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CASE REPORT

A RARE TUMOR OF THE MEDIASTINUM: INFLAMMATORY MYOFIBROBLASTIC TUMOR

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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 wich allow the propre 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.

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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 less 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 reffered 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 homogenous enhancement with central necrosis The above imaging findings were highly suggestive of invasive tumor such thymoma or other mesenchymal tumor of mediastinum. A paraganglioma and Castlesman 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.

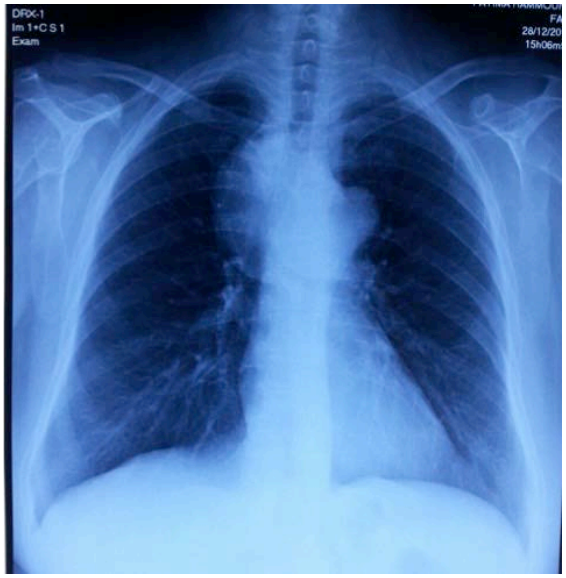

Figure 1: Chest Xray showed a right paratracheal opacity

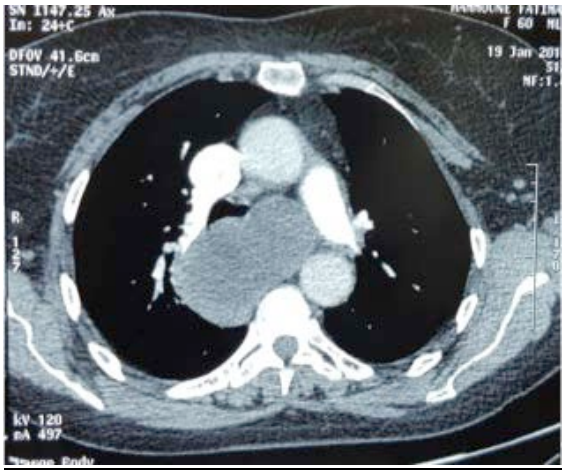

Figure 2 : Chest contrast-enhanced computed tomography (CT) heterogeneously enhanced irregular mass in the middle mediastinum

The resort to surgery was for diagnostic and therapeutic purposes. The patient underwent a right thoracotomy. During the operation it was found that the tumor located in the Barety lodge, encapsuled and firmly adherent to surounding tissues (figure 3).

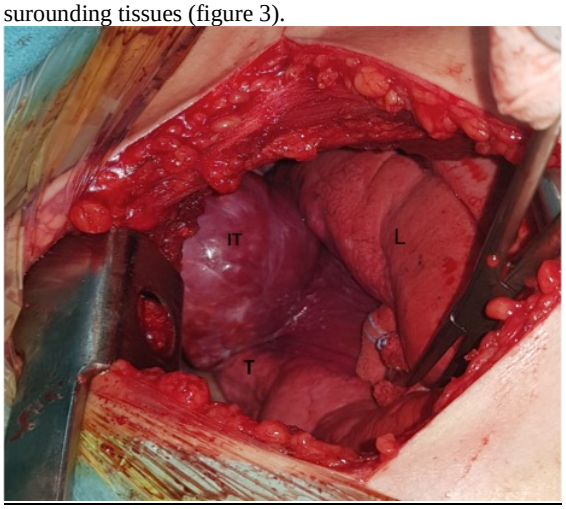

Figure 3 : Per-opératoiry view L : Lung T : Trachea IT : Inflammatory tumor

The tumor was completely removed. Grossly, the mass was well demarcated weighing 180 g and measuring 8x7.5 cm (Figure 4).

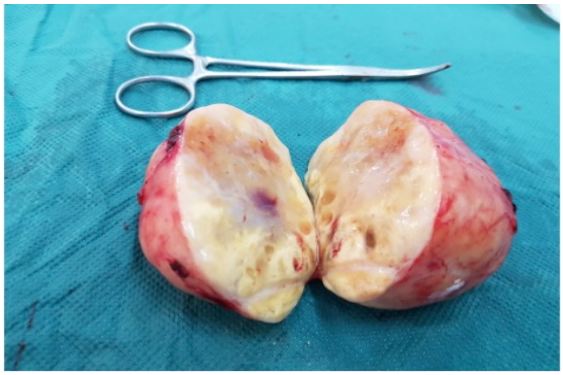

Figure 4 : Specimen piece

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 patiente was discharged 5 days after the operation, and at a 6-month visit, she was well with no evidence of recurrence.

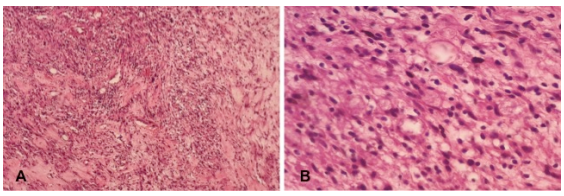

Figure 5: Histologic image of IMT showing myofibroblastic cells and chronic inflammatory cells (lymphocytes, plasma cells, and histiocytes) with areas of myxomatous degeneration (The H/E staining; (A: magnification ×10, B: magnification x40).

Figure 6 : Immunohistochemical study demonstrated (A) Vimentin (+), (B) ALK (+) and (C) CD34 (-)

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 patiente. 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 autors, it is difficult to distinguish IMT from malignant tumors on the basis of small tissue samples obtained from needle biopsy [6, 12] Histologicaly, 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, chemotherapy, and radiation therapy have all been tried. [13, 14] Surgical resection seems to be the treatment of choice. allowing for both proper diagnosis and adequate treatment. [15-17] Complete resection and achieving negative margins leads to excellent outcome. [12,18] Spontaneous regression of IMT has been reported sporadically in lung and other location testifying it inflammatory characteristics. [7]

CONCLUSION

Inflammatory myofibroplastic tumors are rare. The final diagnosis of these benign neoplasme, with agressive behavior, is rarely made before histopathological and immunohistochemical studies. IMT must be resected completely with negative margins in order to avoid récurrence and leading to excellent outcome. The diagnosis of these tumors should be kept in mind and included in the differential diagnosis of mediastinal masses.

AUTHORS' CONTRIBUTIONS

The participation of each author corresponds to the criteria of authorship and contributorship emphasized in the Recommendations for the Conduct, Reporting, Editing, 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.

PATIENT CONSENT

Written informed consent was obtained from the patient for publication of this case report.

COMPETING INTERESTS

The authors declare no competing interests.

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