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Case Report

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A Case of Inflammatory Myofibroblastic Tumour in the Lung: Diagnosis, Management, and Prognosis in a 48-Year-Old Female

Yasir A. Hasan[1*], Fiza Bashir[1], Bibhusal Thapa[1], Allan Pham[2]

Abstract

Background: Inflammatory myofibroblastic tumours (IMTs) are rare, low-grade neoplasms characterized by a proliferation of myofibroblasts with an associated inflammatory infiltrate. IMTs can occur in various anatomical locations, with pulmonary involvement being among the most common. Despite their indolent nature, these tumours can pose diagnostic and therapeutic challenges due to their variable clinical and radiological presentations.

Case Presentation: We present the case of a 48-year-old female who presented with chest pain. Imaging revealed an incidental nodule in the right lower lobe of the lung, initially suspected to be malignant. A biopsy demonstrated spindle cell proliferation with a mixed inflammatory infiltrate, leading to a diagnosis of inflammatory myofibroblastic tumours. The patient underwent a right lower lobectomy, with complete surgical resection confirmed histopathologically. Postoperative recovery was uneventful, and the patient remains disease-free at follow-up 18 months post-surgery.

Conclusion: This case underscores the importance of considering IMT in the differential diagnosis of pulmonary masses, especially in cases with inconclusive imaging findings. Surgical resection remains the cornerstone of treatment, offering both diagnostic confirmation and potential cure. Further studies are warranted to better understand the pathogenesis and optimal management of this rare entity.

Keywords: Inflammatory myofibroblastic tumors; Pulmonary IMT; Rare soft tissue tumour

Introduction

Inflammatory myofibroblastic tumours (IMTs) is a rare soft tissue tumour [1]. IMTs first described by Brunn in 1937, it represents rare form of inflammatory pseudo-tumour which affect mainly children and young people with a prevalence of 0.04% to 0.7% irrespective of race or gender of the world population [2]. Although the aetiology of IMTs is unclear, it is considered a tumour-like lesion with an unpredictable but favourable clinical course [3].

The lungs are the most common site of IMT onset, but it is also known to originate in other anatomic sites such as the retroperitoneum, abdomen, and pelvic cavity. Despite the low metastatic potential, surgery remains the gold standard of care for localized resectable disease. IMTs can recur locally but are rarely metastatic [3,4]. There has been much debate about whether IMTs are reactive lesions or actual tumours. There is now enough evidence

Affiliation:

[1]Division of Surgery, The Northern Hospital, Epping, Victoria, Australia

[2]Anatomical Pathology, The Alfred Hospital, Melbourne, Victoria, Australia

***Corresponding author:**

Yasir A. Hasan, Division of Surgery, The Northern Hospital, Epping, Victoria, Australia

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– including genetic alterations – to prove that the IMTs is a true tumour [5].

There are no obvious causes for IMTs, however; literature hypothetically attributed the causes of IMTs to abnormal body response to tissue damage such as viral infection (EBV, HPV type 8), acid reflux, trauma and over expression of IL-6 [1].

Radiologically, IMTs can present as multiple masses in one anatomical region. The finding can vary from an infiltrating lesion to a well-delineated lesion with different proportions of inflammatory and fibrotic components in the mass. On CT scan, variable attenuation can be noted with persistent and delayed contrast uptake in the fibrotic component of the IMTs. On MRI, the IMTs can show low signal intensity on T1- and T2- weighted images due to fibrosis and restricted diffusion [6].

Histologically, IMTs characterized by predominant spindle cells which are seen as interlacing fascicles among a polymorphous inflammatory infiltrate consisting of mature plasma cells and small lymphocytes [7].

Case Report

A 48-year-old female presented to the hospital with chest pain. Her Hb was 140, WCC was 11 with normal platelets. On further examination a CTPA was suggested which ruled out cardiopulmonary causes but also showed an incidental finding of a 10mm pulmonary nodule in the right lower lobe, with no symptoms. She was an ex-smoker. She had no underlying lung disease and no family history of cancer. On FDG PET, the nodule had poor avidity prompting a Dotatate scan which also showed only low-grade non-specific degree Dotatate activity.

A radial EBUS was done and A lesion was located in the right lower lobe and was solid in appearance. The radial probe was positioned within the lesion and was sampled by brushing. Cytology revealed aggregates of histiocytes with lymphoid material; no acute inflammation/granulomas and no evidence of neuroendocrine tumour.

Her case was presented in the MDM. As no cytological or histological evidence of malignancy was found and the lesion did not have typical radiologically malignant morphology (Image 1), the MDM suggested presenting options of ongoing follow-up and resection to the patient. These were presented to the patient who opted for resection. Due to the deep location of the tumour, she underwent a right lower lobectomy and frozen section. Frozen section showed a cellular proliferation of uncertain lineage, with inflammation however was unfortunately not able to completely rule out malignancy so mediastinal lymph node dissection was also performed.

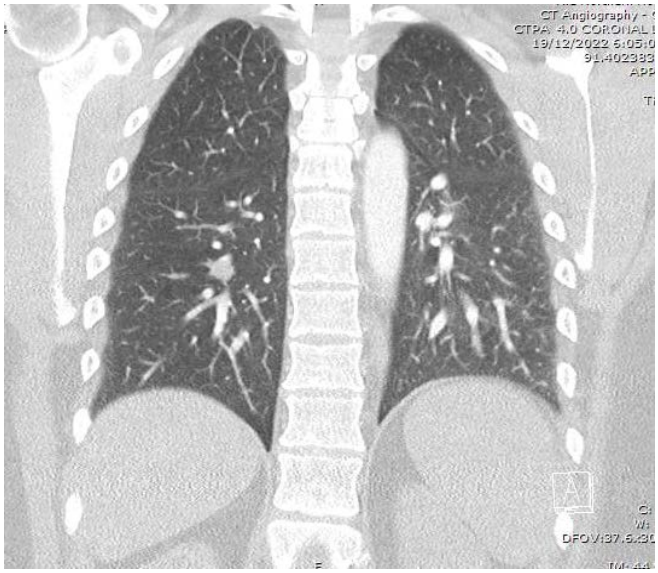

Image 1: Chest CT angiography showing right lower lobe nodule.

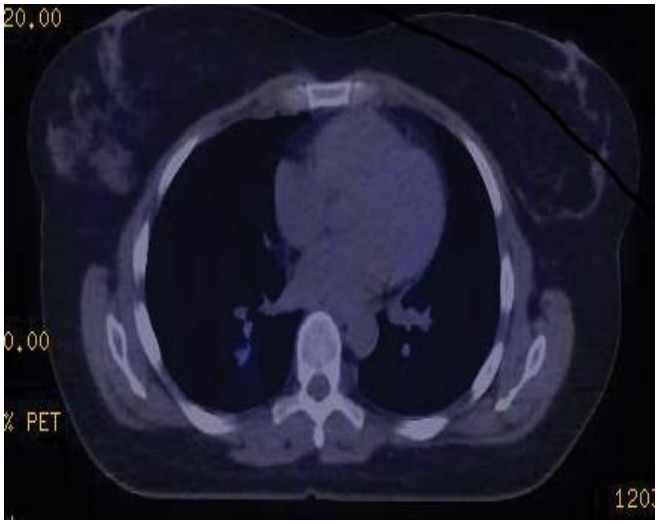

Image 2: Axial section of CT PET scan; right lower lobe 11 mm in size lung lesion shows low degree of Dotatate activity.

Pathological examination of the resected specimen showed a circumscribed and rounded peribronchial tumour, 12 mm in size (Image 3). Nodules of polygonal and spindled cells were accompanied by a very heavy plasma cell-rich inflammatory infiltrate (Image 4), consistent with a diagnosis of IMT. There was positive tumour cell immunohistochemical staining for vimentin, but no significant staining for anaplastic lymphoma kinase protein (ALK) or smooth muscle actin. Regional lymph nodes showed reactive histiocytosis.

The patient made an unremarkable postoperative recovery and was discharged on postoperative day 3. She underwent regular follow-up imaging, which showed no evidence of recurrence or metastasis after 18 months of follow up.

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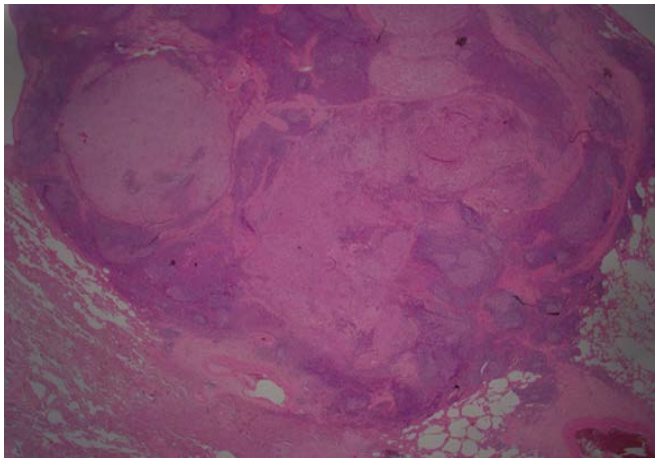

Image 3: A rounded, cellular peribronchial tumour (H&E x 12.5).

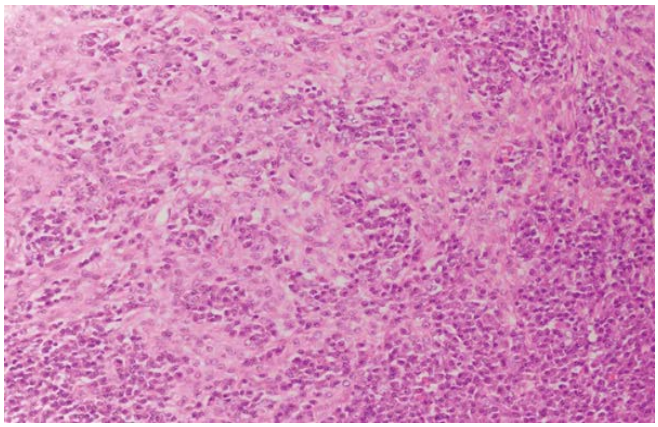

Image 4: Polygonal and spindled tumour cells accompanied by a heavy plasma cell-rich inflammatory infiltrate (H&E x 200).

Discussion

IMT of the lung is a rare neoplasm that can occur in individuals of any age but is most diagnosed in young adults [8]. The clinical presentation is variable but can include symptoms such as cough, chest pain, and shortness of breath.

The pathogenesis of IMT is not yet entirely understood, however; some studies have reported chromosomal aberrations involving the ALK gene, which may play a role in the development of IMT. In the present case, testing for the ALK gene rearrangement was not performed, so it is unclear if this gene was involved.

Surgical resection is the treatment of choice, and the prognosis is generally good, with low rates of recurrence or metastasis. Although there is no standard protocol for follow-up, imaging is typically used to monitor patients for any signs of recurrence or metastasis.

The overall 5- and 10-year disease free survival of 89%; in that series, the only risk factor for poor outcome was recurrence requiring re-operation [9]. The potential intra- and extra-thoracic metastasis of the disease always requires total body CT-scan and PET-CT before surgery and during follow-up [9].

In addition to surgery, other potentially feasible treatments include radiation and chemotherapy [3].

Conclusion

IMT of the lung is a rare neoplasm with an unpredictable clinical course. The diagnosis can be challenging, as the physical and radiological features can resemble other malignant or benign lesions. Surgical resection is the standard treatment, and regular imaging may be used to monitor patients for recurrence or metastasis. Further research is needed to better understand the pathogenesis of this uncommon neoplasm.

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