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Disseminated nocardiosis in a female patient with idiopathic thrombocytopenic purpura: A case report

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CASE STUDY

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ABSTRACT

Nocardiosis is a life-threatening disease if unrecognized and maltreated. We describe a case of disseminated nocardiosis in a patient with idiopathic thrombocytopenic purpura under steroid therapy. She presented with a 2-week history of progressive symmetrical limb weakness, fatigue, and profuse sweating. Brain MRI revealed ring-enhanced lesions at the cerebellum and left parietal lobe with brain oedema. Chest CT revealed a left upper lobe nodule. Aspirate culture confirmed the diagnosis of nocardiosis. We administered antibiotics and dexamethasone to ameliorate the brain oedema. The patient improved clinically after 2 weeks. Follow-up brain MRI showed improvement. Clinicians should consider nocardiosis in immunocompromised patients with non-specific symptoms.

Key Words
Nocardiosis, idiopathic thrombocytopenic purpura, *Nocardia*

Implications for Practice:

1. What is known about this subject?
Nocardiosis is a rare opportunistic infection caused by *Nocardia* species that predominantly affects immunocompromised patients. There are four case reports in the literature.

2. What new information is offered in this case study?
Patients with disseminated nocardiosis can be treated with Bactrim, amikacin, meropenem, and dexamethasone.

3. What are the implications for research, policy, or practice?
Corticosteroids are considered a strong risk factor for disseminated nocardiosis. Radiological and microbiological work-up are mandatory to diagnose this condition.

Background

Nocardiosis is a rare opportunistic infection caused by Nocardia species that predominantly affects immunocompromised patients.[1] Nocardia is a gram-positive, branching, rod-shaped aerobic bacterium from the genus Actinomyces[2] that can be found in the soil, air, water, decomposing vegetation, and other organic matters. Human infections are usually caused through contact with contaminated soil, when bacteria can enter through the respiratory tract or skin wounds.[3] Corticosteroid therapy is considered the most important risk factor, followed by

human immunodeficiency virus infection, solid organ transplant, cancer, chronic pulmonary disease, and autoimmune disease.[4] Corticosteroids have profound effects on the cellular functions of leukocytes, impairing their entry into infection sites, reducing the clearance of bacteria by the reticuloendothelial system, and causing lymphopenia and redistribution of lymphocytes to other sites, affecting T cells more than B cells.[5] Single-organ infection most commonly manifests as lung disease, followed by infection of the central nervous system.[6] Previously reported neurologic symptoms include headaches, visual disturbances, ataxia, seizures, focal neurological deficits, and myelopathy. Early identification of the organism, allowing proper antimicrobial therapy, can be made only through biopsy of the lesion and culture.[7] Herein, we present a case of disseminated nocardiosis in a patient with idiopathic thrombocytopenic purpura (ITP).

Case details

A 60-year-old Saudi female, a housewife, presented with a 2-week history of progressive symmetrical limb weakness associated with fatigue and profuse sweating. The weakness started at the proximal upper limbs and affected both lower limbs three days later. The patient had a history of primary hypertension and dyslipidaemia for 10 years, non-insulin dependent diabetes mellitus for five years, and ITP diagnosed three months previously. She was on corticosteroid therapy with tapering doses; she had received the last dose one week before presentation. She had no history of surgeries, allergies, or blood transfusion. There was no family history of neurological diseases as well as no other family members had developed similar symptoms.

On physical examination, the patient had stable vital signs and was conscious, alert, and oriented to time, place, and persons. Cranial nerve examination revealed pupils equal in size and equally reactive to light, constricting 3mm. The remaining cranial nerves were unremarkable. Motor examination revealed increased muscle tone at the right upper and lower limbs, decreased muscle strength of 2/5 at the bilateral hip flexors and knee extensors and 3/5 at the bilateral ankle dorsiflexors. Deep tendon reflexes were +3 on the right side and +2 on the left side, with a downgoing plantar response bilaterally. Sensory examination was unremarkable for all sensory modalities. There was dysmetria on right finger-to-nose testing and Romberg's sign and gait was difficult to assess.

From the laboratory investigations, there was increased erythrocyte sedimentation rate (ESR) at 115mm/hr and increased C-reactive protein (CRP) level at 5.1mg/L. Other routine blood tests, hepatic and renal function tests, electrolytes, and coagulation function tests were within the normal range. Cerebrospinal fluid (CSF) and serum mycobacterium polymerase chain reaction was negative. CSF analysis findings were as follows: red blood cell count 145, white blood cell count 2700, neutrophils count 85, lymphocytes count 12, glucose 86mmol/L, and protein 180mg/dL. CSF Gram stain was negative. After aspiration from subcutaneous collection in the cervical region, Gram staining showed Gram-positive bacilli and modified acid-fast culture staining revealed Nocardia. The patient was diagnosed with disseminated nocardiosis.

On radiological work-up, chest computed tomography showed a speculated nodule in the left upper lobe with several chest wall intramuscular cystic lesions (Figure 1). Axial T1 magnetic resonance imaging (MRI) of the brain with contrast (Figure 2) revealed lobulated ring-enhanced lesions at the cerebellum and one ring-enhanced lesion at the left parietal lobe surrounded with moderate vasogenic oedema. Sagittal T1 MRI of the cervical spine with contrast (Figure 3) revealed diffuse leptomeningeal enhancement and partially visualized subcutaneous collections at the cervical and thoracic region showing peripheral enhancement.

After receiving the culture result, the patient was started on dexamethasone 4mg intravenously (IV) Q8h in order to decrease the vasogenic oedema, Bactrim 320mg IV Q12h, amikacin 500mg IV OD, and meropenem 2g IV Q8h. The patient improved clinically one week after therapy initiation, as noted by physical examination: the muscle strength of both upper and lower limbs increased to +4/5. The fatigue and sweating reduced. After 13 days of treatment, the ESR decreased from 115mm/hr to 50mm/hr and CRP decreased from 5.10mg/L to 0.20mg/L. Follow-up brain MRI with contrast (Figure 4) after one month showed overall interval regression of the previously seen multiloculated peripherally enhancing lesions with minimal perilesional oedema.

Discussion

Generally, Nocardia is an opportunistic bacterium that mainly infects immunocompromised patients. Our patient was HIV negative, but was receiving methylprednisolone for a year and was thus immunocompromised.

There are only four previous reports on disseminated nocardiosis in patients with chronic ITP.[8-11] The most commonly used immunosuppressant agents were corticosteroids; followed by chemotherapeutic drugs, such

as cyclophosphamide, azathioprine, and methotrexate; calcineurin inhibitors, such as cyclosporine and tacrolimus; and anti-TNF agents.[6]

The first case was published in 1994[8]; the patient had undergone brain surgery due to brain abscess. The authors concluded that nocardial brain abscesses remain difficult to cure; the mortality rate of patients with these abscesses was more than three times higher than that of patients with other bacterial brain abscesses.

The second case was published in 1999[9] and described a patient with Evans' syndrome, a disease requiring long-term immunosuppression, who acquired systemic nocardiosis. The infection was primarily pulmonary, misdiagnosed as tuberculosis, with subsequent haematogenous dissemination to the skin and central nervous system. The cerebral involvement was difficult to prove as the patient presented with stroke-like episodes. After a positive blood culture was obtained, anti-tuberculosis therapy was introduced. "However, the patient's condition deteriorated and he died. Nocardiosis was proven post-mortem in the brain with infiltration of the meninges, lungs, skin, and kidneys.

The third case was published in 2004.[10] The patient had a brain abscess which was aspirated under intensive platelet support. Antibiotics were administered concurrently. Perioperative and postoperative periods were uneventful. The patient started improving three days after the aspiration. He was followed up for 35 months, being on mono-drug therapy with co-trimoxazole the initial 12 months. His muscle weakness improved, and follow-up MRI showed resolution of the abscess with remnant minimal perifocal oedema.

The fourth case was published in 2008.[11] The patient had a history of ITP and disseminated nocardiosis, and showed initial improvement in the skin and lung lesions, but there was no improvement in the cerebral lesions. As there was subsequent neurological deterioration, the right frontal abscess was surgically excised under intensive platelet cover. He initially showed improvement but eventually succumbed to his illness after four months of treatment. Details of the treatment were not provided.

Literature shows that all patients with ITP received steroid therapy in different duration. Although there have been no randomized clinical trials demonstrating the best therapeutic regimen, the best diagnostic method was aspiration and culture. We believe that our combination of different antibiotics and dexamethasone enabled the most effective and fastest treatment for disseminated nocardiosis compared to all other reported cases.

Conclusion

In conclusion, this case demonstrates that immunodeficiency due to HIV or organ transplant is not necessarily the reason behind Nocardia infection. There are only four previous reports on disseminated nocardiosis in patients with chronic ITP, and this case can be added to them. Corticosteroids, even if taken for a short period, can be the reason behind this infection. Thus, the possibility of nocardiosis should be considered in patients with current corticosteroid therapy or history of diseases requiring such treatment.

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CONFLICTS OF INTEREST

The authors declare that they have no competing interests.

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PATIENT CONSENT

The authors, *Foziah A, Mohammed A, Noor A, Leenah T, Ibrahim A, Saeed A, Afnan A,* declare that:

1. They have obtained written, informed consent for the publication of the details relating to the patient in this report.
2. All possible steps have been taken to safeguard the identity of the patient.
3. This submission is compliant with the requirements of local research ethics committees.

Figure 1: High-resolution computed tomography scan of the chest shows a left upper lobe speculated nodule

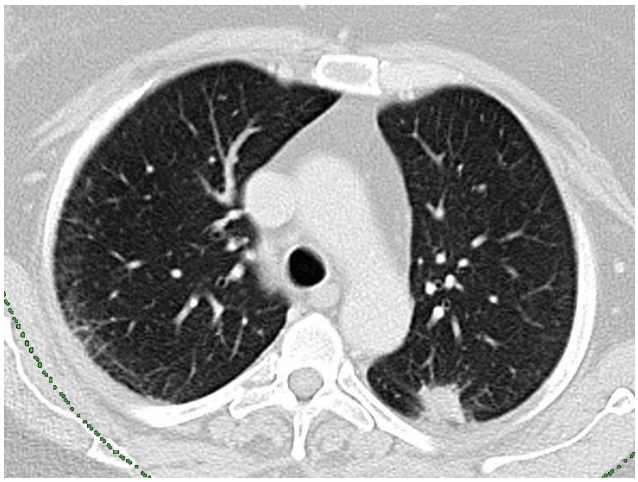

Figure 2: Magnetic resonance image of the brain with contrast (axial, coronal, and sagittal view) revealed multiple multiloculated ring-enhancing lesions at the left parietal region and right superior cerebellar peduncle extending to the mid brain surrounded by vasogenic oedema with mass effect upon the adjacent structures

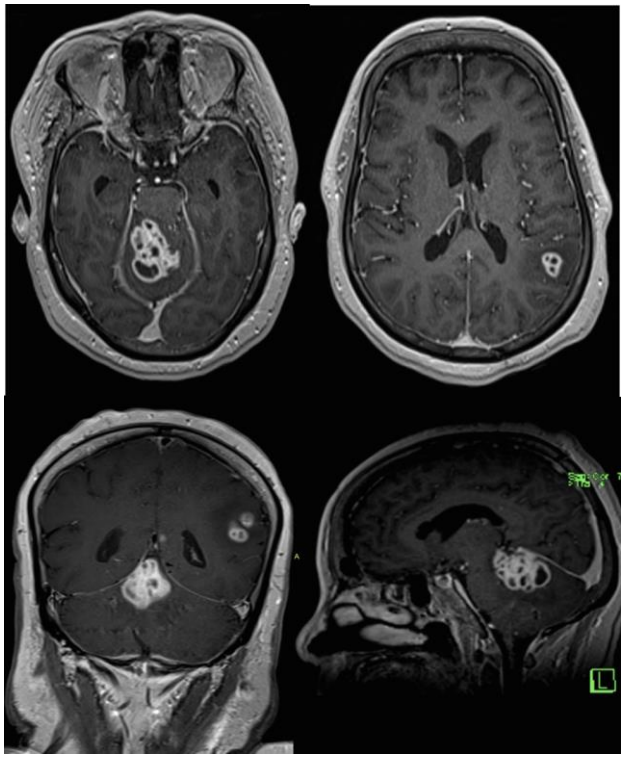

Figure 3: Magnetic resonance post-contrast image of the cervical and thoracic spine (sagittal view) revealed diffuse leptomeningeal enhancement with multiple peripherally enhancing intramuscular and subcutaneous collections within the cervical and thoracic region

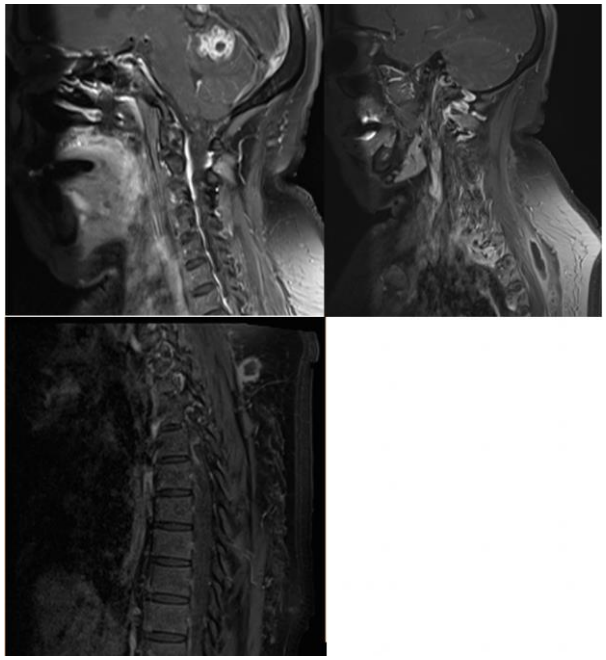

Figure 4: Post-treatment brain magnetic resonance image (axial view) with contrast revealed interval regression of the previously seen multiloculated peripherally enhancing lesions with minimal perilesional oedema