

Ön mediasteninin kondromatöz hamartomu

Anterior mediastinal chondromatous hamartoma

Gülbanu Horzum Ekinci¹, Yelda Tezel², Osman Hacıömeroğlu¹, Serda Kambur Metin³, Ayçim Şen⁴, Çağatay Tezel³, Adnan Yılmaz¹

¹Sureyyapasa Chest Diseases and Thoracic Surgery Training and Research Hospital, Department of Pulmonology, İstanbul, Turkey

²Haydarpaşa Numune Training and Research Hospital, Department of Pulmonology, İstanbul, Turkey

³Sureyyapasa Chest Diseases and Thoracic Surgery Training and Research Hospital, Department of Thoracic Surgery, İstanbul, Turkey

⁴Sureyyapasa Chest Diseases and Thoracic Surgery Training and Research Hospital, Department of Pathology, İstanbul, Turkey

ÖZET

Elli bir yaşında erkek hasta 1 aydır devam eden nefes darlığı yakınması ile başvurdu. Toraksın bilgisayarlı tomografisi, ön mediastende, sol pulmoner arter inen aortanın proksimal bölümüne komşu, 5.4x5.2 cm boyutlarında, düzgün sınırlı, "pop corn" kalsifikasyon içeren kitle gösteriyordu. PET-BT'de lezyonun SUVmaks değeri 2.7 olarak rapor edildi. Sol posterolateral torakotomi yapılarak tümör tam olarak rezekt edildi. Kesin tanı ön mediasteninin hamartomu idi.

Anahtar kelimeler: hamartoma, mediasten, cerrahi

Introduction

Hamartoma is defined as an abnormal mixture of tissue elements or an abnormal proportion of a single element normally present in an organ (1). It can originate from many organs (2). Pulmonary hamartoma is the most common benign tumor of the lung and it comprises 7-14 % of all solitary pulmonary nodules (3). The mediastinal location of this tumor is extremely rare and few cases of mediastinal hamartoma have been reported in the literature (1,2,4-6). We present a case of this rare tumor.

Case Report

A 51-year-old Turkish man presented with dyspnea for one month. He was a nonsmoker and his past medical history was unremarkable. Chest X-ray showed a mass with calcification in the left upper lung field (figure 1).

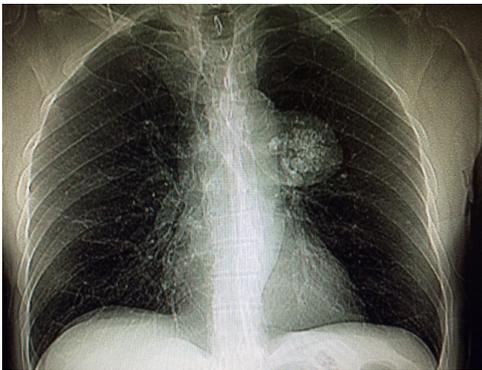


Figure 1. Chest x-ray shows a mass with calcification in the left upper lung field.

SUMMARY

A 51-year-old man presented with dyspnea for one month. Computed tomography scan of the thorax showed a 5.4x5.2 cm well-circumscribed mass with popcorn calcification near left pulmonary artery and proximal descending aorta in the anterior mediastinum. SUVmax value of the lesion on PET-CT was 2.7. Tumor was completely resected through a left posterior-lateral thoracotomy. The final pathological diagnosis was a hamartoma of the anterior mediastinum.

Keywords : hamartoma, mediastinum, surgery

His physical examination and routine laboratory values were normal. Computed tomography scan of the thorax showed a 5.4x5.2 cm well-circumscribed mass lesion with popcorn calcification near left pulmonary artery and proximal descending aorta in the anterior mediastinum. (figure 2)

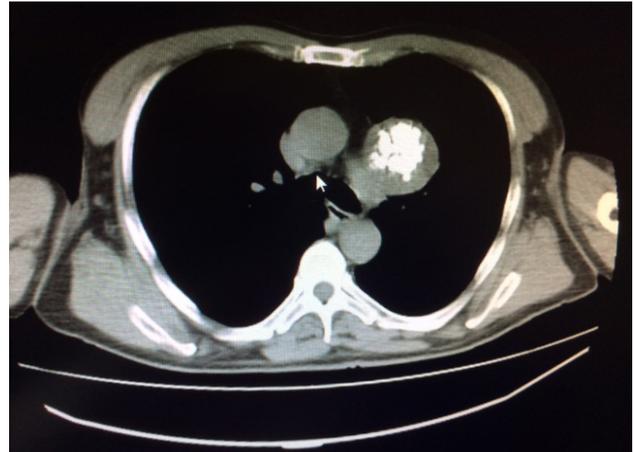


Figure 2. The computed tomography scan of the thorax shows a 5.4x5.2 cm well-circumscribed mass with popcorn calcification near left pulmonary artery and proximal descending aorta in the anterior mediastinum.

Also a 3.4x5.5 cm bullae in the left upper lobe. Bronchoscopy revealed normal endobronchial appearance. Pulmonary function tests were within normal limits (FEV1= 3.33 L 98% - FVC= 4.26 L 99% - FEV1/FVC= 102). The

patient underwent a left lateral thoracotomy. In operation, a large, firm, and yellowish-white colored mass was seen in the anterior mediastinum. The lesion was adjacent to vessels with no evidence of local invasion. The tumor was easily dissected free from the surrounding structures, and was subsequently resected. Also bullectomy was performed. Postoperative course was uneventful, and he was discharged home 2 days after the operation. Macroscopically, the tumor surface was regular and yellowish-white colored with calcification. Its size was 6.5x5.5x5 cm. Microscopic examination revealed a diagnosis of chondromatous hamartoma that consisted of fibrous connective tissue, adipose tissue, and portions of cartilage. At 8 months follow up, there was no evidence of tumor recurrence.

Discussion

Hamartoma is a benign tumor originating from the mesenchymal tissues. The term hamartoma was first introduced in 1904 by Albrecht (7) to describe lesions composed of elements of tissue that are native to an organ but are present in a disorganized array. Its nature has been debated. While this tumor was initially regarded as a development malformation, it is now accepted as a true neoplasm, probably originating from mesenchymal elements (2,6,8).

Hamartoma can occur in any organ, but it is extremely rare in the mediastinum. Few cases of mediastinal hamartomas have been published in the literature (1,2,4-6,9). Grosfeld et al (9) reported that there was only one case of hamartoma among 196 children with mediastinal tumor. This tumor may locate in any mediastinum compartment (1,2,4-6,10). It situated in the anterior mediastinum in our case. Possible mechanism for development of mediastinal hamartomas is unclear. Many authors described a migration theory. They believed that an intraparenchymal hamartoma migrated across the visceral pleura into the mediastinum (1,2,6).

Histologically, hamartomas may include different proportions of mesenchymal elements. Its descriptive name depends on the predominant tissue. Predominant mesenchymal component may be chondroid, fatty, osseous, fibroblastic, or muscle (1,2,8,10). There was a predominant chondroid differentiation in 80% of cases with pulmonary hamartoma (1). Chondromatous (2,6), leiomyomatous (1), and lymphangiomyomatous (10) mediastinal hamartomas were published in the literature. We presented a case of mediastinal chondromatous hamartoma.

Hamartomas are benign tumors. However, it is accepted that there is an association between hamartoma and malignancy. The risk of lung cancer is 6.3 times higher in patients with hamartoma than in the general population. Recurrence may be seen after incomplete resection (8). Thus, complete surgical excision of the tumor is the treatment of choice in the patients with mediastinal hamartoma (1,2,6,10). It was reported that there was no sign of recurrence after surgical resection (1,4,10). The tumor was completely removed through thoracotomy in our case. At 8 months follow up, there was no evidence of tumor recurrence.

In conclusion, mediastinal hamartoma is an extremely rare benign tumor. Complete surgical resection is the treatment of choice because of malignancy and recurrence risk. Though this tumor is rare, it should be considered in

the differential diagnosis of the mediastinal masses.

References

1. Bibas BJ, Terra RM, Fernandez A, Shiang C, Pego-Fernandez PM, Jatene FB. Leiomyomatous hamartoma of the posterior mediastinum. *Ann Thorac Surg* 2010; 89: 304-305.
2. Brichon PY, Pilichowski P, Brambilla E, Coulomb M, Latreille R. Mediastinal chondrohamartoma. *Eur J Cardiothorac Surg* 1987; 1: 176-179.
3. Ganti S, Milton R, Davidson L, Anikin V. Giant pulmonary hamartoma. *J Cardiothorac Surg* 2006; 1: 19.
4. Chatzis AC, Kaklamanis L, Azariadis P. Cystic hamartoma of the posterior mediastinum. *Asian Cardiovasc Thorac Ann* 2014, Feb 11. [Epub ahead of print].
5. Junior RS, de Prospero JD, Gonçalves R, Rivaben JH, Soueff FA. A rare mediastinal tumor: hamartoma. *Rev Col Bras Cir* 2013; 40: 169-171.
6. Gholoum S, Fraser R, Ferri LE. Posterior mediastinal chondromatous hamartoma. *Ann Thorac Surg* 2007; 83: 1528-1530.
7. Albrechts E. ueber hamartome. *Verh Deutsch Ges Pathol* 1904; 7: 153-157.
8. Van Den Bosch JMM, Wagenaar S, Corrin B, Elbers JRJ, Knaepen PJ, Westermann CJJ. Mesenchymoma of the lung (so called hamartoma): a review of 154 parenchymal and endobronchial cases. *Thorax* 1987; 42: 790-793.
9. Grosfeld JL, Skinner MA, Rescorla FJ, West KW, Scherer LR 3rd. Mediastinal tumors in children: experience with 196 cases. *Ann Surg Oncol* 1994; 1: 121-127.
10. Ota H, Kimura Y, Kawai H, Ogawa J. Mediastinal lymphangiomyolipoma in an adult: report of a case. *Surg Today* 2010; 40: 365-368.