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A giant intrapulmonary malignant teratoma - a rare case presentation

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Abstract

Germ cell tumors such as teratomas are unusual and most commonly occur in the gonads. Extra-gonadal teratomas are rare, and the thorax, especially the mediastinum, is the commonly involved extra-gonadal site. Teratoma of the intrapulmonary region is rare and often involves the left upper lobe of the lung. In this report, we describe a case of a 20-year-old male diagnosed with an intrapulmonary malignant teratoma arising from the left lower lobe. Teratomas emerging from the lungs present with vague symptoms like breathing difficulty, cough, and hemoptysis. We report this rare appearance of malignant intrapulmonary teratoma that originated from the left lower zone of the lung.

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Introduction

Teratomas are rare benign germ cell tumors commonly seen in ovaries and testicles, and the thorax is the most common extra-gonadal area of involvement.^{1,2} Mediastinum is the most common site of involvement, but in rare scenarios, intrathoracic teratomas can occur within the lung parenchyma.³ Intrapulmonary Teratoma (IPT) is thought to originate from the third endodermal pouch of the pharynx, an embryonic origin of the thymus, and only a few cases have been reported till now.^{3,4} Indistinct symptoms like chest tightness, cough, and hemoptysis frequently accompany IPT and have a tendency to affect the upper lobe of the lung for reasons that remain unclear.^{1,4} The age of an individual, the presence of immature tissues, and the time of presentation determine the outcome.⁵

Case Report

A 20-year-old male presented to a local hospital complaining of left-sided chest pain and dry cough for 2 years, where he was diagnosed with tubercular pleural effusion based on endemicity, positive Mantoux test, and blunting of left CP angle on chest x-ray (Figure 1A); anti-tubercular chemotherapy (2 months of HRZE and 4 months of HRE) was administered. He had no associated symptoms of hemoptysis, dyspnea, or trichoptysis. Due to poor response to multiple antibiotics, he was referred to our institute for further management.

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On admission, chest x-ray (Figure 1B) showed large homogenous opacity in the left hemithorax with ICD *in situ*. Clinical examination revealed stable vitals, with no lymphadenopathy or testicular mass. On auscultation, breath sounds on the left side were decreased. ICD was removed, confirming its location. Subsequently, CT thorax (Figure 2) delineated a large mixed solid and cystic density lesion occupying 2/3rd of the left hemithorax with small calcified foci. Blood investigations revealed a normal hemogram, and normal abdominal sonography. Tumor markers revealed raised β -human chorionic gonadotropin (20.81 mIU/ml) and LDH (924U/L) while α -fetoprotein was within normal limits, with raised suspicion of malignant germ cell tumor.

To assess the involvement of mediastinum, a whole-body PET-CT scan was done, which showed an FDG avid large mass (SUVmax-3.15) involving 2/3rd of left hemithorax, and FDG avid lymph node in the superior mediastinum, bilateral upper paratracheal region and right supraclavicular lymph node indicating metastasis. As the patient was not completely fit for surgery, an ultrasound-guided transthoracic biopsy of the mass was done, which revealed viable necrotic tumor cells, medium-sized oval and elongated with hyperchromatic nuclei, rosette-like structures were identified, confirming the diagnosis of malignant teratoma (Figure 3).

Surgical resection was deemed unsuitable in this patient due to metastatic spread, as evidenced by FDG avid contralateral upper paratracheal and right supraclavicular lymphadenopathy. He

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was referred to a medical oncologist for initiation of neoadjuvant chemotherapy and further assessment for surgery.

Discussion

Most commonly, teratomas are benign in nature and are composed of tissues originating from pluripotent stem cells that comprise at least two of the three embryonic germ layers: endoderm, mesoderm, and ectoderm.⁶ In 1839, Mohr reported the first case of pulmonary teratoma. The incidence of germ cell tumors increases with age, with a slight female preponderance seen between the second and fourth decade,⁷ while IPTs have equal sex distribution and are predominantly diagnosed between the first and second decade of life.³ The most common site of origin is gonads, followed by anterior mediastinum. Though a majority of intrathoracic teratomas arise from within the mediastinum, they can also arise rarely from an intrapulmonary source and not directly involve the mediastinum.⁸

The formation of an intrapulmonary teratoma is believed to occur when primordial thymic cells migrate along the lung's development. The presence of thymic tissue supports this theory of teratomas seen after surgical resection.⁹ IPTs are usually benign, typically presenting in childhood, and are resectable. However, 30% of them are immature and have a high malignant potential.^{4,10}

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IPTs are manifested with a variety of symptoms: fever, cough, chest pain, hemoptysis, dyspnea, and trichoptysis.⁸ Upper lobes were the most commonly involved sites found radiologically.⁴ Most IPTs are manifested as non-homogeneous, usually well-defined opacity on CXR. A ruptured IPT with bronchial communication on CT appears as a heterogeneous cystic mass containing soft tissue elements (100%), fat (88%), fluid (76%), and punctate calcifications (53%).^{3,4,10,11}

Age and the presence of immature tissue are considered the outcome predictors. Around 20-30% of the teratomas are immature and have a poor prognosis and an aggressive course. A mature teratoma is called a teratocarcinoma or malignant teratoma when a focus of carcinoma or malignant germ cells is seen in the tumor.¹² All cases of suspected teratomas should be surgically operated as teratocarcinomas are aggressive and can be fatal due to local spread or distant metastasis. Chemotherapy may improve the survival in few cases of metastatic teratocarcinoma.^{5,13} Neoadjuvant chemotherapy preceding surgical resection is recommended in patients with Non-Seminomatous Germ Cell Tumour (NSGCTs). Studies recommend using a cisplatin-based chemotherapy regimen for NSGCT as patients have demonstrated a better outcome.¹⁴

In this instance, the prolonged delay in diagnosis extending over approximately two years and the initial misdiagnosis as tubercular pleural effusion, given India's endemic status for tuberculosis, are ascribed to the advancement of the disease. Subsequently, it was diagnosed as

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malignant teratoma and referred to a medical oncologist for further management. Unfortunately, due to unfavorable conditions, the patient was lost to follow-up and succumbed due to the disease.

Conclusions

IPT should be diagnosed and operated on in the early stages as they have a high propensity for malignant transformation. One should always be cautious as rare cases might also present with trivial symptoms, and delay in diagnosis must be avoided. Early diagnosis and proper surgical care are essential for long-term, disease-free survival.

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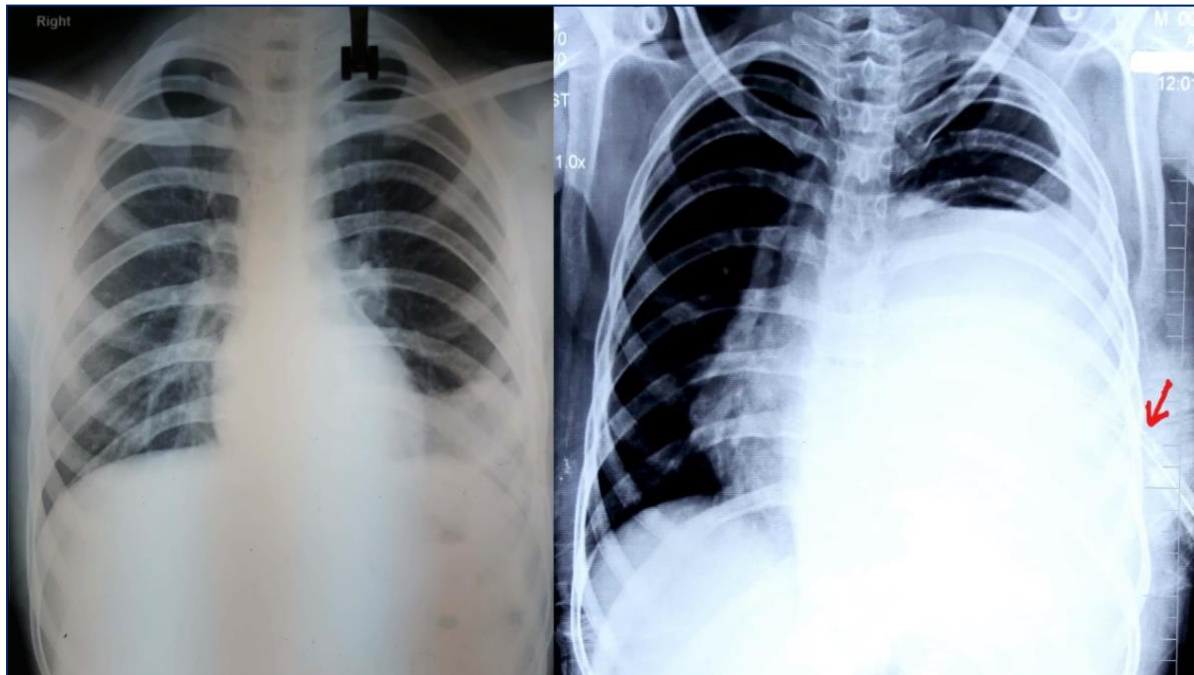


Figure 1. A) Initial chest radiograph showing opacity in left lower lobe; B) chest radiograph showing a large homogenous opacity in the left hemithorax with ICD *in situ* (red arrow).

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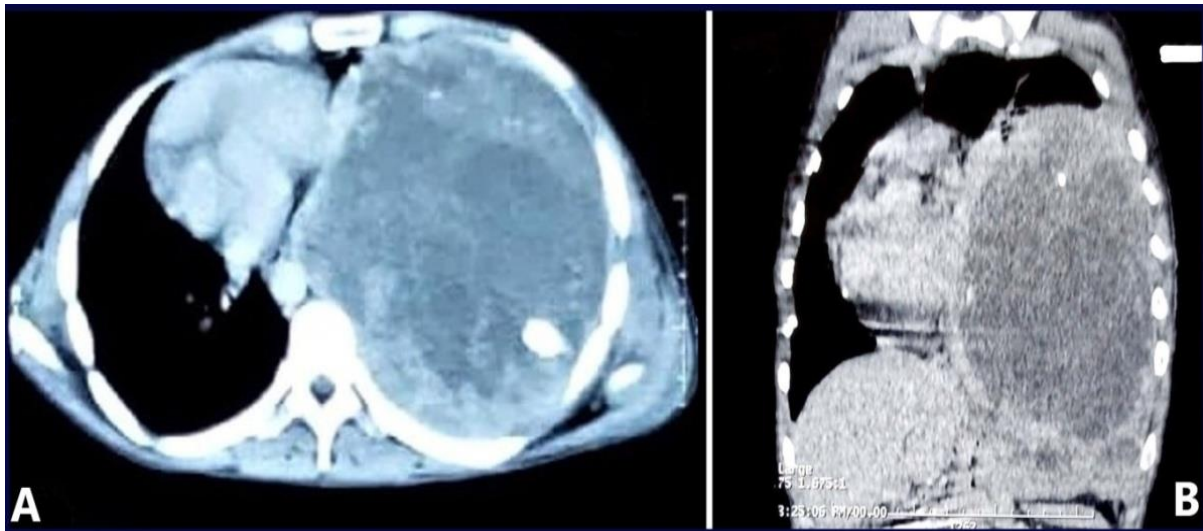


Figure 2. CT Thorax (A, axial view & B, coronal view) showing large mixed solid cystic mass lesion with small calcified foci occupying 2/3rd of left hemithorax with displaced mediastinum to right side.

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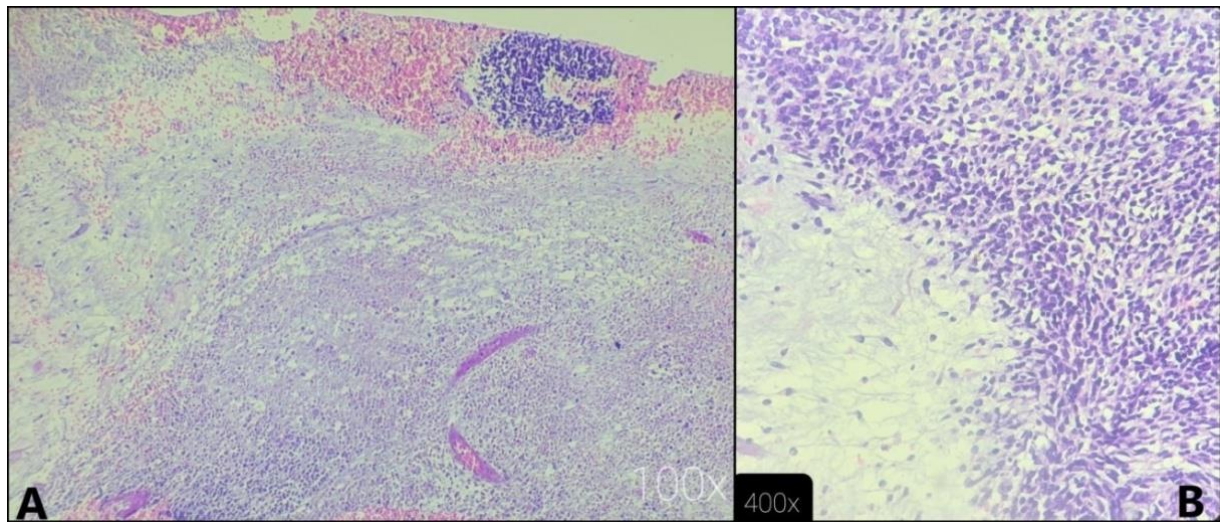


Figure 3. HPE (A-100X & B-400X) showing medium-sized oval and elongated tumor cells with hyperchromatic nuclei & rosette-like structures consistent with malignant teratoma.

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