

## CASE REPORT

# **Bronchogenic cyst, complicated by mediastinitis and contralateral empyema\***

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Despite its less-than-ominous appearance as a mediastinal opaque mass with precise contours, a bronchogenic cyst has significant potential for creating complications. We report a case of severe complications in a 28-year-old male presenting with epigastric pain irradiating to the back. A chest radiograph revealed a well-delimited mass on the posterior-inferior right side of the mediastinum. On the fifth day after onset of the symptoms, the patient developed sepsis resulting from mediastinitis and left pleural effusion consistent with empyema. The patient was submitted to a left thoracotomy for lung decortication and mediastinal drainage and, after a one-week interval, to a right thoracotomy for resection of the infected mediastinal cyst. In view of the inherent risk of complications, mediastinal cyst resection is strongly recommended, even in cases of asymptomatic presentation.

**Key words:** Bronchogenic cyst. Mediastinitis/complications. Empyema, pleural/complications.

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## **INTRODUCTION**

Bronchogenic cysts are the most common congenital cystic lesions found in the mediastinum. Due to their precise contours and liquid contents, radiological images of mediastinal cysts frequently give the impression that they are benign in nature. However, bronchogenic cysts can potentially become symptomatic, cause complications, and even become malignant.<sup>(1)</sup> In a study involving 86 patients submitted to surgery for bronchogenic cysts, 82% presented with specific symptoms or complications.<sup>(2)</sup> The types of symptoms and complications depend upon its location and upon the structures with which the cyst is connected. Since bronchogenic cysts are closely related to the tracheobronchial tree and esophagus, respiratory and esophageal symptoms and complications are more common. However, they may have an atypical presentation when they occur at sites where they are rarely seen, such as the heart<sup>(3)</sup> or diaphragm.<sup>(4)</sup> In light of the severity of the complications that developed in the case presented herein, the following is a cautionary tale regarding the management of mediastinal cysts in general.

## **CASE REPORT**

A 28-year-old male university professor presented to the emergency room at the Hospital Universitário de Brasília with sudden-onset epigastric pain irradiating to the back. The patient characterized the pain as a feeling of tightness and reported an association with eructation. He denied having ingested alcohol or having vomited. He was evaluated on the same day that the symptoms began, and his overall condition was good. Physical examination showed no abnormalities, and his electrocardiogram and laboratory tests were normal. Chest radiography (Figure 1) revealed a well-delineated paramediastinal opaque mass, juxtadiaphragmatic and on the right side.. The patient was medicated and released. He was prescribed an analgesic and a proton pump inhibitor. The patient

continued to have pain and returned to the emergency room two days later. He was re-evaluated, and additional chest radiographs were taken. He was considered clinically stable and the radiography revealed no new findings. The patient was again released, and it was suggested that he schedule a consultation at an outpatient clinic. On the following day, his epigastric and posterior thoracic pain increased, and he returned to the emergency room. After being submitted to chest radiography for the third time, the patient was admitted for investigation and treatment.

On the evening after the day of hospitalization, with esophagoscopy and computed assisted tomography of the chest scheduled for the following day, the patient presented with profuse sweating and severe chest pain, which was irradiating to the left and intensified upon the drawing of a breath. On the following day, after the scheduled tests had been performed, the thoracic surgery team was called to evaluate the case, which was by then complicated with left pleural effusion. The evaluation of the sequence of radiographs revealed progressive enlargement of the paraesophageal cystic lesion on the lower right side. The posterior inferior radiograph taken on the day of admission revealed enlargement of the retrocardiac density, obscuring the distal borders of the thoracic aorta and the left medial border of the diaphragm (Figure 2). When the images were overlaid, it could be seen that the camber to the left of the posterior-inferior mediastinum resulted in the enlargement of the density throughout the majority of the cyst. This was confirmed in the radiograph taken in profile (Figure 3). Chest tomography revealed a thick-walled cystic lesion, with a fluid-fluid level, enlargement of the inferior mediastinum, and pleural effusion with early loculations. Esophagoscopy was normal. The analysis of the pleural liquid revealed 10,000 leukocytes/mL, 80% of which were polymorphonuclear, a protein level of 4.6 g/dL, glucose at 25 mg/dL and a pH of 6.7. An increase in the leukocyte count was observed (from 7,300/mL to 14,300/mL), and 10% were rods.

On that same day, the patient developed sepsis, and clindamycin and ceftriaxone were started. The patient was submitted to a thoracotomy for the exploration of the left hemithorax. Due to a large amount of fibrin and the discovery of suppuration in the mediastinum, the procedure was converted to a left lateral thoracotomy of the lower lobe for early decortication and mediastinal drainage. Three drains were introduced into the pleural space: one in a high lateral position, one in a low paramediastinal position and one between the esophagus and the aorta, directed toward the contralateral cyst. The poor overall condition of the patient, who presented unstable hemodynamics during the procedure, compelled the surgeons not only to alter the course of the surgical procedure, but also, due to anesthesiological considerations, to postpone the removal of the cyst. Following surgery, laboratory testing and clinical examination of the patient revealed rapid resolution of the sepsis. On postoperative day 7, the patient was submitted to right lateral thoracotomy of the lower lobe for removal of the mediastinal cyst. The cyst was inflamed, with firm pleuropulmonary adhesions and purulent contents. There was no indication of any communication with the esophagus or the mediastinum. The patient was released one week later, in excellent condition. The final diagnosis was primary infection of a bronchogenic cyst, complicated by mediastinitis and contralateral empyema.

## DISCUSSION

Bronchogenic cysts result from the abnormal ventral budding of the tracheobronchial tree during embryonic development. They may be located in any part of the mediastinum, but most are found near the tracheal carina and in the medial or posterior mediastinum. Conventional radiographs reveal these cysts as masses with precise contours and homogeneous opacity just below the carina, and they are more easily observed near the right pulmonary hilum. On CT scans, bronchogenic cysts appear as elliptical or spherical masses with imperceptible borders and uniform attenuation coefficients. The coefficient value depends on the cyst contents, which may range from the value for water to that for soft tissues.<sup>(5)</sup> Bronchogenic cysts are usually asymptomatic. However, symptoms resulting from the compression of adjacent structures are quite common in children. In adult surgical candidates, the most common symptoms related to bronchogenic cysts are pain and cough, whereas pain and dysphagia are more common in cases of esophageal cyst.

The significance of the present case lies not as much in the lack of similar reports in the literature as in the warning call represented by the severity of the complications resulting from the cyst. Although

the pain reported by the patient was consistent with the general manifestation in symptomatic cases, the patient was hospitalized because of his insistence, complaining about the persistence of the pain, and not because of any suspicion of complications on the part of the physicians involved. On the day of admission, development of the cyst infection and mediastinitis was evidenced by the enlargement in cyst volume and the widening of the inferior-posterior mediastinum revealed in the Radiographs. However, the patient was not submitted to chest tomography until 36 hours later, despite the fact that, prior to this procedure, the infection invaded the left pleural cavity and his condition worsened. The presence of severe sepsis, a pH of 6.7, and empyema within the first 12 hours after admission demonstrated that the mediastinal cyst was transmitting a bacterial load that could not be ignored. The firm adhesions (seen during the right thoracotomy) between the cyst and the neighboring structures suggested recurrent infection of the cyst and subsequent contamination of the mediastinum and the left pleural cavity.

Due to its location in the lower posterior mediastinum and its close relation to the esophagus, one would have expected the embryonic origin of this cyst to be esophageal, with a double layer of smooth muscle cells and possible communication with the esophagus. However, the anatomopathological study of the cyst revealed that it was coated with pseudostratified ciliated epithelium, had extensive ulcerous areas, and that the walls were composed of smooth muscle fibers with various cartilage foci. This led us to classify it as a bronchogenic cyst. No esophageal communication was found during the surgical procedure, which could explain the recurring infection. This finding would be rare, but not unheard of, in cases of this type of cyst.<sup>(7)</sup> The case reviewed here is of consequence because it demonstrates that bronchogenic cysts may be atypical in location and presentation, and it reinforces the need for surgical intervention, even when there are no symptoms.

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*Figure 1 – Chest radiograph demonstrating inferior right-side paramediastinal opaque mass*

*Figure 2 – Chest radiograph demonstrating volume enlargement of the right-side opaque mass and widening of the inferior mediastinum (retrocardiac)*

*Figure 3 – Chest radiograph in profile demonstrating overlay of the right-side opaque mass and mediastinal widening*