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Case Report

FUJIMOTO'S KIKUCHI DISEASE, IS IT A PART OF FLORID SLE WITH WIDE SYSTEMIC MANIFESTATIONS IN AN ELDERLY FEMALE WITH ABNORMAL LIVER PROFILE? –A RARE CASE REPORT

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ABSTRACT

Kikuchi's disease is a necrotizing lymphadenitis that is prevalent in Asia and is being increasingly recognized in other areas of the world. It usually occurs in women in their late 20s or early 30s and manifests as a posterior cervical adenopathy. It resolves spontaneously, usually over a period of several weeks to 6 months. Its initial clinical appearance is commonly similar to that of a lymphoma, and it can be pathologically misdiagnosed as such. Kikuchi's disease might be associated with systemic lupus erythematosus. We report a case of female aged 54 who presented to us with long standing fever and axillary and cervical lymphadenopathy and after an axillary NL biopsy , She became very sick and subsequent she recovered from her illness after receiving therapy.

INTRODUCTION

Kikuchi's disease, also called Kikuchi-Fujimoto disease, was once known as subacute necrotizing lymphadenitis. It is increasingly recognized as a benign cause of cervical adenopathy in young adults, predominantly women. In 1972, both Kikuchi (1) and Fujimoto *et al* (2) described this disease in the Japanese literature. Five years later, Kikuchi suggested that the cause of necrotizing lymphadenitis was acute toxoplasmic infection. (3) The first case that was identified outside of Japan was published in 1982. (4) In 1985, the first case report appeared in the otolaryngology literature, when Gleeson *et al* described the histologic features and differential diagnosis of Kikuchi's disease. (5) Most reports of Kikuchi's disease have been published in the pathology literature, and there appears to be a paucity of familiarity with this condition among otolaryngologists. Kikuchi's disease usually manifests as a localized cervical adenopathy, primarily in the posterior neck of young women. Its course is benign, and it normally resolves spontaneously in a matter of weeks to 6 months. It can be easily confused with lymphoma, both clinically and pathologically. It presents with localised lymphadenopathy, predominantly in the cervical region, accompanied by fever and leukopenia in up to 50% of the cases. KFD has been rarely described in association with systemic lupus erythematosus (SLE), and its diagnosis can precede, postdate or coincide with the diagnosis of SLE.The cause of Kikuchi's disease is unknown. It might be associated with systemic lupus erythematosus (SLE). The diagnosis is primarily made by tissue biopsy, but there is a report of fine-needle aspiration diagnosis. (6) In this article, we describe a case of Kikuchi's disease, and we review the clinical features of this benign entity.

Case presentation

A 54 years old female was admitted to ED with chief complaints of shortness of breath, recurrent fever, loose stools (6-7episodes) and diffuse abdominal pain since 1 day. She was a k/c/o post OP c/o Choledocal cyst with roux-en-y hepatico jejunostomy, PTBD & stenting and Past history of recurrent cholangitis.

On examination, she was found to have moderate pain at epigastric, left hypochondrium and peri umbilical region, multiple firm to hard palpable axillary lymph nodes.

Investigations

Relevant laboratory results at the time of admission were as follows. White blood cell count was 10.3/mm^3(ref: 4–11),

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hemoglobin was 6.9 g/dL (ref: 11–15.5), urine pus cells were plenty, protein 2+ , urine c/s showed growth of E. coli. procalcitonin was 5.33(2 to 10 : sepsis). LFT showed elevated ALP (460) and GGTP (191). Measured serum potassium was 2.3 mmol/L (ref: 3.5-5.1), and corrected potassium was 4.2 mmol/L. MRI of the abdomen showed mild splenomegaly,no IHBRD, CBD was normal, no obvious obstruction, AKI, B/L basal pleura-pulmonary reaction with basal atelectasis.

Pt developed sepsis with septic shock and severe metabolic acidosis. Then she was shifted to ICU. Creatinine level was elevated (1.85) and she needed dialysis due to acute renal shut down. She received 3 cycles of dialysis and was on ventilator support. Subsequently her Hb and platelet count dropped. Platelet count reached 19,000 and she started gum bleeding. She was then transfused with multiple units of RDP.

Complements-C3 & C4 were very low (39.88 and 6.98 respectively).CRP Quantitative was positive (56). ANA profile showed positive result i.e SS-A Native,Ro-52,SS-B. Lymphnode biopsy confirmed kikuchi Fujimoto's disease. (Figure-1).

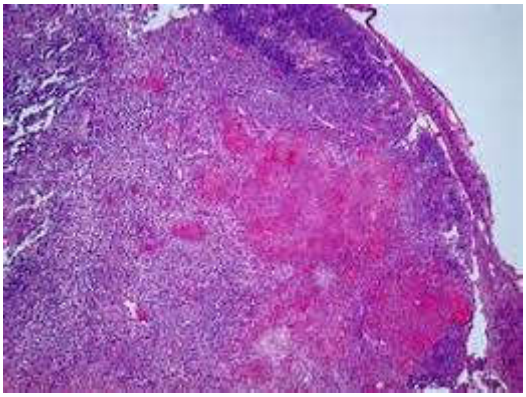

Figure 1

Course in the hospital

Patient's condition deteriorated gradually, She was febrile, tachypneic and was not maintaining adequate saturation. She was shifted to ICU and subsequently intubated in view of very low GCS. SLE profile including other autoimmune panel was abnormal and in favor of Active SLE. Patient's platelet count declined significantly and she started bleeding from gum. Patient was initially started on iv dexamethasone but it was kept on hold due to high count and abnormal renal profiles. She was initiated with high dose of iv immunoglobulin .After a second cycle of appropriate dose of iv immunoglobulin, she rapidly showed signs of improvement. Her platelet count gradually got normalized. she did not require further blood transfusion. Her general conditions improved. She discharged in a stable condition with a low dose Oral steroids.

DISCUSSION

Now this patient had all features of sepsis and source of infection most probable from Urine. Sepsis can explained all her abnormal parameters but her ANA Profiles were quite abnormal as well as Complements levels were proportionately very low and CRP was very high . Lyphnode biopsy showed active kikuchi Fujimoto's disease as shown in the Figure. This may be a very rare clinical scenario where in an elderly Female not in her active reproductive period having such full blown SLE with Lyphnode abnormal histopathology, Subsequently She well responded to high dose of iv immunoglobulin which is clearly indicated in such situation.

Prior Publication

This article has not been published or submitted for publication elsewhere, in whole or in part, before submission to the Case Reports in Critical Care.

Consent

The authors declare that they have provided written informed consent from the described patient for the case report to be published.

Conflict of Interests

The authors declare that there is no conflict of interests regarding the publication of this paper.

Authors'contribution: were involved in the clinical assessment and writing this case report. All authors read and approved the final manuscript.

Abbreviations

ED-Emergency department, GCS-Glasgow Coma Scale, SLE-Systemic Lupus Erythematosus

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