A rare and catastrophic manifestation of mycobacterium Avium complex pulmonary disease: A case report

Enrique O. Ortiz-Diaz

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Case Report: The patient’s clinical setting illustrates an unusual and fatal manifestation of NTM, a left lung necrotizing-cavitating pneumonia in an immunosuppressed host.

Conclusion: Severe necrotizing pneumonia is a rare manifestation of NTM considered after exclusion of other cavitating lung diseases. Also, it adds to the current literature whether anti-neutrophil cytoplasmic antibodies are associated with mycobacterial infections.
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Keywords: Cavitating Lung Disease, Immunosuppression, Non-tuberculous mycobacteria

INTRODUCTION

Non-tuberculosis mycobacterial (NTM) infections in the lung usually present as a chronic fibrocavitary disease, an allergic reaction, or as an extra-pulmonary dissemination in patients with T cell immunosuppression [1]. The case illustrates an uncommon presentation of NTM pulmonary disease in the form of severe necrotizing pneumonia and bronchopleural fistula.

CASE REPORT

A 65-year-old Mexican man with rheumatoid arthritis was admitted to the hospital with left-sided chest pain, productive cough and progressive dyspnea over a week. Progressive pleuritic chest pain, disabling dyspnea (New York Heart Association Class IV) and productive cough were reported 10 days prior to admission. His review of systems was affirmative for chills, rigors, and a 20-pound weight loss in an indeterminate amount of time. Patient had a baseline chronic non-productive cough, which changed in terms of brown phlegm production without blood.

Past medical history was remarkable for long standing rheumatoid arthritis for which he took dexamethasone (10 mg/day) and various over-the-counter non-steroidal anti-inflammatory agents on a daily basis without medical guidance. When the patient arrived to the emergency room, a left pneumothorax was diagnosed by chest radiograph and a 28 Fr chest tube was surgically placed before being admitted to the general medical ward (Figure 1).

His physical examination was significant to find an elderly patient with chronic ill dishevelled appearance, using a nasal cannula at 6 L/min. The patient was afebrile with sinus tachycardia and a blood pressure of 140/65 mmHg. Chest tube was in the left lateral hemithorax. The chest drain had abundant yellow fluid with air bubbles noticeable on passive expiration without coughing. On auscultation of the chest, patient had bilateral mid-to-end inspiratory “velcro” crackles at the bases,
decreased breath sounds on the left anterior and lateral hemithorax along with dullness to percussion in the same area. Heart was found to have regular tachycardia without rub or murmurs. There was non-palpable cervical lymphadenopathy but there was poor dentition without visible jugular venous distention. Abdomen and extremities were unremarkable.

Hematological findings included thrombocytosis and leukocytosis with neutrophilia. Electrolytes were unexceptional. Chest radiographs before and after chest tube insertion are shown in Figure 1. Chest radiograph on the right showing a partially expanded but consolidated left lung, along with deep sulcus sign, a central cavity and a chest tube resting superiorly and medially. A computed tomography of the chest showed hydro-pneumothorax with upper lobe cavities and bibasilar fibrotic changes with honeycombing (Figure 2). Serology reported positive rheumatoid factor (941 units by nephelometry) and anti-myeloperoxidase anti-neutrophil cytoplasmic antibody (MPO-ANCA) by ELISA. 

*Mycobacterium tuberculosis* polymerase chain reaction (PCR) probe returned negative. Bacterial and fungal cultures were finalized as negative. Sputum and pleural fluid acid-fast cultures recognized acid-fast positive organisms finalized as non-tuberculous mycobacteria (NTM), specifically MAC (*Mycobacterium intracellulare*).

Patient received broad-spectrum antibiotics targeting *S. aureus* and *Pseudomonas aeruginosa*. When, acid-fast stains returned positive results, patient was added 4-drug standard therapy for *Mycobacterium tuberculosis* on hospital day-2. When *M. tuberculosis* PCR was found negative. The patient was changed to clarithromycin 500 mg orally twice daily, ethambutol 15 mg/kg daily and rifampin 600 mg orally daily on the fourth hospital day. The patient progressed to severe hypoxemic respiratory failure undergoing orotracheal intubation on hospital day-5 and invasive mechanical ventilation. Patient’s introduction to positive pressure ventilation caused the patient to worsen bronchopleural fistula airflow and die of refractory hypoxemia in the intensive care unit at the sixth day of admission. An autopsy was not obtained.

**DISCUSSION**

The patient presented with an acute-subacute left upper lobe cavitating-necrotizing pneumonia in the setting of steroid-mediated immunosuppression and baseline fibrotic parenchymal abnormalities. The differential diagnosis encompasses bacterial (*Staphylococcus aureus, Klebsiella* species, *Pseudomonas aeruginosa*), fungal (endemic fungi, *Aspergillus*, etc). Other bacterial etiologies causing a more chronic, less clinically catastrophic complications include *Nocardia* and *Actinomyces* species. Parasites such as *Echinococcus* and *Paragonimus* species have been described to cause a similar setting. Since, the patient was born and frequently travelled to Mexico, *Mycobacterium tuberculosis* should be considered as well. This would be an unusual presentation for NTM as explained in the discussion below. Less likely in this case, pulmonary infarction, Caplan’s syndrome (rheumatoid arthritis), vasculitis, (e.g., Wegener’s granulomatosis), and cavitating neoplasms (squamous cell cancer) [2].

**NTM Pulmonary Infections Presentation**

The patient illustrates an unusual manifestation of NTM in the immunosuppressed host. NTM group encompasses over 150 species excluding *Mycobacterium leprae* and *M. tuberculosis* [3]. NTM clinical manifestations depend on the host’s immune competency and geographical location. Over 115 species of NTM have been reported to cause disease in humans [3]. MAC is the most common pathogenic NTM in North America [4]. The estimated NTM associated pulmonary disease prevalence is increasing compared to three decades ago [4]. Classically, NTM clinical presentation ranges from chronic fibrocavitary-nodular disease (most commonly in patients with pulmonary structural abnormalities.
such as chronic obstructive pulmonary disease and bronchiectasis) to allergic disease (hypersensitivity pneumonitis). Both entities are described in patients with non-specific systemic symptoms, dyspnea, cough, specific high resolution computed tomography abnormalities and sputum, bronchoalveolar or pleural culture results (or histological diagnosis) [1, 5]. Considering immunocompromised hosts, specifically with CD4+ T cell deficiency (prototypically acquired immune deficiency syndrome), disseminated or extra-thoracic NTM infection is well described. This subgroup variably presents with fever of unknown origin, gastrointestinal symptoms, generalized lymphadenopathy, cytopenias and less commonly diffuse skin nodules, pustules, and ulcers [1]. Since NTM could be considered a contaminant, specific context and exclusion of other diseases must exist to ascertain the diagnosis [1, 5].

Necrotizing pneumonia is an infrequent presentation of NTM even in immunocompromised hosts. The patient described in the scenario above represents the first reported to have such a severe cavitary-necrotizing pneumonia, bronchopleural fistula causing refractory hypoxemia. Other case reports exist for milder acute presentations [6, 7]. Waller et al. reported an immunocompetent southeastern American woman with microbiological and surgical pathological diagnosis of multi-lobar MAC pneumonia with hypoxic respiratory failure [7]. Asnis et. al. described an immunocompetent gentleman with right upper lobe pneumonia and eccentrically large mediastinal adenopathy. A right paratracheal node biopsy yielded the diagnosis during the patient's hospital course [6].

The described patient's serological studies revealed a positive ANCA-MPO antibody. There is an overlap in symptoms, radiological and pathological findings due to MAC infection and pulmonary vasculitis as they occupy the same differential diagnosis for this presentation. Others have found co-occurrence of ANCA antibodies in chronic suppurrative infections such as Mycobacterium tuberculosis and NTM infections [8–12]. Chaimnuay et al. described an elderly lady with positive ANCA vasculitis and a lung lesion that recovered in culture MAC. Patient responded clinically to anti-mycobacterial treatment without initiating immunosuppressive therapy [8]. Nakayama et al. reported a case of an elderly lady with pulmonary artery vasculitis-stenosis and a right upper lobe cavitary lesion. Sputum culture recovered MAC. The patient was treated with immunosuppressive and antimycobacterial therapy with good clinical-radiological response [9]. There are unanswered questions whether there are associations between the two since it has been only described through case reports.

CONCLUSION

Non-tuberculous mycobacteria should be considered in the necrotizing-cavitating pneumonia differential diagnosis. It is the first report to describe such a severe manifestation of NTM disease. The case adds to the current literature whether anti-neutrophil cytoplasmic antibodies are associated with mycobacterial infections.

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Author Contributions

Enrique O. Ortiz-Diaz – Substantial contributions to conception and design, Acquisition of data, Analysis and interpretation of data, Drafting the article, Revising it critically for important intellectual content, Final approval of the version to be published

Guarantor

The corresponding author is the guarantor of submission.

Conflict of Interest

Authors declare no conflict of interest.

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