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Case Report

Pulmonary Inflammatory Pseudo-Tumor Due to Non-Tuberculous *Mycobacteria*

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Abstract

Inflammatory pseudo-tumor remains a diagnostic challenge because it's a rare tumor and its clinical and radiological manifestations are diverse and non-specific. It's a surgical pathology with good prognosis if completely resected. We present the case of a 43 year-old female with medical history of left eye enucleation due to an inflammatory pseudo-tumor associated to Non-Tuberculous *Mycobacteria*; later she presented with a pulmonary tumor, upper and middle lobectomies were performed with pericardial and parietal pleura resection, pathology studies revealed an inflammatory pseudo-tumor.

Keywords: *Inflammatory Pseudo-Tumor; Non-Tuberculous Mycobacteria; Surgical Pathology; Treatment*

Abbreviations

IPT: Inflammatory Pseudo-Tumor; IMT: Inflammatory Myofibroblastic Tumor; ALK: Anaplastic Lymphoma Kinase

Introduction

Inflammatory pseudo-tumor (IPT) of the lung, first described by Brunn in 1939 [1], the term had been coined by Umiker., *et al.* in 1954 [2] then the World Health Organization officially named it inflammatory myofibroblastic tumor (IMT) [3]. IPT is an uncommon solid tumor, that represents less than 1% of all lung specimen pathologies, usually affects children and young adults [4]. Given the heterogeneity in its pathogenesis, several terms have been linked to inflammatory pseudo-tumor. A clear and simple classification has been reported which divided into non-neoplastic and neoplastic variants [5]. Clinical and radiological findings of IPT are generally nonspecific and differentiation from malignant lung tumors remains a challenge. Complete resection represents the most favorable and recommended diagnostic and therapeutic approach for pulmonary inflammatory pseudo-tumors [5].

Case Report

A 43 year-old female, non-smoker, with medical history of left eye enucleation in 2014 with pathology report revealing an inflammatory peudo-tumor associated to Non-Tuberculous *Mycobacteria* and a positive culture with *Mycobacterium mucogenicum* sensitive to clarithromycin, quinolones and imipenem, therefore she received 10 days of oral moxifloxacin. She presented 3 years later to our hospital with a panuveitis of the right eye; all studies to exclude infection were negative but due to her medical history of Non-Tuberculous *Mycobacteria* infection we decided to perform a full body CT scan that showed an incidental 6 cm lung tumor in the middle lobe (Figure

1A). Our patient received intravenous amikacin, imipenem and oral clarithromycin; however, the lung tumor did not decrease in size after 2 months of therapy. Surgery was advised. A right posterolateral thoracotomy was carried out and upper and middle lobectomies were performed, due to infiltration of the upper lobe, en-block excision with parietal pleura and pericardium was required; pericardial reconstruction was done using bovine pericardium (SJM, Biocor Patch. St. Paul, MN. USA) (Figure 1B). Pulmonary ligament lymph node was enlarged and was resected. Pathology studies revealed an inflammatory pseudo-tumor due to Non-tuberculous *Mycobacteria* (Figure 2), Lowenstein-Jensen culture was negative, probably because of previous antibiotic therapy.

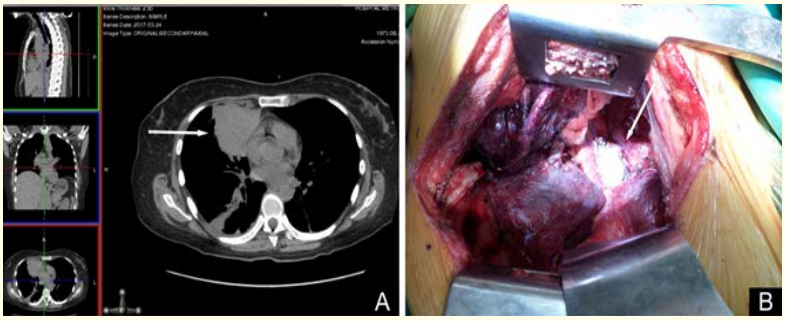

Figure 1: *(A) Computed tomography of the chest showed a right middle lobe tumor (arrow), (B) Right posterolateral thoracotomy, pericardial reconstruction was done using bovine pericardium (St. Jude Medical) (arrow).*

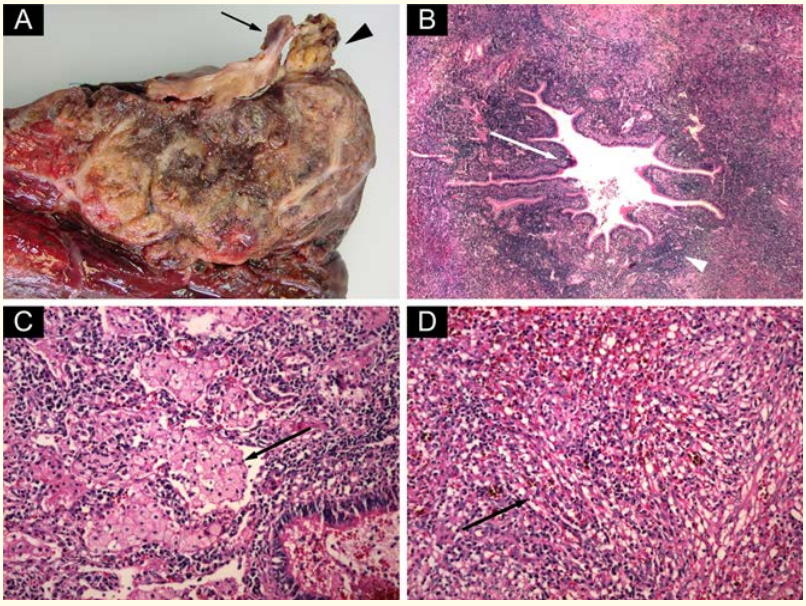

Figure 2: *(A) En-block excision with parietal pleura (arrowhead) and pericardium (arrow), (B) Microscopic appearance of partially collapsed bronchiole (arrow) surrounded by intense inflammatory process with areas of anthracosis (arrowhead), (C) Foamy histiocytes (arrow), (D) Intense inflammatory process that destroys the structure of the pulmonary parenchyma (arrow). (Hematoxylin and eosin staining; B: 10x, C: 20x, D: 40x magnification).*

Tumor-free surgical margins were achieved. Anaplastic lymphoma kinase (ALK) was negative.

The patient's hospital course was uneventful and was discharged home 10 days after surgery with long term antibiotic therapy recommended by infectious disease.

Discussion

Pulmonary inflammatory pseudo-tumor represents a diverse group of pathologies ranging from benign lesions to lesions with more malignant potentials such as inflammatory myofibroblastic tumor (IMT). Inflammatory pseudo-tumor of the lung is a rare entity, constituting around 0.7% of all lung tumors, and 0.04% - 1.2% of all thoracotomies [6].

Most commonly IPT presents in the lung and orbit but not necessarily together [5]; our patient presented the pathology in both locations, which is really infrequent. Other less common locations include: liver, spleen, stomach, breast, esophagus, salivary glands, heart, retroperitoneum, and the central nervous system [3].

Its pathogenesis remains elusive; the inflammation has been attributed to a metabolic disturbance, pulmonary infection (20%-30%), and/or antigen-antibody interaction to an unknown agent [7].

Isolated pathogens include: *Human Herpes Virus, Epstein Barr Virus, Nocardia, Mycoplasma, Actinomycetes*.

IPT occur more frequently in the lower lobes with a predilection for peripheral lung parenchyma and subpleural locations [8], in our patient the tumor infiltration was present in the parietal pleura and pericardium.

ALK (Anaplastic lymphoma kinase) gene has been used as a molecular surrogate to differentiate benign from malignant variation [5]. ALK positive is only observed in IMT patients, and approximately half of this group is ALK positive; in contrast ALK negative is associated with higher rate of metastases and worsen prognosis [9].

Histologically, consists of proliferation of fibroblasts and myofibroblasts intermingled with inflammatory cells including: lymphocytes, histiocytes, macrophages and polyclonal plasma cells [9].

Almost 70% are asymptomatic or with symptoms of other diseases, so they are discovered incidentally, as was the case of our patient [5].

Definitive diagnosis of IPT is mainly achieved by surgical resection, and if it is complete, represents the definitive therapy.

The prognosis is variable, depends on the tumor size and the magnitude of surgical resection. After complete resection the prognosis is favorable, with 5 and 10 years survival of 91% and 77%, respectively. Positive margins after incomplete resection result in recurrence rate from 5% to 25% [10].

Conclusion

Pulmonary inflammatory pseudo-tumor is a rare entity, but it should be considered in the differential diagnosis of pulmonary lesions, most of these tumors are asymptomatic with nonspecific imaging characteristics.

Usually IPT's present in the lung and orbit; the majority show a benign clinical course.

Its pathogenesis remains elusive; Non-tuberculous *Mycobacteria* may be a cause of inflammation in these tumors.

Despite being a benign lesion, its potential for local invasion and recurrence requires a complete surgical resection and long term follow-up is needed.

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