partly yellow and partly black cystic, was seen on the left side of the diaphragm near the spine, away from the lung tissue. (C, D) Case 2: Gross macroscopic specimen. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

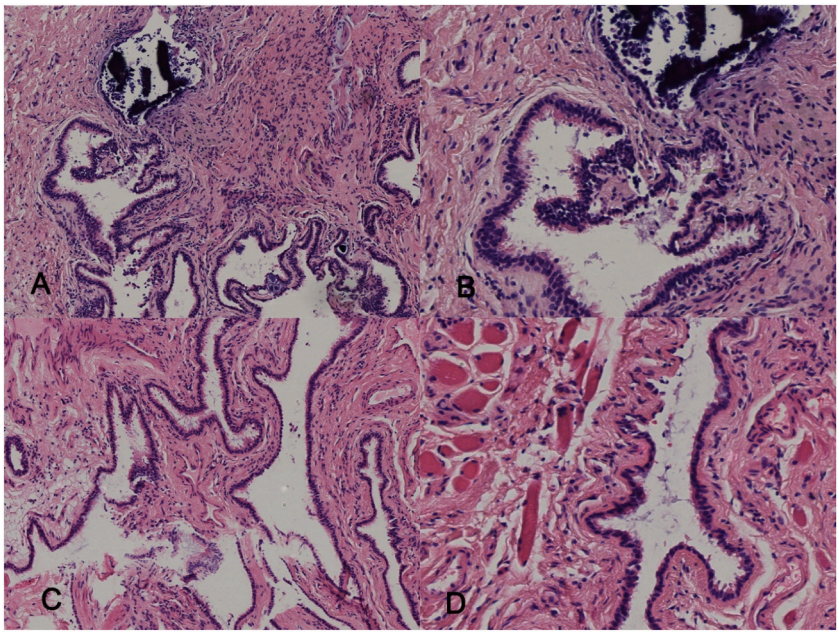

Fig. 3. (A–D) Postoperative pathology indicated the mass consisting of smooth muscle, loose connective tissue, and pseudostratified ciliated columnar epithelium (Haematoxylin and eosin stain, A and B: original magnification × 100; C and D: original magnification × 200).

side of the diaphragm near the spine, showing no correlation with the lung tissue. The cyst was partly yellow and black, with an intact envelope and a narrow base on the surface of the diaphragm. It was dissected off the diaphragm using a linear cutting stapler. Postoperative recovery went well, and the patient was discharged on the fourth postoperative day. BC nature was confirmed by postoperative pathological examination (Fig. 3: C, D). A three month postoperative chest CT performed at the local hospital confirmed there was no recurrence of the cyst. We were informed of this telephonically.

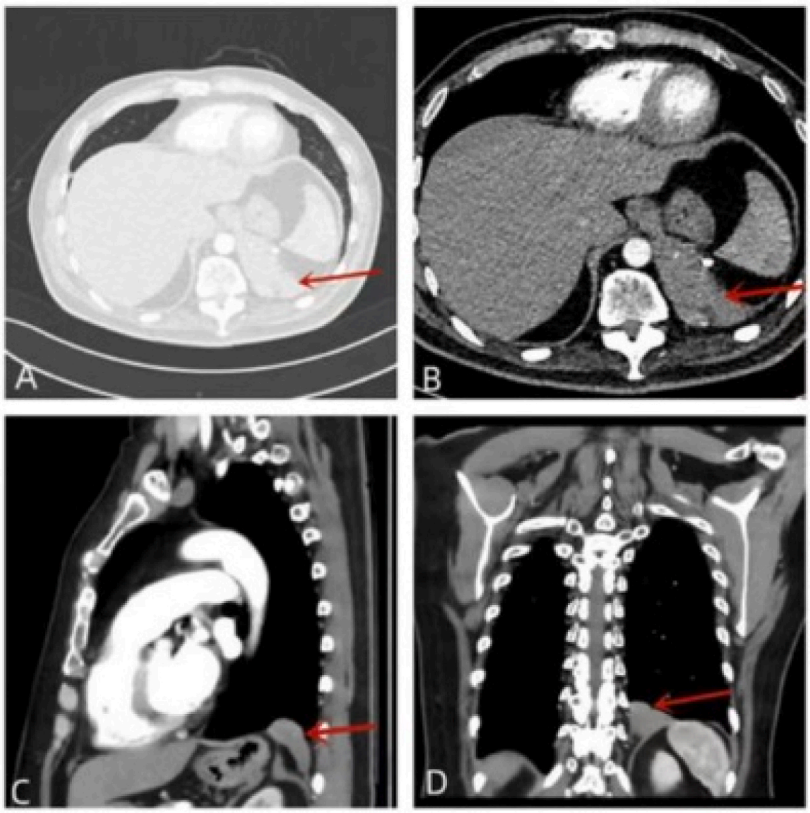

Fig. 4. (A–D) Contrast-enhanced computed tomography revealing a subpleural lesion in the posterior basal segment of the left lower lobe, with a diameter of approximately 4 cm (red arrow). (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

3. Discussion

BCs are extremely rare benign masses that are more commonly found in women [6]. Embryologically, BCs arise from the foregut as a result of bronchial malformations [6]. In clinical practice, BCs are generally located in the lung parenchyma or mediastinum; however, in some rare cases, they may detach and migrate to other areas [1]. Depending on the location of the lesions, BCs can be classified into three types: mediastinal, intrapulmonary, and ectopic, among which the ectopic type has the lowest incidence. Diaphragmatic BCs are very rare. A total of 36 cases of diaphragmatic BCs have been reported in the English literature (1–5) . Diaphragmatic BCs typically occur on the left side and at the angle between the vertebral column and the diaphragm. Most patients with BCs are asymptomatic and are identified inadvertently while being diagnosed with other diseases or through health-check imaging. Usually, clinical symptoms are directly related to the location of the lesions, and patients with ectopic BCs may show a range of symptoms, such as abdominal pain or discomfort, chest pain, dyspnea, and dysphagia [1]. Furthermore, BCs have potential complications, involving infection, cyst rupture, bleeding, and even malignant transformation [1]. Imaging tests are of great clinical value for the early detection of ectopic BCs, but BCs have no specific presentation on CT or MRI scans, and thus these do not provide a conclusive diagnosis [3]. Diagnosing an intradiaphragmatic BC using imaging methods alone can be difficult. Preoperative localization is sometimes challenging, as the diaphragm is a thin and curved structure, and any mass lesion may protrude into the thoracic or abdominal cavity [7]. In case 1, chest CT revealed a left supradiaphragmatic occupancy, and intraoperatively, the mass was found to be located inside the diaphragm and diagnosed as a diaphragmatic cyst. Preoperative imaging of case 2 showed a subpleural lesion located in the posterior basal segment of the inferior lobe of the left lung. Intraoperatively, the mass was found to be a bilocular cyst originating from the diaphragm. A 2016 review by Mubang et al. [8] concluded that while the presumed diagnosis before resection varied widely, BC was most often confused with adrenal masses, as in case 1. Ectopic BCs can be confused with esophageal diverticulum, diaphragmatichernia, neurogenic tumors, or diaphragmatic tumors (6.9). Diagnosing based on clinical manifestations and imaging studies alone is difficult and depends on pathology [10]. The pathology of BCs is characterized by the presence of ciliated pseudostratified epithelium, cartilage, and smooth muscle within the cyst wall [8]. The treatment options include surgical excision and cyst aspiration. Surgical excision is recommended, as the imaging diagnosis is usually inconclusive, and there are reports of malignant transformation [9] and recurrence of BCs following cyst aspiration [7].

3.1. Limitations

The follow-up time was too short to determine long-term survival and recurrence. The follow-up should be continued to determine the long-term benefit of surgery.

4. Conclusion

Ectopic BCs arising from the diaphragm are extremely uncommon. Considering the extreme difficulty in diagnosis by imaging alone, risk of recurrence with cyst aspiration, possibility of malignant transformation, and presence of complications, complete surgical excision is recommended, even in asymptomatic patients. While making a differential diagnosis of abnormal masses in the diaphragmatic region, intradiaphragmatic BCs should be considered. Generally, ectopic BC has a good prognosis for benign tumors if complete surgical resection is performed.

Ethics statement

Ethical review and approval were not required for the study on human participants in accordance with the local legislation and institutional requirements. The patients/participants provided their written informed consent to participate in this study. Written informed consent was obtained from the individuals for the publication of any potentially identifiable images or data included in this article.

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Data availability statement

Data included in article.

CRediT authorship contribution statement

Chao He: Conceptualization, Data curation, Formal analysis, Investigation, Methodology, Supervision, Visualization, Writing – original draft, Writing – review & editing. **Hai-Xiang Yu:** Data curation, Formal analysis, Investigation, Project administration, Resources, Supervision, Validation, Writing – review & editing. **Zheng-Jia Liu:** Data curation, Formal analysis, Investigation, Validation, Visualization, Writing – review & editing. **Cong Yin:** Data curation, Formal analysis, Validation.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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