Pulmonary nocardiosis with superior vena cava syndrome in HIV-infected patient: A rare case report in the world

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Pulmonary nocardiosis with superior vena cava syndrome in HIV-infected patient: A rare case report in the world

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Summary (200 words)

Pulmonary nocardiosis is a common disease among HIV-infected patients. In most
cases, the disease progresses slowly. Here, we present a case whose disease progressed
rapidly. A 35-year-old female with AIDS and superior vena cava (SVC) syndrome who was
lost to follow-up visited our hospital. She presented with a chronic non-productive cough and
her CD4 count was 33 (4%). Her chest x-ray showed opacity in the right upper lobe of her
lung and her sputum acid-fast stain was negative. Anti-tuberculosis agents were prescribed.
Two weeks later, superficial vein dilatation appeared on her chest wall and her chest x-ray
became worse. CT chest showed a mass in her right lung. The size of the mass was 9.6 x 9.8
× 8.3 cm. The mass was heterogeneous. Necrotic mediastinal nodes nearly obliterated the
SVC. Gram-positive beading and branching filamentous organisms were identified in her
sputum by modified acid-fast stain. She was diagnosed with pulmonary nocardiosis. This
diagnosis was confirmed by culture. She had Nocardia beijingensis with SVC syndrome. She
responded to treatment. After two weeks of parenteral agents, we switched her to oral
trimethoprim/sulfamethoxazole which was later followed by antiretroviral agents.
Nocardia is a genus of aerobic actinomycetes responsible for localized or disseminated infections in animals and humans. Humans are infected when they come into direct contact with the organism via the skin or soft tissues or by inhalation (1). Nocardiosis, especially N. cyriacigeorgica, N. nova, and N. farcinica, are the most common organisms that affect immunocompromised patients with acquired immune deficiency syndrome (AIDS), malignancy, or those who have undergone solid organ transplantation (SOT), and in those on long-term steroid therapy (1,2). Pulmonary nocardiosis is the most common clinical presentation of the infection. The onset of symptoms may be subacute to more chronic and can include productive or nonproductive cough, shortness of breath, chest pain, hemoptysis, fever, night sweats, weight loss, and progressive fatigue (3).

Here, we report a case of pulmonary nocardiosis with superior vena cava (SVC) syndrome caused by Nocardia beijingensis in an AIDS patient.

A 35-year-old Thai female AIDS patient, living in Thailand. She was diagnosed with asymptomatic human immunodeficiency virus (HIV) infection 11 years ago during antenatal care. At that time, her initial CD4+ count was 549 cells/μL. She received azidothymidine (AZT) and nevirapine (NVP) during labor and then was lost to follow-up.

Four years later, she came to the hospital because she had a fever and progressive dyspnea for two months. She was diagnosed with disseminated tuberculosis (miliary tuberculosis with hepatosplenic abscess and tuberculous meningitis) and pneumocystis jirovecii pneumonia. She was treated with anti-tuberculous drugs for 10 months and trimethoprim-sulfamethoxazole plus prednisolone for 21 days. After treatment, she completely improved clinically and was prescribed with antiretroviral agents tenofovir, emtricitabine, and efavirenz. She was then lost to follow-up for 3 years.
Three months before readmission, she had a fever with non-productive cough and significant weight loss. Two months before readmission, whenever she coughed, she had chest pain at her right chest. Three weeks before readmission, she came to the hospital with progressive dyspnea. Her chest x-ray revealed a new alveolar infiltration at the right upper lung (RUL) zone. Sputum was collected and subjected to acid-fast staining and polymerase chain reaction (PCR) for mycobacterium tuberculosis. Both tests were negative for mycobacterium tuberculosis. Nevertheless, she was diagnosed with pulmonary tuberculosis and received the standard regimen for tuberculosis.

Three weeks later, she came to the hospital again for her follow-up visit. She still had fever, cough and dyspnea. Chest x-ray result showed that there was a progression of the alveolar infiltration at the RUL (Figure 1). She was found to have a fever of 42°C, respiratory rate of 26 times per minute and room-air SpO2 value of 98%. Physical examination revealed moderately pale conjunctivae, oral thrush, oral hairy leukoplakia, swelling at the right side of the neck, superficial vein dilatation at the chest wall, trachea in midline, equal lung expansion, no stridor, dullness on percussion at RUL, decreased breath sounds at RUL, coarse crepitations at RUL, increased vocal resonance at RUL. Although SVC syndrome was impressed, there was no sign of increased intracranial pressure such as papilledema upon fundoscopic examination. Multiple bilateral cervical, right axilla and right inguinal lymph nodes were swollen.

Complete blood counts showed moderate anemia without leukocytosis (Hb 5.4 g/dL, Hct 17.7%, WBC 9910/uL, N 83%, and L 8.2%). Blood chemistry showed normal renal function and mild hyponatremia (Na 134 mEq/L). Blood culture for bacteria, fungus and mycobacterium tuberculosis were negative. Her CD4+ count was 33 cells/uL (4%) and HIV viral load was 55,488 copies/mL.
Computerized tomography (CT) scan detected a heterogeneous mass size 9.6 x 9.8 x 8.3 cm at RUL adjacent to the necrotic mediastinal nodes, total atelectasis of the RUL, segmental near total obliteration of SVC, azygos vein and total obliteration of the anterior segmental pulmonary artery (Figure 2). Gram positive beaded branching filamentous organism was also found via modified acid-fast staining of her sputum (Figure 3). She was diagnosed with pulmonary nocardiosis. Sputum culture confirmed that she had *Nocardia beijingensis* with SVC syndrome. The species was identified by MALDI-TOF mass spectrometry. The organism was susceptible to trimethoprim-sulfamethoxazole, amikacin and ceftriaxone but resistant to imipenem and ciprofloxacin.

Empirical antibiotic therapy with intravenous trimethoprim-sulfamethoxazole were administered. Three days later, she developed maculopapular rash so the medications were changed from trimethoprim-sulfamethoxazole to intravenous ceftriaxone and amikacin. Consequently, we consulted the immunologist to desensitize her to trimethoprim-sulfamethoxazole. After desensitization, the maculopapular stopped developing. She dramatically responded to treatment.

After two weeks of parenteral agents, oral trimethoprim/sulfamethoxazole was administered followed by antiretroviral drugs. Later, she remained well for over 1 year and came regularly for follow-up at our outpatient clinic.

More than 60% of all reported cases of nocardiosis were associated with significant preexisting immune compromise, ranging from alcoholism and diabetes to chronic granulomatous disease, organ transplantation, and AIDS (1). In Thailand, 80% of nocardiosis
patients have underlying disease of which 34% had AIDS and of those AIDS patients, 42.8% of the cases had underlying diseases (4).

Pulmonary disease is the predominant clinical presentation of nocardiosis and is acquired through inhalation of the organisms from the environment. Any species may cause lung infection, but the most common are *N. cyriacigeorgica, N. nova, and N. farcinica*. The onset of symptoms may be subacute or chronic and include one or more of the following signs: productive or nonproductive cough, dyspnea, hemoptysis, fever, etc (1). The most common features are single or multiple nodules and airspace consolidation. Notably, cavitation coupled with nodules, masses or consolidations were observed in most patients. Other findings that have been reported are Ground Glass opacity (GGO), air bronchograms, bronchiectasis, lymphadenopathy, pleural effusion and pericardial fluid (4,5).

Pulmonary nocardiosis may occasionally complicate advanced immunodeficiency virus (HIV) infection, where it often presents with alveolar infiltrates that progress rather than that of cavity disease. There are few reports of patients with pulmonary nocardiosis presented with SVC syndrome (6,7). All patients had immunocompromised status but not HIV infection. In a previous Thai study, 17.7% of the patients had *N. beijingensis* (8). This report showed a rare case of *N. beijingensis* that was presented with SVC syndrome in an HIV-infected patient. To our knowledge, this is the first case of pulmonary nocardiosis from *N. beijingensis* in Thailand and also the first case in the world diagnosed with pulmonary nocardiosis and co-infected with HIV presenting SVC syndrome.

Trimethoprim-sulfamethoxazole is the mainstay of treatment for nocardiosis. Clinical improvement is generally evident within 3 to 5 days after initiation of appropriated therapy as shown in our patient. Duration of treatment is generally prolonged to minimize risk of disease relapse. Immunosuppressed patients and those with central nervous system (CNS) disease
should receive at least 12 months of antimicrobial therapy with appropriate clinical monitoring tests (1,3).

In conclusion, we report a case of pulmonary nocardiosis in an AIDS patient who presented with SVC syndrome. This is uncommon and should be considered in differential diagnosis. Specific therapy resulted in a cure of the infection and a long-term state of clinical well-being.

**Conflict of Interest:** None to declare
References


Figure legends

**Figure 1.** Chest x-ray taken three weeks before readmission (left), was compared to the one taken three weeks later (right) showed that there was infiltration in the right upper lung.

**Figure 2.** The CT chest showed a 9.6 x 9.8 x 8.3 cm heterogenous mass in the right lung with necrotic mediastinal nodes nearly obliterating the SVC.

**Figure 3.** Sputum modified acid-fast staining yielded a positive result.
Figure 1