

Case Report

Aspergilloma or intrapulmonary teratoma - time to rethink

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ABSTRACT

Intrapulmonary teratoma (IPT) is a rare type of extra gonadal germ cell tumor which can often be misdiagnosed due to its non-specific presentation. Complete surgical resection should be done because of its malignant potential. We report a case of 33 years old female previously treated as tuberculous osteomyelitis of sternum. She presented with chest pain and sudden hemoptysis. Based on the radiological picture she was diagnosed as complex aspergilloma of lung with a thick walled cavity. Hemoptysis was controlled with supportive management. On surgery right upper lobe was densely adherent to the anterior chest wall. Exploration of the cavity revealed a 6×4 cm whitish mass with non-pigmented hair. Right upper lobectomy and wide excision of the mass was performed. Histopathology was consistent with the diagnosis of IPT with no malignant cells.

Keywords: Intrapulmonary teratoma, Aspergilloma, Hemoptysis

INTRODUCTION

Teratomas are benign tumors, consisting components of three germinal layers ectoderm, endoderm and mesoderm.¹ In adults, germ cell tumor (GCT) make up 10-20% of all anterior mediastinal masses.¹ Intrapulmonary teratomas (IPTs) are rare. They can occur at any age but mostly seen in the third decade of life with equal predilection for both genders.^{2,3} In the lung, teratoma may present as metastasis from gonadal GCT such as seminoma, embryonal cell carcinoma, yolk sac cancers and choriocarcinoma.¹

We would like to report a case of IPT presenting as a cavitary intraparenchymal lesion treated previously as aspergilloma lung.

CASE REPORT

A 33 years old female, was treated with antitubercular therapy three years ago for anterior chest wall sinus secondary to tuberculous osteomyelitis and abscess

(Figure 1). She developed chest pain and occasional hemoptysis over the course of last two years. Chest X-ray showed heterogenous opacity in right middle zone with air crescent sign, pathognomonic of aspergilloma lung. Contrast computed tomography of chest (CT) showed mixed density lesion involving anterior segment of right upper lobe with peripheral air density (Figure 2).

She was being treated for aspergilloma lung pertaining to her history and radiological findings. She presented with chest pain and sudden moderate hemoptysis for which supportive care was started. Initial management comprised of continuous cardiorespiratory and hemoglobin monitoring, nursing in right lateral decubitus position and transfusion of blood products. As there was a risk of massive hemoptysis, surgical intervention was planned. Right posterolateral thoracotomy was performed under single lung ventilation to protect the contralateral lung. Intraoperatively, there were significant dense adhesions between anterior segment of right upper lobe of lung and the anterior chest wall. Adhesionolysis was done with

Ligasure^R. A thick walled cystic cavity over anterior segment of right upper lobe was noted. Upon opening the cavity, a 6×4 cm whitish tumor with non-pigmented hair was noted (Figure 3).



Figure 1: Anterior chest wall abscess with discharging sinus and osteomyelitis of sternum (initial CT chest with tuberculous osteomyelitis and abscess of sternum).



Figure 2: Computed tomography showing lesion in right upper lobe with heterogeneous intracavitary mass.

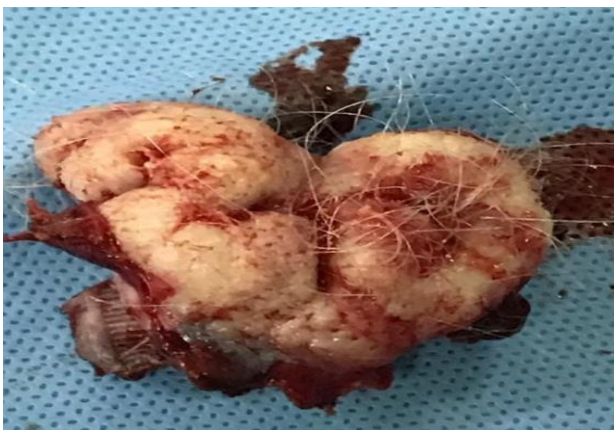


Figure 3: Mass extracted with non-pigmented hair.

Tumor was excised with wide margins and separated from mediastinum followed by right upper lobectomy. Post-operative recovery was uneventful. Histopathological examination favored our diagnosis composed of component from all three germ layers including adipose tissue, cartilage, blood vessels, nerves, pancreatic tissue and stratified ciliated columnar respiratory epithelium, there was no evidence of immature teratomatous elements or malignancy.

DISCUSSION

Teratomas are the most common mediastinal GCTs with 80% of mature type and are slow growing benign tumors.^{1,3} IPTs are rare with only 65 cases reported till now.³ They mostly involve the left upper lobe. Only few cases have been reported with involvement of right upper lobe as seen in our case. They are believed to arise from third pharyngeal pouch.^{3,4} Derivatives from any of the three germinal layers can be found within the tumor mostly including hair, bone and teeth. Other tissues include respiratory epithelium, pancreatic or gastric tissues, cartilages and muscles.⁴

Clinical presentation can range from chest pain, cough, fever, bronchiectasis and pneumonia.^{1,2} Although rare trichoptysis (expectoration of hair) is a characteristic feature of IPT and indicates communication with airway but is reported to occur in only 13% of cases.¹⁻³ Erosion of the tumor into bronchus can cause hemoptysis thus can be life threatening.⁵ It is believed that the proteolytic or digestive enzymes secreted by intestinal mucosa or pancreatic tissue found in the teratoma can lead to enzyme-induced erosions causing rupture into airway, mediastinal structure and even through the skin to form draining fistulae.⁶ A previous history of chest wall sinus and hemoptysis in our patient who was treated as tubercular sinus with osteomyelitis of sternum, strongly favors enzymatic erosion of tumor into surrounding tissue and bronchus when studied retrospectively. This observation was further supported by presence of pancreatic tissue in the excised tumor. The cavitation of the upper lobe was clinically and radiologically consistent with aspergilloma lung and had been treated as such for a year.

Potential complications of IPT include rupture of tumor, life threatening hemoptysis, airway compression and malignant transformation.^{2,7}

In the absence of calcifications, bones or cartilages, correct preoperative diagnosis of IPT based upon radiology is difficult. It is not unusual to misdiagnose the condition especially in areas with high burden of tuberculosis as in our case.⁴ CT scan remains investigation of choice and findings of IPT includes a heterogeneous lobulated opacity containing soft tissue, fat, fluid, and/or calcification. Cavitation, peripheral translucency or air crescent mimicking aspergilloma lung and consolidation are other features.^{5,8} Air within a cavity distinguishes intrapulmonary from mediastinal teratomas.⁸ All cases of

IPTs, must be evaluated for gonadal GCTs, to rule out metastases.¹ Furthermore, this case illustrates that any thick-walled lung cavity with air crescent mimicking an aspergilloma lung should have IPT as a possible differential diagnosis.

Complete surgical resection is the aim of the treatment for IPT due to its malignant potential in 25% of the cases.^{6,7} Complete excision of the tumor might be difficult in cases with rupture where adhesions around the tumor are found. In such cases, subtotal resection can be carried out to prevent injuries to surrounding vital structures.⁶ Surgical excision is compulsory to avoid undesirable complications such as massive to life threatening hemoptysis, increase in size with malignant change and acute respiratory distress syndrome.³

Histopathologic study of the resected specimen should also be looked for immature tissue, other germ cell component, carcinoma and sarcoma as these may coexist.¹ These findings were not present in our case, hence diagnosis of mature teratoma was confirmed.

Prognosis depends on the presence or absence of immature tissue and age of the patient.¹ Mature teratomas have excellent prognosis with low recurrence rate.¹ In malignant teratoma, adjuvant combination chemotherapy may result in improved survival.⁵

CONCLUSION

IPT can mimic an aspergilloma lung and be easily missed where tuberculosis is endemic and necessitates consideration as a differential diagnosis for any thick walled cavitary lung lesion with inhomogeneous density. Early recognition and prompt surgical management saves the patient from life threatening hemoptysis.

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