An atypical presentation of bronchogenic cyst

Atypická prezentace bronchogenní cysty

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Summary
Background: Bronchial cysts (BCs) can be difficult to diagnose because of non-specific site of occurrence and heterogeneous density of cyst content in some patients. We present herein a BC case with such nonspecific findings. ***Case:*** A 23-year-old man referred to our hospital because of an abnormal chest image during a mass-screening. CT revealed a cystic nodule in the posterior mediastinum. The content of the cystic lesion was heterogeneous, measuring 30–100 Hounsfield Units, and it was surgically resected and diagnosed as a BC. ***Conclusion:*** Diagnosis of BC could be difficult in patients like this, who have cysts that occur in relatively rare sites such as the posterior mediastinum, or whose cyst contents are highly dense and heterogenous. If atypical findings are observed on images, it is necessary to actively perform histological diagnosis to differentiate it from other diseases.

Key words
bronchogenic cyst – mediastinal tumor – computed tomography – resection

Souhrn
Východiska: Bronchiální cysty (BC) mohou být u některých pacientů obtížně diagnostikovatelné kvůli nespecifickému místu výskytu a heterogenní hustotě obsahu cyst. V tomto článku prezentujeme případ BC s takovým nespecifickým nálezem. ***Případ:*** Muž ve věku 23 let byl odeslán do naší nemocnice kvůli abnormálnímu obrazu hrudníku při plošném screeningu. CT odhalilo cystický uzel v zadním mediastinu. Obsah cystické léze byl heterogenní, měřil 30–100 Hounsfieldových jednotek a byl chirurgicky resekován a diagnostikován jako BC. ***Závěr:*** U pacientů, jako je tento, může být diagnostika BC obtížná. Cysty se u nich vyskytují v relativně vzácných lokalitách, jako je zadní mediastinum, nebo obsah cysty je velmi hustý a heterogenní. Pokud jsou na snímcích pozorovány atypické nálezy, je nutné aktivně provést histologickou diagnostiku, aby se odlišila od jiných onemocnění.

Klíčová slova
bronchogenní cysta – tumor v mediastinu – výpočetní tomografie – resekce

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Introduction

Bronchogenic cysts (BCs) are rare benign congenital disease. This disease develops mainly in young subjects, and most of they have incidentally been discovered during treatment of other diseases or on chest radiograph in mass-screening in hilum and middle mediastinum [1–8]. Since the contents of BC are liquid, many BCs have CT values up to 20 Hounsfield units (HU) [1,5,8], but some BCs have shown CT values up to about 50 HU [5,6]. Herein we show a case with BC developed from the posterior mediastinum. As the density of the contents were high and heterogenous, it was excised and diagnosed as BC. Although rare, we report our clinical experience as it might provide some suggestions for the treatment of patients who will have a similar course in the future.

Case report

A 23-year-old man was referred to our hospital due to abnormal finding on chest radiograph in mass-screening. He had no medical history. Physical examination on admission was unremarkable. There were no abnormalities in blood and respiratory function tests. A chest radiograph on admission revealed a nodule adjacent to thoracic vertebrae (Fig. 1). A chest CT scan showed smooth margin nodule with 3 cm in diameter in the left posterior mediastinum (Fig. 2). The density inside the nodule was heterogeneous, with the lowest density being 4.0 HU, the highest being 90.0 HU, and the average density being 51.9 HU (Fig. 2). There were no irregularities or thickened areas in the cyst wall. As the contents were high and heterogenous density for a benign cyst, we considered it necessary to histologically differentiate the presence or absence of malignant components, and the patient underwent resection with video-assisted thoracic surgery (VATS). VATS revealed a hemispherical raised lesion with a smooth surface in the posterior mediastinum. There was no redness or bleeding in the area covering the lesion. The lesion was excised with an 'en bloc'. When the excised lesion was incised, jelly-like contents flowed out, but it was not possible to analyze this content. A hyaline-like stroma was observed inside the cyst wall. The inner surface of the cyst was covered with ciliated columnar epithelium and there were sites where lymphocytes gathered. Cartilage tissue was confirmed in some parts (Fig. 3). Based on the above, a diagnosis of BC was made. Post-operative course was unremarkable. Two years have passed since the initial episode, there has been no recurrence and the patient is doing well.

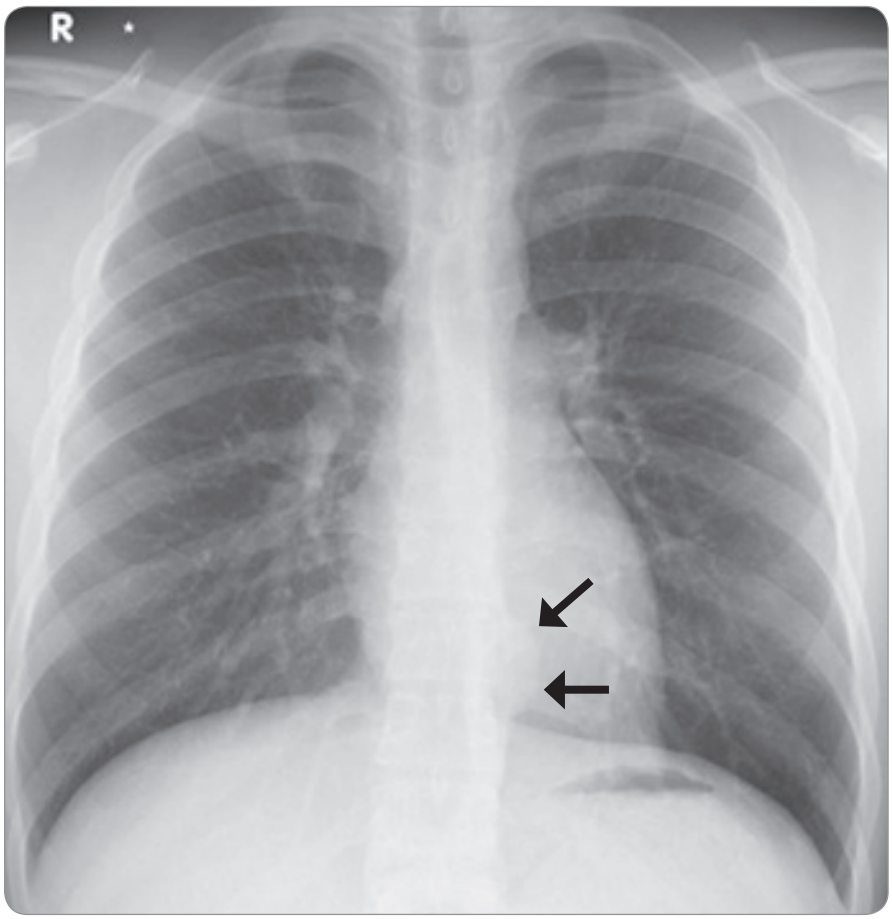

Fig. 1. Chest radiograph on admission revealed a nodule adjacent to the thoracic vertebrae (arrows).

Discussion

BC is a relatively rare benign disease that occurs as a result of abnormal budding, separation, or migration of the tracheal primordium during the embryonic period [7]. Pathologically, a BC has a structure similar to a bronchial wall, and the cyst wall contains smooth muscle, bronchial glands, and cartilage tissue, and the lumen of the cyst wall is lined with ciliated columnar epithelium, however, there are not many patients who have all of these structures [8]. Most of BCs occurred in the mediastinum and the hilum [1–4], but were also known to originate from the lungs and the retroperitoneum [9, 10]. Many studies reported that BCs originating from the mediastinum developed more frequently from the anterior and middle mediastinum, and less frequently from the posterior mediastinum [2–4]. In a report by Ma et al., out of 66 patients with BCs, 38 patients had BCs that originated from the anterior mediastinum, and 12 had BCs that developed from the posterior mediastinum [2]. Kawaguchi et al. showed that in only 1 of 12 patients BC originated from the posterior mediastinum [3]. According to a report by Muramatsu et al., they treated 13 mediastinal BCs, and 5 of them originated from the posterior mediastinum [5]. However, some reports have shown that BC occurring in the posterior mediastinum is not necessarily rare [11,12]. Martinod et al. reported

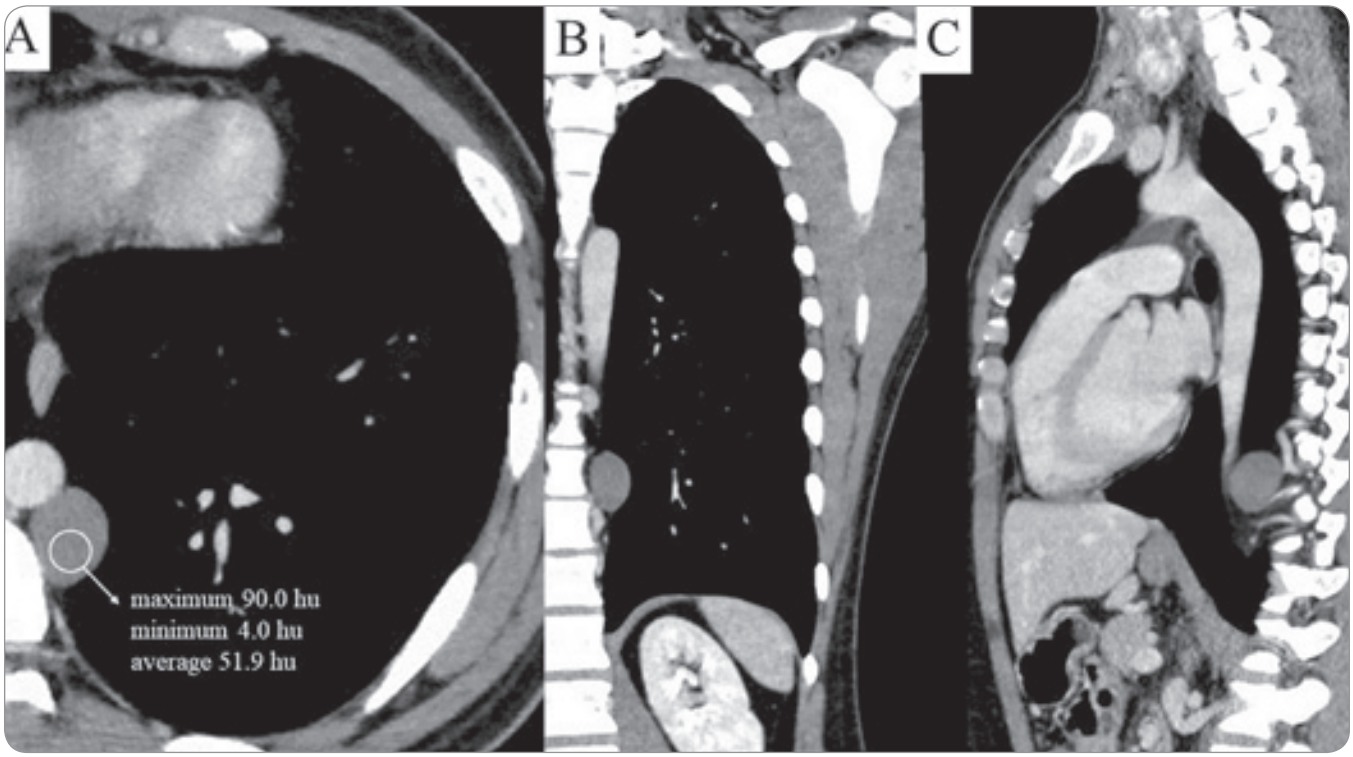

Fig. 2. Chest CT scan shows a smooth margin heterogenous nodule with 3 cm in diameter in the posterior mediastinum: transverse view (A), coronal view (B), sagittal view (C).

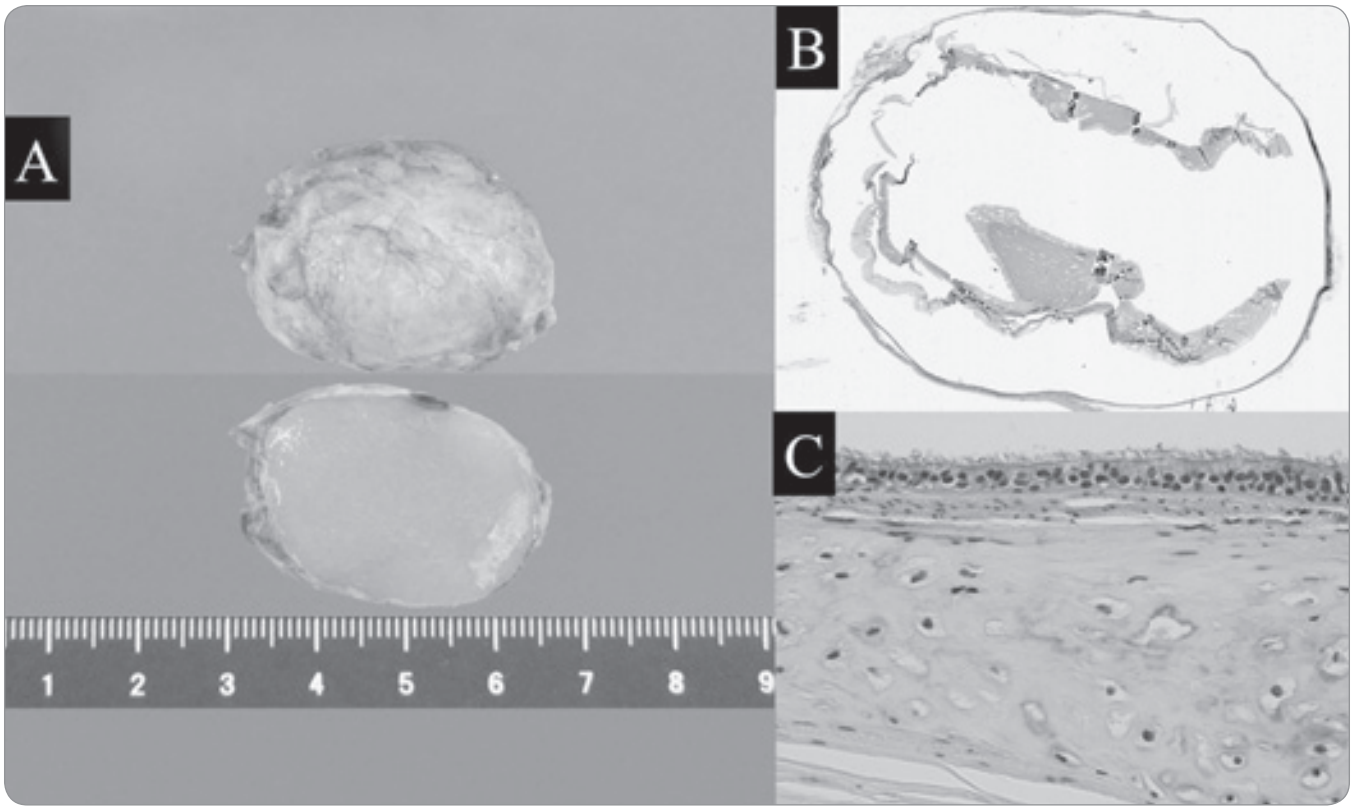

Fig. 3. Surface of the resected lesion (A-upper). The size of the cyst was 35 × 25 × 20 mm. Cut surface of the lesion (A-lower). A hyaline-like stroma was observed inside the cyst wall. Low-power photomicrograph (B) and high-power photomicrograph (C) after formalin fixation. The inner surface of the cyst was covered with ciliated columnar epithelium and there were sites where lymphocytes were gathered. Cartilage tissue was confirmed in some parts.

that 10 out of 20 patients with mediastinal onset had posterior mediastinal onset [11]. Fernandez et al. reported that the development of bronchial cysts from the posterior mediastinum was not extremely rare [12]. We should be careful not to exclude BC from the differential diagnosis even if it arises from atypical locations, including the posterior mediastinum.

CT scan provides important information in the imaging diagnosis of BC. Although BC is a 'cyst', it is important to note that there have been patients in whom the density inside the cyst is not necessarily 'water density' [1,6,13–16]. Vos et al. reported a patient with BC of heterogeneous density located in the posterior mediastinum [15]. Their patient had a 20 HU BC of homogeneous density. In addition to this patient, they reported 3 patients with posterior mediastinal BCs. The CT densities of those cysts were 20, 30, and 50 HU, respectively. Gaeta et al. indicated that chronic entrapped mucus collections, due to water reabsorption and higher protein content, can have CT densities higher than 20 and reaching even 130 HU [6]. Very recently, Gu et al. showed CT density of BCs [1]. According to them, CT densities of 83 BCs were 36.3 ± 1.9, 29.7 ± 7.9, and 37.5 ± 4.6 HU [1]. They concluded that misdiagnosis occurs frequently when CT densities exceed 20 Hu [1]. These fluctuations in CT values were due to the fluid components inside the cyst having high calcium concentration, high protein concentration, hemorrhage, high viscosity, and inflammation [17]. If CT densities of internal structures are high, it might be difficult to differentiate from solid tumors. Some previous researchers described that the accuracy rate of BCs images was approximately 60–70% [13,14].The BC in our patient had atypical CT imaging findings for BCs, including development from the posterior mediastinum and high internal density and heterogeneity.

When a BC is suspected, the diagnosis must be confirmed in the presence of several atypical findings, such as location of origin, high CT value, and heterogeneous CT density. Even magnetic resonance imaging and PET/CT cannot confirm the contents of the cyst or degree of an atypicality of the cells in the cyst wall. In such cases, resection and aggressive confirmation of the pathological diagnosis must be recommended.

Conclusions

Although rare, there are patients with BCs that exhibit atypical location and morphology, as observed in our patient. For patients with atypical characteristics, treatment options, including surgical treatment, must be carefully considered. Close follow-up will be required even after resection.

Statement of Ethics

This study was approved by the institutional ethics committee of our institute (No. 1639). Written comprehensive informed consent at the time of admission for obtaining pathological specimens was obtained from the patient.

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Authors' contributions

HM and HS designed the study. HM, SH, YM, SO, GO, KI,HK, NT and HS collected data. HM and HS prepared the manuscript. All Authors approved the final version for submission.

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