CASE REPORT

A RARE TUMOR OF THE MEDIASTINUM: INFLAMMATORY MYOFIBROBLASTIC TUMOR

Pr. Adil Arsalane¹, MD ; Pr. Hicham Fenane², MD ; Dr. Amine Azami³, MD ; Dr. Simohamed Mzouri⁴, MD ; Pr. Abdelfetah Zidane¹, MD.

¹ Department of Thoracic surgery, Military Teaching Hospital Ibn Sina , University Cadi Ayaad, Morocco.
² Department of Thoracic surgery, Teaching Hospital Mohammed VI, University Cadi Ayaad, Morocco.
³ Department of Histopathology, Military Teaching Hospital Ibn Sina, University Cadi Ayaad, Morocco.
⁴ Department of Pneumology, Military Hospital Agadir, Morocco.

ABSTRACT

Introduction: Inflammatory myofibroblastic tumors (IMTs) arising in the mediastinum is rare. Their etiology remains unknown and their diagnosis is often overlooked before the use of surgery which allow the proper diagnosis and adequate treatment.

Case report: We report a case of 56-year-old woman that had a mediastinal mass discovered after a long complains of chest discomfort. Chest contrast-enhanced computed tomography (CT) showed a heterogeneously enhanced mass in the middle mediastinum. The diagnosis was confirmed by histopathology and immunohistochemical study after surgical resection through a thoracotomy. The patient was well and had no recurrence 6 months after surgery.

Conclusion: The diagnosis of IMT should be kept in mind and included in the differential diagnosis of mediastinal masses.

KEY WORDS: Inflammatory myofibroblastic tumor, mediastinum, surgery.

Correspondence: Pr Adil Arsalane, Department of thoracic surgery, Military Teaching Hospital Ibn Sina, University Cadi Ayaad, Marrakech, Phone: + 212 639174901 Email: arsalaneadil@gmail.com

This is an open access article distributed under the Creative Commons Attribution 4.0 International, which permits unrestricted use, distribution, and reproduction in any medium, provided the original work is properly cited.

INTRODUCTION

Inflammatory myofibroblastic tumor (IMT) is a rare benign tumor that most commonly occurs in the lung and orbit [1]. Middle mediastinum is an extremely rare location. Its origin is unknown, but recent studies have shown that it is a true tumor rather than a reaction process. [1-4] Its clinical and radiological manifestations are non specific. That's why diagnosis is difficult to establish prior to surgery or at least an ultrasonography-guided needle core biopsy. [5, 6, 7] We report an IMT in the mediastinum, for which only the histopathological findings after surgery have confirmed the diagnosis.

CASE REPORT

A 56 year old women with a history of right thoracic discomfort for 3 months, was referred to our department for further evaluation and eventual surgical treatment. She denied any history of other health problems. On admission, there was no fever nor other abnormalities on physical examination. His laboratory tests revealed a serum C-reactive protein concentration of 6.1 mg/dL and a normal count of white blood cell (WBC). Initial chest roentgenogram (figure 1) showed an abnormality in the right paratracheal region. Chest contrast-enhanced computed tomography (CT) (figure 2) showed a heterogeneously enhanced irregular mass in the middle mediastinum. The tumor had no calcification. After contrast administration, this tumor showed moderate homogeneous enhancement. The above imaging findings were highly suggestive of invasive tumor such thymoma or other mesenchymal tumors of mediastinum. A parangangioma and Castleman disease were also evoked. The dosage of serum tumor markers, including alpha-fetoprotein, human chorionic gonadotropin, carcinoembryonic antigen, and Cancer Antigen 19-9 were within normal limits.
The tumor was completely removed. Grossly, the mass was well demarcated weighing 180 g and measuring 8x7.5 cm (Figure 4).

The capsular surface was smooth. On cut section it was lobular, and a yellowish grayish firm mass with haemorrhagic and myxoid changes. Histologically, the tumour was composed of admixture of prominent chronic inflammatory cells including lymphocytes, plasma cells and histiocytes, and spindle-shaped cells with pale eosinophilic cytoplasm, oval nuclei, fine chromatine and inconspicuous nucleoli (Figure 5). Immunohistochemically, it was positive for vimentine (figure 6A), ALK-1 (figure 6B) and negative for CD34 (figure 6C). The morphological features along with immunophenotypical characteristics of the lesion support the diagnosis of inflammatory myofibroblastic tumour. The patient was discharged 5 days after the operation, and at a 6-month visit, she was well with no evidence of recurrence.

DISCUSSION
Inflammatory myofibroblastic tumor (IMT) is a rare disease most commonly found in the lung, liver, or spleen. however, its occurrence in the mediastinum is rare [8, 9]. The cause, pathogenesis, and long-term prognosis of inflammatory myofibroblastic tumor are unclear . [5] The (IMT) was initially reported as “inflammatory pseudotumor:” as a result of an exaggerated immunologic response by proliferated spindle cells and primary myofibroblasts to injury, inflammation, or infection. But it is now viewed as a true neoplasm because it can invade adjacent structures. [3,4]There are no specific signs, radiologic
manifestations, or symptoms related to IMT. These tumors could often be accompanied by elevated serum C-reactive protein and/or an increased WBC count, reflecting the inflammatory characteristics of this tumor. This laboratory parameters were normal in our patient. For these reasons, it’s impossible to make an accurate diagnosis prior to operation. [4, 10, 11] A definitive diagnosis is made based on the histopathological findings from either a resected tumor or a needle biopsy. [7] But from some authors, it is difficult to distinguish IMT from malignant tumors on the basis of small tissue samples obtained from needle biopsy [6, 12] Histologically, these tumors are characterized by the presence of a proliferation of spindle-shaped cells surrounded by chronic inflammatory cell infiltration. Immunohistochemical study is helpful in diagnosing and distinguishes IMT from other types of tumors, which usually show positive staining for smooth muscle actin and vimentin. [6] Anti-inflammatory therapy, 

AUTHORS’ CONTRIBUTIONS
The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the Recommendations for the Conduct, Reporting, and Publication of Scholarly work in Medical Journals of the International Committee of Medical Journal Editors. Indeed, all the authors have actively participated in the redaction, the revision of the manuscript and provided approval for this final revised version.

REFERENCES
[1] Almadi A, Rami M, Khattala K, Chater L, Harmouch T, Atmani S, Affifi A, Bouabdallah Y: Pseudotumour inflammatoire pulmonary chez un enfant. J Pediatr Puericulture 2011; 24: 69–71.
[2] Zhang Y, Dong ZJ, Zhi XY, Liu L: HU M: Inflammatory myofibroblastic tumor in lung with osteopulmonary arthropathy. Chin Med J 2009, 122:3094–3096.
[3] Fletcher CD, Unni KK, Mertens F. Pathology and genetics: Tumors of soft tissue and bone. World health organization classification of tumors. Lyon: IARC Press; 2002. p. 94-95.
[4] Berman M, Georgiou GP, Schonfeld T, Feinmesser M, Horev G, Vidne BA. Pulmonary inflammatory myofibroblastic tumor invading the left atrium. Ann Thorac Surg 2003; 76:601-603.
[5] Chen C H, Lin R L, Liu H C, Chen C H, Hung T T, Huang W C. Inflammatory Myofibroblastic Tumor Mimicking Anterior Mediastinal Malignancy. Ann Thorac Surg 2008; 86: 1362–1364.
[6] Meng X, Wang R. Inflammatory myofibroblastic tumor occurs in the mediastinum. Journal of Cancer Research and Therapeutics - October-December 2013 - Volume 9 - Issue 4
[7] Sugiyama K, Nakajima Y. Inflammatory myofibroblastic tumor in the mediastinum mimicking a malignant tumor. Diagn Interv Radiol 2008; 14:197–199
[8] Yamaguchi M, Yoshino I, Osogawa A, et al. Inflammatory myofibroblastic tumor of the mediastinum presenting as superior vena cava syndrome. J Thorac Cardiovasc Surg 2003; 126: 870 – 872.
[9] Gorospe L, Fernandez-Gil MA, Torres I, et al. Misleading lead: inflammatory pseudotumor of the mediastinum with digital clubbing. Med Pediatr Oncol 2000; 35: 484–487.
[10] Melloni G, Carretta A, Ciriaco P, et al. Inflammatory tumor of the lung in adults. Ann Thorac Surg 2005; 79: 426–432.
[11] Agrons GA, Rosado-de-Christenson ML, Kirejczyk WM, et al. Pulmonary inflammatory tumor: radiologic features. Radiology 1998; 206:511 – 518.
[12] Abdennadher M, Kolsi M, Khabir A, Abdelmalek M, Boudaoura T, Frikha I. Tumeur myofibroblastique pulmonaire : intérêt de la chirurgie première. Rev Mal Respir 2005 ; 22 : 1043-1047.
[13] Berger A, Kim C, Haqstrom N, Ferrer F. Successful preoperative treatment of pediatric bladder inflammatory myofibroblastic tumor with anti-inflammatory therapy. Urology 2007; 70: 372.e13-15.
[14] Bishop MK, Warmer BW, Dehner LP, Kriss VM, Greenwood MF, Geil JD. Successful treatment of inflammatory myofibroblastic tumor with malignant transformation by surgical resection and chemotherapy. J Pediatr Hematol Oncol 2003; 25:153-158.
[15] Cerfolio RJ, Allen MS, Nascimento AG. Inflammatory tumors of the lung. Ann Thorac Surg 1999; 67:933–936.
[16] Melloni G, Carretta A, Ciriaco P, et al. Inflammatory tumor of the lung in adults. Ann Thorac Surg 2005; 79: 426–432.
[17] Bahadori M, Liebow AA. Plasma cell granulomas of the lung. Cancer 1973; 31:191–208.
[18] Lee HJ, Kim JS, Choi YS, Kim K, Shim YM, Han J. Treatment of inflammatory myofibroblastic tumor of the chest: The extent of resection. Ann Thorac Surg 2007; 84:221-242.