**CASE STUDY**

**Bronchogenic cyst presenting as mediastinal mass with pleural effusion**

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Bronchogenic cysts are congenital abnormalities resulting from embryological budding of the bronchial tree; they often arise near the carina [1–3]. Mediastinal bronchogenic cysts are frequently detected incidentally, on routine chest radiographs. They manifest as smooth and well-circumscribed masses near the carina [2, 3].

We describe two cases of bronchogenic cysts that appeared as mediastinal mass with pleural effusion, without pulmonary infection. To our knowledge, this radiological manifestation, in association with bronchogenic cysts, has not previously been reported.

**Case reports**

**Case 1**

A 49 year old man presented with a one month history of dysphagia. Chest radiography revealed right mediastinal mass and ipsilateral pleural effusion. Oesophagoscopy revealed a marked extrinsic compression of the oesophagus. Fibreoptic bronchoscopy showed an infiltration of the mucosa of the right intermediate bronchus. Biopsies were negative.

Chest computed tomography (CT) scans showed a mediastinal tumoral mass with tissular density (35 Hounsfield units (HU)) and a right pleural effusion (fig. 1a and b). CT guided needle biopsies were negative. The diagnosis of necrotic mediastinal malignancy with pleural effusion was suspected.
The patient underwent mass resection by right thoracotomy. The pleural effusion was evacuated (400 mL of serosanguinous fluid), and the mediastinal mass was excised. The mass was adherent to the posterior pericardium, the right inferior pulmonary vein, and the intermediate bronchi. The oesophagus was not distinguishable from this mass. Greyish white noninfected fluid was evacuated from the cyst.

A pathological study confirmed multilocular bronchogenic cyst without neoplasm cells.

After surgery the patient progressed well and received a normal diet; he did not complain of difficulty in swallowing.

**Case 2**

A 58 year old woman presented with a persistent dry cough for 10 days, sometimes associated with shortness of breath. Clinical examination was normal. A chest radiograph showed a right mediastinal mass.

Fibreoptic bronchoscopy examination confirmed extrinsic narrowing of the right intermediate bronchus with calibre reduced by 50%.

On unenhanced chest CT scan (fig. 2a), the mass appeared oval and had a homogeneous high density, evaluated at 65 HU. It was located in the middle mediastinal compartment, in the subcarinal area; and symmetrically compressed the right intermediate bronchus. The posterior cyst wall was calcified. A small right pleural effusion was observed. After injection of contrast media (fig. 2b), the mass itself was not enhanced. Enhancement of its right edge suggested thickening of the inflammatory mediastinal pleura or a lung collapse around the mass.

Magnetic resonance imaging (MRI) demonstrated a homogeneous high signal intensity on both T1-weighted and T2-weighted sequences (fig. 2c and d). The pleural effusion had a low signal intensity in T1-weighted imaging and a bright signal intensity in T2-weighted imaging (Fig. 2c). The mass symmetrically compressed the right pulmonary artery.

The patient underwent mass resection by right thoracotomy. Sero-fibrinous pleural effusion (800 mL) was evacuated. Aspiration of the cyst content yielded a greyish white fluid. Bacteriological studies were negative. During dissection, the intermediate bronchus was injured, creating a small defect in the membranous wall. This defect was successfully closed without further complications. An elliptical piece of pericardium was removed because of adherence. The entire azygos arch and the right branch of vagus nerve were sacrificed.

**Fig. 2.** – Case 2. a) Computed tomography (CT) scan without contrast media, mediastinal windows, shows an oval mass with homogeneous high density (65 UH), peripheral calcification, and a right small pleural effusion. b) CT scan with contrast media, mediastinal windows, shows a ring enhancement which is probably related to mediastinal pleura inflammation. c) Sagittal spin echo T1-weighted image shows the mediastinal mass with a high signal intensity and its anatomical relations. d) Axial spin echo T2-weighted image also shows a high signal intensity of the mass (like cerebrospinal fluid) and a pleural effusion.
Pathological examination supported the diagnosis of bronchogenic cyst. There was found to be an inflammatory infiltration of the external cyst wall.

Three months later, the chest roentgenogram was seen to be normal.

**Discussion**

Bronchogenic cysts are rare cystic malformations, with walls similar to those of normal bronchi and with respiratory epithelial lining [2].

Radiographic and CT scan characteristics of bronchogenic cysts are well-defined [2–5]. On chest radiographs they are round, well-circumscribed, unilocular or multilocular masses. CT density may vary from typical water density (0–20 HU) to high density (89–99 HU), as illustrated in our cases [6]. MRI appearance is of a mass of low (grey to black) or high (white to gray) signal intensity in T1-weighted imaging, and bright (white) signal intensity in T2-weighted imaging [7, 8]. In T1 weighted images, fat, proteinaceous and haemorrhagic fluids appear in white; and water appears with a low signal intensity. In T2-weighted images water, with or without proteins, appears with a high signal intensity. In our case (Case 2), the cyst appeared with a high signal intensity in T1- and T2-weighted images; these signal abnormalities are related to an inflammatory fluid or a high protein content. CT and MRI studies are of value to clarify the relationship between the mass and the adjacent mediastinal structures before thoracotomy.

To our knowledge, pleural effusion in association with bronchogenic cyst has not previously been described. In our first case (Case 1, 1985) pleural effusion was admitted as a radiological sign against the diagnosis of bronchogenic cyst. Mediastinal mass with pleural effusion was an argument for malignancy, such as lymphoma or lung cancer. The interpretation was not the same in the second case (Case 2, 1993).

Serofibrinous or serosanguinous fluid was evacuated at surgery. These pleural effusions were probably related to an inflammatory reaction of the pleura to the bronchogenic cyst. Such inflammation could be due to the large size of the masses, perhaps with the onset of rupture. In our two cases, bronchogenic cyst excision was difficult because of dense pericystic adhesions. This inflammatory reaction was observed by means of CT scans using contrast media, as an enhancement in the cyst edge.

Percutaneous needle aspiration can be used for diagnostic purposes, or even therapeutically, if thin fluid is present within the cyst [9]. This technique for diagnosis was used in the first case and was not helpful.

Because of dense pericystic adhesions to adjacent organs, surgical excision of mediastinal bronchogenic cysts can be hazardous. A watching and waiting attitude seems inadvisable in a patient with a mediastinal cyst, even if the cyst is not symptomatic. This approach could lead to the development of complications, that tend to make operation more difficult. Other arguments for a more aggressive treatment, with total excision of these lesions, include reported cases of malignant degeneration and symptomatic recurrence [2, 3, 9].

In conclusion, the two cases presented are particularly interesting because they are the first reported cases of bronchogenic cyst with pleural effusion. Surgical treatment of these two cases was difficult. Difficulties were due to adherence to mediastinal structures, independently of the para-oesophageal characteristic of the first case. Pleural effusion could have a predictive value for these surgical difficulties.

**References**