International Journal of Healthcare Sciences ISSN 2348-5728 (Online)
Vol. 6, Issue 2, pp: (218-220), Month: October 2018 - March 2019, Available at: www.researchpublish.com

Kikuchi Disease: Announcement for an Atypical Presentation in 9 Years Old Boy: A Rare Case Report

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Abstract: **A 9 years old boy presented with atypical and new presentation of Kikuchi disease with extensive generalized lymphadenopathy with kidney involvement in radiological studies. The disease is established by the combination of clinical presentation, laboratory testing, radiological findings and histo-pathological confirmation. As many reports, which gave different trails of management, we chose not to use any medications such as antibiotics and steroids as there was no causative organism identified and there was no need for urgent intervention. Spontaneous resolution within weeks to months is the nature of Kukuchi disease.**

1. INTRODUCTION

Kikuchi disease, also named as histiocytic necrotizing lymphadenitis was first identified in 1972 in Japan. Most commonly affected gender is female in the adult population and male in pediatrics. The most common clinical presentation is cervical lymphadenopathy with or without fever. Extra-systemic manifestations as weight loss, myalgia, arthralgia and skin rashes can be noted (1). A good number of case reports presented unusual presentation for Kikuchi disease as cutaneous manifestation. (1) Here, we discuss a case of a boy who presented with atypical feature of Kikuchi inform of extensive generalized lymphadenopathy with extra nodal involvement.

2. CASE REPORT

A 9 years old Saudi man, presented with 2 months' history of painless submandibular lymphadenopathy, easy fatigability, and subjective weight loss. There were no other complaints. He had been previously fit and well and was on no medication. On examination, he was pale but otherwise looked well. He was afebrile and hemodynamically stable. The significant finding was generalized firm mobile painless lymphadenopathy with a large one (4*3) in submandibular area. There were 2 cm smooth no tender hepatomegaly and 6 cm splenomegaly. A blood test revealed hypochromic microcytic anemia with high erythrocyte sedimentation rate (42) and high lactate dehydrogenase (488). Blood glucose urea creatinine, sodium, potassium and bicarbonate level were normal. Blood, urine, and stool cultures were negative. Epstein-Barr virus IgG was reactive. Antinuclear antibody (ANA) was (1:40), Double strand DNA (dsDNA) (67.28) C3 complement {1.650}, C4 complement {0.492}.

Contrast-enhanced CT to neck-chest-abdomen & pelvis support the diagnosis by showing extensive generalized enlarged lymph nodes involving the neck, bilateral axillary lymph nodes. Chest/mediastinum, both sides of the lung, Abdomen, and inguinal regions. Remarkable splenomegaly with multiple variable size hypodense lesions; mild hepatomegaly with hypodense lesions; both kidneys showed satisfactory contrast perfusion with multiple small cortical hypodense lesions. Many of the lymph nodes mentioned above displayed necrosis along with enlargement. [Figure 1]

Left cervical lymph node biopsy confirmed the diagnosis of Kikuchi disease by demonstrating a fragmented lymph node tissue, follicular hyperplasia, and extensive necrosis consistent with histiocytic necrotizing lymphadenitis (Kikuchi disease) with no evidence of malignancy. [Figure 2]

Immunohistochemical studies showed that most of the lymphocytes were CD3 positive and less amount are CD20 positive, which are concentrated mainly in the follicles. The pattern mainly was consistent with the the reactive process. Numerous scattered cells were positive for CD68 consistent with histiocytes. CD30 was negative.

3. DISCUSSION

Kikuchi disease, also named as necrotizing histiocytic lymphadenitis is a rare, self-limiting disease with a benign course. At first, it was the disease of young women but many reports clearly approved the opposite and showed it can affect both genders and all races and age groups (1).

Cervical lymphadenopathy is the most common manifestation of Kikuchi disease (1). Our patient unusually presented with not only cervical lymphadenopathy but his whole lymphatic system was affected, with neck, axillary, chest/mediastinal, abdominal including spleen and inguinal lymphadenopathies, along with extra-nodal involvement in both kidneys. There were few cases reported by having generalized lymphadenopathy in the adult population but not sufficient data in the pediatric age group. (2) There was one case reported of Kikuchi disease which had renal involvement, acute renal failure (ARF), as part of its presentation as the serum creatinine level was {72 mg/dL}, BUN {231mg/dL}, Na {141 mEq/L} and K {6.1 mEq/L}. Renal biopsy showed tubular degenerative change. Different from our study, the ARF was part of severe systemic manifestations added to pericarditis with cardiac tamponade, pneumonitis and hepatitis in 24 years old male who was significantly improved with prednisone (3). Our patient had renal involvement under CT scan with contrast as multiple small cortical hypodense lesions without clinical or laboratory association. Laboratory test results were good to exclude any cause of cervical lymphadenopathy except for EBV IgG EBNA which was positive in our patient. A study was published in 2014 investigating the relationship between Kikuchi disease and EBV in both adults and pediatrics revealed that EBV reactivity was found more in children (26.7%) who were diagnosed with Kikuchi disease than adults (6.7%). (4). And one interesting study was comparing the frequency of detection of human herpes viruses in 30 cases of Kikuchi disease to EBV, HHV6, HHV7 and HHV8 and the results revealed that EBER was commonly detected in comparison to the other viruses and it was frequently found positive in necrotizing lymph nodes of Kikuchi cases more than Reactive Lymphoid Hyperplasia cases. These results strongly encourage the idea of that Kikuchi disease could be secondary to infectious causes mainly EBV. (5). As there is no clear etiology for Kikuchi disease, there will be no defensive treatment. Many studies agreed not to interfere with medication and they were content only with serial follow-up appointments (6). The use of antibiotic is not advisable as theirs no causative microorganism identified. As in our patient, we did not give any medication, as there was no clear need for any intervention. In fact, schedule of a serial follow-up appointments to track the prognosis of the disease and the improvement of clinical manifestations and laboratory results was the plan.

4. CONCLUSION

Kikuchi disease is a rare disease with no definitive etiology. We had a case with of Kikuchi disease with renal involvement in radiological imaging without clinical nor laboratory association and extensive generalized lymphadenopathy reaching in some lymph nodes 5 cm. although, Kikuchi disease is a self-limited disease benign course that present with atypical feature needs highly index of suspicion.

LEARNING POINTS

1-KFD is a self-limited disease benign course that present with atypical feature needs highly index of suspicion.

2-KFD could be secondary to infection mainly (EBV and human herpes viruses) and need more studies to prove that.

COMPETING INTERSTS

The author declare that they have no competing interest.

CONSENT

Written informed consent was obtained from the patient and his mother for publication of this case report and any accompany images. A copy of the written consent is available for review.

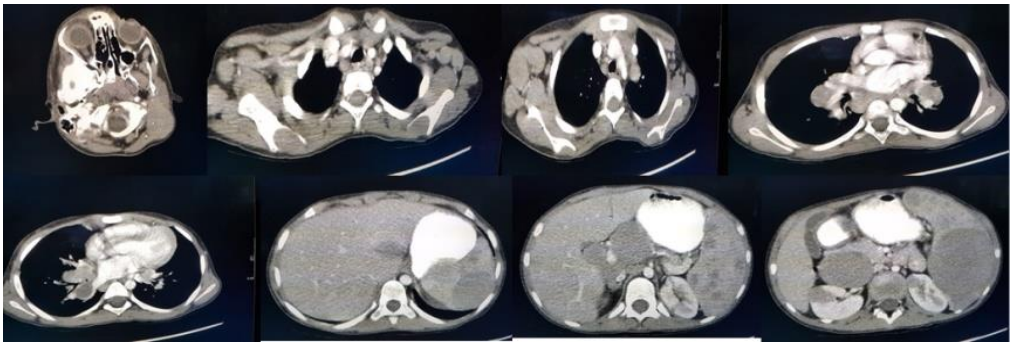

Figure 1: Computed Tomography scan of neck, axillary, chest/mediastinal, abdominal, inguinal in a patient with Kikuchi disease.

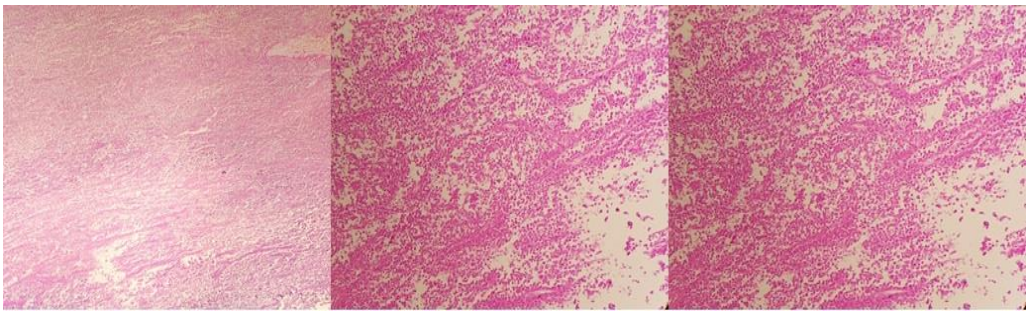

Figure 2: Left cervical lymph node biopsy confirmed the diagnosis of Kikuchi disease.

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