

**Case
Report**

Intrapulmonary Bronchogenic Cyst in the Thoracic Cavity: A Case Report

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This case report presents an intrapulmonary bronchogenic cyst exhibiting a unique shape. The patient was a 19-year-old man who had been diagnosed with a posterior mediastinal tumor by computed tomography and magnetic resonance imaging, 2 years previously. The imaging revealed that the tumor was located on the left side of the posterior mediastinum and was 45 × 25 mm in size. As the size and shape of the tumor did not change in the 2 years after its detection, surgical extraction was planned. Preoperative diagnosis was, firstly, a neurogenic tumor originating in the posterior mediastinum. Surgical findings revealed that the tumor formed a bridge between the visceral pleura of the left lower lobe and the chest wall, and most of the tumor was located in the thoracic cavity. Pathological diagnosis was intrapulmonary bronchogenic cyst. An intrapulmonary bronchogenic cyst with a unique shape, as observed in this case, is very rare. Although preoperative imaging could predict the tumor size, it could not confirm where the tumor originated. Surgical resection of this type of tumor, which is diagnosed preoperatively as a posterior mediastinal tumor, is a superior strategy for precise diagnosis and treatment.

Keywords: intrapulmonary bronchogenic cyst, mediastinal tumor, video-assisted thoracic surgery

Introduction

A bronchogenic cyst is a common congenital cystic disease. This disease is usually classified into 2 types; mediastinal bronchogenic cysts and intrapulmonary bronchogenic cysts. This disorder often causes symptoms in adults and certain surgical interventions are recommended.¹⁾ Most cases of bronchogenic cysts can be precisely diagnosed preoperatively by using computed tomography (CT) or magnetic resonance imaging (MRI).^{1,2)} In this report, we present a case of an

intrapulmonary bronchogenic cyst with a unique shape, which originated from the peripheral parenchyma, and most of which was located in the thoracic cavity.

Case Report

The patient was a 19-year-old man with an unremarkable medical history. The lesion in question had been already diagnosed as a benign posterior mediastinal tumor during his prior hospital visit 2 years previously. Thoracic CT had revealed a tumor located on the left side of the posterior mediastinum that was 45 × 25 mm in size (**Fig. 1a**), and MRI had revealed an overall high-intensity tumor both on T1- and T2-weighted images (**Figs. 1b, 1c**), which indicated that the tumor involved mucus. The patients wanted to receive treatment for the tumor; therefore, he was referred to our hospital for surgical intervention. Contrast-enhanced CT revealed that no remarkable change had occurred in the size or form of the tumor over the 2 years

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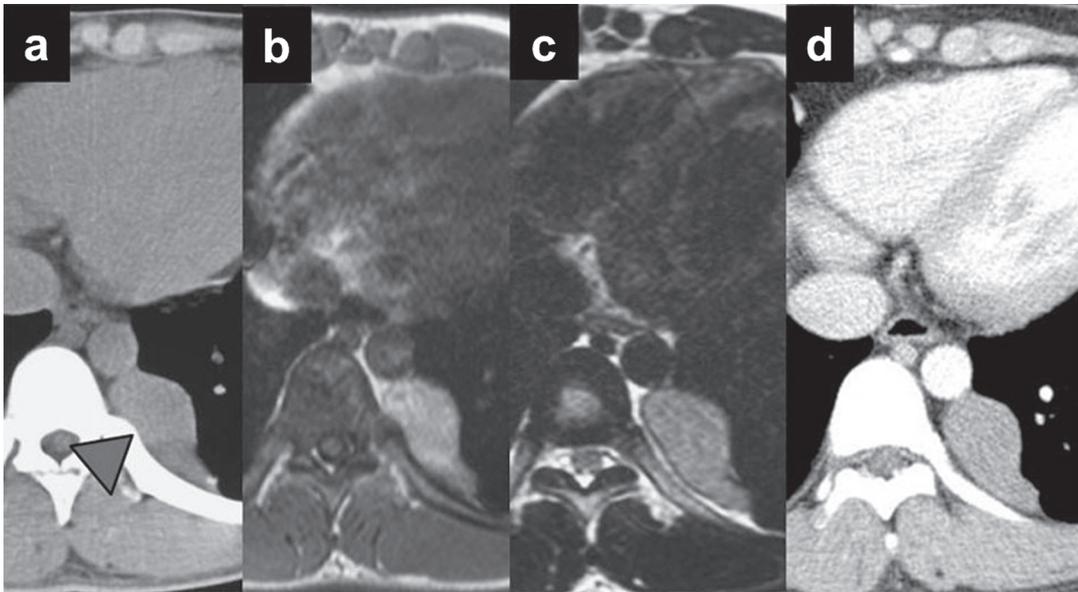


Fig. 1 Preoperative imaging. (a) Simple computed tomography (CT) performed 2 years previously revealed a posterior mediastinal tumor (arrowhead). (b) and (c) are T1- and T2-weighted magnetic resonance imaging (MRI) images, respectively, obtained 2 years previously. MRI images revealed that the tumor involved mucus. (d) Contrast CT performed just before the surgical intervention revealed that the tumor did not change in shape or size.

after its diagnosis (**Fig. 1d**). We made the preoperative diagnosis of neurogenic tumor that originated from the sympathetic trunk or the intercostal nerve. Although mediastinal bronchogenic cyst was also the differential diagnosis because of the cystic finding, the tumor was distant from the bronchus.

The patient underwent video-assisted thoracic surgery. An access port was established at the 9th intercostal space, and 2 thoracic ports were inserted at the 8th and 10th intercostal spaces. The targeted tumor was detected easily; it formed a bridge between the peripheral parenchyma of the left lower lobe and the chest wall, and most of the tumor was located in the thoracic cavity (**Fig. 2a**). The tumor was separated from the pulmonary side by using an endostapler, moreover, the tumor was separated from the chest wall side by using an electric knife. No connections were identified between the tumor and the intercostal nerve or its branches on the chest wall. The tumor was completely resected, and the patient was discharged with no complications. The tumor was a single follicular cyst arising from the pulmonary parenchyma and was filled with gelatinous contents (**Fig. 2b**). Microscopic findings (**Fig. 3**) revealed that the inner wall of the tumor was covered with pseudostratified ciliated epithelium displaying squamous metaplasia. The tumor wall also

included a smooth muscle layer. Thus, the pathological diagnosis was intrapulmonary bronchogenic cyst.

Discussion

Bronchogenic cysts are more often present in the mediastinum, and one-third of these cysts are located in the lung parenchyma.³⁾ The distinguishing features of this case are that this intrapulmonary bronchogenic cyst arose from the peripheral parenchyma and that the major part of the tumor was located in the thoracic cavity and fused to the chest wall. In other words, the tumor formed a bridge between the left lower lobe and the chest wall. Although bronchogenic cysts that are fused to the diaphragm have been reported,^{4,5)} few cases of these cysts with such a unique shape have been reported. Bronchogenic cysts arise from abnormal budding of the tracheobronchial tree. They develop from the ventral primitive foregut between 3 and 7 weeks of gestation in fetal lines. Lung budding proceeds in a centrifugal manner. Mediastinal bronchogenic cysts develop before the fourth week of fetal life, whereas, intrapulmonary bronchogenic cysts develop after the fourth week of fetal life.^{3,6,7)} The bronchogenic cyst in our patient developed in the extreme peripheral lung, in approximately the seventh week of fetal life. It is

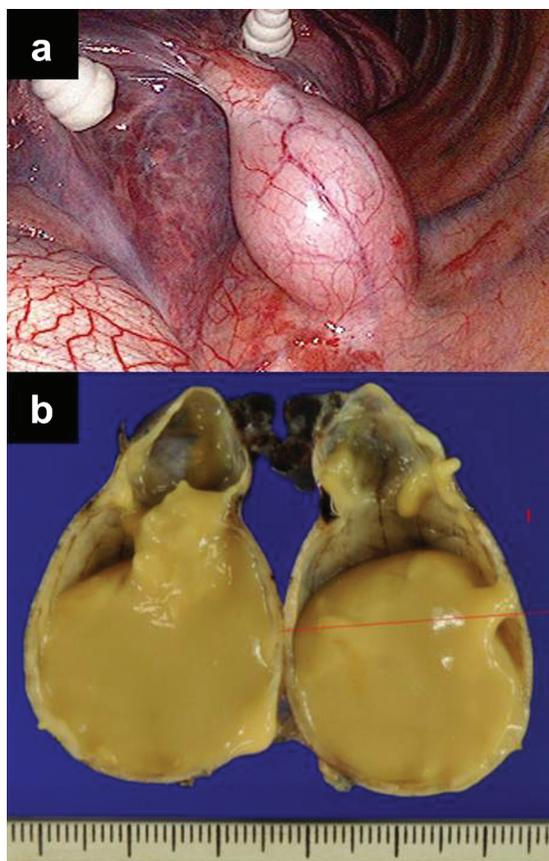


Fig. 2 (a) Surgical findings showed that the major part of the tumor was located in the thoracic cavity and the tumor formed a bridge between the left lower lobe and the chest wall. (b) The sectioned excised tumor, which was filled with gelatinous contents.

believed that the period or pattern of occurrence of the bronchogenic cyst in fetal life in our patient is similar to that of intradiaphragmatic bronchogenic cysts; however, their details are unknown.

Because intrapulmonary bronchogenic cysts usually exist in the pulmonary lobe, the decision to perform lobectomy is often a crucial component of the surgical intervention. Fortunately, the tumor in our case could be resected easily because of its unique shape. Conversely, because of the unique shape of the tumor, our preoperative diagnosis regarding its origin was incorrect. Although some authors have reported that CT and MRI are very useful for the diagnosis of bronchogenic cysts,^{1,2)} it is not easy to preoperatively predict the origin of tumors that surround the vertebrae, chest wall, and visceral pleura.

The tumor in our case did not change in size or form for 2 years. This indicates that the tumor was benign.

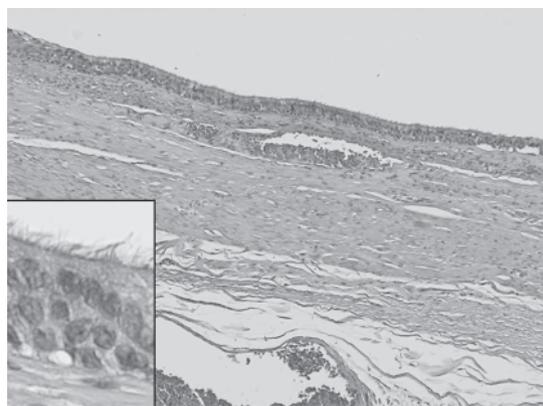


Fig. 3 Histology of the tumor. The inner wall was covered with pseudostratified ciliated epithelium. The tumor wall also included a smooth muscle layer (hematoxylin and eosin stain, $\times 20$ original magnification). The inset shows a higher magnification image of the ciliated epithelial cells.

The preoperative diagnosis was a neurogenic tumor or bronchogenic cyst located in the posterior mediastinum. However, because of the uncertainty of the preoperative diagnosis and the possibility of malignancy, surgical excision was considered as an acceptable strategy with the patient's permission. In our patient, only the surgical procedure revealed the unique shape and location of the bronchogenic cyst.

Conclusion

We presented a case of an intrapulmonary bronchogenic cyst growing into the thoracic cavity. Surgical resection of tumors located around the posterior mediastinum or chest wall is a superior strategy for precise diagnosis and treatment.

Disclosure Statement

The authors do not have a conflict of interest.

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