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Case Report Open Access

Case report: Persistent generalized mycobacterial spindle cell pseudotumor of lymph nodes in a young man with newly diagnosed AIDS

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Abstract

Background

Differential diagnosis of persistent generalized lymphadenopathy in a young man with newly diagnosed AIDS remains challenge.

Keywords: Disseminated Mycobacterium avium-intracellulare complex; spindle cell pseudotumor; AIDS; persistent generalized lymphadenopathy; Kaposi sarcoma

Introduction

Differential diagnosis of persistent generalized lymphadenopathy in a young man with newly diagnosed AIDS remains challenge. The current case report is a unique presentation of persistent generalized mycobacterial spindle cell pseudotumor of lymph nodes that initially began as a diagnostic challenge for the healthcare providers involved in the patient's care. The overall aims of this case report include raising awareness of a common HIV related lymphadenopathy that can present a diagnostic challenge; to review recent evidence including radiographic features, diagnostic considerations, and treatment of disease; and finally, to emphasize an appropriate diagnostic strategy.

Case presentation

A 34-year-old male tour guide complained of general malaise, soreness, intermittent fever for 1–2 months, appetite loss and weight loss (approximately 4–5 kg). He experienced chest discomfort on the day of the visit, but he did not appear to be severely ill. The temperature, blood pressure, pulse rate, and respiratory rate were 37.6°C (99.6°F), 112/64 mmHg, 102 beats/min, and 15 counts/min, respectively. The results of the physical examination were normal except for enlargement of the left tonsil without a pus coating. Blood test results revealed a hemoglobin level of 12.8 g/dL; a white blood cell (WBC) count of 4030/μL, consisting of 56% neutrophils and 1% band neutrophils; and a platelet count of 107000/μL. His urine sample and sputum specimen did not reveal any specific findings. The additional laboratory data revealed that the levels of blood urea nitrogen, creatinine, alanine aminotransferase, aspartate aminotransferase, and C-reactive protein were 12 mg/dL, 0.76 mg/dL, 74 IU/L, 49 IU/L, and 0.208 mg/dL, respectively. The total bilirubin levels and direct and indirect bilirubin levels were not measured. A chest X-ray revealed a mottled opaque shadow with suspicion of a focal infiltrative lesion in the left upper lung field. The patient was homosexual for years and not yet married, but he did not remember when his last sexual activity had occurred. The results of additional laboratory testing were as follows: anti-HAV IgM negative, HBsAg positive, anti-HBsAg negative, anti-HCV negative, Human immunodeficiency virus (HIV) screening test (EIA method) positive, positive Western blotting of HIV, absolute CD4 count 69, and absolute CD8 count 657, HIV viral load test (by using RT-PCR) 776000 copies/mL, serum Cryptococcus Ag negative, STS-RPR 1:2, and TPPA 1:1280. A computer tomography (CT) of the chest revealed the following: (a) multiple mildly enlarged lymph nodes at the renal and infrarenal para-aortic regions, (2) moderate splenomegaly, and (3) three solid lung nodules (each >4 mm) distributed in the right upper, right middle, and left lower lobes of the lungs (Figure 1). He was diagnosed to have acquired immunodeficiency syndrome (AIDS) and latent syphilis at the first visit to emergency department but the nature of the multiple lung nodules and lymphadenopathies have not yet been determinates. He was treated with anti-retroviral therapy (ART), benzathine penicillin at first and was instructed to seek treatment from an infectious diseases specialist within the next 7 days.

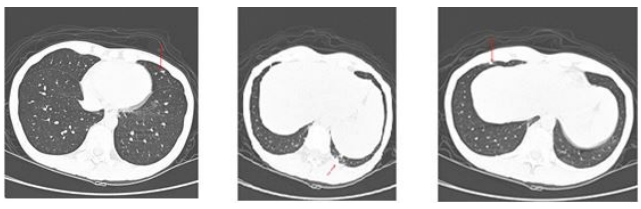
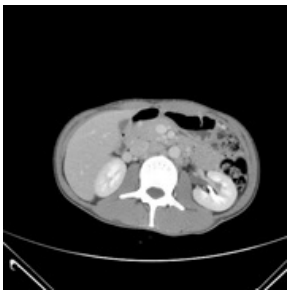


Figure 1: Chest CT: Three solid lung nodules (each>4mm).

However, the patient continued to experience general malaise and night fever in the following days until his outpatient department

(OPD) visit. We checked CMV viral load test revealed that the viral load was 5096 IU/mL. The patient's energy levels and mood improved after oral valgancicyclovir prescribing. Two months later, the HIV viral load and CD4 count both decreased to 66 copies/mL and 52, respectively. Moreover, the results of his blood test revealed a decreasing trend in the following parameters: hemoglobin level (8.4 g/dL), WBC count (2250/μL consisting of 57% neutrophils, consisting of 17% lymphocyte and 24% monocytes), and platelet count (76000/μL). He did not exhibit any bleeding tendencies such as bruising easily or ecchymosis, gum bleeding, or hematochezia. He also denied any bone pain. Bone marrow culture and biopsy were performed and the pathology revealed mildly hypercellular marrow with interstitial plasmacytosis and histiocytic aggregates. It showed negative results for periodic acid–Schiff (PAS), Gomori methenamine silver (GMS), and acid-fast staining. No hematological malignancies were observed. Three months after the prescription of ART, the patient noted several erythematous papules over his right forearm. Drug eruption or Kaposi Sarcoma (KS) was considered the appropriate cause. Hence, skin biopsy was performed immediately. One week after the skin biopsy, the report revealed KS. During these intervening periods, the patient still experienced intermittent fever, appetite loss, and general malaise. To follow-up the lymphadenopathies, the patient received abdominal CT examination. The abdominal CT examination showed the progression in size of the para-aortic lymphadenopathies (Figure 2). HIV related lymphoma should be considered at this stage. The patient hesitated to undergo an invasive procedure for intra-abdominal lymph node biopsy for further definite diagnosis. After discussion with a general surgeon, deep neck lymph node biopsy was considered a viable option because it is less invasive and the deep neck lymph nodes are easier to approach than the para-aortic lymph nodes. Finally, the patient agreed to undergo a deep neck lymph node biopsy (figure 3).



Before ART

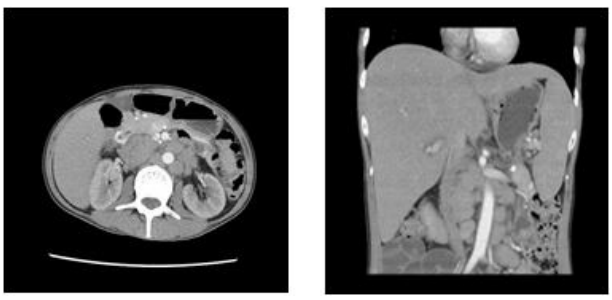


Figure 2: CT of the abdomen before and after ART.

3-4 months after ART, progression in size of the para-aortic lymphadenopathies

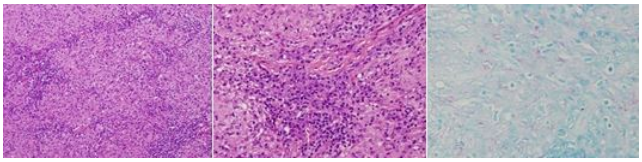


Figure 3: Pathology of the deep cervical lymph nodes.

Microscopically, the specimen showed diffuse proliferation of sheets and confluent trabeculae throughout the lymph nodes. It consisted of ovoid-to-plump spindle cells with appreciable amounts of eosinophilic cytoplasm. Notably, both acid-fast and GMS stains highlighted numerous intracellular bacilli. Well-formed granulomas or multinucleated giant cells were not observed.

Disseminated Mycobacterium avium-intracellulare complex infection complicated with generalized lymph nodes spindle cell pseudotumor was diagnosed finally due to bone marrow culture which was done prior to deep cervical lymph node biopsy yield mycobacterium avium-intracellulare complex (MAC). Clarithromycin 500 mg po bid and ethambutol 1200 mg po qd were prescribed for at least 12 months until the radiological diminution of para-aortic lymphadenopathies.

Discussion

Mycobacterial spindle cell pseudotumors are rare, benign lesions caused by the local proliferation of spindle-shaped fibro-histiocytic cells without the formation of epithelioid granulomas in response to mycobacterial infection. The pathological characteristics of pseudotumors resemble those of some mesenchymal neoplasms, particularly Kaposi sarcoma7 (KS). The morphological features that favored the diagnosis of KS over mycobacterial spindle cell pseudotumor were the prominent fascicular arrangement of spindle cells and slit-like spaces, the lack of granular and acidophilic cytoplasm, and the presence of mitoses[1]. Without staining for acid-fast bacilli, the histological distinction of pseudotumors from other spindle cell lesions, including malignancies, is difficult. Mycobacterial spindle cell pseudotumors have been reported in various sites such as the lymph nodes [2, 3] bone marrow [2], lungs [4, 5] nasal cavity [6], brain [7], skin [8], appendix vermiformis [9], spleen[10], and plantar fascia [11]. These tumor-like lesions can be confused clinically as well as radiologically for cancerous tumors. They were first described in 1963 as a variant form of lepromatous leprosy, in which spindle cell proliferation is prominent [12].

Disseminated MAC infection complicated with persistent generalized lymphadenopathy (PGL) is extremely rare in Taiwan to which tuberculosis is endemic. These observations suggest the role of cross-protective immunity from childhood Bacillus Calmette–Guérin or latent tuberculosis [13-14]. We cannot conclude whether our patient initially had disseminated MAC disease when he received the diagnosis of AIDS. Regimens for prophylactic treatment and therapy for MAC infection are different. Currently, several diseases such as lymphoma, KS, acute and chronic lymphocytic leukemia, HIV infection related PGL, syphilitic lymphadenitis, fungal infection, and mycobacterial infection, must be properly differentiated. The major decision in HIV

lymphadenopathy remains distinguishing between patients who should be observed, those who should undergo fine needle aspiration, and those who should undergo an open biopsy [15]. In this case, ART prescription successfully suppressed HIV replication within 2 months. However, the patient's immunodeficiency status remained (CD 4 count 52), which may have increased the risk of exposure to various opportunistic infections or HIV-associated malignancies such as lymphoma with/without bone marrow involvement and Kaposi sarcoma. Biopsies of the lung nodule, para-aortic lymph nodes, and bone marrow may be required for a definite diagnosis. However, the need for simultaneous biopsies on several parts is questionable.

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Conclusion

We reached the final diagnosis of disseminated MAC disease complicated with a generalized lymph node mycobacterial spindle cell pseudotumor after performed bone marrow biopsy, bone marrow mycobacterial culture, and deep cervical lymph node biopsy; however, they could have arrived at the diagnosis and started appropriate treatment earlier.

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