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Case report

Ruptured mediastinal mature teratoma with shigellosis empyema: A case report

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ABSTRACT

Introduction and importance: Mediastinal mature teratomas are often benign and asymptomatic, but ruptured mediastinal mature teratoma is rare and induces severe complications.

Case presentation: A male, 23 year old, complained shorthness of breath, right chest pain, fever, and cough. Radiological examination (X-ray, CT-Scan and MRI) showed mediastinal teratoma and pleural effusion. The patient received supportive therapies, including oxygen, symptomatic therapy, antibiotics, and lateral thoracotomy. Empyema culture was positive for *Shigella dysenteriae*. He was discharged after thirty-six days of hospitalization in good clinical condition.

Clinical discussion: Perforation of mature teratoma is a rare but severe complication. Ruptured mediastinal teratoma can cause extensive pleural adhesions and empyema, making it challenging to perform VATS because of the risk of bleeding and damage to adjacent organs during surgery.

Conclusion: Ruptured mediastinal mature teratoma has a good prognosis post-surgical partial resection despite tumor attachment to the pericardium and heart.

1. Introduction

Teratomas are germ cell tumors originating from pluripotent embryonic cells that have differentiated into tissues with two or more germ cell layers, namely ectodermal, endodermal, and mesodermal [1,2]. In general, these tumors are benign. Mature teratomas consist of fully differentiated tissue from one or more germ cell layers but have an irregular arrangement [3]. Meanwhile, mediastinal teratoma is a rare neoplasm (8–13 % of all mediastinal tumors and only 1–10 % of germ cell tumors occur in the mediastinum) [4]. Mediastinal teratomas result from pluripotent germ cells that fail to migrate from the mediastinum to the gonads during the early developmental phase [5]. The incidence of teratomas is about 1 in 4000 live births [4,6]. Perforation of mature teratoma is a rare but severe complication. This study reported an Indonesian male with ruptured mediastinal mature teratoma based on SCARE 2020 guidelines [7].

2. Presentation of case

A male, 23 years old, complained shorthness of breath, right chest pain, fever, and cough without phlegm for 1 week. He also complained of a low-intake diet, fatigue, vomiting, and weight loss of 2 kg in 2 months. He denied night sweats. The patient previously had no health problems and a history of tumors in his family. The shorthness of breath evaluation showed SpO2 of 92 % without oxygen therapy, whereas when using a nasal cannula of 4 L/min, the SpO2 value increased to 96 %. Thorax physical examination showed a chest pain scale of 8 and an inspection of asymmetries lung development (slower right lung). Meanwhile, palpation of decreased fremitus, percussion of dim, and auscultation of Ronchi in 2/3 lower right lung.

Radiological examination supported mediastinal teratoma, in which a chest X-ray showed the mediastinal tumor in the right lung (Fig. 1). A thorax CT scan showed a mass (size of 8.7 × 6.7 cm) in the right superior mediastinal similarly mediastinal teratoma (Fig. 2). Abnormal laboratory examination included white blood count (WBC) of 32,210/μL, neutrophile of 91.9 %, lymphocytes of 2.9 %, alanine aminotransferase (ALT) of 140 u/L, aspartate aminotransferase (AST) of 56 u/L, C-reactive protein (CRP) of 28.4 mg/dL, and albumin of 2.98 g/dL. Meanwhile, blood gas analysis (BGA) showed respiratory acidosis with moderate hypoxemia (pH of 7.32, pCO2 of 49 mmHg, pO2 of 70 mmHg, HCO_3^- of

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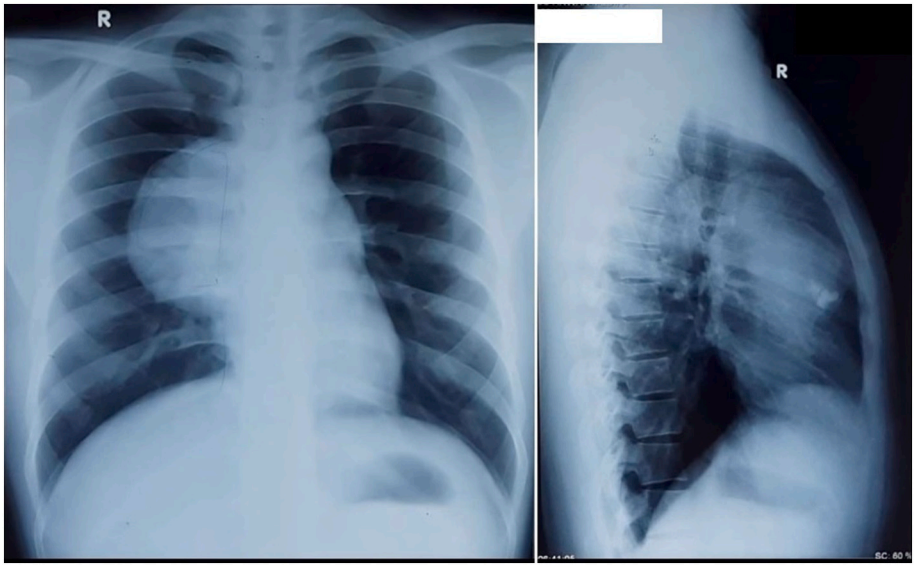

Fig. 1. Chest X-ray showed the mediastinal tumor in the right lung.

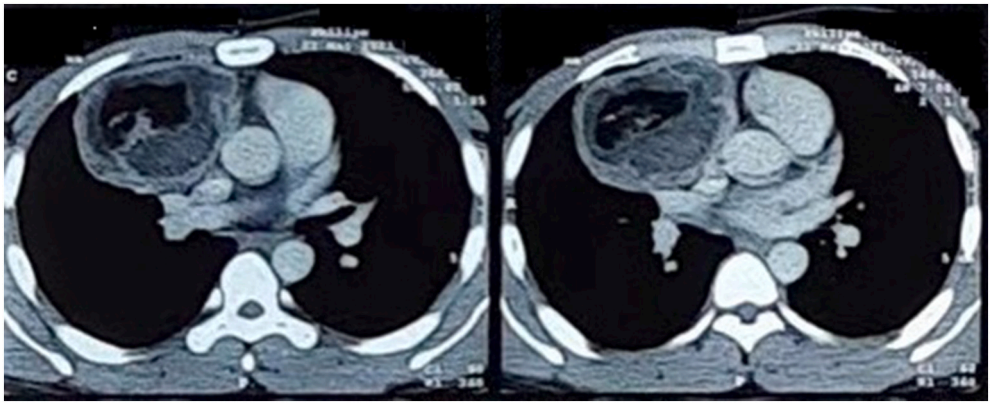

Fig. 2. Thorax CT scan showed a mass in the right mediastinal of 8.7 × 6.7 cm. Characteristic mass included thick-walled cysts with fat, solid, and calcified components.

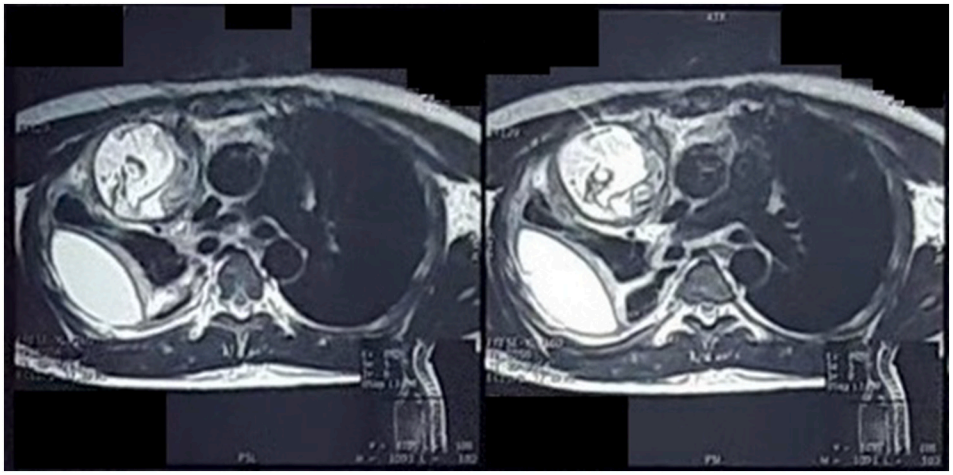

Fig. 3. Thoracic MRI showed a solid mass (5.7 × 7.7 × 8.7 cm) with cysts and fat component, peritumoral bleeding, and Loculated pleural effusion in the right lobe.

25.3 mmol/L, BE of −0.9 mmol/L, and SO2 of 92 %), which he used nasal cannula with 2 L/min doses. The patient received O2 of 6 L/min (simple mask), NaCl 0.9 % of 1000 mL/day, levofloxacin of 750 mg/day for 7 days, codeine of 10 mg/8 h, paracetamol of 500 mg/8 h, lactulose of 15 mL/8 h, curcuma of 20 mg/8 h, and albumin capsule of 500 mg/8 h.

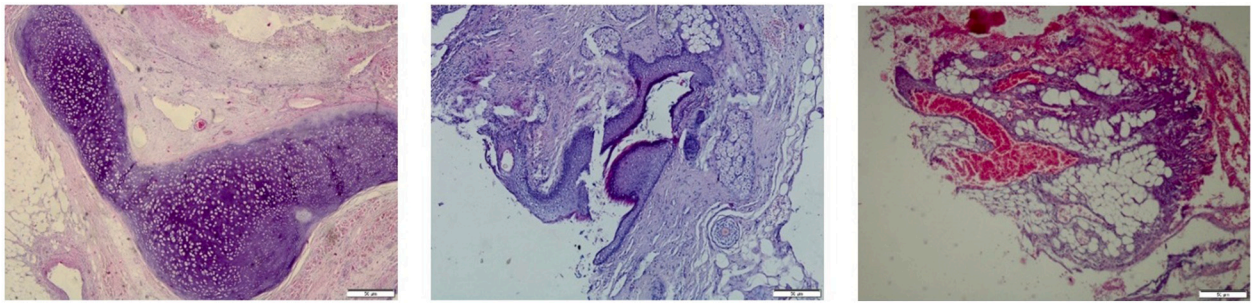

Fig. 4. Tumor tissue consists of squamous epithelium, sebaceous glands, hair follicles, adipose tissue, cartilage tissue, and mucinous glands.

On the 4th day, the patient felt decreased chest pain (pain scale of 6) and SpO2 of 98 % with a simple mask of 6 L/min. Meanwhile, BGA value showed an average with moderate hypoxemia (pH of 7.41, pCO2 of 42 mmHg, pO2 of 79 mmHg, HCO_3^- of 26.6 mmol/L, BE of 2.0 mmol/L, and SO2 of 96 %). The patient had an increase in procalcitonin of 3.57 ng/mL. Chest X-ray and ultrasound showed pleural effusion in the right lung. On the 8th day, the patient was still experiencing increased procalcitonin (3.57 ng/mL). Sputum culture results showed *Streptococcus viridans*. The patient's antibiotic therapy was changed to ceftazidime of 1 g/8 h for 7 days and O2 of 3 L/min (nasal cannula) while the others were continued. On the 21st day, alpha-fetoprotein (AFP) of 0.2 ng/mL, beta human chorionic gonadotropin (β-HCG) of 0.39 mlU/mL and thoracic magnetic resonance imaging (MRI) showed a solid mass with the cystic and fat component, peritumoral bleeding with well-defined margins was also found (Fig. 3).

On the 27th day, the patient underwent mediastinal tumor thoracotomy and found loculated empyema in the right hemithorax and adhesions in all three lobes of the right lung. Mediastinal tumors were found in 2 locations as anteromedial and inferior medial extra lobes. Mediastinal tumors could not be entirely incised because the pericardium and heart border the posterior wall of the tumor. On the 28th day, the fluid pleural analysis showed exudate with a pH of 9, mononuclear (MN) of 10 %, polymorphonuclear (PMN) of 90 %, a cell number of 8.38 × 103/μL, glucose of 63 mg/dL, and protein of 7.1 g/dL. The patient received metamizole of 1 g/8 h, metoclopramide of 10 mg/8 h, and nebulizer (ventolin of 2.5 mg/8 h) after surgery as additional therapy. On the 32 days, empyema culture analysis was *Shigella dysenteriae* sensitive to antibiotic drugs such as cefoperazone-Sulbactam, meropenem, piperacillin-tazobactam, cotrimoxazole, and piperacillin. The antibiotic patient changed to cefoperazone-sulbactam of 1 g/12 h and metoclopramide changed to omeprazole of 40 mg/12 h. On the 33rd day, the histopathology of tumor tissue found no signs of malignancy, concluding that the mediastinal teratoma was mature (Fig. 4).

On the 6th month post-surgical, thoracic MRI showed a residual mass of 3.4 × 3.6 × 3.3 cm, adherent to the vena cava and right atrium with well-defined borders in the right anteromedial mediastinal. The patient has no complaints, is in stable condition and has no problem daily activities. On the 1st years post-surgical, the patient had no complaints and thoracic MRI showed size and location teratoma of stable (no metastasis), similar to 6 months ago.

3. Discussion

The radiological manifestations of ruptured teratoma on CT examination depend on the space in which the rupture occurred. On CT scan, ruptured teratomas show inhomogeneous density in each internal compartment, possibly related to mixing extravasated components between compartments. Other characteristics of ruptured teratoma include fatty lumps at the perforation site, consolidation or atelectasis in the lung near the lesion, and pleural effusion [8]. Pleural effusion may be the most common CT finding in ruptured mediastinal teratoma. Pericardial effusion, especially in patients with teratoma adhering to the pericardium, suggests tumor rupture into the pericardium [9].

MRI is an adjunct to CT scanning in diagnosing mediastinal lesions [10]. MRI is useful for further evaluating mass infiltration. MRI can demonstrate heterogeneous signal intensity containing a mixture of fat, water, soft tissue, and calcifications [8]. MRI is dominant over CT scan in evaluating spread through the tumor capsule and infiltration of adjacent structures with obliteration of fat tissue. MRI is superior to CT in differentiating cystic lesions from solid lesions. Thoracic MRI examination provides a more detailed and often definitive evaluation of mediastinal masses than CT because it is more detailed in providing information about the characteristics of tumor tissue. Macroscopic fat tissue can be determined on CT scans and MRIs, but microscopic intracellular fat is only detected on MRI [10,11].

Examination of tumor markers is one of the modalities that can differentiate between benign and malignant teratoma. In patients with benign mature mediastinal teratoma, serum levels of AFP and β-HCG are within the normal range [5,12]. Benign teratomas (pure mature teratomas) do not secrete AFP and β-HCG, so there is no increase in AFP and β-HCG levels in benign mature teratomas. Increased AFP and β-HCG may indicate malignancy [5,13]. Serum levels of AFP, β-HCG, and LDH are well validated for noninvasive diagnostics of Germ Cell Tumors. Significant elevations in serum AFP or β-HCG, each suggesting a significant component of a yolk sac tumor or choriocarcinoma, exclude the diagnosis of a pure mature teratoma or seminoma. During the diagnosis and follow-up of mediastinal teratoma, it is necessary to monitor serum AFP and β-HCG levels [5,14].

Tumor perforation can present with various symptoms depending on the location of the tumor [2]. Tumors can perforate and enter the pleural cavity, pericardium, lung parenchyma, tracheobronchial tract and extraordinary vessels, causing life-threatening complications [15]. Pleural effusion is the most common finding on CT scans of ruptured mediastinal teratoma. A pericardial effusion suggests a rupture of the tumor into the pericardium in a patient with a teratoma adhering to the pericardium [8,9]. The tumor may rupture and invade the tracheobronchial tree, causing hemoptysis and trichoptosis. This condition results in severe complications [2,8]. Ruptured mediastinal teratoma can cause extensive pleural adhesions and empyema, making it challenging to perform VATS because of the risk of bleeding and damage to adjacent organs during surgery [16].

Shigella bacteremia is a rare condition. The prevalence is estimated at 0.4–7.3 % of Shigella infections in the adult population, and the mortality rate is 21 %. The risk factors for shigella bacteremia are patients with immunodeficiency, diabetes, leukemia, sickle cell anemia, malignancy, HIV, cirrhosis, changes in intestinal integrity and transplantation. Shigella bacteremia is more common in the pediatric population, with a prevalence of 5–12 % of all infections and a mortality rate of 46 %, especially in malnourished children in countries with poor socioeconomic conditions [17]. Shigella can cause bacteremia without gastrointestinal symptoms such as diarrhea, nausea, vomiting and abdominal pain [9]. Shigella replicates in the cytoplasm of phagocytic cells and moves from cell to cell without being exposed to the extracellular environment and thus is protected from immune-mediated

clearance. Shigella can induce programmed cell death of macrophages and induce the release of IL-1β, thereby stimulating a local inflammatory response [18].

Complete surgical resection is the primary modality for treating mediastinal teratoma [5,8]. Patients with benign mediastinal teratoma have a good prognosis after surgical resection. Complete surgical excision is the treatment of choice to prevent complications such as compression of adjacent structures, rupture, or malignant transformation [5,11]. The most common surgical excision method is median sternotomy because of its excellent exposure. However, if the tumor affects organs in the hemithorax, the surgical method of choice is lateral thoracotomy [8,11].

Complete surgical resection is sometimes not possible due to the large size of the tumor and severe adhesions due to inflammation in the surrounding tissue [1,5]. Complete surgical resection of a moderate-sized tumor, round mass with well-defined walls, and no invasion of adjacent structures yielded promising results without complications and recurrence. In contrast, extensive tumors, filling almost the entire chest cavity and adhering to surrounding organs, pose a risk of damage to surrounding organs [2,5].

4. Conclusion

A male, 23 years old, complains of short breathlessness, right chest pain, fever, and cough. He is diagnosed with mediastinal teratoma based on a chest X-ray confirmed, thorax CT scan, thoracic MRI and histopathological. However, mature teratoma is confirmed based on histopathological and thoracotomy. Shigellosis empyema is a rare complication of ruptured mediastinal teratoma. Mediastinal mature teratoma resection cannot be wholly excised because of the attachment between the tumor, pericardium, and heart.

Consent

Written informed consent was obtained from the patient or/and guardian for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Ethical approval

Not applicable.

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Author contribution

All authors contributed toward data analysis, drafting and revising the paper, gave final approval of the version to be published and agree to be accountable for all aspects of the work.

Guarantor

Farah Fatma Wati is the person in charge of the publication of our manuscript.

Research registration

N/A.

Declaration of competing interest

All authors declare no conflict of interest.

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