

A case of pleural mature teratoma masquerading as empyema thoracis

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SUMMARY

Mediastinal masses are diagnostically challenging due to their varied presentations. Teratomas are a rare cause. We report a case of a young woman with chronic cough and weight loss, initially treated for tuberculosis (TB) based on pleural fluid analysis. Thoracic CT revealed a large left-sided empyema. After thoracotomy and decortication, histopathology confirmed a mature teratoma. We believe this is the first report of a teratoma of the pleura which manifested as a tuberculous empyema.

INTRODUCTION

Teratoma is a unique type of tumor consisting of mature or immature cell originating from more than one germ cell layer, sometimes all three.¹ The majority of germ cell tumors develop in the gonads, with only approximately 5% being extragonadal. Among extragonadal cases, 80% are located in the mediastinum.² Adult patients with mediastinal teratoma usually are asymptomatic and only detected when they cause compressive symptoms. Pleural effusion is rather a rare presentation and only reported in a few cases.³

CASE PRESENTATION

A 19-year-old woman presented to a district hospital with a chronic cough and a 10 kg weight loss over a month. She had no fever or TB contact. Examination revealed reduced breath sounds in the left lung, and a chest X-ray showed left-sided pleural effusion. She was treated with oral antibiotics and followed up as outpatient for TB workup.

She was subsequently admitted to a tertiary hospital for persistent pleural effusion. A chest tube was inserted, and pleural fluid analysis revealed transudative fluid. TB workup was mostly negative, except for elevated pleural fluid ADA (138.73 U/L). Cytology was negative for malignancy, and she was empirically treated for TB. Intrapleural streptokinase was administered due to minimal chest drainage and persistent effusion (Figure 1A).

A CT scan two weeks later showed a large, loculated left pleural empyema with small hydropneumothoraxes in the lower part, causing subsegmental collapse of the left lung lobes, and right tracheal and mediastinal shift. Fat density lesions in the effusion suggested pleural lipomatosis (Figure 2). She was referred for decortication.

She underwent emergency left thoracotomy and decortication in our centre. Intraoperatively, a thick-walled multiloculated cortex was found at the left lung hilum, displacing and entrapping the upper and lower lobes. The loculated cortex contained clear fluid, with two cystic collection of purulent material (Figure 3C, D). Few loculations contained "cheesy" material (Figure 4), which was aspirated. Specimens were sent for culture, TB workup, and histopathology.

Postoperatively, a chest X-ray showed good lung expansion (Figure 1B), and she was discharged on day 5. She recovered without complications. Intraoperative specimens were negative for TB, so anti-TB medications were discontinued.

Histopathology confirmed a mature teratoma arising from the pleura, with no mediastinal involvement (Figure 5 A-D). It contained cysts lined by squamous and columnar epithelium, as well as adipocytes, glands, smooth muscle, and pancreatic tissue, with no signs of malignancy. The final diagnosis was mature pleural teratoma.

She was scheduled for follow-up in 3 months and pelvic imaging to rule out occult gonadal neoplasm.

DISCUSSION

The term "teratoma" was coined by Virchow in 1863, originating from the Greek word "teras," which translates to "monster." Teratomas are tumors typically comprised of various cell types derived from all three embryonic layers, namely ectoderm, mesoderm and endoderm.¹

Teratoma commonly arise from gonads, whereas only 1 to 5% of them arise from extragonadal sites such as mediastinum, pineal area, sacrococcygeum.⁴ Among the extragonadal teratoma, 80% of them are located in the mediastinum, more commonly anterior mediastinum. If the teratoma is made up of well-differentiated components, then it is a mature teratoma. A mature teratoma located in the mediastinum is typically benign, yet it harbors the potential for malignancy.² Benign mediastinal teratomas occur with equal frequency in both women and men, whereas malignant teratomas are more prevalent among men.⁵ In this reported case, the patient is a young female who had a teratoma, which originated from pleura only, not other organs in the mediastinum. To date, there is no literature

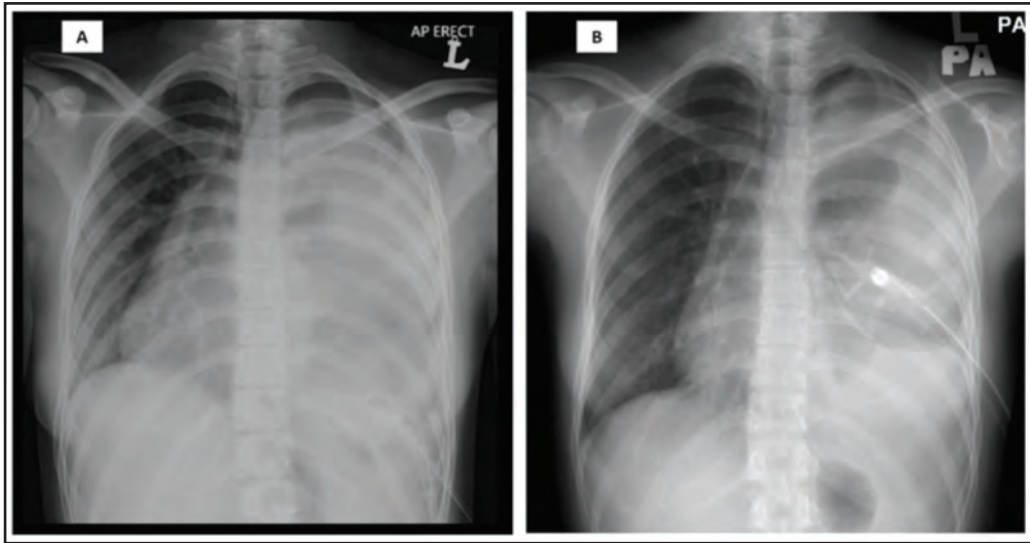


Fig. 1: CT scan coronal view, showed large loculated left pleural empyema with small locules of hydropneumothoraces at the lower part, with right tracheal and mediastinal shift

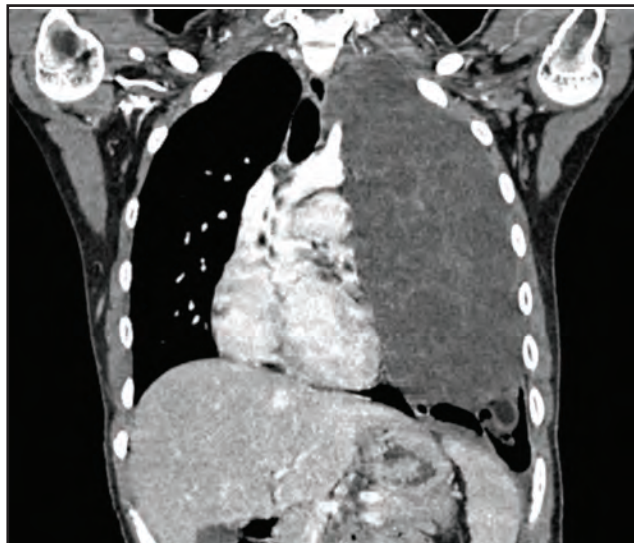


Fig. 2: CT scan coronal view, showed large loculated left pleural empyema with small locules of hydropneumothoraces at the lower part, with right tracheal and mediastinal shift

that reported on pleural teratoma manifesting as tuberculous empyema.

Benign mediastinal teratomas are usually incidentally discovered on chest radiographs during examinations. Symptomatic patients usually are due to the mass effect exerted by the mediastinal teratoma. Patient may also exhibit expectoration of hair, which is considered a pathognomonic symptom. Nevertheless, this distinctive symptom is very rare and typically a late presentation following tumor rupture into the tracheobronchial tree.⁶ Laboratory tests often yield normal results, and serum levels of human chorionic gonadotropin (hCG) and alpha-fetoprotein (AFP) are consistently within normal ranges in patients with benign teratomas.⁷ Pleural effusion and

empyema is considered rare presentation for mediastinal teratomas and limited to case reports only. There is also lack of data available on the its nature of pleural fluid.

The modality for assessing mediastinal teratomas is a CT scan thorax, because it offers superior evaluation of their location, extension, and vascularity. Shameem et al. highlighted that the presence of a fat-fluid level is considered pathognomonic for teratomas.¹¹ In the scenario of tumor rupture, pancreatic enzymes are reported to be useful, with associated findings including fat globules at the rupture site, airspace opacities, such as consolidation or atelectasis in adjacent lung parenchyma, and pleural effusion. Magnetic Resonance Imaging (MRI) is useful in cases where there is mass infiltration, demonstrating signal intensities of fat,

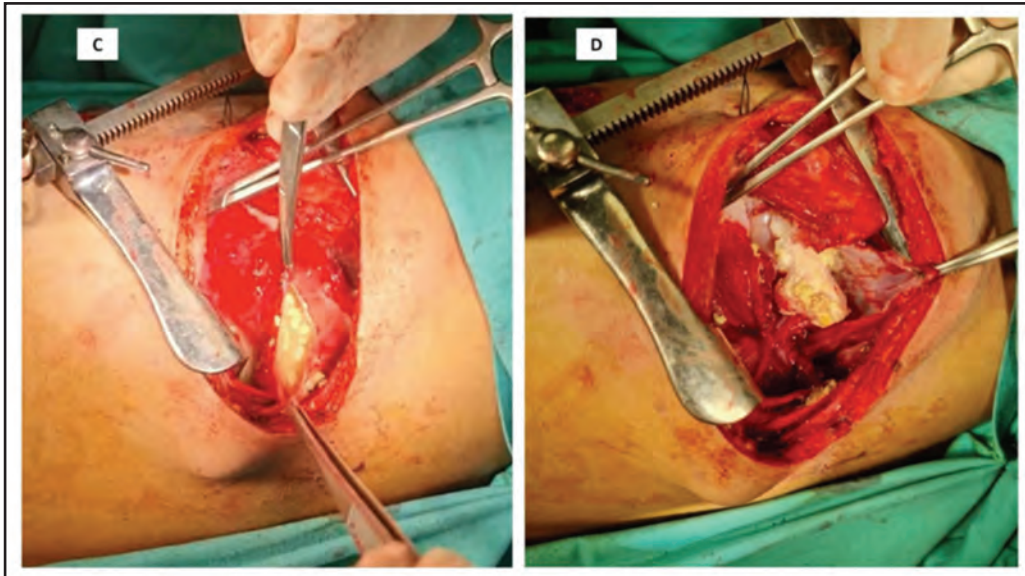


Fig. 3C & D: These figures showed intraoperatively noted purulent loculations with “cheesy” material



Fig. 4: This figure shows the decortication specimen, multilobulated with yellowish cheesy material seen.

fluid, and calcification. CT-guided biopsy is performed to determine the benign or malignant nature of teratomas, especially when there is pericardial involvement which warrants urgent intervention.⁸

The ideal management for benign mediastinal teratoma is always surgery aimed at complete excision of the tumor.⁹ This yields an excellent prognosis, with nearly 100% survival rates. Surgery serves the benefit of both establishing the diagnosis and ensuring long-term cure, thereby reducing the likelihood of recurrence. The preferred surgical technique is median sternotomy due to its accessibility, while lateral thoracotomy is beneficial in cases involving extension into the hemithorax.¹⁰

CONCLUSION

Mediastinal mature teratoma represents a rare and benign disease. Patients are typically asymptomatic, but rarely may present themselves as pleural effusion. Distinctive features on CT scan and MRI aid in diagnosis and assessment of disease extent. Biopsy serves to confirm the diagnosis and exclude any immature elements suggestive of malignancy. In cases where pleural effusion lacks an evident cause, considering teratoma as a potential diagnosis, even in cases of transudative effusion, is essential. Regardless of its presentation, prompt surgical excision offers a curative approach for mature teratoma. As of date this is the first reported case of mature teratoma from pleura alone which manifests as empyema thoracis.

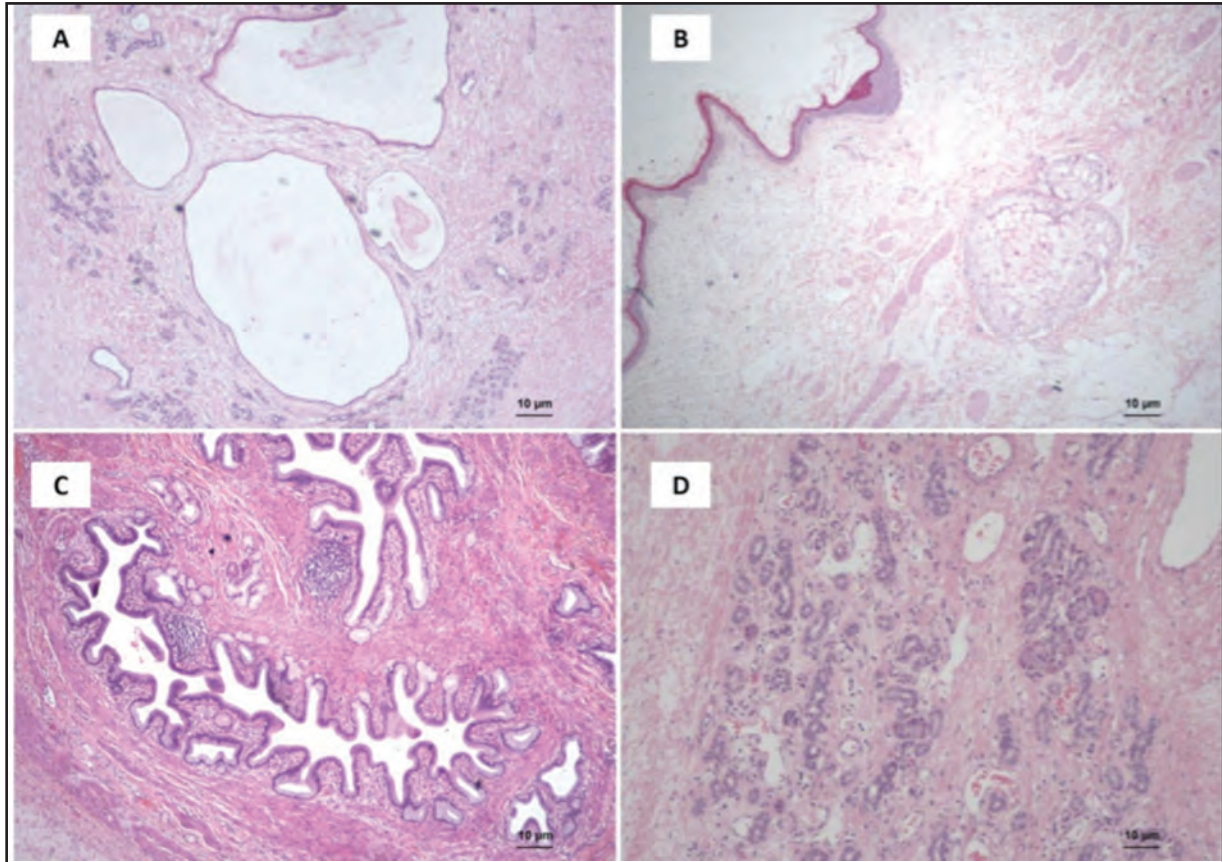


Fig. 5: Hematoxylin and eosin (H and E) stain section of the cortex showed features of mature teratoma: (A) Multiple cystically dilated structures partly lined by squamous epithelium and columnar epithelium. (B) Skin, pilosebaceous unit and adipocytes. (C) Glandular epithelium and smooth muscle bundle (D) Pancreatic ducts and acini

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DECLARATION

The authors have no conflict of interest to disclose.

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