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Case report

# Symptomatic mediastinal mass in a 32-year-old male

A 32-year-old male, known to have atopic eczema, presented with a 3-day history of acute pleuritic chest pain and shortness of breath. No traumatic event had occurred. The pain was described as sharp and posture dependent, and was worse when leaning backwards. The patient had no palpitations, oedema or radiated pain. Dyspnoea was present both at rest and during exercise, without the presence of wheezing, haemoptysis or purulent sputum. He had no history of fever, cold shivers, weight loss or perspiration. His nutritional state was normal and he had no symptoms of nausea, vomiting or diarrhoea. For 3 days he had experienced dysphagia during meals. The patient was working full-time as a national courier and did not visit foreign countries for his work and did not travel outside Europe. The patient owned a dog but he had no specific contact with (farm) animals. He never smoked and never consumed alcohol. His physical condition, before the start of the present symptoms, was excellent due to daily exercises. On clinical examination our patient was conscious and orientated. He had a normal temperature of 36.9 ºC, a respiratory rate of 16 breaths·min$^{-1}$, blood pressure of 115/75 mmHg, heart rate of 68 beats·min$^{-1}$ and an oxygen saturation of 95% on room air. Both percussion and respiratory sounds were reduced over the right hemithorax, without crackles or rhonchi. He had no pitting oedema around the ankles. A chest radiograph was performed (figure 1).

**Task 1**
Describe the chest radiograph taken on admission (figure 1).

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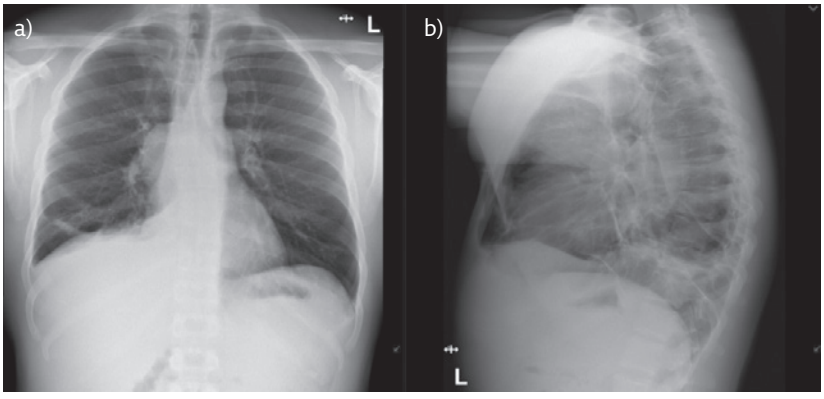


**Figure 1** *Chest radiograph showing a) a posterior–anterior and b) a lateral view.*

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**Most bronchogenic cysts are found incidentally and clinicians should be aware of an atypical case presentation. Total surgical resection is the treatment of choice of a bronchogenic cyst, especially in symptomatic patients.** https://bit.ly/3uQrFXo

**Answer 1**
On the posterior-anterior view the chest radiograph shows a rounded mass at the level of the right hilum, projecting in the middle mediastinum on the lateral view. Furthermore, there is a pleural effusion of the right hemithorax.

His blood test showed the following: haemoglobin 7.5 mmol·L$^{-1}$, white cell count 8.7×10$^{9}$ cells·L$^{-1}$, platelets 386×10$^{9}$ cells·L$^{-1}$, C-reactive protein 74 mg·L$^{-1}$ and D-dimers 2155 µg·L$^{-1}$. Kidney and liver function values and arterial blood gas test were normal.

A contrast enhanced computer tomography (CT) scan was performed (figure 2).

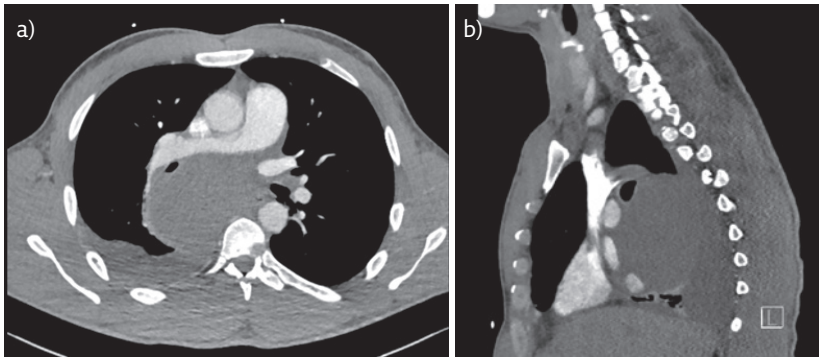


**Figure 2** *Computed tomography of the chest at mediastinal window. a) Axial view and b) sagittal view.*

**Task 2**
Describe the CT scan of the thorax taken on admission (figure 2).

**Answer 2**
The CT of the thorax shows an ovoid-shaped lesion in the middle inferior mediastinum with small inclusions of gas. In accordance with the chest radiographs, pleural effusion is seen in the right hemithorax.

A homogenous mass of 7.0×8.0×10.0 cm with a density of 30 HU in the middle inferior mediastinum was found, with extension to the posterior mediastinal component. Pleural fluid and post-obstruction consolidations of the right hemithorax were present. Pulmonary embolisms were excluded. Diagnostic puncture of the pleural effusion revealed an exudate. Pathological analysis of the pleural effusion showed the presence of inflammation without bacterial involvement and no malignant cells.

**Task 3**
What is the differential diagnosis of this patient?

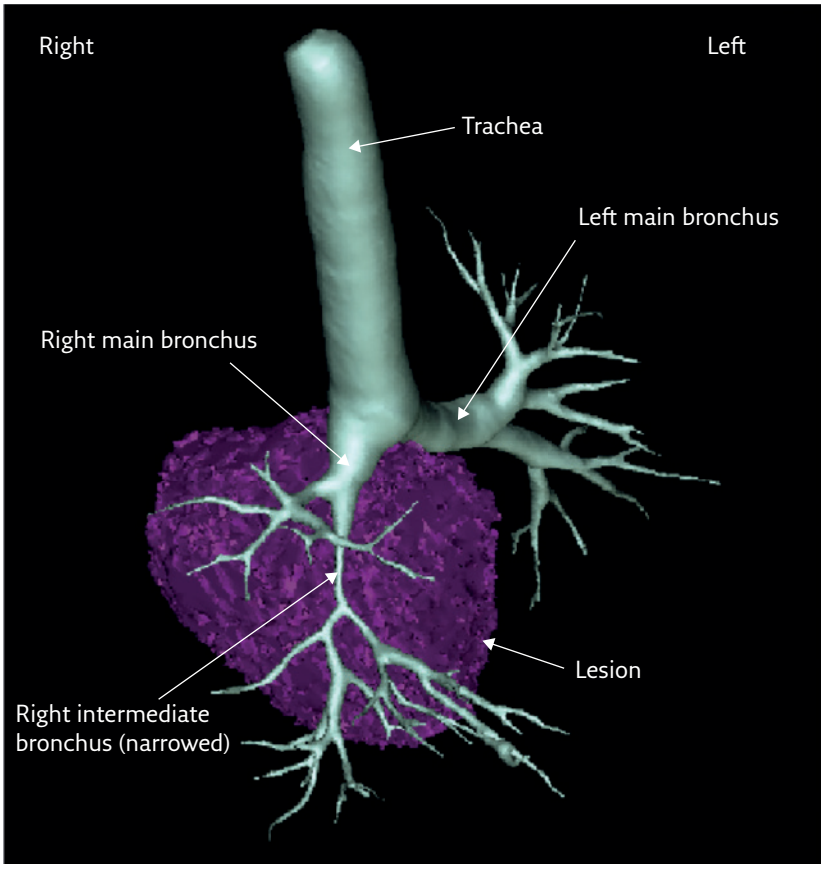


**Figure 3** *3-dimensional reconstruction (computed tomography) of the lesion, with a narrowed right intermediate bronchus as a result of compression by the lesion.*

**Answer 3**
The tumour is located in the middle inferior mediastinum in a young adult male, therefore the differential diagnosis is oesophageal duplication cyst, bronchogenic cyst, lymphangioma (of the thoracic duct), pericardial cyst, abscess, cystic teratoma or a lymphoma. Based on the location of the mass, thymoma, thymic carcinoma, teratoma, thymic cyst, germ cell tumour or schwannoma are less likely. A neoplasm in a young nonsmoker is not excluded but unusual.

Although, conventional radiological imaging was not conclusive, compression of the right intermediate bronchus was seen on a 3-dimensional reconstruction of the CT scan (figure 3). The compression of the intermediate bronchus in combination with elevated C-reactive protein was suspicious for a post-obstructive pneumonia. The patient was empirically started on amoxicillin/clavulanate and the chest pain was relieved by a combination of paracetamol, ibuprofen and oxycodone.

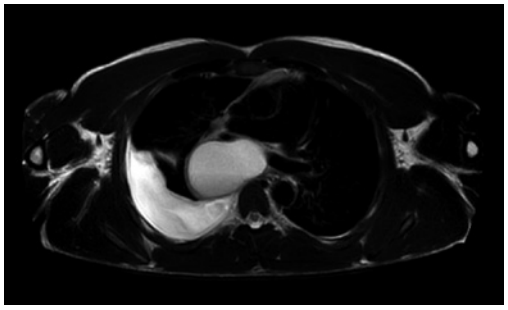

**Figure 4** *Magnetic resonance imaging scan of the chest (axial view, T2-weighted imaging).*

Although the lesion was suspected to be cystic, the presence of pleural fluid is usually not seen with cystic lesions. To rule out a solid mass a magnetic resonance imaging scan was performed (figure 4). The axial T2-weighed image at the level of the pulmonary trunk showed pleural fluid on the right side of the hemithorax and a well-demarcated mediastinal lesion with high T2 signal, which indicated a cystic nature. The T2 signal content of the cystic lesion was lower than that of the pleural fluid, which can be due to protein rich fluid. Furthermore, the lesion showed some shading and contained debris. These findings were suspicious for a cystic nature. No pericardial fluid was found. The size of the mediastinal lymph nodes was normal.

Echocardiography showed compression of the mass on the left atrium, normal left ventricular function (ejection fraction 62%), increased right ventricular pressure (30 mmHg) and no pericardial fluid. A new incomplete right bundle branch block and negative T-segments in the anterior–lateral leads were present on the 12-lead ECG. Cardiac biomarkers were normal. Spirometry was performed after adequate pain management and was suspected for a restrictive defect: forced expiratory volume in 1 s (FEV1) 64%; FEV1/forced vital capacity (FVC) 80% (lower limit of normal 71%); FVC 65%; and transfer coefficient of the lung for carbon monoxide 98%.

**Task 4**
What intervention is preferred in this patient?

**Answer 4**
Complete surgical resection of the cystic lesion.

In this patient, a possible explanation for the pleural effusion in combination with elevated C-reactive protein serum level could be a rupture of the cyst, although the shape of the cyst was not strongly suggestive for rupture on radiological imaging. An alternative cause might be pulmonary congestion due to local compression of the right inferior pulmonary vein and left atrium. Complete resection of the cyst was the treatment of choice. Although a diagnostic biopsy was performed in several case reports, biopsy is not preferred when an infected cyst is suspected. Potentially, the biopsy needle could trigger a rupture or mediastinitis [1].

Within 2 weeks after the onset of the symptoms, the lesion was resected under general anaesthesia. First, the patient was placed in a left lateral decubitus position. Single lung ventilation was performed. A 10 mm trocar was introduced in the fifth intercostal space for inspection by video-assisted thoracoscopic surgery. Visualisation of the cystic lesion was sub-optimal so the incision was extended to a muscle-sparing thoracotomy without the use of a rib spreader. 500 mL of clear yellow pleural fluid was evacuated. The parietal pleura was hyper vascularised making it difficult to distinguish the mass from the lung parenchyma. Because the wall of the lesion was fragile, the fluid inside was evacuated before further resection was continued. Tissue of the mass was meticulously peeled off the lung and oesophagus, which was later covered with parietal pleura. There was no invasion in the pericardium or lung. The mass was totally removed (figure 5). After resection, the pleural space was rinsed with 6 L povidone-iodine and sodium-chloride 0.9%. Total surgical time was 180 min. The patient lost 400 mL of blood during surgery as a result of oozing. A 20-French pleural drain was inserted and was removed the day after surgery.

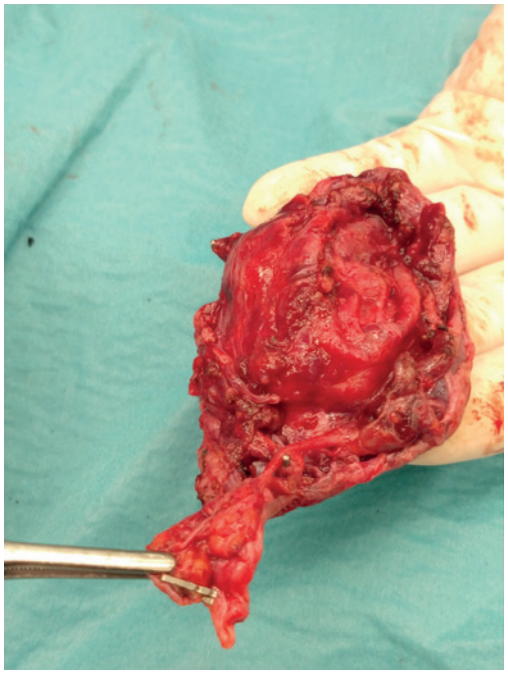

**Figure 5** *A cystic lesion of 5 cm was surgically removed by video-assisted thoracic surgery.*

A cyst with a diameter of 5 cm was resected and sent for pathological analysis. The wall of the cyst contained fibrotic connective tissue with respiratory epithelium, characterised by typical ciliated columnar epithelium (figure 6). The cystic fluid and pleural fluid, which were evacuated during surgery, had signs of chronic inflammation without the presence of malignant cells. Microbiological analysis was performed and showed no involvement of bacteria, fungi or yeast.

**Task 5**
What is the most likely diagnosis?

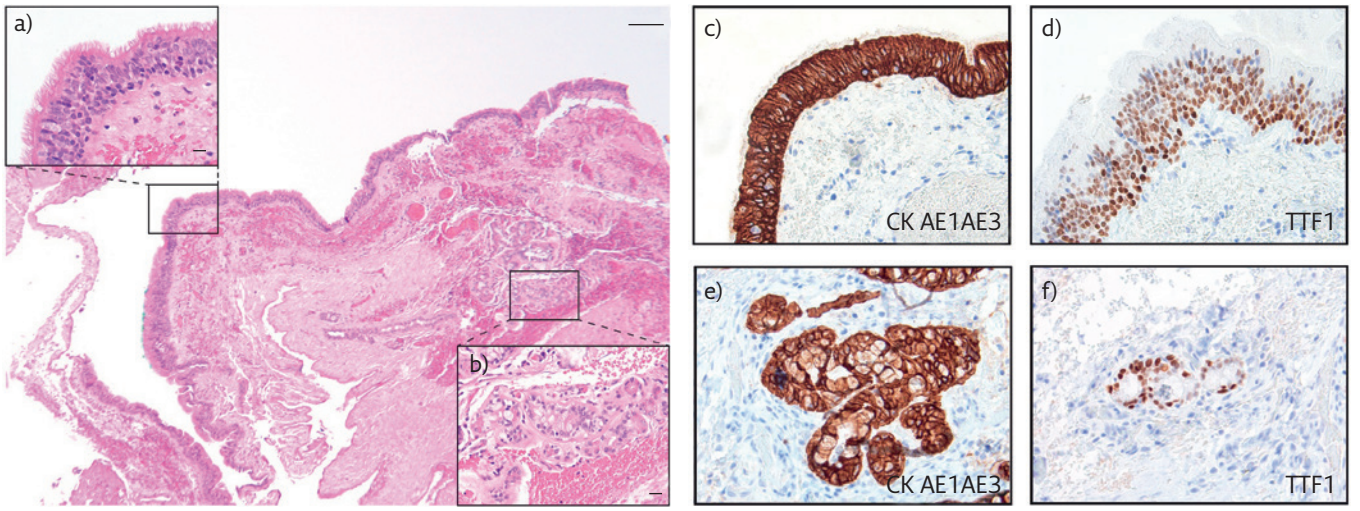


**Figure 6** *Histology of the resected cyst. Cyst lined by respiratory type epithelium (haematoxylin and eosin staining, scale bar = 200 µm). a) The epithelium is characterised by pseudostratified columnar and ciliated cells (haematoxylin and eosin staining, scale bar = 50 µm). b) Seromucous glands are located in the underlying fibrotic cyst wall (haematoxylin and eosin staining, scale bar = 50 µm). c) Positive cytoplasmic staining for an epithelial marker (CK AE1/AE3-immunostaining 40x magnification) and d) positive nuclear staining for a pulmonary/bronchial marker (TTF1 immunostaining, 40x magnification). e) Positive cytoplasmic staining for an epithelial marker (CK AE1/AE3-immunostaining, 40x magnification) and f) positive nuclear staining for a pulmonary/bronchial marker (TTF1 immunostaining, 40x magnification).*

**Answer 5**
A bronchogenic cyst was the final diagnosis.

The patient recovered well and was discharged from the hospital 3 days after surgery. 2 weeks after discharge, the patient was seen at the outpatient clinic for follow-up and he had made a full recovery. He no longer suffered from chest pain or shortness of breath and used paracetamol sporadically after discharge.

## Discussion

This case report describes a symptomatic adult male with a bronchogenic cyst in the middle inferior mediastinum with compression on the left atrium, oesophagus and right intermediate bronchus. The chest pain associated with the pleural fluid can be explained as pleuritic pain due to pleural inflammation. Bronchogenic cysts are rare congenital malformations, often found by coincidence on radiological imaging. A bronchogenic cyst is a duplication of the primitive pulmonary primordium and is formed from the foregut during the early stage of gestation [2]. Although the most common location of a bronchogenic cyst is the mediastinum, it can develop in an ectopic location along the pathway of the foregut [3]. Up to 85% of bronchogenic cysts are located in the mediastinum and 12% in the parenchyma of the lung, while locations like the pericardium, neck and abdomen have been reported as well [4]. Regardless of the location, a bronchogenic cyst is lined by a bronchus-type epithelium. The prevalence of bronchogenic cysts ranges from 1 per 42000 to 1 per 68000 according to two hospital series [5]. Bronchogenic cysts are more frequently diagnosed in men and can remain undiscovered for decades. Though rare, a bronchogenic cyst is the most common primary cyst of the mediastinum accounting for 50–60% of all mediastinal cysts and 10–15% of all mediastinal tumours [6]. According to Tiwari *et al*. [7], the most frequent localisation of a mediastinal bronchogenic cyst was the middle mediastinum (79%), followed by the posterior mediastinum (17%) and very infrequently by the anterior mediastinum (4%). In children, bronchogenic cysts are mainly discovered due to symptoms or in this case recurrent pulmonary infections [6].

Although many bronchogenic cysts are found incidentally, they can lead to symptoms like chest pain, dyspnoea, fever, cough and haemoptysis. In rare cases, oesophageal compression can lead to dysphagia. Post-obstructive pneumonia is not uncommon in adults and children [8]. Pulmonary located bronchogenic cysts are more likely to be symptomatic than mediastinal bronchogenic cysts and 86.4% of symptomatic patients have a complicated cyst [9]. The case we present is rare because the patient had symptoms suspicious of pericarditis or pulmonary embolism, although the dysphagia and pleural fluid were atypical features in this regard, and red flags in a young adult.

Surgical resection of a bronchogenic cyst is indicated for three reasons: 1) to confirm the diagnosis; 2) to prevent the development or progression of symptoms and complications;

and 3) to avoid potential malignant transformation [10, 11]. Malignant transformation of a bronchogenic cyst is very rare and only a few well-documented cases have been reported. These cases reported histological findings of transformation of a bronchogenic cyst to an enteric adenocarcinoma, bronchioalveolar carcinoma, adenocarcinoma, large cell carcinoma and squamous cell [12]. Even a case of a carcinoid tumour nested inside a bronchogenic cyst has been described [13].

Although a malignant transformation is rare, complete resection of a symptomatic mediastinal cyst is recommended to relieve symptoms and to confirm an adequate diagnosis [10–14].

## Conclusion

A bronchogenic cyst is a rare malformation and develops early in life. Most bronchogenic cysts are found in children, while adult cases are less frequently seen. In our case, the atypical presentation of dysphagia in combination with shortness of breath, acute chest pain and pleural fluid in a relatively young adult patient triggered the clinicians to perform additional clinical and radiological evaluation. Total surgical resection is the treatment of choice for a bronchogenic cyst, especially in symptomatic patients, to confirm the diagnosis and release symptoms.


### Affiliations

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### Consent

The patient signed informed consent for this case report.

### Conflict of interest

None declared.

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