

Giant bronchogenic cyst causing severe dyspnea: a case report

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ABSTRACT

Background: Bronchogenic cysts (BCs) are cysts formed by abnormal budding of the tracheobronchial tree during embryological development and are less common in adults. They are often located in the mediastinal region or lung parenchyma, but they can rarely be seen extrathoracically. BCs may not be detected radiologically on direct radiography most of the time. However, the diagnosis can be made by thorax Computer Tomography in 97% of lesions. Although they are usually benign, they can lead to severe airway symptoms in patients if they grow large enough to compress the lungs or airway. After this stage, surgical treatment should be planned to prevent recurrences with extraluminal compression and tracheal narrowing that may lead to negative consequences by making the right treatment plan.

Case Presentation: In our case, although we recommended the operation at the age of 18, surgical excision was planned for the patient, who did not accept it and developed dyspnea symptoms during follow-up and increased the size of the BC seen on thoracic CT during follow-up.

In our patient, hybrid thoracotomy was decided to intervene effectively in the close proximity of the lesion to the superior vena cava and possible complications.

Conclusion: Although BCs are usually asymptomatic, surgical excision should be performed because they may cause compression symptoms to the respiratory tract due to an increase in size and recurrence may occur if they are not completely excised. Minimally invasive methods such as video-assisted thoracoscopic surgery and robot-assisted thoracoscopic surgery for surgical resection, or wedge resection and lobectomy with thoracotomy may be considered a good choice for intraparenchymal BCs.

Keywords: Congenital bronchogenic malformation, dyspnea, tracheal deviation.

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Background

Bronchogenic cysts (BCs) are cysts formed by abnormal budding of the tracheobronchial tree during embryological development and are less common in adults. It constitutes 50%-60% of all mediastinal cysts. They are often located in the mediastinal region or lung parenchyma, but they can rarely be seen extra thoracically [1]. BCs are reported more commonly in the right paratracheal area and male patients [2].

Mediastinal BCs are also divided into five types according to their location: paratracheal, carinal, paraoesophageal, hilar, and various. Although they are usually benign, they can lead to severe airway symptoms in patients if they grow large enough to compress the lungs or airway. BCs can also appear as cystic lesions that form an air-liquid level or appear as a homogeneous mass. If it is seen as more dense, it becomes much more common to confuse it with other diseases such as hydatid cysts and tumors [3,4]. There are also studies in the literature showing that

malignant transformation due to BCs is observed, albeit rarely [5]. For these reasons, surgical treatment should be planned to prevent recurrences with extraluminal compression and tracheal narrowing that may lead to negative consequences by making the right treatment plan in the early period [1,6].

We aimed to present our case report for the management of our treatment plan after a patient with a large BC presented with mild shortness of breath and presented with severe shortness of breath after refusing to accept the recommended surgery.

Case Report

An 18-year-old male patient was admitted to the cardiology outpatient clinic on 25.04.2023 with complaints of palpitations, chest pain, and shortness of breath. In the patient, who could not find a cardiological cause to explain his complaints in the examinations, thorax Computer Tomography (CT) was planned for the patient

after opacity compatible with a mass lesion was detected in the right ac upper zone in the direct chest X-ray. A thorax CT scan for further evaluation revealed a paratracheal mass lesion in the right posterior mediastinum. This lesion is present on CT; it was reported as “a homogeneous well-circumscribed mass lesion with soft tissue density, measuring 69 × 47 mm in size at the widest part of the right superior mediastinum” (Figure 1).

The pathology report of the patient, who underwent a transthoracic biopsy in May 2023 for the diagnosis of the mass, was reported as epithelial cells whose nature could not be fully selected and contaminated squamous cells.

Pet CT was planned for further evaluation of the mass that could not be diagnosed by transthoracic biopsy. In the June 2023 Pet CT, it was reported that benign pathologies were considered in the foreground for a 72 × 55 mm hypometabolic mass lesion in the paramediastinal area in the apicomeral part of the upper lobe of the right lung.

The patient, who continued to complain of shortness of breath and palpitations, was recommended an operation by us for both diagnosis and treatment of the existing undiagnosed mass. The patient, who did not want to have surgery and was not followed up for 1 year, applied to us again in May 2024 with the complaint of shortness of breath, and an up-to-date tomography was requested.

May 2024 thoracic CT: 80 × 60 mm mass lesion in the right paratracheal area, slightly enlarged compared to previous imaging (Figure 2).

The patient, whose complaint of shortness of breath became evident and the mass was found to be enlarged in his current examinations, was operated on by us on May 14, 2024, upon his acceptance of the operation. Mediastinal cystic lesion excision was performed with hybrid thoracotomy. Hybrid thoracotomy was decided to ensure safe dissection of areas tightly adhered to the mediastinum, due to the tight adhesion of the lesion to the vena cava superior, to intervene early and effectively in the vena cava superior during endoscopic intervention,

and to ensure safe dissection of the areas tightly adhered to the mediastinum due to the possibility of recurrence when residual tissue is left. Due to the fact that the mass was adherent to the vena cava superior, the cyst contents were aspirated first and then the outer surface was excised (Figures 3 and 4). To prevent recurrence, the part with tight adhesion to the mediastinum was completely excised by blunt dissection, paying attention to the integrity of the vascular structures (Figure 5). The patient was followed up with tube thoracostomy for 3 days postoperatively. He was discharged 1 day after the drain was terminated.

Pathology report of the patient: Macroscopically, a thin-walled uniloculated mucin-filled cystic lesion was characterized as a cystic lesion containing pseudostratified ciliated and goblet cells, and seromucinous glands on the wall, smooth muscle and hyaline cartilage (Figure 6).

(A thin-walled uniloculated mucin-filled cystic lesion was characterized as a cystic lesion containing

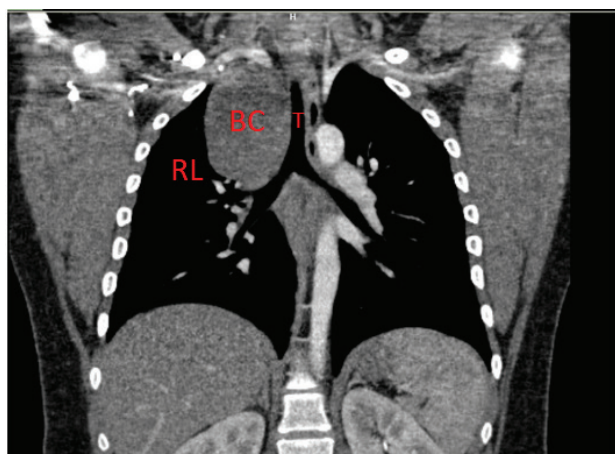


Figure 1. Thoracic CT coronary section image in the preoperative examination of a patient diagnosed with BC (BC: Bronchogenic Cyst T: Trachea RL: Right Lung).

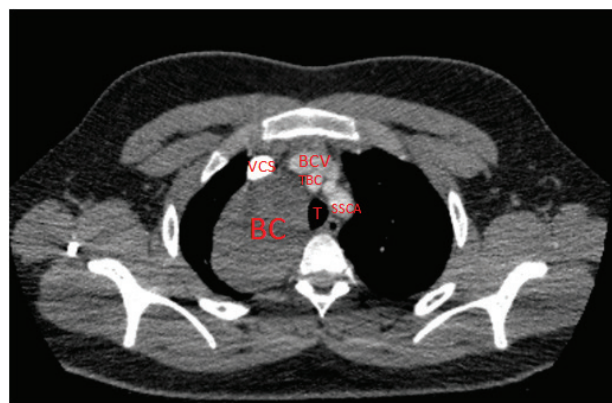


Figure 2. Thoracic CT axial section image in preoperative examination of a patient diagnosed with BC (BC: Bronchogenic Cyst, T: Trachea, VCS: Vena Cava Superior, BCV: Vena Brachiocephalicus, TBC: Truncus Brachiocephalicus, SCA: Arteria Subclavian).

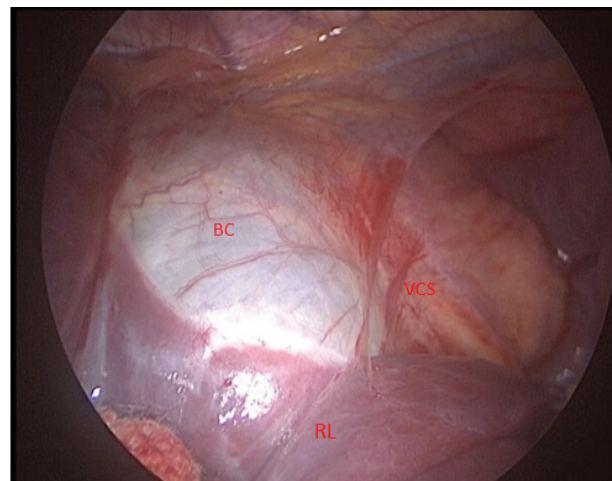


Figure 3. Intraoperative image of our patient who underwent surgery for BC excision (BC: Bronchogenic Cyst, VCS: Vena Cava Superior, RL: Right Lung).

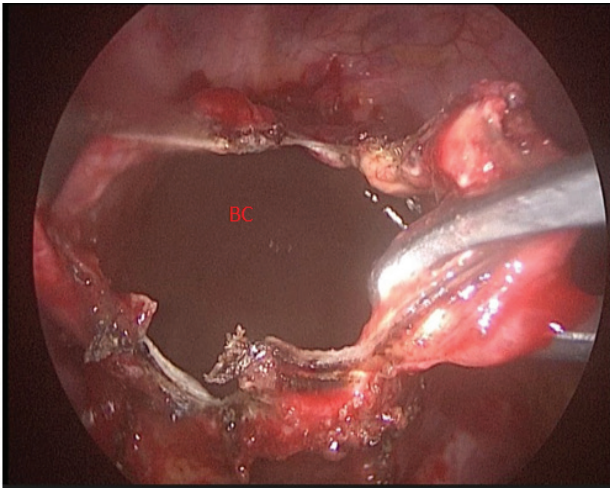


Figure 4. Intraoperative photography of BC contents.

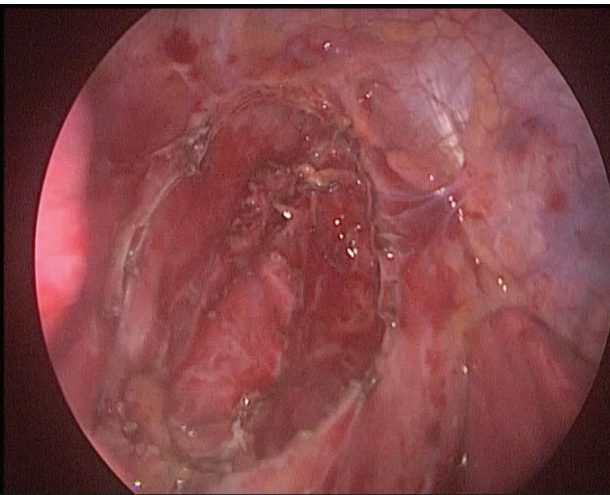


Figure 5. Intraoperative image of the surgical field remaining after excision of the cyst.

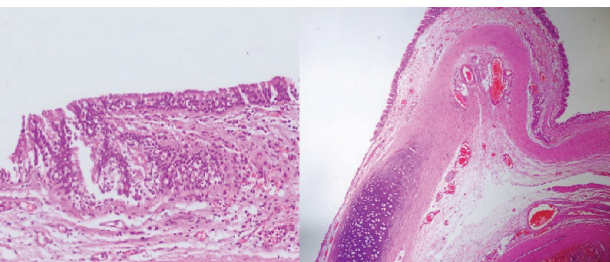


Figure 6. Examination of sections taken from the pathology specimen with H&E staining.

pseudostratified ciliated and goblet cells, and seromucinous glands on the wall, smooth muscle, and hyaline cartilage.)

Discussion

BCs resulting from abnormal ventral budding of the tracheobronchial tree constitute 5%-10% of pediatric mediastinal masses and 15%-20% of mediastinal masses among all age groups [3].

BCs may not be detected radiologically on direct radiography most of the time. The diagnosis can be made by Thorax CT in 97% of lesions. In the preoperative period, thorax CT and MR are also used to have an idea about the structure, localization, neighborhood, and invasion of the cyst with the surrounding tissues [7].

BCs can also appear as cystic lesions that form an air-liquid level or appear as a homogeneous mass. If it is seen as more dense, it becomes much more common to confuse it with other diseases such as hydatid cysts and tumors [3,4]. Although it is diagnosed incidentally due to its asymptomatic course, it may rarely cause hemoptysis, cough, and dyspnea. Symptomatic conditions are more common in younger patients, especially because the thoracic structures are more flexible and the airways are smaller. Cyst structures with respiratory distress. As a result of compression on the aforementioned flexible airways, life-threatening symptomatic conditions can occur with the clinic of cyanosis, stridor, and infection. If they reach large enough to compress the lungs and airways, they can cause severe airway obstruction-related symptoms, so they should be surgically performed in the early period [6].

Close follow-up of the lesions is also recommended in the literature, as compression symptoms (dyspnea, wheezing), hemoptysis, perforation (empyema, mediastinitis), and malignant transformation may develop in diagnosed cases if they are not followed up.

For surgical resection, minimally invasive methods such as video-assisted thoracoscopic surgery (VATS) and robot-assisted thoracoscopic surgery or wedge resection and lobectomy with thoracotomy can be seen as a good choice for intraparenchymal BCs [1,4,6].

In our case, the patient who did not accept the operation even though it was detected in the early period was usually asymptomatic, but there was a need for surgical excision for the mass that grew during the follow-up, causing tracheal deviation, shortness of breath and palpitations, and the cyst was completely excised.

In our case, the treatment was planned by us with VATS for the purpose of a minimally invasive surgical approach, and it was continued as mini thoracotomy due to the fact that the BC was in the form of an excessively inflated, hard mass and its close proximity to the vena cava superior.

Although our case was a cystic lesion, we aimed to present it because it caused severe airway symptoms.

What is new?

BC, which is generally asymptomatic and considered a benign pathology detected incidentally, increased in size to the point of causing compression symptoms after non-follow-up in our case and as a result, the patient was admitted to the hospital with shortness of breath, which is a rare condition in the literature. For this reason, the patient required surgical excision.

List of Abbreviations

BC	Bronchogenic Cyst
CT	Computer tomography
MRI	Magnetic resonance imaging
VATS	Video Associated thoroscopic surgery

Conflicts of interest

The authors declare that they have no conflict of interest regarding the publication of this case report.

Funding

Yes.

Consent for publication

Written consent was obtained from the patient.

Ethical approval

Ethical approval is not required at our institution to publish an anonymous case report.

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Summary of case

1	Patient (gender, age)	Male, 18
2	Final diagnosis	BC
3	Symptoms	Dyspnea
4	Medications	Surgical treatment was applied, there was no need for additional medical treatment.
5	Clinical procedure	The patient who underwent surgical resection did not need additional medical treatment. Follow-up continues without any problems
6	Specialty	Pulmonary medicine