Case Report

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A rare case of intrapulmonary teratoma

Atish N. Bansod*, Neha S. Awachar, Rohan K. Umalkar, Manjunath C, Jantli, Priyanka C. Tayade, Ishwar G. Patil

Department of General Surgery, Indira Gandhi Medical College and Hospital, Nagpur, Maharashtra, India

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*Correspondence: Dr. Atish N. Bansod.

E-mail: atish6267@gmail.com

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ABSTRACT

Benign cystic teratomas of the lung are extremely rare. We diagnosed such case in 22 years old male suffering from heaviness in left side of chest for 2 years with occasional cough with expectoration for 1 year. Patient suffered typical symptom of white hair in expectoration. Diagnosis was confirmed on computed tomography thorax and Fine needle aspiration cytology. Teratoma was located in anterior segment of left upper lobe and was treated with left upper lobectomy. Histopathology was done and was implicational mature teratoma.

Keywords: Intrapulmonary teratoma, Trichoptysis, Benign neoplasm

INTRODUCTION

Intrathoracic teratomas nearly always occur in the mediastinum. Although malignant counterparts are described, teratomas are more often benign than malignant. Intrapulmonary teratomas (IPT) have rarely been reported since Mohrs description of this entity in 1830 (around 100 cases are reported in world literature till 2016).1 Intra pulmonary teratomas are found in children and adults with the age range reported being 5 to 68 years and most ordinarily found in 2nd decade. Intra pulmonary teratoma have predilection for upper lobes on each side and it is proposed that it develops in relation to the thymus, as a derivative of 3rd pharyngeal pouch.² The presence of Intra pulmonary teratoma could also be due to separation or displacement of thymus during early embryogenesis. Intrapulmonary teratomas are extremely rare compared to similar lesions occurring in ovary, testis, sacrum, retroperitoneum and mediastinum.⁵

CASE REPORT

A 22 years old male presented with complains of heaviness in left side of chest for 2 years and occasional cough with expectoration for 1 year. A very unusual thing was that he complained of white hair coming in his

expectoration. No other complaints like fever, weight loss. He had no addictions and no significant past or family history. On examination his vitals were normal. On respiratory system examination he had coarse crackles over left upper zone. Rest of the systemic examination was normal including external genitalia.

On radiological study X-ray revealed a well-defined soft tissue large opacity in left upper lobe (Figure 1).

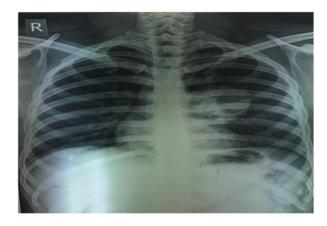


Figure 1: Pre-operative chest x-ray showing well defined opacity in left upper lobe.

Contrast enhanced computed tomography thorax was done which showed a large thin-walled space occupying lesion of size 5.7 by 5 by 5.6 cm involving the anterior segment of left upper lobe (Figure 2).



Figure 2: Contrast enhanced computed tomography thorax was done which showed a large thin-walled space occupying lesion of size 5.7 by 5 by 5.6 cm involving the anterior segment of left upper lobe.

It appeared to be adherent to left anterolateral subcoastal pleura laterally and mediastinal pleura medially starting at the level of main pulmonary trunk and extending along left ventricular surface. The lesion showed heterogenous density containing soft tissue elements like fat. Space occupying lesion consisted of central predominant cystic component with tiny peripheral air pockets with marginal thin peripheral enhancement of capsule with no pleural extension. Perilesional mediastinal or inflammatory changes were present. No mediastinal lymphadenopathy, pleural effusion or thickening of pleura was noted. Computed tomography thorax suggested space occupying lesion in left upper lobe anterior segment most probably an infected bronchogenic cyst, benign epithelial cyst. Serum beta human chorionic gonadotropins and alpha fetoprotein were normal. Ultrasonography abdomen and scrotum was also normal. Guided Fine needle aspiration cytology smear showed blood, fair number of neutrophils, large number of anucleate squamous cells and some mature squamous cells and occasional multinucleated giant cells, cytological evidence of malignancy was not seen. Thus, fine needle aspiration cytology was done suggested possibility of benign cystic lesion lined by squamous epithelium/dermoid cyst/differentiated teratoma. With a positive history of trichoptysis, X-ray and computed tomography features of left lobe space occupying lesion and fine needle aspiration cytology report he was diagnosed as a case of left lung upper lobe teratoma.

Patient was posted for left sided thoracotomy with left upper lobectomy. Intra-operatively there was a mass in

left upper lobe anterior segment which was adherent to pericardium, which was separated. As the mass was in close proximity to left upper lobe bronchus, a left upper lobectomy was done (Figure 3).



Figure 3: Post operative specimen gross appearance showing left upper lobe, as teratoma in anterior segment of left upper lobe was densely adherent to pericardium left upper lobectomy was done.

Pathological gross specimen was of size 6×5×6 cm. The cut gross section showed predominantly cystic consistency with thick cheesy white material also known as pultaceous material along with bunch of hairs. No bone, cartilage or teeth identified on gross specimen (Figure 4).



Figure 4: Post operative specimen cut opened showing tufts of hairs and pultaceous material suggestive of teratoma.

Microscopic findings in histopathological specimen showed all three-germ layer component. It composed of respiratory epithelium, apocrine and eccrine sweat glands, stratified squamous epithelium and hair follicles and cartilage. No immature element pre-set and no atypia in any mature component was seen. And on the basis of histopathological findings the diagnosis of mature intrapulmonary teratoma was established.

Patients post-operative course was uneventful. His chest tubes were removed on third post-operative day and patient was discharged on fifth post-operative day. Post-operative chest-X ray shows near total complete compensatory expansion of left lower lobe (Figure 5). Post operatively patient is doing well. He has been advised 6 monthly follow up initially and yearly follow up after 2 years.

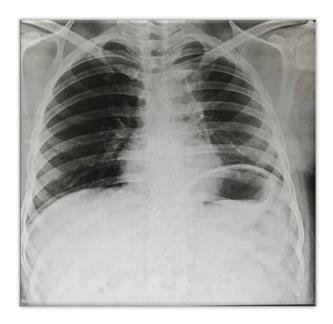


Figure 5: Post operative chest X-ray of the patient with complete expansion of remnant left lower lobe.

DISCUSSION

In the above case the differential to the diagnosis could be immature teratoma or metastasis from the primary gonadal teratoma. The microscopic findings all threegerm cell component with no immature element to rule out immature intrapulmonary teratoma. Other sites of primary teratoma were ruled using clinical examination, ultrasonography abdomen and gonads along with normal hormonal studies (serum beta human chorionic gonadotropins and alpha feto protein).

Mature teratomas are the foremost common histological sort of germ cell tumours, followed by seminomas. Germ cell tumours are predominantly found in the gonads, while the anterior mediastinum is the most common extragonadal site. The first case of pulmonary teratoma was reported by Mohr. Germ cell tumours in the lung

occur typically in the second to fourth decades of life with alittle female preponderance.²

Intrapulmonary teratomas typically range from 2.8 to 3 cm in diameter and are cystic and multiloculated but may rarely be predominantly solid.² In 42% of the cases, the cysts are in continuity with bronchi, and have an endobronchial component resulting in haemoptysis or expectoration of hair or sebum.³ Histopathologically, IPT are similar to other benign cystic teratomas having ectodermal, endodermal and mesodermal elements.² Due to presence of various tissues within intra pulmonary teratoma, these can produce digestive or proteolytic enzymes, making them more prone to rupture. Clinically, patients with intrapulmonary teratomas present with pain (52%), haemoptysis (42%) and cough (39%).²

The most specific symptom is trichoptysis or expectoration of hair (13%).² Turna et al, Istanbul, Turkey have coined the term 'Bronchotrichosis' for presence of hair in expectoration or bronchoscopy and called it as a diagnostic sign.⁴ Bronchiectasis occurs in 16% of cases and may delay the recognition of the pulmonary tumor.⁵ Radiologically, lesions are typically cystic masses often with focal calcification.² Computed tomography accurately estimates the density of all elements such as soft tissue (in virtually all cases), fluid (88%), fat (76%), calcification (53%) and teeth.⁶ Magnetic resonance Imaging is valuable in detecting the anatomic relation to mediastinal and hilar structures.

Surgical resection is the treatment of choice and lobectomy leads to a long recurrence-free survival.⁷ The other reason for surgical resection of intrapulmonary pulmonary teratoma is high incidence (30%) of malignant teratomas. Malignant teratoma is defined by the presence of immature tissue within the teratoma, rather than metastasis or infiltration and the prognosis being extremely poor.⁸

CONCLUSION

Intrapulmonary Teratoma is an extremely rare tumour, trichoptysis is a diagnostic feature and surgical resection is the definitive treatment.

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