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CASE REPORT

Kikuchis Disease with Myocarditis: A Rare Case Report

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Abstract

Kikuchis disease is a rare, self limiting disease characterized clinically by fever with tender regional lymphadenopathy with characteristic histopathological features on lymph node biopsy. It is a rare disease with unkown incidence with very few reported cases. It is most prevalent in young asian population equally affecting both the sexes. The disease is usually self limiting ,in rare cases complications such as aseptic meningitis, cerebellar ataxia, encephalitis, myocarditis, acute renal failure can occur. We report a case of 28 year old male presented with fever and generalized lymphadenopathy with features suggestive of myocarditis.

Keywords: Kikuchi disease; lymphadenopathy; myocarditis

Introduction

Kikuchis disease is a rare, self limiting disease characterized clinically by fever with tender regional lymphadenopathy with characteristic histopathological features on lymph node biopsy [(1)]. It is a rare disease with unknown incidence with very few reported cases in the literature. The disease is most prevalent in young Asian population equally affecting both the sexes. The disease is usually self-limiting, the clinical symptoms resolve within few months without any specific treatment. The cause of Kikuchis disease is unknown, histopathology helps in differentiating this benign condition from lymphomas, systemic lupus erythematosus and infective lymphadenitis. In rare instances complications such as aseptic meningitis, encephalitis, cerebellar ataxia, panuveitis, acute renal failure, and myocarditis can occur [(2)]. We report a case of 28-year-old male presented with fever with generalized lymphadenopathy with features suggestive of myocarditis.

Case Report

A 28-year-old male presented with the complaints of fever and painful swellings over neck, axilla, and groin since 8 days.

Fever was high grade, intermittent type with no diurnal variation. There was no history of cough, loss of weight, loss of appetite, skin rash, pain abdomen, vomiting, diarrhea, joint pain. No history of any drug intake prior to the onset of the symptoms, No history of similar complaints in the past. Personal and family history were not significant. On examination multiple, tender, mobile lymph nodes were palpable bilaterally in neck, axilla, inguinal region, the largest measuring approximately 3 x 4 cm in size.

Rest of the clinical examination was unremarkable. Laboratory work up at the time of admission revealed hemoglobin of 11 g/dl, total count of 19,210 cells/cu mm, platelet count of 80,000 cells/cu mm, peripheral smear showed normocytic normochromic anemia with leucocytosis with thrombocytopenia. ESR was elevated (95 mm/h), serum procalcitonin was elevated (7.61 mcg/l). Fever profile for Dengue, Malaria, Ricketssia, Typhoid were negative. Liver function tests and renal functions tests were within normal limits. ECG, chest X-ray, USG abdomen did not reveal any abnormality. Blood culture did not yield any growth. Serology for HIV was non-reactive, HbsAg and HCV were negative. Mantoux test was negative. ANA was negative. Exicisional biopsy of cervical lymph node was done, which showed effaced architecture of lymph node with patchy, well circumscribed areas of necrosis with areas of karyorrhectic nuclear debris were present. Plenty of lymphocytes, histocytes, plasma cells and apoptotic cells were seen. Neutrophils and eosinophilis were absent. Section did not show any atypical cells. Hence histopathological diagnosis of histiocytic necrotizing lymphadenitis was made. On clinic pathological correlation, a diagnosis of Kikuchis disease was made. During 3rd day of admission patient developed an episode of hypotension, ECG showed sinus tachycardia with nonspecific ST-T changes. CPK-MB was elevated (31 U/L). Echo was normal.

Features were suggestive of Myocarditis. Patient was started on steroids and tapered slowly over 4 weeks.

Discussion

Kikuchis disease is a benign, self-limiting condition characterized by regional tender lymphadenopathy, with or without fever and night sweat [3].

Described independently in 1972 by Kikuchi and Fujimoto et al., this subacute necrotizing lymphadenitis has been recognized in Asia and sporadically in Western countries [4]. Affected patients are most often young adults under the age of 30 years, equally affecting both the sexes. The pathogenesis of Kikuchi's disease is still not fully understood. It has been proposed that the primary event may be the activation of T-lymphocytes and histiocytes [5]. Here, the proliferating T cells enter the cycle of apoptosis, which may form the areas of necrosis in lymph nodes and then the histiocytes clear the apoptosed cellular debris and fragments. Our patient was a young male presented with fever and generalized lymphadenopathy for 8 days.

Histopathology of lymph node biopsy showed features of histiocytic necrotizing lymphadenitis. On clinicopathological correlation the diagnosis of Kikuchis disease was made. The clinical features of Kikuchis disease mimic lymphomas, HIV, SLE, Infectious mononucleosis, tuberculosis. Kikuchis disease is under- diagnosed in clinical practice, as majority of cases with fever and lymphadenopathy without biopsies might have been wrongly diagnosed as viral infections. Supportive measures are the mainstay of therapy as no treatment is required in most cases of Kikuchis disease. However patients may develop severe systemic complications like aseptic meningitis, encephalitis, cerebellar ataxia, myocarditis, panuveitis, and acute renal failure. Our patient developed an episode of hypotension with ECG changes of sinus tachycardia and nonspecific ST-T changes and elevated levels of CPK-MB were suggestive of myocarditis. Thus Kikuchis disease might have systemic complications. Anticipating such complications and timely interventions accordingly is needed in such rare instances.

Conclusion

Kikuchis disease is rare, high index of suspicion helps in early diagnosis. Kikuchis disease should be kept as differential diagnosis in a case of fever with lymphadenopathy. Histopathology of the lymph node biopsy helps in confirmation of the diagnosis. The disease is usually self-limiting, complications like aspetic meningitis, encephalitits, cerebellar ataxia, myocarditis, panuveitis, acute renal failure do occur in rare instances. Anticipating such complications and timely intervention accordingly helps in reducing the mortality of the patient.

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