CASE REPORT

A Rare Case of Intrapleural Teratoma – Mimicking as Empyema Thoracis

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ABSTRACT

Teratoma is a type of germ cell tumor that derived from all the three embryonic layers which are endoderm, mesoderm and ectoderm. The commonest site of extragonadal germ cell tumor is at the mediastinum, which accounts for 50-70% of all mediastinal tumor. Intrapleural teratoma is exteremely rare, with only one previously reported case to date. Teratoma is usually a slow growing tumor, and symptoms typically presented late as a result of compression or obstruction to the surrounding structures. Due to its rare occurrence, intrapleural teratoma can pose a diagnostic challenge for clinicians. Familiarity with the presentation and imaging findings is therefore of great values which can guide the diagnosis and later the specific management plans. This case report highlights the rare case of intrapleural teratoma and summarizes the presentations and imaging findings of intrapleural teratoma which was initially misdiagnosed as empyema thoracis. Several learning points from this case were outlined.
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INTRODUCTION

Teratoma is a type of germ cell tumor that derived from all the three embryonic layers which are endoderm, mesoderm and ectoderm. The commonest site of extragonadal germ cell tumor is at the mediastinum, which accounts for 50-70% of all mediastinal tumor. Intrapleural teratoma is exteremely rare, with only one previously reported case to date. Teratoma is usually a slow growing tumor, and symptoms typically presented late as a result of compression or obstruction to the surrounding structures. Due to its rare occurrence, intrapleural teratoma can pose a diagnostic challenge for clinicians. Familiarity with the presentation and imaging findings is therefore of great values which can guide the diagnosis and later the specific management plans. This case report highlights the rare case of intrapleural teratoma and summarizes the presentations and imaging findings of intrapleural teratoma which was initially misdiagnosed as empyema thoracis. Several learning points from this case were outlined.

CASE REPORT

A 33 years old lady, non-smoker and was previously well presented with history of intermittent cough for the past 1 year. There were also episodes of night sweats with chills over the past 1 year, with worsening symptoms especially during the nighttime. She started experiencing episodes of shortness of breath for the past 2 months with worsening of her symptoms for 2 days prior to admission. No history of hemoptysis. Otherwise, there were no constitutional symptoms and no history of recent travels. She had completed two doses of Covid-19 vaccination.

At the presentation to the Emergency and Trauma Department (ETD), patient was in distress, with blood pressure of 157/105 mmHg and tacyhcardic with heart rate of 129 beats per minute. Her oxygen saturation (SpO2) level is 90-91% under room air. No fever was documented. Chest auscultation showed reduced breath sound at the left lower-mid zone. Otherwise, normal heart sound with no murmurs, abdomen was soft with no palpable mass and no palpable enlarged lymph nodes.

She was immediately put on high flow oxygen mask, and the SpO2 improved to 95%. Arterial blood gas taken on high flow oxygen mask and it showed mixed acute respiratory failure with compensated respiratory acidosis: pH 7.38 (normal value: 7.35-7.45), pCO2 52mmHg (normal value: 35-45mmHg), pO2 80mmHg (normal value: 75-100mmHg), HCO3- 30.8mEq/L (normal value: 22-26mEq/L).

Bedside ultrasound scan in the Emergency Department showed Inferior Vena Cava is <50% collapsible (indicating normal central venous pressure), heart displaced to the right side with good cardiac contractility and no D sign (no evidence of right ventricular pressure or volume overload).

Chest X-ray revealed a left sided pleural effusion with mediastinal shift to the right side. Ultrasound of the left thorax showed complex massive left sided pleural effusion with debris within. Left chest tube size 32Fr was inserted over the left lower thoracic region, and approximately 1400mls of thick curdy fluids with sediments were drained. Pleural fluid biochemical results showed exudative pleural effusion, not chylothorax; with pH 6.046 (normal value: 7.60-7.64), protein 61 g/L (normal value: 10-20 g/L), protein ratio 0.84 (normal value < 0.5), Lactate Dehydrogenase (LDH) 400U/L, LDH ratio 1.87 (normal value: <0.5), glucose 0.2mmol/L (normal value: <3.3 mmol/L), cholesterol 1.61 mmol/L (normal value: 3.5-6.5 mmol/L) and triglycerides 0.46 mmol/L (normal value <2mmol/L). Pleural fluid Adenosine Deaminase (ADA) was normal at 1.37IU/L, making the diagnosis of tuberculous fluid less likely. Pleural fluid cytology result showed degenerated cells and debris mixed with some inflammatory cells and blood in the background with no atypical cells seen. Bacterial and fungal culture, together with the biohazards screening were negative.

Total white cell count is mildly elevated with 11.1 x 109 /L and C-Reactive Protein (CRP) were raised at at 319 nmol/L (normal value < 47.6nmol/L). The rest of the biochemical profiles were normal. Tumor markers in particular alpha-fetoprotein (AFP) and the beta-human chorionic gonadotropin (beta-hCG) were all within normal limit.

Patient was treated empirically as left sided empyema thoracis with Intravenous (IV) antibiotics (IV Metronidazole and IV Ceftriaxone). Over the course of the next three days, approximately 2000cc of fluid were further drained. Patient improved symptomatically, and oxygen was eventually weaned down.

Contrast Enhanced CT Thorax were performed and it showed large residual amount of complex left pleural effusion with air locules and mild enhancement of the pleural lining (Fig. 1-3). Overall findings are suggestive of infective causes. No evidence of lung or mediastinal mass. Respiratory physician decided to administered 6 doses of intrapleural (IP) streptokinase following the CT findings. However, minimal fluid of less than 50mls were drained daily post IP streptokinase with poor re-expansion of the left lung.

This case was subsequently referred to cardiothoracic team for decortication of the left empyema thoracis. Left thoracotomy and decortication were performed, and intraoperative findings showed hair like structures

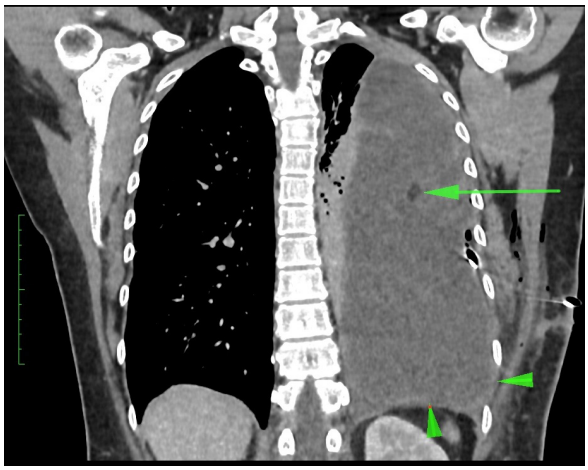

Figure 1: CECT Thorax coronal view. Fat attenuated lesion, attenuation of -70 HU (green arrow) within the massive left pleural effusion. Enhancing left pleural lining (green arrowhead)

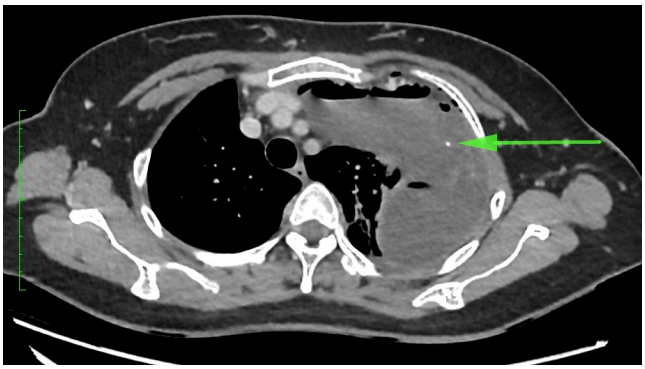

Figure 2: CECT Thorax axial plane. Focus of coarse calcification (green arrow) within the left pleural effusion

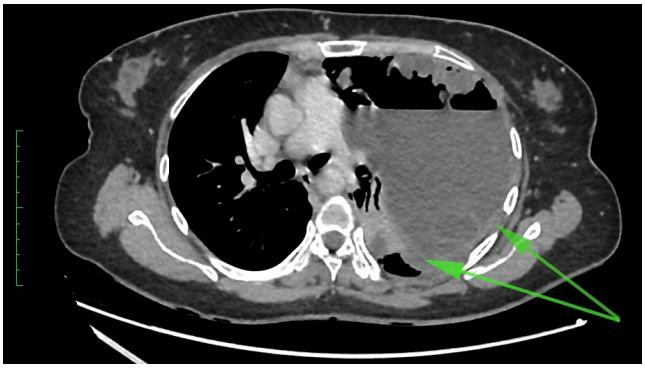

Figure 3: CECT Thorax axial plane. Thick soft tissue densities along the pleural lining (green arrow)

within purulent pleural fluid with slough and fibrous strands. Both the parietal and visceral pleural linings over the left upper and lower lobes were thickened.

Parietal pleural was biopsied and sent for histopathological examination (HPE). Microscopy findings showed fragments of fibro collagenous tissue, some were covered by stratified squamous epithelium. There were cystically dilated spaces and glands within the stroma lined by abundant cytoplasmic mucin with basally located nuclei. Some of the cells exhibit Paneth cell metaplasia and goblet cells. These cells were

positive for CK7, CDX2 and focally positive for CK20. In some areas, there was inflamed granulation tissue, stromal hemorrhage, neutrophilic micro abscesses with collection of foreign body-type multinucleated giant cells and lamellar keratinous material. Scattered moderate lymphoplasmacytic and neutrophilic infiltrate was noted with some foamy histiocytes. Lobules of mature adipose tissue, skeletal muscle fibers and neuroglial tissue were also present. No evidence of malignancy seen. Overall the HPE of the parietal pleura was consistent with intrapleural teratoma.

Surgical removal of mature teratoma is almost always curative. This patient was scheduled for CT Thorax follow up at 6 months interval to look for evidence of recurrence and post operative changes.

DISCUSSION

Extragonadal germ cell tumors made up approximately 1-5% of the entire germ cell tumor. The conventional theory is that extragonadal germ cell tumors originated from the primordial germ cells was misplaced during their movement to the gonads. Commonest site for extragonadal germ cell tumor are at mediastinum, accounting to 50-70% of all germ cell tumor, follow by retroperitoneal region that accounts for 30-40% of the germ cell tumor (1). Teratoma is the most common type of germ cell tumor that develops extragonadally.

Intrapleural teratoma without mediastinal component is extremely rare. To the best of our knowledge, up to the time of writing, this is only the second reported case of intrapleural teratoma with absence of mediastinal component. The previously reported case was also seen in a young lady who presented with acute chest pain with shortness of breath; and the CT imaging revealed a complex cystic mass within the left pleural cavity with pleural effusion. The HPE diagnosis was confirmed post thoracotomy removal of the cystic mass (2). In this case, CT imaging was only performed after the chest tube insertion; and it did not show any evidence of encapsulated tumor or cystic mass in mediastinum and the pleural cavity. We can only postulate that either there was spontaneous rupture of the intrapleural teratoma leading to the acute presentations or the tumor was ruptured iatrogenically during the chest tube insertion.

There were several cases of mediastinal germ cell tumor mimicking pleural effusion or empyema as their initial presentation, especially when the mass is predominantly of cystic component (3). In one of the cases, it was misdiagnosed as tuberculosis at presentation and was treated on anti-tuberculosis for 6 months. The diagnosis of mediastinal mass was only made after the CT Thorax revealed a huge cystic mediastinal mass (4). However, in all these cases, the mediastinal mass were clearly evident on the CT imaging whereas in our case the mediastinum was totally clear, with no evidence of mass.

Several setbacks leading to the misdiagnosis of this patient were identified. These include the pleural fluid analysis, which showed inflammatory exudation; and raised CRP, which is an inflammatory marker. However, the most important factor is likely due to the imaging findings in which the CT Thorax showed complex left sided pleural collection with enhancement of the left pleural lining which was consistent of empyema. All these findings were incorrectly attributed to empyema thoracis. Retrospectively, this patient had never had documented fever during presentation and throughout the admission. The infective marker, total white cell count was only mildly elevated in this patient. Accordingly, a retrospective review of the CT images of this patient showed small fatty attenuated lesion; attenuation of -60 to -70HU with scattered coarse calcifications can be seen within this encapsulated left pleural collection. Some part of the pleural lining was also vastly thickened, which may suggest some soft tissue component.

CONCLUSION

Diagnosis is a dilemma for this case. Considering the clinical presentations with the findings from multimodal diagnostic tools including radiological, microbiological, and histological favour towards diagnosis of phaeohyphomycosis. Since these fungal infections are subclinical in presentation, the importance of imaging and histology should be emphasized to achieve appropriate treatment, prevent recurrence, and avoid functional and aesthetical impairments.

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