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Giant Mediastino-Pulmonary Hydatid Cyst with Endobronchial and Tracheal Extension and Probable Vertebral Involvement: A Case Report

R. C. M. Manzika Dzoussi[1*], N. Ait Said[1], N. Bakri[1], Y. Bouktib[1], A. Elhajjami[1], B. Boutakioute[1], N. C. I. El Ganouni[1]

[1]Department of Radiology, Ar-Razi Hospital, Mohammed VI University Hospital, Marrakech, Morocco

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***Corresponding author:** R.C.M. Manzika Dzoussi
Department of Radiology, Ar-Razi Hospital, Mohammed VI University Hospital Centre, Marrakech, Morocco

Abstract **Case Report**

Mediastinal hydatid disease is rare, and extension of a hydatid cyst into the central airways is exceptional. We report a 67-year-old man from a rural area, a smoker of 15 pack-years, who presented with low-volume hemoptysis, chronic dyspnea, chest pain and general deterioration lasting two months. Contrast-enhanced chest computed tomography (CT) showed an enlarging, multiloculated, non-enhancing cystic mass of the left hilum and lower lobe (99 × 81 × 110 mm, previously 68 × 64 × 97.5 mm) extending into the anterior and middle mediastinum. The mass abutted the left pulmonary artery and descending aorta and displaced the left superior pulmonary vein. Material continuous with the cyst completely filled the left main and lobar bronchi and extended to the carina and distal trachea, with luminal narrowing, complete left lung atelectasis and right-to-left transmediastinal herniation. A well-defined lytic lesion of the posterior left aspect of the L2 vertebral body eroded the pedicle base and bulged into the spinal canal. Anti-*Echinococcus* serology was positive. This case shows that a complicated hydatid cyst can mimic necrotic lung cancer in an older smoker. In endemic areas, a non-enhancing multiloculated mediastino-hilar cyst should prompt serology, and CT should map the airway, vascular and skeletal extent before surgery.

Keywords: Echinococcosis, Hydatid Cyst, Mediastinal Cyst, Pulmonary Echinococcosis, Bronchial Obstruction, Tracheal Stenosis, Spinal Echinococcosis, Computed Tomography.

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INTRODUCTION

Cystic echinococcosis is a zoonosis caused by the larval stage of *Echinococcus granulosus* and remains endemic in North Africa and the Mediterranean basin [1–3]. The liver is involved in more than 65% of cases and the lung in about 25% [4], while intrathoracic extrapulmonary locations account for about 7.4% [5]. Mediastinal involvement is rare, representing less than 0.1% of all hydatid disease [6]. In surgical series of intrathoracic hydatidosis, mediastinal cysts accounted for 3.4% and 5.7% of operated patients; most were intact and occurred in young adults [6, 7]. They usually present with compression of the airway, vessels, phrenic nerve or spine [8–11]. Complicated cysts are a recognized pitfall, as rupture, infection and pericystic inflammation can make them resemble a necrotic tumor on CT and PET-CT [12].

We report a giant, enlarging mediastino-pulmonary hydatid cyst in a 67-year-old smoker, with endoluminal extension into the left main bronchus, carina and distal trachea and a probable vertebral localization. We describe the CT findings and discuss the radiologist's role in diagnosis and surgical planning.

CASE REPORT

A 67-year-old man from a rural area, a smoker of 15 pack-years, was referred by the thoracic surgery department of Ar-Razi Hospital for low-volume hemoptysis, chronic dyspnea and chest pain. Symptoms had lasted two months and were accompanied by asthenia, anorexia and weight loss. A previous chest CT, of unknown date, had already shown a left-sided cystic lesion.

Chest CT was acquired helically with 1-mm sections, before and after intravenous iodinated contrast. It showed a multiloculated cystic mass centered on the left hilum and the posterior and lateral basal segments of the left lower lobe (Figure 1). The content was

heterogeneous with fluid attenuation, and neither the wall nor the septa enhanced. The mass measured 99 × 81 × 110 mm (anteroposterior × transverse × craniocaudal), compared with 68 × 64 × 97.5 mm on the previous CT. It extended into the anterior and middle mediastinum and was in close contact with the left pulmonary artery and its basal branches. It displaced the left superior pulmonary vein and abutted the left inferior pulmonary vein; both veins remained patent. The fat plane with the descending aorta was lost in places.

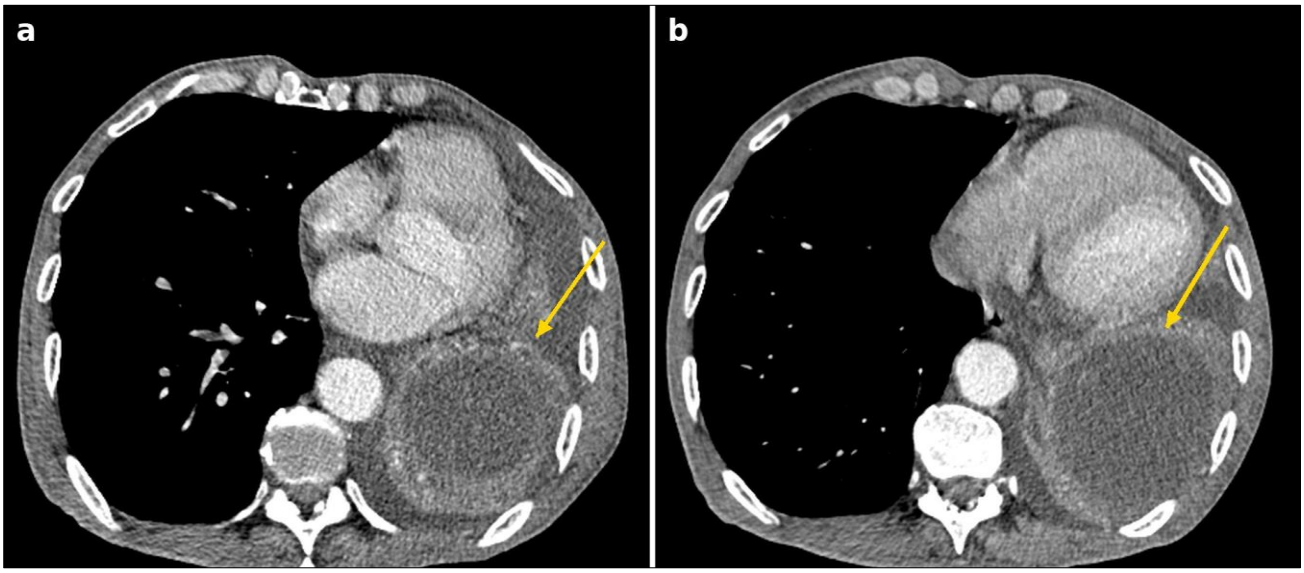


Figure 1: Contrast-enhanced axial chest CT, mediastinal window, at the level of the cardiac ventricles (a) and 3 cm lower (b). A large, well-circumscribed cystic mass in the left lower lobe has fluid attenuation, heterogeneous content and a thin non-enhancing wall (arrows). It is surrounded by the enhancing, completely atelectatic left lung and the left pleural effusion

Material continuous with the cyst completely filled the left main bronchus and the upper and lower lobar bronchi. It extended to the carina and distal trachea, narrowing the tracheal lumen, and caused complete atelectasis of the left lung (Figure 2). Associated findings were a large left pleural effusion, right-to-left transmediastinal herniation, paraseptal and centrilobular emphysema of the right lung, 5-mm micronodules of the right upper lobe and atheromatous calcification of the thoracic aorta. There was no significant mediastinal lymphadenopathy.

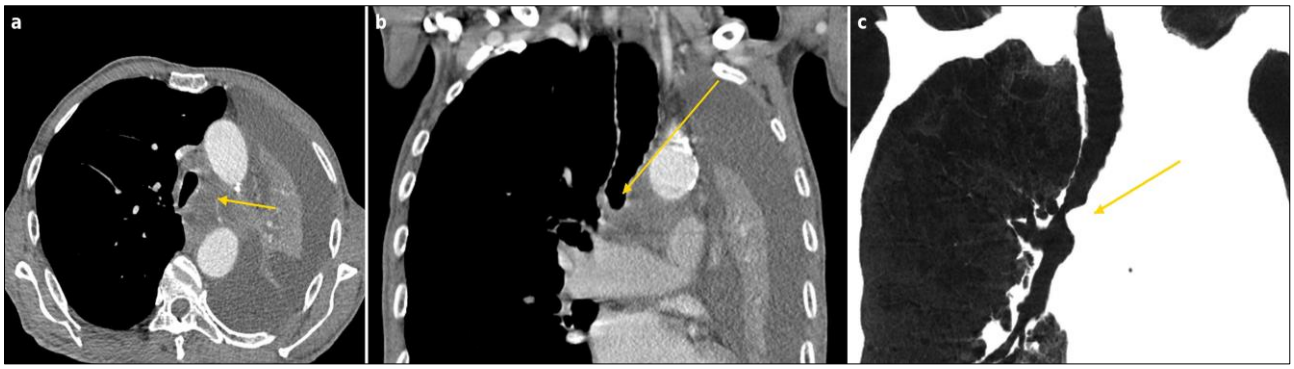


Figure 2: Airway involvement. (a) Axial contrast-enhanced CT at the carina: the left main bronchus is replaced by soft-tissue material (arrow). (b) Coronal reformation: the trachea is displaced to the right and its distal lumen is narrowed (arrow); only the right main bronchus is aerated. (c) Curved coronal minimum-intensity projection (10-mm slab following the trachea): the right bronchial tree is patent, whereas the left main bronchus contains no air (arrow). Note the right-to-left transmediastinal herniation of the emphysematous right lung.

Sections through the upper abdomen showed a thin perisplenic effusion. At the L2 level, a well-defined, rounded lytic lesion of fluid attenuation with a thin sclerotic margin involved the posterior left aspect of the vertebral body. It eroded the base of the left pedicle and bulged into the spinal canal (Figure 3).

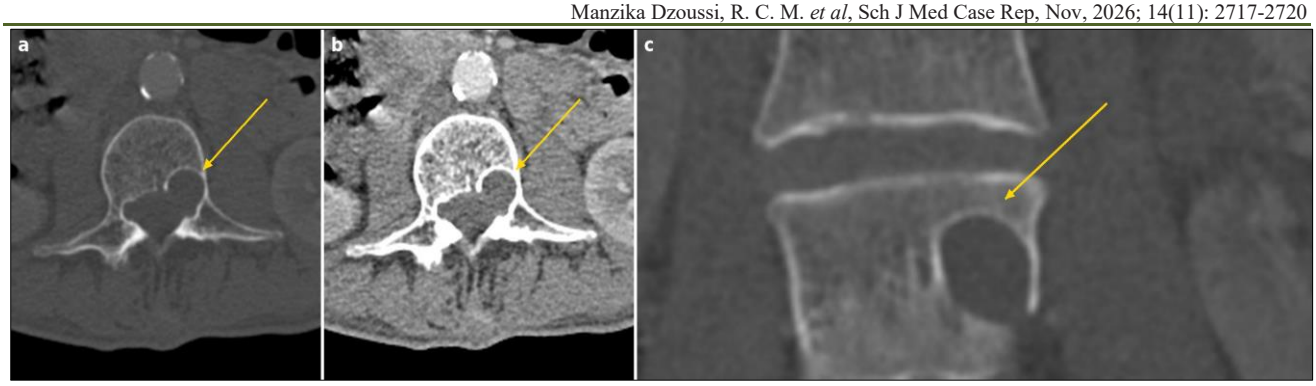


Figure 3: L2 vertebral lesion. Axial CT in bone (a) and soft-tissue (b) windows and coronal reformation in bone window (c). A well-defined, rounded, fluid-attenuation lesion with a thin sclerotic margin lies at the posterior left aspect of the vertebral body, erodes the base of the left pedicle and bulges into the spinal canal (arrows)

Anti-*Echinococcus* serology was positive, confirming the diagnosis of hydatid disease. Bronchoscopy, treatment and follow-up data were not available. Written informed consent for publication was obtained from the patient. We confirm that any aspect of the work covered in this manuscript that has involved human patients has been conducted with the ethical approval of all relevant bodies and that such approvals are acknowledged within the manuscript.

DISCUSSION

This case combines three uncommon features: a mediastino-pulmonary location, endoluminal extension into the left main bronchus, carina and trachea, and presentation in an older smoker with hemoptysis and weight loss suggesting lung cancer. Primary mediastinal cysts are thought to arise from embryos that bypass the hepatic and pulmonary filters, whereas secondary involvement results from contiguous growth of a pulmonary or pleural cyst [6-13]. The epicenter in the left hilum and lower lobe favors a pulmonary cyst extending into the mediastinum. Its size, multicompartment spread and the patient's age contrast with surgical series, in which mediastinal cysts were mostly intact and found in patients under 50 [6, 7].

CT is the reference modality for thoracic hydatid disease [1]. An intact cyst is a well-defined, fluid-attenuation lesion with a thin non-enhancing wall; multivesicular cysts show septa or daughter cysts, and rupture produces the crescent, onion-peel and water-lily signs [1-14]. The absence of enhancement is the key discriminator from a necrotic tumor, which usually shows an enhancing solid rim or nodule [1]. This matters because complicated hydatid cysts can be FDG-avid, with a mean SUVmax of 8.8 in one series, so PET-CT does not reliably exclude malignancy [12].

Rupture into the bronchial tree is the most frequent complication of pulmonary hydatid cysts, reported in 20–40% of cases [4]. It usually produces a cavity with an air–fluid level and expectoration of cyst contents, and may cause anaphylaxis or fatal respiratory failure [14, 15]. In our patient, the airway was filled from the left main bronchus to the carina and distal trachea. Fistulization with migration of membranes into the airway and extrinsic compression with intraluminal debris are both possible; the hemoptysis and complete atelectasis favor a cysto-bronchial communication. Bronchoscopy, when feasible, is the best test to identify membranes in the lumen. Mediastinal cysts can also compress the trachea, superior vena cava and pulmonary arteries [9, 11]. Here, the loss of the fat plane with the descending aorta predicts a difficult dissection.

The vertebral lesion suggests multifocal disease. Bone hydatidosis is rare, and the spine is one of its recognized sites [1-16]. Without a true pericyst, the parasite spreads along trabecular spaces and produces well-defined expansile lytic lesions that may extend into the spinal canal [1-17], posterior mediastinal cysts can also invade the spine directly [10]. In a 67-year-old smoker, the differential diagnosis includes metastasis, myeloma, aneurysmal bone cyst or giant cell tumor, tuberculous spondylitis and a nerve sheath tumor. The thin sclerotic margin favors a slow-growing benign process. MRI, which is superior to CT for assessing intraspinal extension [1-17], was not available.

Serological tests are less sensitive for pulmonary than for hepatic cysts [18], but positive serology with typical imaging is accepted as diagnostic [2]. Surgery combined with perioperative albendazole is the treatment of choice; for mediastinal cysts, partial pericystectomy is acceptable when dissection near vital structures is dangerous [2-19]. Albendazole alone is reserved for inoperable patients [2, 3]. The main limitations of this report are the absence of histological confirmation, bronchoscopy, MRI and follow-up, and the fact that the previous CT could not be reviewed.

CONCLUSION

A mediastino-pulmonary hydatid cyst with endobronchial and tracheal extension is a rare, potentially life-threatening presentation that can closely mimic necrotic lung cancer in older smokers. In endemic areas, an enlarging, multiloculated, non-enhancing cystic mass should prompt serology. The radiologist should

map the airway, vascular, pleural and skeletal involvement that determines the surgical strategy, and recommend MRI for any lytic vertebral lesion extending into the spinal canal.

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