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Case Report

A Mature Teratoma of the Mediastinum Revealed by an Intra-Pleural Rupture: A Rare Entity

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Abstract

Teratomas are tumors composed of tissues from at least two of the three primary germ layers, ectoderm, mesoderm and endoderm. Mediastin is the most common extra-gonadal site for germ cell tumors including mature teratomas. Although this is an important differential diagnosis of anterior/medium mediastinal masses in young adults and various atypical presentations have been reported. However, pleural effusion secondary to the rupture of a benign germ cell tumor is quite rare and the nature of pleural effusion in such rupture cases has not been examined in detail in the literature. We report here a case of mature cystic teratoma coexisting with pleural effusion, which is quite rare and makes this case a very interesting learning curve.

Keywords: *Teratoma; Mediastinal Masses; Pleural Effusion*

Introduction

Mediastinal tumors include thymoma, lymphoma, metastases and teratoma. The mediastinal teratoma is an uncommon tumor (about 3 to 12% of mediastinal tumors).

Most mediastinal teratomas produce no symptoms; they are most often associated with the compression of neighborhoods structures.

Teratomas occur mainly in young adults; with approximately equal incidence in men and women.

We present the case of a patient with a mature mediastinal teratoma revealed by purulent pleural effusion.

Clinical Case

A 27-year-old divorced patient, native and resident in a rural area around Marrakech-Morocco, a professional housekeeper, with no particular pathological background, exposed to passive smoking, notion of risky sexual relations, presented itself in our training for Sadoul stage IV stress dyspnea associated with moderate intensity left chest pain in the tip of the side of the chest radiating interscapularly with a productive cough bringing back purulent blood striated sputum in a context of febrile sensation and sagging general condition evolving since 01 months of its admission after a cooling episode.

On examination, the patient was conscious, hemodynamically and respiratorily stable, no sign of respiratory struggle, fever at 38.5°C, ODS2 at 94% in ambient air, FR at 20cpm, FC 90bpm, BP at 120/70 mmgh, a weight of 65 kg or a BMI at 23.6 kg/m^2, objective pleuropulmonary examination, a liquid pleural effusion syndrome of the entire left chest without further detectable anomaly.

The chest radiograph showed a homogeneous dense opacity of pleural type without aerial bronchogram with positive silhouette sign pushing the mediastinum elements towards the contralateral side (Figure 1).

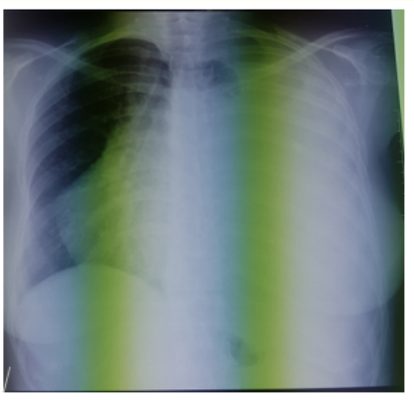

Figure 1: *Chest x-ray on admission.*

A pleural puncture was performed objectivizing a viscous purulent liquid requiring thoracic drainage at the 4th left intercostal space using a Joly 28F drain bringing back 1000 cc of the liquid, the radiological examination showed a persistence of the pleural image (Figure 2 and 3).

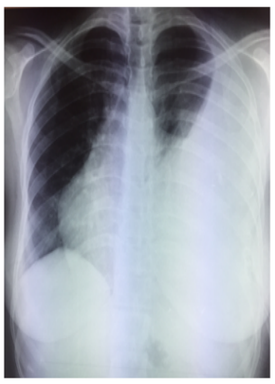

Figure 2: *Chest x-ray after posterior drainage.*

Figure 3: *Purulent pleural fluid.*

The study of pleural fluid:

- Cytological analysis shows the presence of many poly-morpho-nuclear structures, some altered without suspicious cells.
- Biochemical analysis showed a protein level of 87 g/l, adenosine deaminase 26.6 IU/L, amylase 210 IU/L, LDH 1820 U/L,
- Bacteriological and parasitic analysis: Gram staining, Ziehl-Neelsen staining, pyogenic culture and mycobacterial culture, all negative. The existence of a scolex and an echinococcus hook could not be demonstrated. Xpert MTB/RIF does not detect any DNA sequences specific for Mycobacterium tuberculosis in pleural fluid.

Chest ultrasound showed the presence of a pleural effusion with evidence of a floating pleural mass.

The tumour markers of mediastinum (alpha-fetoprotein and human chorionic beta gonadotropin) were both normal.

HIV serology was negative.

The thoracic CT allowed to individualize a left pleural lesional process measuring 10/15 cm of tissue and calcium fat component associated with a left pleural effusion of great anechoic abundance supplemented by a thoracic MRI objectifying a radiological aspect in favour of a solidocystic teratoma at the mediastinum's expense (Figure 4).

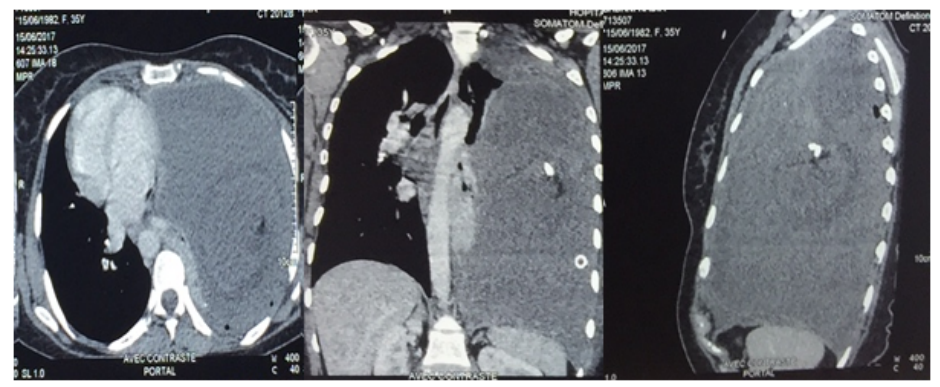

Figure 4: *Scanning aspect of the teratoma.*

Bronchoscopy showed a diffuse 2nd degree inflammatory state without detectable endobronchial lesion. Bronchial aspirations did not reveal any specific abnormalities.

Subsequently, we referred the case to our thoracic surgery department, the patient underwent a left posterolateral thoracotomy under general anesthesia, the exploration showed a huge left basithoracic solidocystic mass adhering to the lower left lobe and based on mediastinal implantation, the mass was resected entirely in monoblock.

Histopathology of the excised mass showed solidocystic structures bordered by laminated squamous cell carcinoma, sebaceous material containing a tooth, fatty tissue, pancreatic tissue and mature glial tissue associated with pilious follicles and muscle fibres. The final diagnosis was a mature multi-vascular mediastinal teratoma (Figure 5).

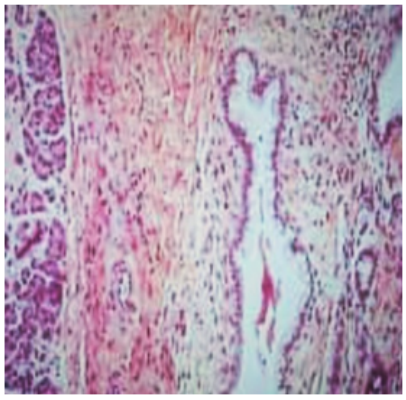

Figure 5: *Histology of the mature teratoma.*

The patient was followed up regularly in our department with very good clinical and radiological improvement.

Discussion and Conclusion

The first case of pulmonary teratoma was reported by Mohr in 1839. The anterior mediastinum is the most common extragonadal site for germ cell tumours and mature teratomas are the most common histological type of primary mediastinal germ cell tumours.

They usually occur in the middle of the body, which is the germ cell migration pathway during embryogenesis; their migration may be lost along the way to their respective organs, resulting in the development of tumours later in life.

In fact, several theories exist; one of them suggests that the benign teratoma is derived from the region of the third cleft or bronchial pocket. A second theory is that these tumours originate from germ nests of cells along the urogenital crest that did not migrate to the gonads during embryological development.

Benign teratomas are often asymptomatic and are not covered on the chest x-ray for unrelated reasons, but sometimes they give symptoms leading the patient to consult. If symptoms are present, it will be because of the mass effect caused by the mediastinal teratoma [1].

Teratomas are more common in young adults. About one-third of patients remain asymptomatic while symptoms develop when cysts become infected and eroded in the pleural (respiratory distress and chest pain) or pericardial (cardiac tamponade) or bronchial space (hemoptysis, trichoptysis or expiration of sebaceous material) [2].

Radiologically, cystic teratomas are generally smooth, rounded and well defined, while solid varieties are more lobed and asymmetric. CT can clearly identify soft tissue, fat and calcification (even fully formed teeth and bones), making it one of the few mediastinal tumours that can be diagnosed with confidence before surgery [3].

Laboratory tests are often normal and serum levels of human chorionic gonadotropin and alpha-fetoprotein are always normal in patients with benign teratoma [4].

Several mechanisms explaining the rupture of a teratoma in the surrounding organs have been mentioned: local ischemia related to the progressive growth of the tumor and responsible for a neighbourhood necrosis or infection that can weaken the wall of the teratoma. The most attractive hypothesis is that of "autolysis" through proteolytic enzymes secreted by certain well-differentiated tissues composing the teratoma. Pancreatic tissue is probably the most common cause [5].

It is interesting to note that pleural effusion resulting from the rupture of a benign germ cell tumour is quite rare. The study by Choi., *et al*. in 17 patients revealed a preoperative rupture with concomitant pleural effusion in < 25% of patients. There is no detailed report on the nature of pleural effusion activity in such cases of rupture. In some cases, a high level of carcinoembryonic antigen was observed in the pleural fluid, and in others, a high level of amylase. However, no conclusive description was provided. Our case showed exudative pleural effusion with a marginal increase in amylase levels. Now, our case is worth mentioning in the perspective of factors that require strong clinical suspicion to predict mature mediastinal cystic teratoma as a potential etiology of undiagnosed pleural effusion [6].

Surgical excision is the treatment of choice for mediastinal teratomas [7].

The goal of surgery is to achieve complete removal of the tumor without damaging associated or adjacent structures. This requires careful and meticulous surgical dissection.

The prognosis of mature mediastinum teratomas is excellent after complete resection. Recurrences are rare, especially those related to incomplete resection [8,9].

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