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Case Report Open Access

Intradiaphragmatic Bronchogenic Cysts: A Case Report and Literature Review

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Abstract

Bronchogenic cysts are a congenital developmental abnormality arising from the ventral foregut and are found most commonly in the mediastinum. However, several case reports indicated that bronchogenic cysts occur rarely within the diaphragm. Mubang et al. reported a case of intradiaphragmatic bronchogenic cyst and summarized 21 cases previously reported in the English literature. Most cases had several clinical symptoms, including back and/or chest pain, cough, etc. Here, we report a case of intradiaphragmatic bronchogenic cyst detected incidentally on chest computed tomography (CT) in an adult male patient without any clinical symptoms.

Keywords: Bronchogenic cysts; Diaphragm; Intradiaphragmatic bronchogenic cysts

Case Report

A 68-year-old man who suffered from pharyngeal cancer had treatment histories including initial operation, subsequently radiotherapy, and chemotherapy over the previous 2 years. He had ceased treatment for cancer because of deterioration of his systemic condition. He was hospitalized frequently due to gastrostomy problems and pneumonia. When hospitalized with pneumonia, chest computed tomography (CT) revealed a boundary clear cystic lesion in his left diaphragm, which was considered to be a benign tumor based on the contrast pattern, and not metastasis of pharyngeal cancer (Figure 1). He died due to worsening of his underlying illness and pneumonia. An autopsy was performed and a cystic lesion was detected in the diaphragm. The pathological findings revealed linear cystic lesions with cuboid or pili-bearing columnar epithelium (Figure 2A and 2B), which were positive for thyroid transcription factor-1 (TTF-1) on immunohistological staining (Figure 2C). These findings confirmed the diagnosis of intradiaphragmatic bronchogenic cyst [1-3].

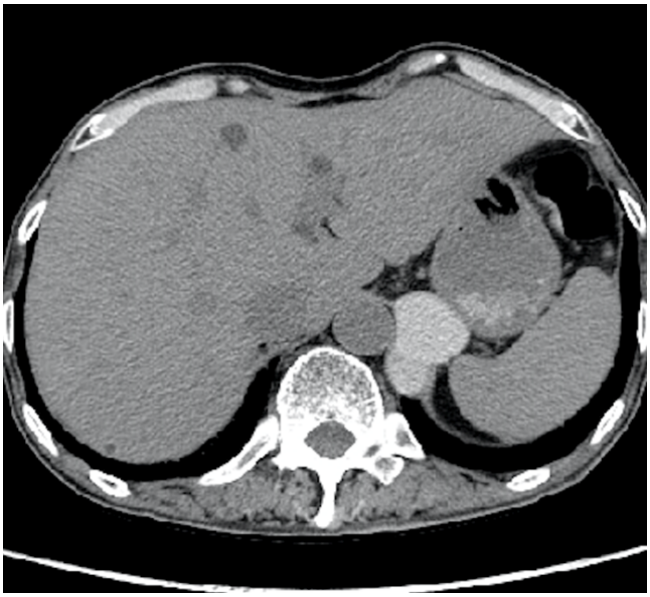

Figure 1: Chest computed tomography revealed a boundary clear cystic lesion in the left diaphragm.

Discussion

Bronchogenic cysts, which are congenital cysts that develop due to abnormal germination in early gestation, frequently occur in the mediastinum. They develop from abnormal buds pinched off the primitive tracheal anlage or the bronchial tree after the third week of embryonic life [1,2]. If the abnormal buds are pinched off and subsequently migrate before fusion of the diaphragm components at the end of the sixth week of embryonic life, the cysts may also be found in the subdiaphragmatic region [1]. Mubang et al. reported that intradiaphragmatic bronchogenic cyst tended to be more common in females and to be present on the left side [3]. Saper et al. reported that mediastinal bronchogenic cysts were frequently found in men [4]. Intradiaphragmatic bronchogenic cyst is extremely rare, and the present case indicates that bronchogenic cysts can develop in the diaphragm. In addition, intradiaphragmatic bronchogenic cyst should be included as a differential diagnosis of unusual masses in the diaphragm.

Many symptoms have been reported for bronchogenic cyst, with pain as the most common presenting symptom in patients [5]. Other symptoms include fever (due to infection), cough, and dyspnea [3,4,6]. However, our patient did not have symptoms and the lesion was discovered incidentally. Mubang et al. reported that 14% (4/21) of patients with intradiaphragmatic bronchogenic cysts were asymptomatic [4].

Imaging studies have an increasingly important role in the diagnosis of intradiaphragmatic bronchogenic cysts. Consistent with the characteristics of mediastinal bronchogenic cysts, CT and MRI indicated homogenous soft tissue and hypoenhancing masses [3]. The fluid in these cysts has an average CT density of 30-100 Hounsfield units [1,4,6]. However, the clinical and radiological findings of intradiaphragmatic bronchogenic cyst are confusing because of its rarity. Furthermore, surgical resection and pathological examination

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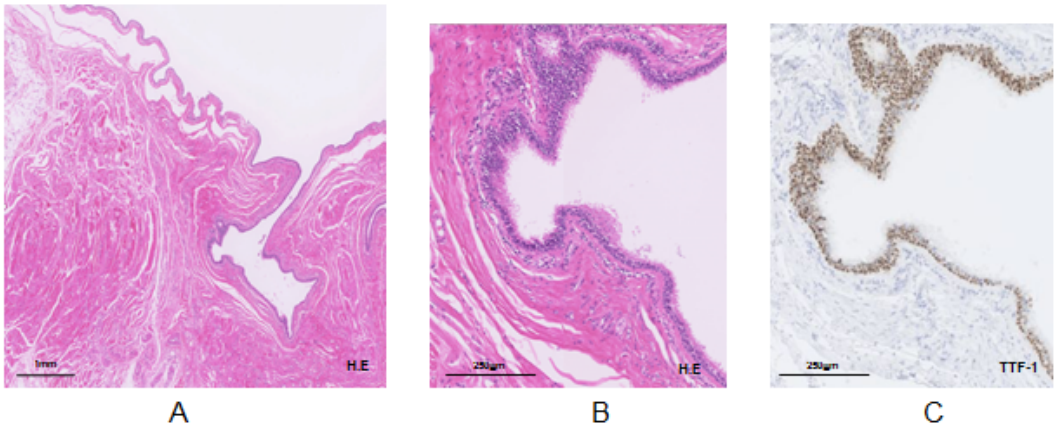

Figure 2: Pathological examination revealed lined cystic lesions with pilus columnar epithelium or cuboid epithelium (A: ×40 and B: ×100). These epithelial cells were positive for thyroid transcription factor-1 (TTF-1) on immunohistological staining (C).

remain necessary for accurate diagnosis of intradiaphragmatic bronchogenic cyst [7]. The pathological hallmarks of bronchogenic cysts are the presence of ciliated pseudostratified columnar epithelium, cartilage, and smooth muscle within the cyst wall [3]. Malignancy has been reported to arise from bronchogenic cysts [3,7,8], so surgical resection is recommended for all suspected bronchogenic cysts in operable candidates.

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