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Case report

Right upper lobe intrapulmonary mature cystic teratoma. An unusual location with unusual associations and a review of the literature

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ABSTRACT

Introduction: Teratomas are rare neoplasms made up of mature embryonic germ cell layers. Extra-gonadal teratomas are rare and primarily in the mediastinum. Mature cystic teratomas (MCT) are slow-growing benign tumors accounting for a small number of mediastinal germ cell tumors. Patients with MCT are often asymptomatic, and even most symptomatic intrapulmonary teratomas present with vague and non-specific symptoms such as cough, hemoptysis, and chest pain.

Case presentation and discussion: A Yemeni patient presented for two years with dry cough, shortness of breath, and stitching right-sided chest pain. Plain X-ray showed pleural thickening associated with two opacities in the right lung- one located centrally in the upper lobe and another peripherally- which were confirmed with a CT scan. During right-sided decortication with resection of the two masses, extensive adhesions were found between the right pleura and the chest wall. The adhesions were released, and right-sided decortication led to the removal of the peripheral cystic mass attached to the pleura. Histopathology reported the presence of squamous epithelium lining with ectodermal components such as hair follicles, sebaceous glands, keratin debris, fatty, and fibromuscular tissue.

Conclusion: Intrapulmonary teratomas are rare tumors, with less than 100 cases reported in the past few years. All the reported cases are in the left lung lobes, yet our case is on the right side. Although the right-sided mature cystic teratoma is even scarcer, physicians should have a high index of suspicion for teratoma when encountering abnormal lesion in the right side of the lung confirming it either by CT or MRI.

1. Introduction

Teratomas are rare germ cell tumors composed of well-differentiated tissues originating from 2 or 3 germ cell layers such as hair, sebaceous glands, or teeth [1]. They are common neoplasms of the gonads. Mediastinal mature cystic teratomas (MCT) are relatively scarcer and account for approximately 10%–15% of mediastinal masses [2]. The very few cases of intrapulmonary teratomas (IPT) that exist have been reported to occur within the lung parenchyma. Intrapulmonary teratomas have a predilection for the left upper lung lobe [3]. Patients with IPT usually present with vague and nonspecific symptoms such as chest pain, persistent cough, and/or hemoptysis, making the diagnosis indistinguishable from other causes [4]. A rare sign that has been reported and owed to bronchial communication of the teratoma is called trichoptysis (expectoration of hair) and is found in 13% of all IPTs [5]. Chest X-rays are the initial imaging modality in diagnosing mediastinal teratomas. At the same time, computed tomography (CT) scan is the next step in evaluation for specifying the nature, location, and relationship of the tumor to the other structures. Recently MRIs have shown a superior diagnostic power over Conventional CT scans, especially in identifying spread through the tumor capsule and to adjacent structures [6].

2. Case presentation

A 34-year-old male Yemeni patient presented for two years with fever, sweating, dry cough, shortness of breath, and stitching right-sided

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chest pain. Plain X-ray showed pleural thickening associated with two opacities in the right lung. The first was located centrally in the right lung's upper lobe and the other peripherally. CT scan revealed a right-sided central irregular parenchymal mass attached to the thickened pleura with another lesion in the upper lobe of the right lung. Differential diagnoses included thymoma, thymic carcinoma, or lymphomas, and a plan for right sided decortication with resection of the two masses was undertaken (Fig. 1).

Intra-operatively, a right thoracotomy was done, yet extensive adhesions were found between the right pleura and the chest wall. The adhesions were released, and a right-sided decortication led to removal of the peripheral cystic mass, which was attached to the pleura. Resection of the centrally allocated mass with full capsule was then done without the need for a right upper lobectomy as the mass showed free margins with no parenchymal attachment. The masses were sent for histopathology for a definitive diagnosis (Fig. 2).

Histopathology report revealed the presence of squamous epithelium lining with ectodermal components such as hair follicles, sebaceous glands, keratin debris, fatty, and fibromuscular tissue. No immature tissues were detected, leading to a final diagnosis of mature cystic teratoma. The second peripheral mass was found to be an abscess, and cultures of the content revealed purulent nature. Post-operative antibiotics were prescribed; however, the patient was discharged after a week of hospital stay without any complications. The patients after then were followed up at 3 and 6 months with chest x-ray and tumor markers which came free.

3. Discussion

Germ cell tumors in the thorax are either primary tumors de novo developed at the thorax or secondary tumors metastasized from the gonads [7]. The most common extragonadal site for germ cell tumors in adults is the mediastinum especially in the lungs- intrapulmonary teratomas (IPTs). IPTs are a very rare germ-cell tumor with only 65 cases reported in English and Japanese literature in 1996 with the first case reported in 1939. Most IPTs reported are in the left upper lobe; however, right sided teratoma has only reported once in literature and our case is the second one [2].

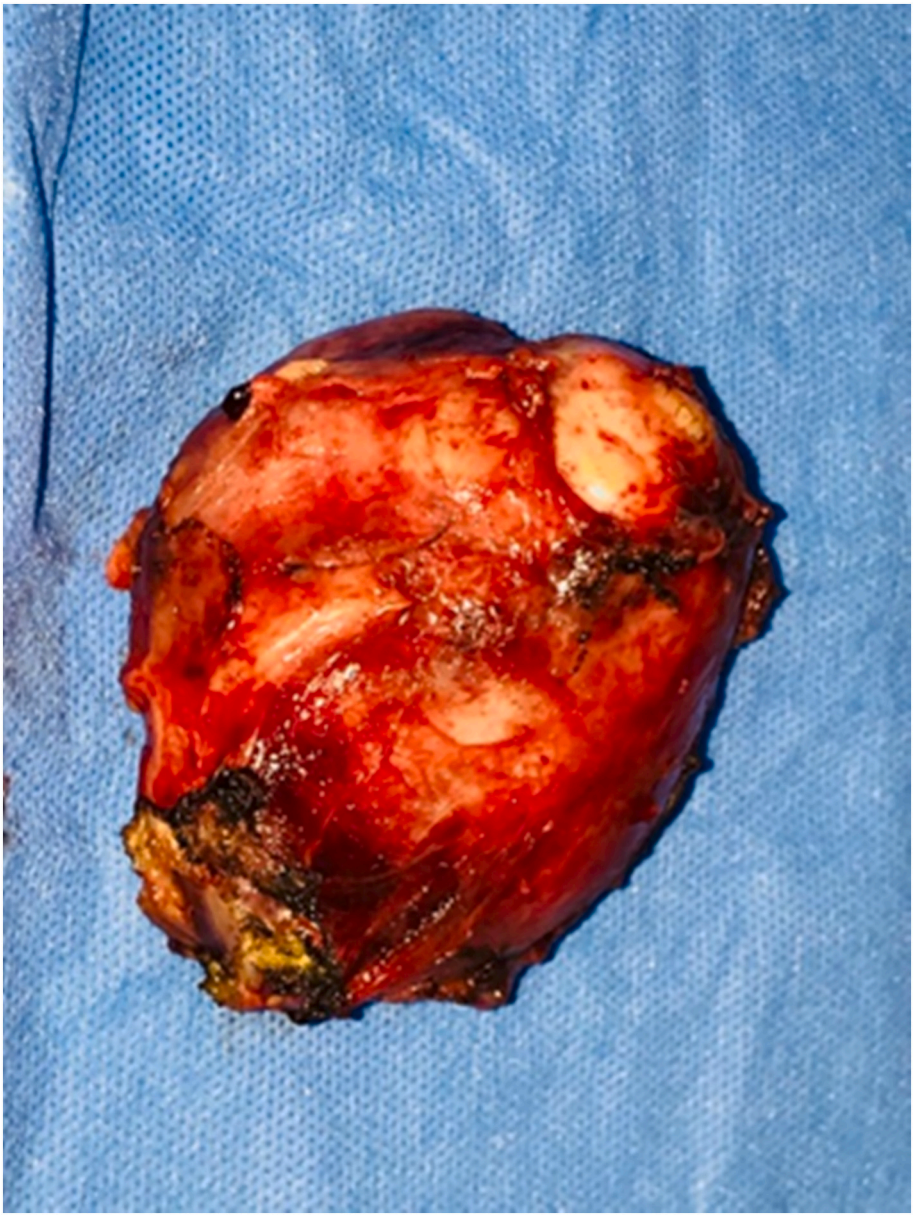

Fig. 1. A well-circumscribed mass with free margins removed from the upper lobe of the right lung (mature cystic teratoma).

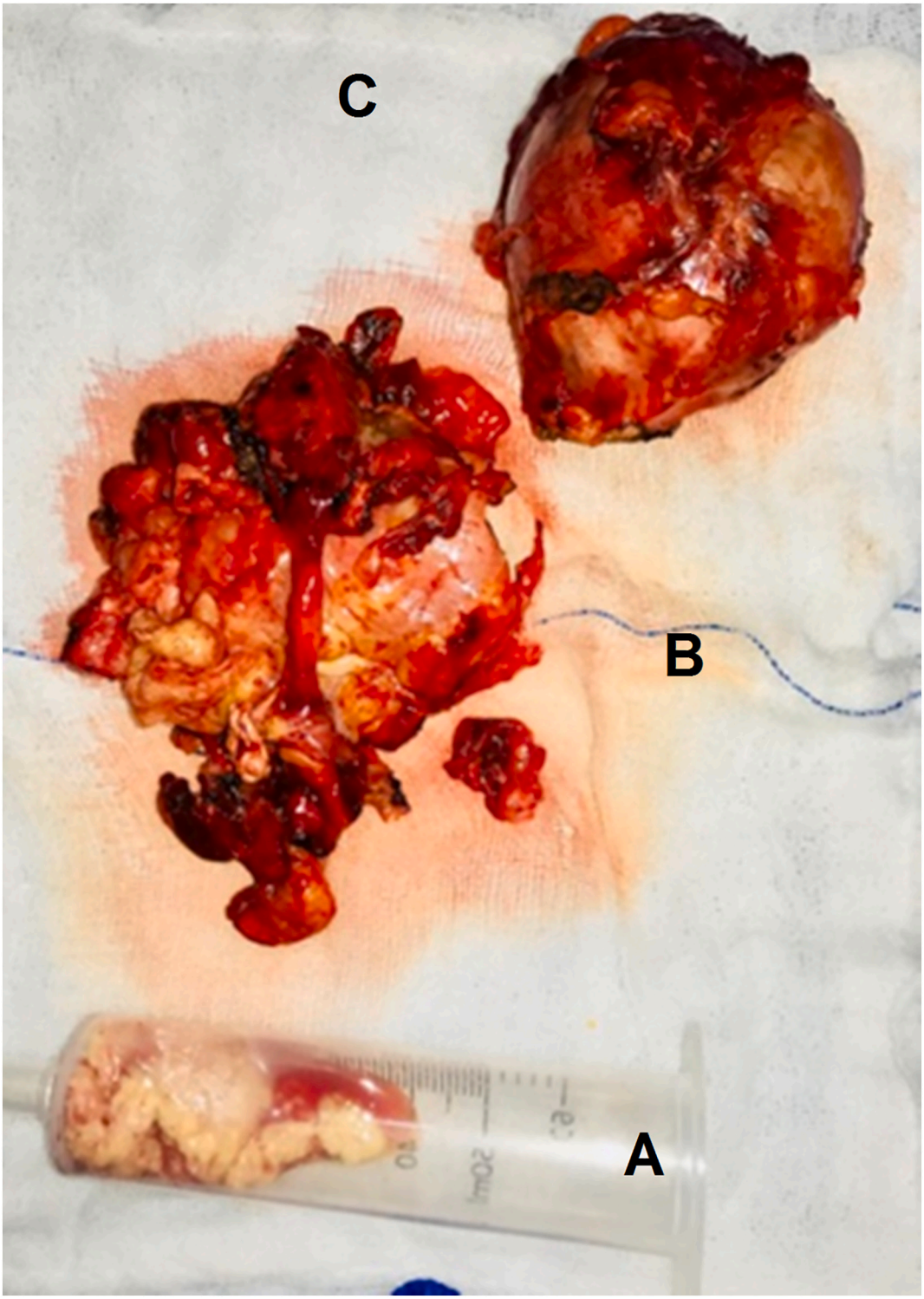

Fig. 2. (A) Peripheral abscess content, (B) a piece of the Right pleura with the attached cyst (C) the upper lobar mature cystic teratoma.

Most teratomas are benign and slowly growing, yet with no relationship between the size of the teratoma and malignancy [6,8]. Although teratomas may be present in any age group, IPTs usually affect patients in their first and second decades of life. IPTs manifest equally in males and females, unlike malignant mediastinal germ cell tumors, which affect males more than females [6]. The diagnosis of IPTs is challenging due to several differential diagnoses, including thymoma, thymic carcinoma, thyroid, or parathyroid neoplasm [8]. Furthermore, symptoms of IPTs can vary according to multiple factors, including their size, location, and tissue type of which they are comprised [9]. The patient, in this case, complained of dry cough, which is considered the most common symptom of IPTs, as well as hemoptysis, chest pain, and recurrent infections, which is considered the most reason for hospital admission in teratoma patients [10]. Moreover, IPTs can be associated with a wide scale of complications, such as pneumonia, bronchiectasis, abscess formation, and superimposed infections [10].

In diagnosing mediastinal teratomas, although conventional chest radiographs still play a significant role in the initial evaluation,

computed tomography (CT) scan is essential for specifying the nature, location, and relationship of the tumor to the surrounding structures. In addition, CT role lays in its distinguishing role between tumors originating in the mediastinal areas from those that invade from lungs or other organs. Recently, ongoing developments in magnetic resonance imaging (MRI) are considered an adjunct to CT scan in diagnosing mediastinal lesions. Moreover, MRI was more dominant than CT scan in evaluating the spread through the tumor capsule and adjacent structures infiltration by fat plane obliteration [10].

On CT scans, mediastinal mature teratomas are often well-defined showing internal heterogeneous appearances owing to the presence of fat, fluid, soft tissue, or calcifications. Furthermore, these lesions can have an internal proteinaceous or hemorrhagic fluid component with high density on non-contrast images. Mediastinum teratomas can manifest in different forms on CT scans, as reports of CT imaged mediastinal teratomas revealed 53% involved calcifications, 65%–75% noted fat attenuation, and 85%–90% had areas of fluid attenuation [9]. As the absence of fat cannot exclude a germ cell tumor from the differential diagnosis, the presence of solid components in the tumors can be the predictor of malignant risk. In this study, the benign mature teratoma had the major components of fat, proteinaceous fluid, and some small areas of solid tissue. The hyperattenuating (up to 100 Hounsfield units) pre vascular lesions on CT can be benign hemorrhagic, proteinaceous cysts, or solid necrotic lesions [6].

Teratomas have several complications, such as respiratory distress, hemorrhage, pneumothorax, or fistula formation with the aorta, the esophagus, or bronchus [10]. One of the main complications of teratomas is rupturing into the pleural space or pericardium, which can be diagnosed radiologically if the wall of the lesion appears ruptured with associated effusions or lung consolidation [11], yet no complications were noted in our case except for the on-top abscess formation. Complete surgical excision is the treatment of choice for mediastinal teratoma because of possible complications. A minimally invasive approach via thoracotomy is usually preferred over mediastinotomy, as it decreases the hospital stay and further improves the quality of life. Failure to surgically remove the tumor can lead to severe fatal complications, including excessive hemoptysis, tumor enlargement and spread, adult respiratory distress syndrome, and eventual death [12].

4. Conclusion

IPTs are rare tumors, with less than 100 cases reported in the past few years. The previous cases reported are in the upper lobes without proposed explanation yet- mostly in the left upper lobes. Right mature cystic teratoma is a scarcer event. Associated with many complications, IPT's most common presenting manifestation is recurrent infections. Thus, we recommend physicians to have a high suspicious index for teratoma when encountering an abnormal upper lobe mass associated with recurrent infection picture even in the right lobe.

Abbreviations

IPT intrapulmonary teratomas

Consent to publish

Written informed consent was obtained from the patient 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.

Availability of data and materials

All data generated or analyzed during this study are included in this published article or its supplementary information files.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

The case report was performed following the declaration of Helsinki and approved by Ain-Shams University Hospital ethics committee- Ain Shams institute- according to the international guidelines and ethics.

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Guarantor

A.A. is the corresponding author and the guarantor of this manuscript.

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CRediT authorship contribution statement

All the authors (A.A., A.F., S.K., M.E., S.E.) have shared work in collecting, analyzing, and writing the research paper. S.E. and A.A. were major contributors in writing the manuscript, and A.A. was the assistant in the operating surgery. All authors read and approved the final manuscript.

Declaration of competing interest

All authors declared no conflict of interests.

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