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Case Report

Bronchogenic cyst at unusual location

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ABSTRACT

Bronchogenic cyst results from abnormal bronchial budding. Thin-stalked mobile bronchogenic cysts are rare and sometimes radiologically mimic mass lesion, making preoperative diagnosis difficult. We present a 12-year-old boy with a preoperative diagnosis of intraparenchymal cystic lung lesion misled by radiology. We performed a mini-thoracotomy revealing a thin stalked mobile elongated cyst that arose from the right inferior pulmonary ligament, confirmed as a bronchogenic cyst in histopathology.

1. Introduction

A bronchogenic cyst is a rare congenital malformation due to abnormal bronchial budding that accounts for 13%–15% of congenital cystic lung disease [1,2]. Most of the cysts are usually found in the middle mediastinum [2]. We report a patient with a solitary pedunculated bronchogenic cyst, which is usually rare, emphasizing on proper evaluation as it created confusion with a intraparenchymal lesion.

2. Case

A 12 years old, apparently healthy boy referred to us with the complaints of occasional mild, diffuse, dull aching, non-radiating, right-sided chest pain for a few months having a non-resolving shadow in the chest x-ray without any other associated factors. He has no remarkable past medical history, and there was no relevant family history. General physical and systemic examination revealed no abnormality.

We performed a new chest x-ray that showed a tubular inhomogeneous opacity in the medial part of the right lower lung field (Fig. 1). CECT chest further delineated the lesion and revealed a well-defined, thin-walled cystic lesion about 8.5 × 3.6 × 4.5 cm within the right lower lobe (Fig. 2). Post-contrast scan shows mild marginal enhancement of the lesion. Routine blood workup was within normal limits.

A right mini-thoracotomy was performed through the fifth intercostal space with anticipation toward the right lower lobectomy, considering a parenchymal lesion. However, the opening of the chest cavity revealed a tubular, narrow-based pedunculated, mobile, pinkish, cystic smooth lesion arising from the mediastinum around the area of the inferior pulmonary ligament (Fig. 3). There was no adhesion with surrounding structures, and the lesion was completely mobile. The entire lesion was removed just ligating the stalk. The lesion contained thick mucus.

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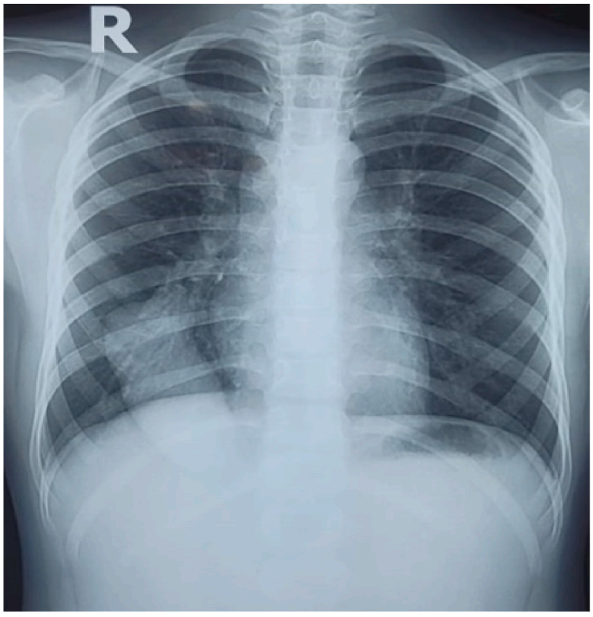

Fig. 1. Chest Xray shows tubular shadow in the right lower lung field away from mediastinum.

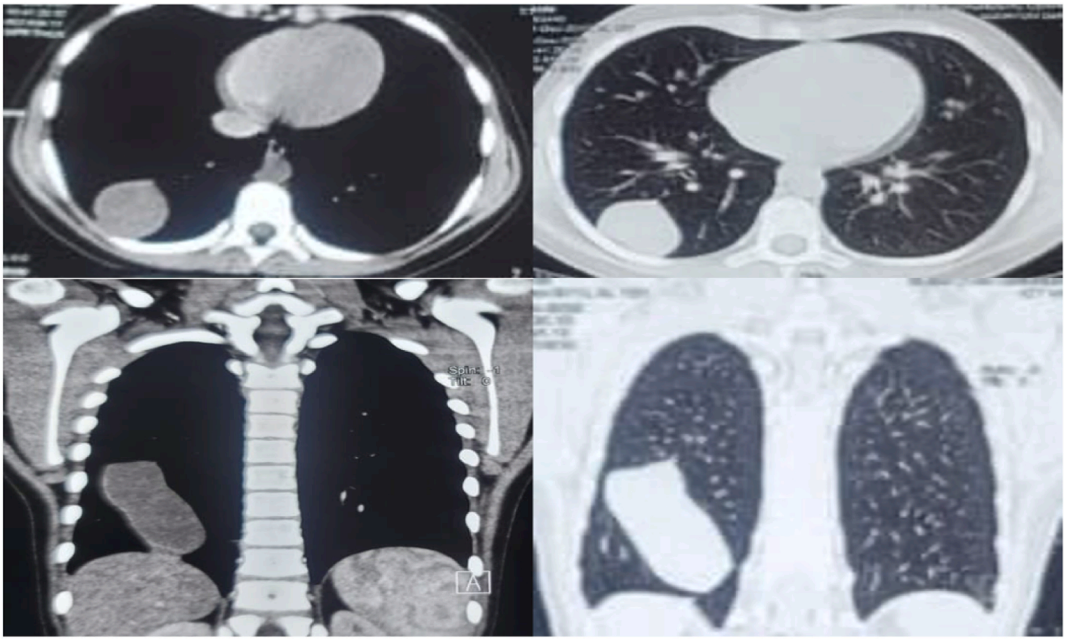

Fig. 2. CECT scan demonstrates tubular cystic lesion within right lower lobe.

Histopathological examination confirmed that the lesion was a bronchogenic cyst, which was lined by respiratory epithelium with a variable amount of seromucinous gland, cartilage and smooth muscle (Fig. 4). The post-operative event was uneventful. The patient was discharged without any complication on the 5th Post-operative day. He has no recurrence after 6 month follow up.

3. Discussion

Bronchogenic cysts are congenital lesions originating from abnormal budding of the primitive ventral foregut [1,2]. Though the exact epidemiology is unknown, it is estimated that it may affect 1/42.000-1/68:000 people [3]. These lesions are usually solitary and lined by either squamous or ciliated columnar epithelium. However, they may be filled with fluid or mucus but do not contain air unless they are instrumented or become infected. Although they can appear in several other places, such as the inferior pulmonary ligament, retroperitoneum, and neck, Bronchogenic cysts primarily take mediastinal and parenchymal forms. Approximately two-thirds

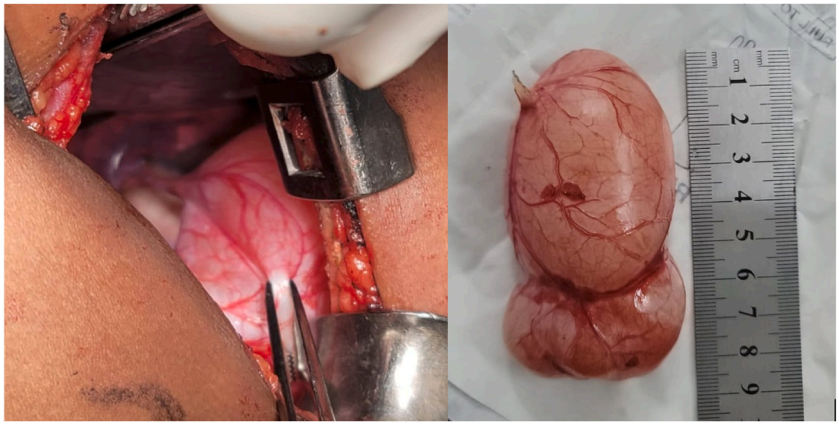

Fig. 3. (A) Operative view of the mass in the right hemithorax, (B) resected specimen of the mass with stalk.

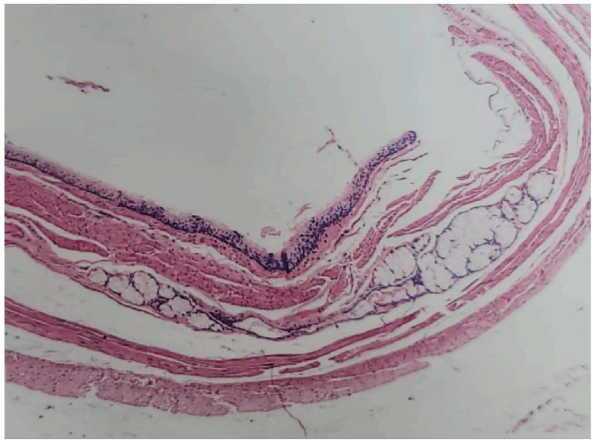

Fig. 4. Microscopic view of the cyst wall.

of cysts are found in the mediastinum [4–6]. The location depends on the embryologic stage of development at which the anomaly occurs. Parenchymal lesions are usually more peripheral [2]. In our case, the cyst was connected with the mediastinum with a thin stalk, and the rest of the cyst was mobile within the right chest cavity, which is a very unusual presentation.

Bronchogenic cysts can progressively enlarge during fetal growth and over time after birth. Many patients with cysts are asymptomatic. Symptoms may develop at any age due to the mass effect and the compression of mediastinal airways, vascular structures, and the esophagus. Symptoms often include coughing, wheezing, dyspnea, respiratory distress, dysphagia, arrhythmia, venacaval compression, infection, hemoptysis, hemothorax and pneumothorax [7–9].

Bronchogenic cysts can be diagnosed as a thoracic mass on prenatal ultrasonography. Plain chest radiography is often diagnostic and reveals around or ovoid, homogeneous soft-tissue mass protruding from or contained within the mediastinum [9,10]. Computed tomography (CT) is an effective imaging tool for diagnosing bronchogenic cysts. The cysts are often thin-walled and show wall enhancement on contrast administration. The cysts can show a water density (0–20 Hounsfield units [HU]) but more commonly display higher density (20–90 HU) attenuation. This variation in attenuation is because the contents of the cysts may be watery fluid, hemorrhagic fluid and thick mucoid fluid [10,11]. Magnetic resonance imaging (MRI) can also be a very effective tool to help differentiate high-attenuation cysts from soft-tissue masses. High-attenuation Bronchogenic cysts are typically iso- or hyperintense to cerebrospinal fluid on T1 and T2 sequences [11]. Sometimes it is challenging to differentiate the cyst from an intraparenchymal lesion, as in our case, which was connected with the mediastinum with a narrow pedicle, and most of the lesion was in the chest cavity.

Bronchogenic cysts should be resected due to the risk of secondary infection, ulceration and malignant transformation [8,12,13]. Temporary maneuvers such as cyst aspiration are not definitive treatments and should be followed by resection. A symptomatic patient needs an early resection, and asymptomatic patients can be evaluated as outpatients and undergo an elective resection [14,15]. A strict follow-up is required in children with a history of recurrent respiratory infection and lung hypoplasia [16].

4. Conclusion

In conclusion, a bronchogenic cyst is a rare congenital malformation of the tracheobronchial tree, whereas a pedunculated variety is rarer. Often it is asymptomatic, and findings are incidental. Symptoms may appear in different age groups. Complete removal of the

cyst is the surgical goal. Detailed assessment of any suspicious bronchogenic cyst is necessary to identify the nature and origin of the lesion keeping in mind the pedunculated variety prior to embarking on any surgical treatment.

Ethics approval and consent to participate

Ethics approval has been taken from the ethical board of National Institute of Diseases of the Chest and Hospital, Dhaka, Bangladesh. Parents of the patient have given consent for the study.

Consent for publication

Consent for publication has been obtained from parent of the patient and copy has been stored.

Availability of data and materials

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

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Authors' contributions

Mosharraf Hossain was the chief of the unit, treating this patient and supervised the report writing. S M Tajdit Rahman (TRT) and Kazi Munzerin Akhter carried out the patient diagnosis. TRT contributed to regular follow-up of the patient, data collection from the in-hospital admission period. Morshedul Islam made the preliminary draft for the case report. TRT gave it a final form. Md. Zahidul Islam helped in writing the draft. All authors were involved in the final reviewing and approving of the final manuscript.

Declaration of competing interest

We do not have any financial and non-financial competing interests.

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List of abbreviations

CXR	Chest X-ray
CECT	Contrast enhanced computed tomography
CT	Computed tomography

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