Intra-pulmonary Teratoma: A Rare Case

Ajit Vigg¹, Sanjeev K. Khulbey², Sanjay Kumar Agarwal², Vijay Dikshit², A. Sathpathy³, U. Srinivas³, G. Swarnalata⁴, Tejal Modi⁴ and A.S. Hemalatha¹

Departments of Respiratory Diseases¹, Cardio-thoracic Surgery², Anaesthesia³ and Pathology⁴, Apollo Health City, Hyderabad, India

[Indian J Chest Dis Allied Sci 2012;55:155-157]

CLINICAL SUMMARY

A 28-year-old female, home-maker presented to our department with a four-year history of intermittent episodes of cough and haemoptysis. She had no history of fever, weight loss or expectoration. Physical examination was unremarkable.

INVESTIGATIONS

Routine haematological and biochemical investigations were normal. Chest radiograph (posterior– anterior view; Figure 1) showed a well-defined opacity in the right lower zone with no satellite lesions and no radiological evidence of calcification.



Figure 1. Chest radiograph (posterior-anterior view) showing a well-defined opacity in the right lower zone.

Computed tomography (CT) of the chest showed a multiseptate cystic lesion (6.5cmx6.5cm) with thick irregular and calcific densities within it seen in the right paracardiac region abutting the right atrium (Figure 2).

Fibreoptic bronchoscopy did not reveal any remarkable findings. A right lateral thoracotomy was performed under general anaesthesia. A solid well



Figure 2. Contrast enhanced CT of the chest showing a multiseptate cystic lesion in the right para cardiac region abutting the right atrium (axial and coronal views).

capsulated tumour measuring 6cm in diameter was found in the medial aspect of the right lower lobe densely adherent to the pericardium.

[Received: July 24, 2012; accepted after revision: November 21, 2012]

Correspondence and reprint requests: Dr Ajit Vigg, Vigg Villa, 553, Road No. 3, Banjara Hills, Hyderabad-500 034 (Andhra Pradesh), India; Phone: 9440952871; e-mail: drajitvigg@gmail.com

Enucleation of the lesion was performed without any damage to the surrounding tissues. Histopathological examination of the cut-section of the lesion revealed multilocular cyst filled with pultaceous material, hair and few grey white to yellow solid areas (Figure 3A).

Microscopic examination showed a cystic mass lined by keratinised squamous epithelium along with adenexal structures. Large dilated glands lined by ciliated columnar epithelium were seen along with mature glial tissue. Mature adipose tissue was seen with lymphoid follicles and aggregates of siderophages, multi nucleate histiocytes (Figure 3B).

The post-operative period was uneventful and the patient was discharged on the 5th post-operative day with complete recovery.



Figure 3. Cut-section of the specimen (A) showing a multilocular cyst filled with pultaceous material, hair and few grey white to yellow solid areas; (B) microscopic examination of the cyst showing lining by mature squamous epithelium with adipose tissue in the wall (Hematoxylin and Eosin stain×10).

DIAGNOSIS

Intra-pulmonary teratoma right lower lobe.

DISCUSSION

Teratomas are tumours consisting of tissues derived from more than one germ cell line. Criteria for pulmonary origin are: exclusion of a gonadal or extra-gonadal primary site and origin entirely within the lung. These lesions originate from the third pharyngeal pouch. Primary intra-pulmonary teratoma is rare.¹

Intra-thoracic teratomas almost always occur in the mediastinum, but occasionally, these may be found in the lung as intra-pulmonary teratomas. Mature teratomas are the common histological type of germ cell tumours, followed by seminomas. Germ cell tumours are predominantly found in the gonads, while the anterior mediastinum is the most common extra- gonadal site.² Most intra-pulmonary teratomas have been diagnosed in the first or second decade of life, though the age ranging from 10 to 68 years.¹⁵ The first case of intra-pulmonary teratoma was reported by Mohr in 1839.³ A total of 67 cases have been reported in the literature from 1939 to 2007, including 35 from Japan and 7 from Korea, and the rest are in the English literature.⁴

There are different theories postulated for the genesis of intra-thoracic teratomas. According to Joo and Colleagues⁵ intra-thoracic teratomas including mediastinal and intra-pulmonary teratoma have common genesis and are thought to originate from displaced thymic tissue of the third pharyngeal pouch. Another study indicates that intra-pulmonary teratomas arise from aberrant thymic tissue. It is proposed that the primodial teratomatous focus in the potential mediastinum is caught up by the respiratory outgrowth and hence locates within the lung.⁵ Another theory suggested that primary pulmonary germ cell tumours represent an unusual differentiation of somatic cell line.⁶ There are a few case reports of intra-pulmonary teratoma from India.⁷⁻¹¹

Clinically, patients with intra-pulmonary teratomas present with chest pain (52%), haemoptysis (42%), and cough (39%). The most specific symptom is trichoptysis or expectoration of hair (13%) which was not seen in our case. Bronchiectasis occurs in 16% of cases.¹² Some patients may present with pyothorax as reported by Tandon and Colleagues.¹³

Two-thirds of intra-pulmonary teratomas occur in the upper lobes, usually in the left upper lobe.¹⁴ Occurrence in the right lower lobe, as in the present case, is uncommon.

Radiologically, an intra-pulmonary teratoma usually presented as a lobulated mass, but may be seen as a cavitary lesion, a consolidation, or as a peripheral translucency. In the present case it was seen as a multiseptate cystic lesion on CT of the chest. A cavity with peripheral translucency is a distinguishing feature of intra-pulmonary teratomas from mediastinal teratomas.¹⁵ This feature indicates air within the cavity secondary to a bronchial communication.

CT demonstrates discrete areas of different densities due to soft tissue, high local fat content, or punctate calcification, or a combination of these, and is extremely valuable to detect a ruptured teratoma. In a ruptured teratoma, the internal density becomes heterogeneous, the tumour margin becomes irregular, and the fat component shows a bursting configuration.¹⁵ Magnetic resonance imaging is another emerging method that can be used preoperatively to diagnose a pulmonary teratoma.¹⁶

Histologically, intra-pulmonary teratomas may contain any tissue originating from one of the three germinal layers. A large proportion (approximately 30%) of teratomas are of the immature type and therefore have malignant potential.¹⁵ Tumours with pancreatic tissue are prone to rupture owing to enzymatic reactions. Due to its malignant potential and possibility for rupture, surgical resection of an intra-pulmonary teratoma is advocated; options may range from segmentectomy to pneumonectomy.¹⁵

In conclusion, an intra-pulmonary teratoma is an exceptionally rare tumour. The diagnosis is made on the radiologic imaging, especially a CT that demonstrates calcification, cavitations, and peripheral translucent areas. Trichoptysis is the only clinical feature that can be diagnostic. Due to its potential for rupture or malignancy, surgical removal is indicated.

REFERENCES

1. Walrond ER, Prussia PR. Pulmonary teratoma. West Indian Med J 1987;36:39-42.

- 2. Morgan DE, Sanders C, Mcelvein RB. Intrapulmonary teratoma: a case report and review of the literature. *J Thorac Imag* 1992;7:70-77.
- 3. Collier FC, Dowling EA, Plot D, Schneider H. Teratoma of the lung. *Arch Pathol* 1959;68:138-42.
- Rana SR, Swami N, Mehta S, Singh J, Biswal S. Intrapulmonary teratoma: an exceptional disease. *Ann Thorac Surg* 2007;83:1194-6.
- Joo M, Kang YK, Lee HK, et al. Intrapulmonary and gastric teratoma: report of two cases. J Korean Med Sci 1999;14:330-4
- 6. Miller RR, Champagne K, Nevin Murrey RC. Primary pulmonary germ cell tumor with blastomatous differentiation. *Chest* 1994;106:1595-6.
- Sushama A, Gomathy S, Rajendran VR, Vasu CK. Mature intrapulmonary teratoma: a case report. *Indian J Radiol Imag* 2003;13:31-2.
- Saini ML, Krishnamurthy S, Kumar Rekha V. Intrapulmonary mature teratoma. *Diagn Pathol* 2006;1:38.
- Rawat J, Saini S, Raghvanshi S, Sindhwani G, Kesarwani V. Intra-pulmonary teratoma presenting with tricoptysis. Indian J Chest Dis Allied Sci 2011;53:237-9.
- Madusudhan KS, Sharma R, Gadodia A, Kumar A. Spontaneous rupture of benign mediastinal teratoma: a report of 2 cases. *Indian J Med Peadiatr Oncol* 2012;33:123-5.
- 11. MH Beg, Kumar G, Abdul Samad, Shahbaz A. Intrapulmonary mature teratoma. *JK Practitioner* 2012;17:64-65.
- 12. Colby TV, Koss MN, Travis W. Atlas of Tumour Pathology: Tumours of the Lower Respiratory Tract, (Third Series). Armed Forces Institute of Pathology;1994:pp 487-9.
- Tandon S, Kant S, Singh AK, Sinha KN, Chandra T, Agarwal PK. Primary intra-pulmonary teratoma presenting as pyothorax. *Indian J Chest Dis Allied Sci* 1999; 41:51-5.
- 14. Day DW, Taylor SA. An intrapulmonary teratoma associated with thymic tissue. *Thorax* 1975;30:582-7.
- 15. Zenker D, Aleksic I. Intrapulmonary cystic benign teratoma: a case report and review of the literature. *Ann Thorac Cardiovasc Surg* 2004;10:290 -2.
- Iwasaki T, Iuchi K, Matsumura A, Sueki H, Yamamoto S, Mori T. Intrapulmonary mature teratoma. *Jpn J Thorac Cardiovasc Surg* 2000;48:468-72.