CASE REPORT

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Rare case of intrathoracic mature cystic teratoma in rural area children: a case report

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ABSTRACT

Introduction: Teratoma is a tumor that often occurs in the gonad area, but can also occur in rare cases in the extragonadal area, such as in the intrathorax. In this paper, we present the experience in diagnosing and managing a case of teratoma in a child, which was first mistaken for an abscess. This study aims to report one of the rare cases of mature cystic teratoma in a rural area in Indonesia and discuss its clinical presentation, modality examination, therapy, and outcome, thereby adding insight into providing an accurate diagnosis of this condition.

Case description: A 13-year-old boy with a chief complaint of shortness of breath that had been felt for 1 year, accompanied by pain in the upper right abdomen, weight loss, and fatigue. The right lung vesicular base sound was decreased. Chest x-ray and computed tomography were performed with suspicion of an abscess. An exploratory thoracotomy was performed. A cystic mass containing hair and pus was found. The mass was attached to the pleura and completely evacuated by dissection. Histopathology examination of the mass specimen confirmed a mature cystic teratoma. The patient was discharged only with complaints of mild pain. The patient had no more symptoms during the post-operative follow-up.

Conclusion: Intrathoracic mature teratoma is a rare tumor that may mimic more common conditions, making timely and accurate diagnosis difficult. Surgical resection remains the treatment of choice, and clinicians in tuberculosis-endemic regions should maintain high suspicion to avoid misdiagnosis and improve outcomes.

Keywords: intrathoracal teratoma, intrathoracal tumor, mature cystic teratoma, mature teratoma.
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INTRODUCTION

Mature teratomas are commonly found in young adults in the second to fourth decades of life, but they can also be found in children.[1] About 50% of children and 66% of adults typically have asymptomatic mature mediastinum teratomas. Usually, they are discovered by chance on a chest X-ray.[2] Germ cell tumor (GCT) of the teratoma type are produced from primitive germ cells that do not fully migrate during the early stages of embryonic development. GCT in children is an uncommon condition that accounts for just 1% to 3% of all pediatric tumors. Twenty percent of pediatric GCT are malignant, but the remainder are benign.[3] Benign teratomas, seminomas, and embryonal tumors (malignant teratoma or nonseminomatous GCT) are the three categories of GCT based on the kind of cell. Benign GCT consists of mature teratoma and mature teratoma with an immature component of <50%.[4]

Teratomas in rural communities are often associated with witchcraft, miscarriages, and immoral acts due to the presence of a lump resembling a fetus or accompanied by hair and teeth. Well-differentiated descendants from at least two of the three germ cell layers—the ectoderm, mesoderm, and endoderm—make up mature teratomas. Ectodermal elements can be represented by skin, teeth, and hair. Mesodermal elements can be represented by bone, cartilage, and muscle. Meanwhile, endodermal elements can be represented by pancreatic tissue, gastrointestinal epithelium, and bronchial epithelium.[4]

Teratomas occur more frequently in the gonads (testes and ovaries) but can also occur in extragonadal locations (15%), including intracranial, cervical, mediastinal, retroperitoneal, and sacrococcygeal.[5] There are some reports of teratoma occurring in the hemithorax.[6–11] This case report describes a rare intrathoracic mature cystic teratoma in a child from a rural area in Indonesia, highlighting its clinical presentation, diagnostic challenges, management, and outcome. It is crucial in the context of rural healthcare, where limited access to advanced diagnostic tools often leads to misdiagnosis, highlighting the necessity of increased clinical knowledge of uncommon thoracic tumors, including mature cystic teratoma.

CASE DESCRIPTION

A 13-year-old boy came to the thoracic and cardiovascular surgery polyclinic with a chief complaint of shortness of breath that had been felt for about 1 year. Complaints were accompanied by pain in the upper right abdomen, weight loss, and fatigue. Previously, the patient was referred from another hospital with suspicion of right pleural effusion. The patient had no history of pulmonary tuberculosis medication, history of smoking, or history of other pulmonary disease. The patient

comes from a financially disadvantaged family. There is no history of tuberculosis medication in the family but the father and the brother of the patient were smoker. From physical examination, it was found that the patient is underweight with a BMI of 14.3 kg/m^2, a weight of 32 kg, and a height of 151 cm. The right lung vesicular base sound decreased. Lymphadenopathy was not found. From the chest X-ray, an opaque shadow with a sharp border was found in the middle to lower right lung field with suspicion of abscess or mass, accompanied by scoliosis thoracalis vertebrae (Figure 1).

Furthermore, an intravenous contrast-enhanced computed tomography (CT) scan was performed. A round, thin-walled hyperdense lesion in the superior segment, laterobasal segment to posterobasal segment of the inferior lobe of the right lung, with a hypodense lesion of air density appearing. It was found accompanied by right pleural effusion (Figure 2). Pre-operative blood test showed anaemia (Hb 9.6 g/dL) and thrombocytosis (472 × 10^3/μL).

Therefore, with suspicion of lung abscess, an exploratory thoracotomy was performed by right posterolateral thoracotomy through the fifth intercostal space. Pleural effusion and a pinkish, well-circumscribed, cystic mass measuring 8cm x 8cm x 7 cm were found in the right hemithorax (Figure 3). The middle and lower lobes had been squeezed by the solid, rigid, immovable mass that was connected to the parietal pleura. After being opened up, the cystic mass, which contained hairs and a thick, yellowish fluid, was aspirated. Following the cystic mass's decompression, the mass was completely removed using both blunt and sharp dissection. During surgery, the compressed lung was fully re-expanded. A thoracic drainage tube measuring 24 Fr was inserted. The procedure went smoothly. The mass's tissue samples underwent further analysis. Histopathological analysis revealed that the cyst had normal nuclei and was bordered with stratified squamous epithelium (Figure 4). The lump was confirmed to be a mature cystic teratoma by the observation of lymphocytes, bone, hair follicles, and sebaceous glands infiltrating the subepithelial fibrocollagen connective tissue. From the analysis of the pleural effusion, the Rivalta test was negative, and the acid-fast bacilli test was negative.

The patient was treated together with the paediatric department and given administration of antibiotics (IV Metronidazole and IV Ceftriaxone), and anti-tuberculosis therapy (Rifampicin, Isoniazid, Pyrazinamide, and Ethambutol) based on a clinical diagnosis of pulmonary tuberculosis despite of negative result of the microbiology test. Post-operative day 1 chest x-rays showed good lung expansion (Figure 5). The patient was discharged on post-operative day 3 with complaints of mild pain. The patient had no more symptoms during the post-operative day 10 follow-up in the thoracic and cardiovascular surgery polyclinic. For treatment of the scoliosis, the patient was planned for physiotherapy and referred to the neurosurgery department.

DISCUSSION

The exact cause of teratomas remains unknown. The theory suggests that teratomas are caused by abnormal differentiation of fetal germ cells originating from the yolk sac. Regular migration of these primordial germ cells leads to tumors in the gonads, while abnormal migration leads to extragonadal tumors.[12] This tumor is an embryonal neoplasm, which arises from the three layers of the blastodermis and develops into a solid or cystic mass with a variety of tissue pictures when totipotent germinativum cells remain unchecked throughout embryological development. According to earlier epidemiological research, the majority of these teratomas occur during the second and fourth decades of life.[13] The patient in this case is in that range.

Mature teratomas are asymptomatic in 50% of children and 60% of adults.[5] Benign teratomas are often discovered on chest radiographs obtained for unrelated reasons.[14] These tumors have a slow growth rate, thus remain asymptomatic until they

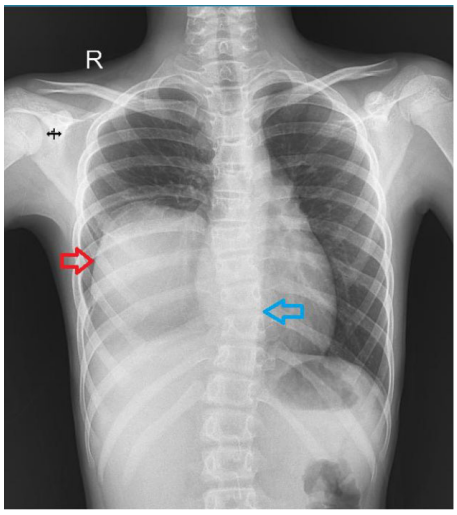

Figure 1. Pre-operative chest x-ray showed an opaque shadow with a sharp border in the middle to lower right lung field with suspicion of abscess or mass (red arrow), accompanied by scoliosis thoracalis vertebrae (blue arrow).

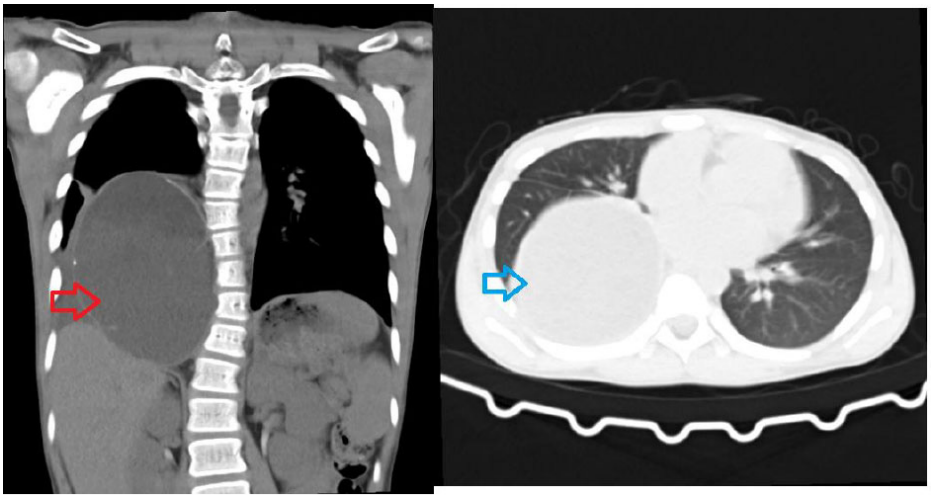

Figure 2. Pre-operative computed tomography scan: coronal view of the mass without contrast-enhanced (red arrow) and axial view of the mass with contrast-enhanced (blue arrow).

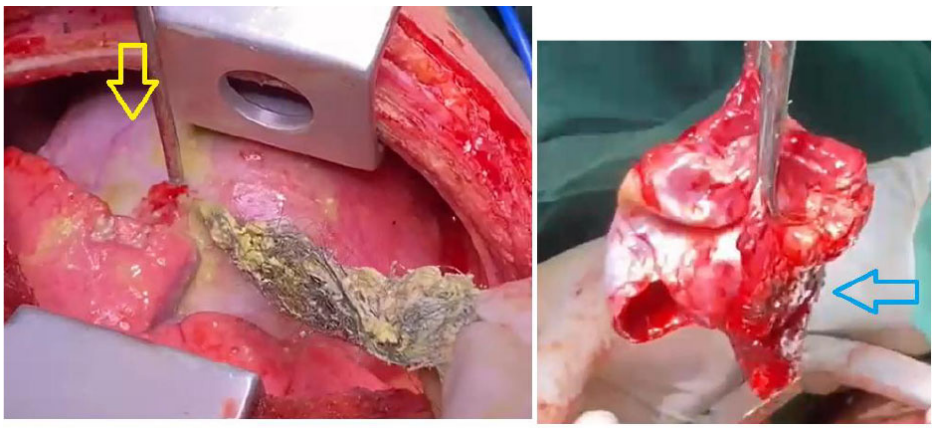

Figure 3. Intra-operative pictures of the cystic teratoma: Prior to dissection, the cystic mass was opened up then hairs and yellowish thick fluid was aspirated (yellow arrow). The mass was evacuated entirely after decompression of the cystic mass (blue arrow).

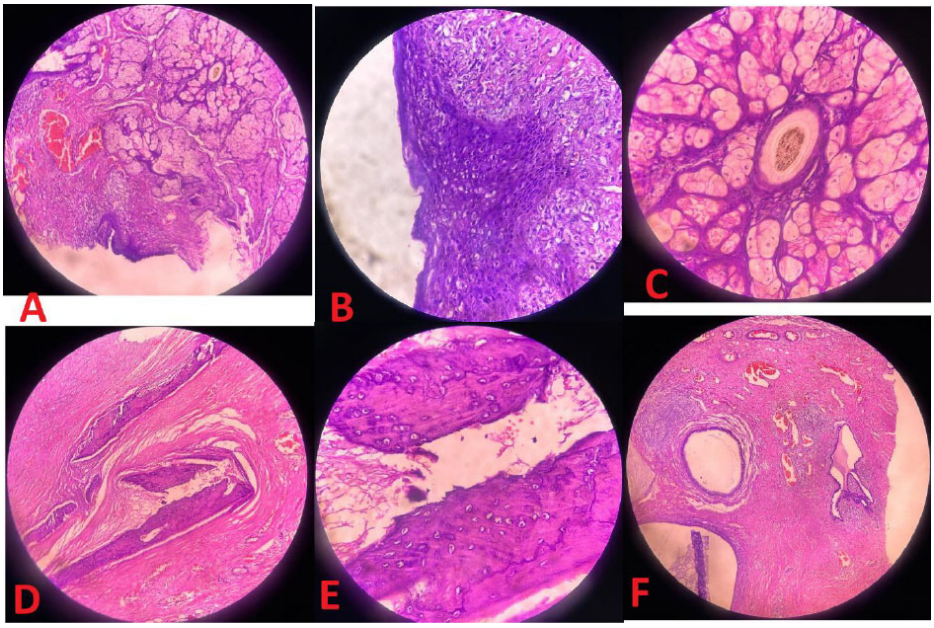

Figure 4. Histopathological analysis. A. ectodermal elements, B. stratified squamous epithelium, C. hair follicles and sebaceous glands, D. mesodermal elements, E. bone, and F. gland tissue.

reach a large size, causing vital organ compression and obstruction. The tumor's mass effect and persistent pneumonia are the causes of the associated symptoms, which include coughing, shortness of breath, or discomfort in the chest, back, and shoulders. Children and newborns experience respiratory discomfort more frequently than adults do, typically due to the size of the lesions that occupy space.[2] As a result of the tumor and airways connecting, patients may seldom experience expectorate hair (trichoptysis), which is regarded as a pathognomonic sign.[4] Our patient presented with shortness of breath and pain in the right upper abdomen. Other studies reported patients with spinal deformity or scoliosis due to the mass compression effect, which also happened to our patient.[15,16]

Diagnostic approach in teratoma is by physical examination of mass enlargement, laboratory findings, radiologic imaging, and histopathology anatomy examination. One of the laboratory techniques that can distinguish between benign and malignant teratomas is the analysis of tumor markers. Serum Alpha-Fetoprotein (AFP) and beta-human chorionic gonadotropin (β-HCG) values are within acceptable limits in patients with benign mature teratoma.[16,17] AFP and β-HCG are not secreted by benign teratomas, also known as pure mature teratomas. Therefore, benign adult teratomas do not exhibit elevated levels of AFP and β-HCG. Elevated β-HCG and AFP might be a sign of cancer.[16] For non-invasive GCT diagnostics, serum levels of AFP, β-HCG, and Lactate Dehydrogenase (LDH) have been well verified. A pure mature teratoma or seminoma cannot be diagnosed when there are considerable increases in blood AFP or β-HCG, each of which indicates a significant component of a yolk sac tumor or choriocarcinoma.[17] In our case, we do not examine the AFP nor β-HCG levels due to the unavailability of the test in our rural area and we had to refer it to a bigger centre with a long time for the results to come out and high cost.

A well-defined tumor with calcification may be seen on a chest x-ray in 26% of instances with adult teratoma.[18] Although they are uncommon on X-rays, well-formed teeth or bone strongly imply the diagnosis. The preferred imaging technique for suspected GCT is computed tomography (CT) with intravenous contrast since it can precisely identify the mass and describe its various densities. The link with the surrounding structures can also be shown on a CT scan. Benign teratomas often have sharp edges and a rounded shape. Usually cystic, lesions frequently include localized calcification. When the tumor is located in the thoracic inlet or thoracoabdominal region, the MRI is more useful in detecting invasion of adjacent tissues.[18] Heterogeneous signal intensity including a mixture of soft tissue, calcifications, fat, and water can be seen on MRI. When assessing dissemination through the tumor capsule and infiltration of nearby tissues with obliteration of adipose tissue, MRI is superior than CT scan. When it comes to distinguishing solid lesions from cystic lesions, MRI is better than CT. 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.[17] MRI is not accessible in our rural area, thus we did a contrast-enhanced CT scan

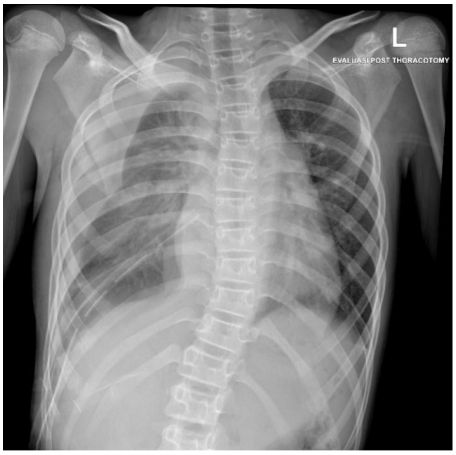

Figure 5. Post-operative chest x-ray showed complete excision of the mass and expansion of the right lung.

in our case, and showed a thin-walled hyperdense lesion with a hypodense lesion of air density appearing. It is with suspicion of an intrathoracic/pulmonary abscess.

The tumor's macroscopic size falls between 2.8 and 30 cm. Although it can be a solid, encapsulated tumor, it is typically multilobulated and cystic. The tumor in this instance measured 8x8x7 cm. The diagnosis of teratoma must be confirmed by histopathological analysis. Histopathology is characterized by the presence of mature tissue components from the three germ layers, skin and appendageal structures, respiratory epithelium, lobules of mature adipose tissue, islands of hyaline cartilage, and salivary gland tissue.[6] Sebaceous substances and mucus-secreting columnar epithelial cells often line the cyst. The cyst wall contains muscle, skin glands, hair follicles, and other tissues. An immature mediastinal teratoma histologically contains immature ectodermal, mesodermal, and ectodermal elements.

Since adult teratomas are often firmly encapsulated, surgical excision is the conventional therapy for them. Mature teratomas have a fairly excellent prognosis after surgical removal. In a research including 45 cases of adult teratomas, every patient who underwent surgical removal was cured.[18] Survival and resectability can be limited by the size of the tumor and its invasion of adjacent structures. A moderate-sized tumor with spherical, well-defined walls and no invasion of nearby tissues was completely surgically removed, and the outcomes showed promise in terms of no problems or recurrence. On the other hand, large tumors that enlarge almost the whole chest cavity and adhere to nearby organs run the danger of causing harm to such organs.[17] Recurrence is uncommon and mostly occurs when the tumor is not completely removed. Verifying that histopathology does not include any immature components is crucial. Chemotherapy is necessary for any immature component.[18]

Due to the high incidence of tuberculosis in Indonesia, which is 387 per 100,000 population, tuberculosis abscess was the first to be included in the differential diagnosis.[19] It may be challenging to differentiate radiologically a mediastinal abscess from GCT. Due to the clinical symptoms of the patient, such as short of breath, fatigue, and weight loss that mimicked the clinical symptoms of tuberculosis infection, the paediatrician decided to observe the symptoms while given the patient anti-tuberculosis medication. The patient received a tuberculosis regimen, consisting of Rifampicin, Isoniazid, Pyrazinamide, and Ethambutol, on the treatment day in the hospital until post-operative day 10, and plans to evaluate the patient in the pediatric polyclinic.

This case report has several limitations. First, the histopathology specimens were not adequately documented, and pathology images could not be included because they were referred to an external laboratory, limiting the visual support for the diagnosis. Second, tumor marker levels such as AFP and β-HCG were not assessed due to resource constraints; these markers could have strengthened the diagnostic confirmation and differentiation between benign and malignant germ cell tumors. Third, this is a single case report, which inherently limits the generalizability of the findings to broader clinical practice.

CONCLUSION

Intrathoracic mature teratoma is a rare tumor that often mimics other conditions, making accurate diagnosis challenging. A rigorous diagnosis strategy including clinical examination, laboratory tests, imaging, and histological confirmation is necessary. With a great prognosis, surgical resection is still the preferred course of therapy. Clinicians, especially in tuberculosis-endemic regions, should maintain a high index of suspicion when encountering atypical respiratory cases to prevent misdiagnosis and ensure timely surgical management.

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DISCLOSURES

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Conflict of Interest

The authors do not have a conflict of interest to declare.

Author Contribution

The manuscript was conceptualized, designed, and overseen by MRF, RM, and HAA. The study is carried out by RM and HAA. The data is interpreted by MRF, RM, and HAA. The final draft of the paper is prepared by all authors, who consent to its submission to this journal.

Ethical Consideration

The patient consented to the publication of this case for research purposes or in a journal.

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