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Case Report

Kikuchi-Fujimoto disease in a young male post-COVID -19 vaccination

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ABSTRACT

Kikuchi disease (Kikuchi-Fujimoto disease) is one of the rarest forms of lymphadenopathy. It is also known as necrotizing lymphadenitis. The patients usually present with fever and lymphadenopathy. Here is a case of Kikuchi-Fujimoto disease presented with fever and cervical lymphadenopathy. Radiological imaging revealed matted lymph nodes. Fine needle aspiration of the lymph node revealed granulomatous inflammation. Acid Fast stain was negative and further investigations ruled out tuberculosis. A histopathological biopsy of the lymph node confirmed the diagnosis of Kikuchi-Fujimoto disease. The patient was treated with medications and the patient's condition improved in follow-up. Tuberculosis must be ruled out in such conditions as the symptoms and radiological and FNAC findings mimicking tuberculosis. The diagnosis of Kikuchi-Fujimoto disease is made by histopathology and confirmed by immunohistochemistry.

1. Introduction

Kikuchi-Fujimoto disease is one of the rarest forms of lymphadenopathy. It is also known as necrotizing lymphadenitis. This is more common in adults but can occur in children with an average age of second to fourth decade of life. In adults, females are commonly affected and in paediatric age groups males are more commonly affected.[1,2] The etiology of Kikuchi disease is unknown but it can be triggered by some infections mostly viral and also by autoimmune diseases. Studies suggest that there is association with COVID-19 and Kikuchi disease.[3,4] Kikuchi disease is seen in patients with active COVID -19 and also may develop post covid. The Kikuchi disease is also seen in patients who have been administered with COVID -19 vaccination.[5,6] The usual symptoms of patients with Kikuchi disease are fever with cervical lymphadenopathy. Very few cases present with generalized lymphadenopathy. We describe a young male presenting with multiple enlarged cervical lymph nodes. Fine needle aspiration needle showed a history of granulomatous lymphadenitis and Kikuchi-Fujimoto disease was diagnosed in histopathological examination and confirmed by immunohistochemistry.

2. Case History

A 19 year old male, came to the hospital with a history of fever and enlarged cervical lymph node. Ultrasound neck revealed bilateral cervical nodes with few matted, necrotic nodes. Fine needle aspiration of the lymph nodes revealed a cellular smear consisting of chronic inflammatory infiltrate of histiocytes, lymphocytes admixed with ill-defined granulomas consisting of epithelioid cells and lymphocytes in an extensive necrotic background. FNAC findings were suggestive of granulomatous lymphadenitis

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and the smear was negative for acid fast stain. Other investigations such as sputum and gene-xpert studies were done to rule out tuberculosis. One lymph node was surgically removed and it was sent for histopathological examination. On gross examination, the node was 3 x 2 cm in size and externally capsulated, cut surface showed grey white areas. Microscopic sections revealed lymph node with fairly preserved architecture and reactive follicles composed of small lymphocytes admixed with immunoblasts in clusters and singly scattered. Focal areas of necrosis was seen along with histiocytes and nuclear debris. No definitive granuloma was made out and the sections were negative for Acid fast stain. The diagnosis of Kikuchi-Fujimoto disease was suggested and was confirmed by immunohistochemistry. Post-surgery the patient was medically managed and his condition improved well on regular follow up.

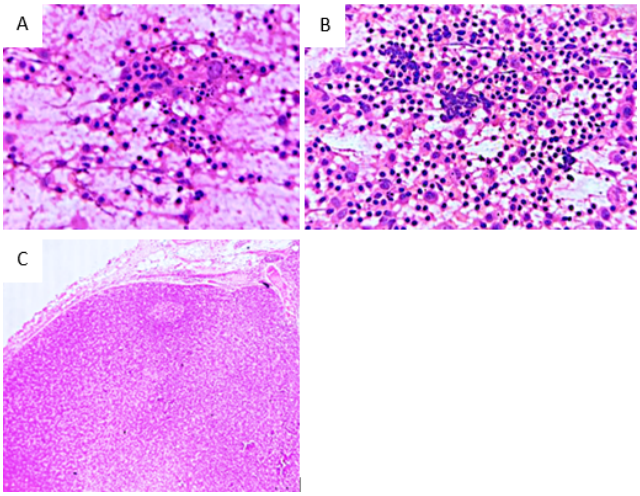

Figure 1: A: 40x Photomicrograph showing crescentic histiocytic cells in a granular background; **B:** 40x Photomicrograph showing necrotic debris; **C:** 4x-photomicrograph showing effaced node with alternating pale and dark areas.

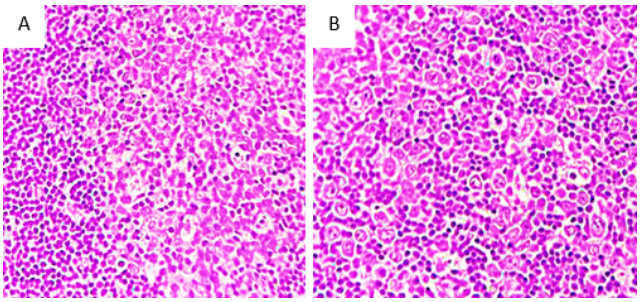

Figure 2: A: 10x- Photomicrograph showing karyorrhectic debris with central necrosis; **B:** 40x- Photomicrograph showing phagocytic cell with eosinophilic cytoplasm and cresent shaped nuclei

3. Discussion

Kikuchi disease is one of the rare diseases which occurs most commonly in the South East Asian population. Among the affected patients the disease is common in young adults with a mean age of 20-30 years of age. Kikuchi disease is more common in females than in males with a female to male ratio of 2-4:1.[7]

Kikuchi disease occurs due to an unknown etiology but few studies show that Kikuchi disease can be triggered by viral infections or can be due to an autoimmune etiology. Few case reports suggest that Kikuchi disease can be triggered due to COVID -19 infection[3,4] and COVID -19 vaccinations.[5,6] The usual presentation of patients with Kikuchi disease will be acute in onset with complaints of fever, cervical lymphadenopathy which is usually unilateral.[8,9] Lymphadenopathy is observed in almost all the cases with Kikuchi disease and majority of the cases present with tender cervical lymphadenopathy and few cases may also have generalized lymphadenopathy.[10]

The diagnosis of Kikuchi disease is based on the histopathological findings followed by fine needle aspiration cytology. Possibility of tuberculosis and lymphoma must be considered and ruled out. Histological findings include preserved nodal architecture with areas of coagulative necrosis and absence of neutrophils.[11] Immunohistochemistry must be done to confirm the diagnosis of Kikuchi disease. The positive expression of immunohistochemical markers such as CD 68 and CD 4 suggests Kikuchi disease.[12]

The treatment of Kikuchi disease is supportive management along with steroid therapy. Though the disease resolves on its own, regular follow up must be done to know the condition of the patient and for disease recurrence.[13]

4. Conclusion

Kikuchi disease is rare self-limiting disease, though it is more common in eastern and western part of world. This disease should be always considered as differential diagnosis clinically and histomorphologically while reporting lymphoma in post pandemic era to avoid expensive molecular investigations

5. Source of Funding

None.

6. Conflict of Interest

None.

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