

Mediastinal Teratoma With Coexisting Parenchymal Pulmonary Cystic Lesion

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A double-located mediastinal and intrapulmonary cystic teratoma is a rare condition to be considered by thoracic surgeons. Clinical or radiologic diagnosis of a ruptured mediastinal teratoma into adjacent structures may be highly suggestive. An atypical presentation may indicate cautiousness for complete surgical excision. We report the case of a 14-year-old girl presenting with chronic chest pain. The radiologic work-up showed a large cystic mediastinal tumor and a heterogeneous intrapulmonary left upper-lobe lesion. We discuss the radiologic differential diagnosis of this atypical double-located thoracic tumor and the surgical strategy for complete excision.

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Cystic teratomas are usually unique germ cell tumors that are located in the anterior mediastinum [1]. They typically appear as uncomplicated encapsulated lesions that consist of fluid, calcifications, or fat tissue, or both [1]. Acute rupture is rare and mostly occurs into adjacent structures such as the pleural space, pericardium, and lung [2, 3]. Clinical presentation combined with specific computed tomography (CT) features usually leads to an easy diagnosis. We discuss here the main differential diagnosis and surgical strategy in the case of a double-localized mediastinal and intrapulmonary cystic tumor with unspecific chronic symptoms.

A 14-year-old girl from Algeria was referred to our center for a voluminous anterior mediastinal tumor discovered at chest roentgenogram in the context of chronic chest pain. A chest CT scan showed a multiloculated, rather homogenous mediastinal cystic lesion measuring 90×55 mm (Fig 1). A second bullous cystic lesion was visualized in the left upper lobe (Fig 2), with pleural thickening and suspicion of a communication cord between the 2 lesions. There was no mediastinal adenomegaly. Results of echinococcosis serologic analysis were negative. On the basis of the patient's origin and the radiologic findings, we suspected a diagnosis of mediastinal hydatid cyst ruptured into the lung.

Surgical en bloc excision was planned, with particular care to avoid any tumor fragmentation, and hypertonic

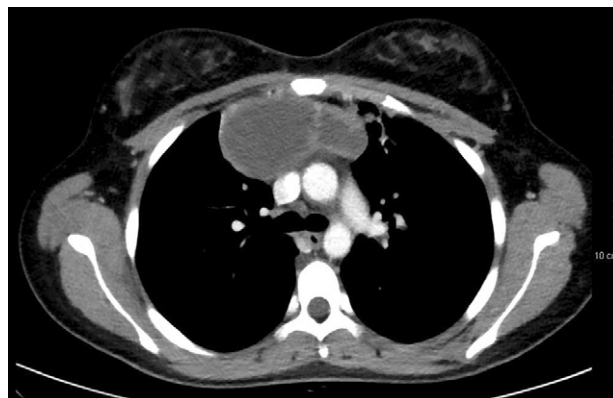


Fig 1. A chest computed tomography scan in the mediastinal window shows a multilobular mass in the anterior mediastinum.

saline solution was used in the operative field. A median sternotomy revealed a 10-cm, solid, nonhomogenous mediastinal mass in contact with the 2 phrenic nerves. Both pleurae were opened, revealing dense bilateral adhesions.

The lesion was first dissected, with both phrenic nerves preserved throughout their mediastinal course. A frozen section of the surrounding tissue showed no signs of malignancy, allowing us to preserve both nerves. The innominate vein and both thymic horns were dissected free of the tumor. The en bloc resection required partial pericardial excision. No tumor fragmentation occurred during this procedure. This was followed by an atypical pulmonary resection of the left upper lobe (wedge resection). The pulmonary lesion was indeed well defined and located in the anteroapical segment.

Postoperative histopathologic examination showed a unilocular cyst, filled by a yellowish fluid and a grumous material mixed with hair. Although adhesions were present between the cyst and the surrounding thymic, pericardial, and pleural structures, no sign of macroscopic infiltration was observed. The tumor was microscopically composed of mature, adult-type tissues, including squamous and respiratory-type epithelium, sweat glands,

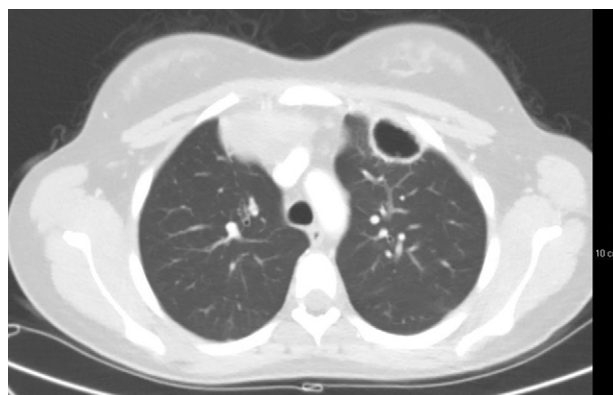


Fig 2. A chest computed tomography scan in the parenchymal window shows a bullous cystic lesion in the upper left lobe associated with pleural thickening.

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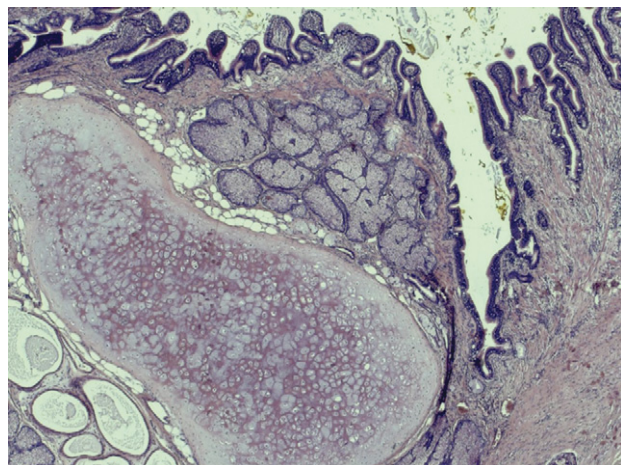


Fig 3. A photomicrograph shows mature teratoma with respiratory-type tissue, skin appendages, mature cartilage, and fat (hematoxylin and eosin stain, original magnification $\times 25$).

sebaceous glands, mature cartilage, and hair follicles (Fig 3). There was neither necrosis nor hemorrhage. No immature (embryonic or fetal) or germinal- or somatic-type malignant features were observed in the tumor. The tumor was surrounded by a fibroinflammatory tissue rich in lymphocytes, histiocytes, and foreign-body giant cells. The diagnosis of mature cystic teratoma was confirmed. A subpleural cystic lesion, filled with a yellowish material, was found in the parenchymal resection. The lung cyst microscopically corresponded to a foreign-body reaction against the hair and fat (Figs 4 and 5).

The patient's postoperative course was uncomplicated, with transient elevation of the left diaphragm that resolved spontaneously by postoperative week 2.

Comment

We report the case of a patient with a mature mediastinal teratoma with secondary intrapulmonary localization.

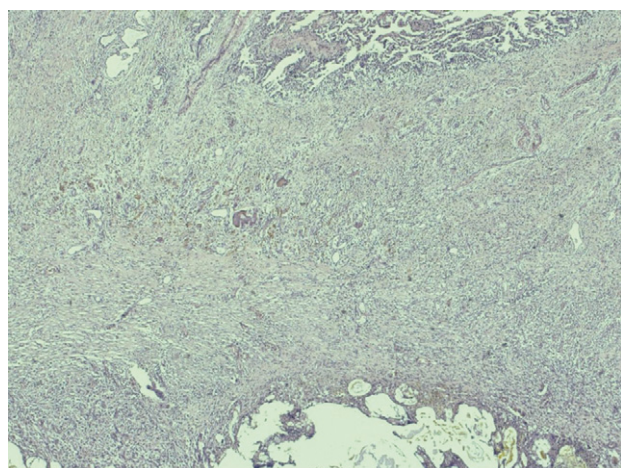


Fig 4. A photomicrograph shows lung parenchyma supplanted by fibroinflammatory tissue (hematoxylin and eosin stain, original magnification $\times 25$).

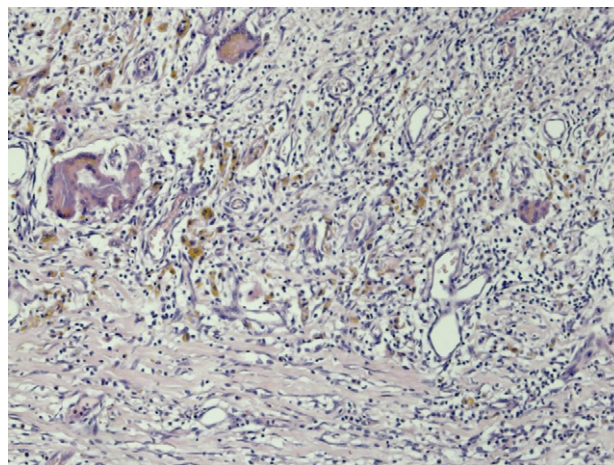


Fig 5. A higher magnification illustrates the important foreign-body giant cell reaction (hematoxylin and eosin stain, original magnification $\times 100$).

Teratomas are the most common mediastinal germ cell tumors [1]. Symptoms of uncomplicated mediastinal teratomas include chronic cough and chest pain. Rupture is the major complication. Acute pain is a sign of such complication that can occur in the pleura [2], bronchial tree [3], or even the pericardial space [4]. Mediastinal teratoma rupture into the lung has been very rarely described [5]. Trichoptysis (hair expectoration) is rare but specific of this complication [2]. Finally, teratomas can also be hemorrhagic [6].

In our case report, despite the presence of a secondary pulmonary tumor, the patient's chronic chest pain was not clearly suggestive of acute rupture of a mediastinal primitive lesion. Therefore, clinical diagnosis for cystic tumors should also include double-located hydatid cysts, because these tumors essentially affect adolescents in endemic areas and are poorly symptomatic until growth or rupture [7]. Main symptoms also include chest pain and cough. The rupture of a mediastinal lesion into the pleural cavity is a rare but previously described pathology [7].

Because the serologic test is not specific for a diagnosis, chest CT is considered the most efficient diagnostic method [8]. Hydatid disease rather typically presents as homogeneous air-filled fluid cavity masses with air-fluid levels [8]. A preoperative diagnosis is mandatory because of the perioperative measures to prevent the surrounding tissues from being flooded by cystic material and the recommendations for a conservative operation.

The double-located cystic lesion in this patient, rather homogenous at CT and clinically poorly suggestive of a complicated lesion, had to be managed with caution. This presentation was first suggestive of double-located hydatid disease, not excluding a diagnosis of complicated teratoma. In such an atypical presentation, surgical management should consider systematic use of intraoperative hypertonic saline solution and a

complete tumor excision to avoid tumor fragmentation and spillage.

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Thymoma Originating in a Giant Thymolipoma: A Rare Intrathoracic Lesion

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Thymolipoma is a rare, slow-growing, benign tumor that arises from the anterior mediastinum and corresponds to 2% to 9% of all thymic neoplasms. We present the case of a 49-year-old man who had a large heterogeneous mass with areas of soft tissue and fat tissue located on the anterior mediastinum and right hemithorax. After resection, histologic analysis confirmed the diagnosis of a giant thymolipoma containing solid components that corresponded to thymomas B1, B2, and B3. We discuss the occurrence of an atypical variant of thymolipoma containing three types of thymomas inside.

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Thymic neoplasms are rare tumors that account for less than 1% of all adult malignancies [1]. The primary epithelial neoplasms of the thymus are thymoma and thymic carcinoma, the first being more common. According to the World Health Organization, thymomas are histologically classified in five subtypes with prognostic significance: A, spindle cells; AB, mixed spindle cells and lymphocytes; B1, predominance of lymphocytes; B2, mixed lymphocytes and epithelial cells; and B3, predominance of epithelial cells.

A 49-year-old man complained of weakness and shortness of breath in the last year, with worsening of the symptoms in the last month. He reported a prior incidental diagnosis of an asymptomatic mediastinal mass. No relevant abnormalities were noted on the hematologic or biochemical studies. Microbiologic analysis of sputum was negative.

A chest roentgenogram showed almost complete opacification of the right hemithorax, with a contralateral shift of mediastinal structures and inferior displacement of the right hemidiaphragm (Fig 1). A computed tomography (CT) scan demonstrated a large heterogeneous mediastinal lesion, with areas of soft tissue and fat tissue confirming the extensive involvement of the anterior mediastinum and right hemithorax (Fig 2). This lesion displaced the mediastinal structures to the left and the diaphragm inferiorly, leading to nearly complete collapse of the right lung. The evaluation for distant metastases using positron-emission tomography–CT was negative.

The patient was referred for surgical treatment, and the lesion was entirely resected. The resected mass consisted of a partially encapsulated fatty mass that measured 34.3 × 23.4 × 14.6 cm and weighted 4.1 kg (Fig 3). Microscopically, the mediastinal lesion comprised an admixture of approximately 50% unremarkable mature adipose tissue and approximately 50% of normal thymic parenchyma along with interspersed areas of thymomas subtypes B1, B2, and B3 (Fig 4). Sections taken from the fatty areas of the mass showed mature fatty tissue harboring scattered foci of unremarkable thymic tissue containing Hassall corpuscles and calcifications.

The solid mass was divided by a thin fibrous band emanating from the fibrous capsule. It comprised a varied aspect of plump epithelioid cells having pale chromatin, prominent nucleoli, and pink ill-defined cytoplasm, associated with mature lymphocytes representing areas of thymomas subtypes B1, B2, and B3 according to the predominant cell finding and the World Health Organization classification [1]. Mitotic figures were scarce. Hemorrhage and necrosis were eventually found. No evidence of squamous, glandular, or sarcomatoid differentiation was present. The resection margins and mediastinal and hilar lymph nodes were negative. The final diagnosis was giant thymolipoma containing components of thymoma subtypes B1, B2, and B3.

The patient was discharged 1 week later in excellent condition. No further therapy was given, and the patient was scheduled for follow-up.