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Case Report

Tubercular Mycobacterial Spindle Cell Pseudotumour: A Case Report

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Abstract

Pseudotumour is a benign inflammatory lesion. Mycobacterial spindle cell pseudotumour (MSP) is a rare pseudotumour. It is a benign proliferation of spindle-shaped histiocytes containing acid-fast mycobacterium, commonly reported in immunocompromised patients. MSP is usually associated with mycobacterium avium complex (MAC). Here, we present the case of a 38-year-old gentleman with acquired immune deficiency syndrome (AIDS) who presented with low-grade fever for 1-month duration. Clinically, he had generalised lymphadenopathy. Chest X-ray showed miliary infiltration in bilateral lung fields. Lymph nodal biopsy showed spindle-shaped histiocytes filled with acid-fast bacilli on Ziehl-Neelsen (ZN) stain, suggestive of MSP. Immunohistochemical (IHC) stains were positive for CD68, S-100 and negative for CD31, which are consistent with MSP. Polymerase chain reaction (PCR) of the biopsy tissue was positive for MTB. Highly active antiretroviral therapy (HAART) was continued and anti-tubercular therapy (ATT) was started. The fever resolved within two weeks and there was a resolution of lymph nodal swelling by 6 weeks. The diagnosis of MSP associated with mycobacterium tuberculosis (MTB) makes our case interesting. It is of utmost importance to differentiate MSP from Kaposi's sarcoma (KS) and other pseudotumours and to know whether it is of tubercular or non-tubercular origin, as the treatment is entirely different.

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Keywords ● Acquired immunodeficiency syndrome ● Biopsy ● Mycobacterium tuberculosis (MTB) ● HIV ● Spindle cell

What's Known

- The mycobacterial spindle cell pseudotumour (MSP) is a rare inflammatory pseudotumour.
- MSP is usually associated with mycobacterium avium complex (MAC).

What's New

- A rare case of MSP associated with mycobacterium tuberculosis (MTB) is presented.

Introduction

Mycobacterial spindle cell pseudotumour (MSP) is a rare inflammatory benign pseudotumour which was first reported by Wood et al. in the skin of a post-cardiac transplantation patient.[1-3] Clinically as well as pathologically, it mimics Kaposi's sarcoma (KS) and other mesenchymal tumours.[2,4,5] MSP is commonly reported in patients who are immunocompromised, have human immunodeficiency virus (HIV) infection, undergone organ transplantation, and those receiving prolonged immune suppressing medications.[4,6] It is rarely reported in neonates following BCG vaccinations.[7] As per most of the case reports, it commonly involves the lymph node and skin.[4,5] Other rare organs reported to be involved are the lung, brain, nasal septum, spleen.[8-10] Most of the reported cases of MSP are associated with mycobacterium avium complex (MAC) and other non-tubercular mycobacterium.[2] MSP associated with mycobacterium

tuberculosis (MTB) is very rare, which makes our case interesting.[2,7]

Case Presentation

A 38-year-old gentleman presented to us for fever and weight loss of two months duration. He had a significant background history of having multiple sex partners and had no other co-morbidities. His vitals were normal, systemic and general physical examinations were normal except for low body mass index (BMI) of 17.5 kg/m^2. On subsequent evaluation, he was found to have acquired immune deficiency syndrome (AIDS) with cluster of differentiation 4 (CD4) cell count of 50 cells/mm^3. He was started on highly active antiretroviral therapy (HAART) comprising of tenofovir, lamivudine, and efavirenz. His fever subsided with improvement in general well-being till one month back, when he again presented to us, this time with generalised lymphadenopathy and low-grade fever. On examination, he was febrile with the temperature of 100ºF, had normal blood pressure (112/74 mmHg) with a heart rate of 96/min and respiratory rate of 18/min. He was malnourished with body mass index (BMI) of 18 kg/m^2. General physical examination showed pallor with multiple, bilateral, cervical and inguinal lymphadenopathy. The lymph nodes were non-tender, mobile, firm in consistency and maximum 3×3 cm in size. Examination of the respiratory system, cardiovascular, per-abdominal and central nervous system were within normal limits. Complete blood count showed normocytic, normochromic anaemia (10.4 g/dl) with mild neutropenia (32×10^9/L) and normal platelet count (180×10^9/L). Chest X-ray showed miliary infiltration in bilateral lung fields. Lymph nodal biopsy showed spindle-shaped histiocytes, filled with acid-fast bacilli on Ziehl-Neelsen (ZN) stain, suggestive of MSP (figures 1A-1C). Immunohistochemical (IHC) stains were positive for CD68, S-100 and negative for CD31, which were consistent with MSP (figures 1D, 2A, and 2B). Polymerase chain reaction (PCR) of the biopsy tissue was positive for MTB. Subsequently, he underwent bone marrow biopsy, which also showed epitheloid granuloma suggestive of tubercular marrow infiltration. Hence, HAART was continued and anti-tubercular therapy (ATT) was started with isoniazid, rifampin, ethambutol, and pyrazinamide along with pyridoxine supplementation. Fever resolved 2 weeks after the initiation of ATT and there was a resolution of lymph nodal swelling by 6 weeks.

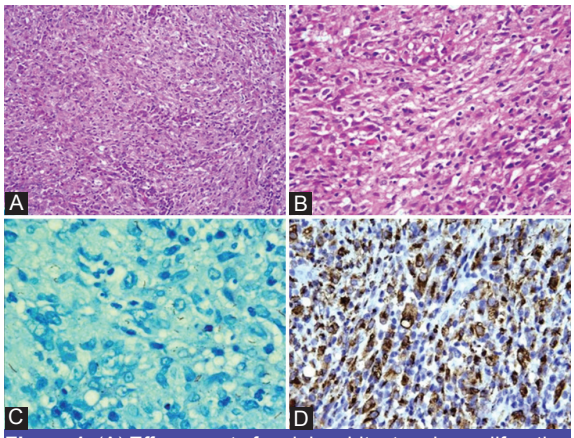

Figure 1: (A) Effacement of nodal architecture by proliferation of spindle-shaped histiocytes (100×), (B) High power shows spindle-shaped histiocytes with scattered lymphocytes (400×), (C) Ziehl Neelsen stain shows many acid fast bacilli (1000×) and (D) IHC shows spindle-shaped cells with diffuse positivity for CD68 (400×)

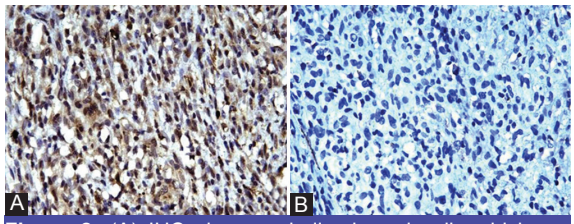

Figure 2: (A) IHC shows spindle-shaped cells which are positive for S100 (400×) and (B) negative for CD31 (400×)

Discussion

The exact pathophysiology of MSP is still unclear. There are various proposed hypotheses that it may be either a host immune response or a part of inflammatory cytokine response to the pathogen that leads to localised proliferation of spindle cells and colonization of the mycobacterium.[1] MSP mimics other benign as well as malignant tumours. Among the malignancies, Kaposi's sarcoma is very closely mimicked by MSP. Histopathological examination (HPE) of the biopsy tissue provides vital clues to differentiate MSP mimicking other pseudotumour and Kaposi's sarcoma. Microscopic examination shows typical spindle cell arrangement of histiocytes and ZN stain shows histiocyes filled with acid-fast mycobacterium. IHC establishes the histiocytic nature of the spindle cell and thereby helps to differentiate MSP from Kaposi's sarcoma. While MSP is positive for CD68, S-100, and negative for CD31, CD34, Kaposi's sarcoma is positive for CD31, CD34 and negative for CD68, S-100, and ZN stain.[2] It is very important to differentiate Kaposi sarcoma from MSP, as Kaposi sarcoma has a well-known association with HIV infection and coexistence of MSP and Kaposi's sarcoma has also been

reported in medical literature.[2] For management, HPE plays a major role as it not only establishes the diagnosis of MSP but aetiology also, so that early anti-microbial therapy can be initiated to prevent further complication. With adequate antimicrobial therapy, MSP can be reversible.

The role of surgical therapy has not been well established. Surgery is required for preventing compressive symptoms in a space-occupying lesion of vital organs as well as for definitive diagnosis by HPE.[6,9,10] The recurrence rate following medical as well as surgical therapy is still unclear due to the lack of follow-up and literature review. Further studies are required to know the exact pathophysiology, reason behind localised proliferation, role of surgical excision, and risk of recurrence following surgery. MAC and other non-tubercular mycobacterium are most commonly associated with MSP.[2,11,12] The index case of MSP was unusually associated with MTB, which is also rare. In future, as more cases of MSP would be reported, especially from the tuberculosis endemic countries like India, tubercular MSP might emerge as a commoner variant compared to non-tubercular MSP. We also expect that much of the unattended and unknown facts regarding MSP will be clarified systematically.

Conclusion

MSP should be considered in the differential diagnosis of tumours with spindle cell proliferation. For management, it is very important to differentiate MSP from Kaposi's sarcoma, other pseudotumours, and whether tubercular or non-tubercular. Tubercular MSP is likely to be more common as compared to non-tubercular MSP in the areas with high prevalence of tuberculosis.

Conflict of Interest: None declared.

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