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Case Report

Kikuchi-Fujimoto disease: a case report

Prameyratna R. Kadam*

General Surgry, Karad, Maharashtra, India

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***Correspondence:**
Dr. Prameyratna R. Kadam,
E-mail: dr.prameyratnakadam@gmail.com

ABSTRACT

Kikuchi-Fujimoto disease is unique because it mimics serious illnesses yet has a benign course, requiring awareness to avoid misdiagnosis. Its novelty lies in its mysterious origin, possible autoimmune associations, and evolving clinical profile in the era of new infections and immunologic events. Given the potential for recurrence and its association with autoimmune diseases like systemic lupus erythematosus, further research is needed to clarify its pathogenesis and guide standardized treatment protocols. A 19-year-old Marathi (Indo-Aryan) female presented with multiple, painless, gradually enlarging neck swellings and occasional low-grade fever without systemic symptoms. Similar complaints occurred in childhood. On examination, firm, mobile, non-tender lymph nodes were found in the posterior neck triangle, with no skin changes or lymphadenopathy elsewhere. Fine needle aspiration cytology (FNAC) suggested reactive lymphadenitis, and antibiotics were ineffective. An excision biopsy was performed, and histopathology confirmed the diagnosis of Kikuchi-Fujimoto disease. The patient was treated with symptomatic treatment with good results. The aim of the case report is to increase awareness of this disease so that it can be recognized early and prevent misdiagnosis and subsequent over treatment. The best way to diagnose this non-specific lymphadenopathy is to do a biopsy of the lymph node. Once diagnosed, it can be managed conservatively as it is mostly self-limiting.

Keywords: Kikuchi-Fujimoto disease, Histiocytic necrotizing lymphadenitis, Cervical lymphadenitis, Lymphadenopathy

INTRODUCTION

Kikuchi-Fujimoto disease or Histiocytic necrotizing lymphadenitis is an uncommon illness that typically has a benign, self-limiting course but can resemble dangerous conditions like lymphoma or infected lymphadenitis. Its importance stems from the possibility of incorrect diagnosis, which could result in needless tests and treatments. Its etiology is yet unknown, but there have been reports of potential links to autoimmune diseases such systemic lupus erythematosus. In young patients with unexplained cervical lymphadenopathy, Kikuchi-Fujimoto illness should be taken into consideration. Early diagnosis with lymph node biopsy can prevent overtreatment and allow for proper conservative care. Since clinical and laboratory presentation typically mimics circumstances requiring time-consuming and expensive diagnostic and therapeutic treatments, early diagnosis is essential.[1]

CASE REPORT

A 19-year-old Marathi (Indo-Aryan) female patient came with chief complaints of multiple swellings over both sides of the neck. The swellings were insidious in onset, gradually progressive, and not associated with any pain or discomfort. There was a positive history of evening rise of fever on few occasions, and which was not accompanied by chills or rigors. There wasn't a history for loss of appetite or loss of weight. The patient had similar complaints in childhood and during that episode the symptoms were relieved on taking medications for which no documentation was available. There was no history of similar complaints in the family.

On examination, she was averagely built and nourished and was afebrile. There were two swellings on the left side of the neck and three swellings on the right side of the neck; all the swellings were situated in the posterior triangle of the neck. There were no skin changes visible over the swellings.

On palpation, all the swellings were firm, matted, mobile, and non-tender. The temperature over the swellings was normal. The largest lesion was 3 cm large and the smallest being 0.5 cm. There were not any palpable swellings in the axilla or the inguinal region (Figure 1).

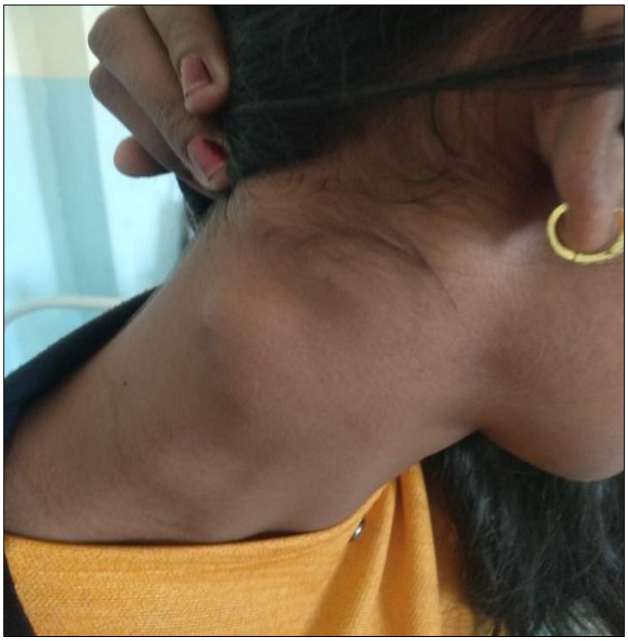

Figure 1: Posterior triangle lymphadenopathy.

A fine needle aspiration cytology (FNAC) was performed which reported the swellings to be reactive lymphadenitis without any confirmatory diagnosis. A provisional diagnosis of reactive lymphadenitis was made, and a course of antibiotics was prescribed. The medications had no effect on the lesions. After discussions with the patient, it was decided to perform an excisional biopsy under local anesthesia. A minimally enlarged lymph node of size 0.5 cm was excised and sent for histopathological examination. The histopathology report confirmed the diagnosis to be Kikuchi Fujimoto disease.

Histopathology

An FNAC was performed which reported the swellings to be reactive lymphadenitis without any confirmatory diagnosis. After discussions with the patient, it was decided to perform an excisional biopsy under local anesthesia. An enlarged lymph node of size 0.5 cm was excised from the posterior triangle and sent for histopathological examination. The histopathology report confirmed the diagnosis to be Kikuchi Fujimoto disease.

Treatment, outcome and follow up

The case was discussed in a multi-disciplinary team meeting, and a management plan of conservative treatment was agreed. This included thorough counselling regarding the disease, management options, and need for long term follow-up. She was prescribed with non-steroidal anti-inflammatory drug and anti-pyretic medications as required for symptom control. The patient reported a gradual decrease in the size of the swellings and absence of further fever episodes. The patient was followed regularly for two months, and showed complete remission of the left cervical lymphadenopathy, along with reduction in the size and number of right cervical groups of lymph nodes.

DISCUSSION

Kikuchi-Fujimoto disease, which is also known as the histiocytic necrotizing lymphadenitis, is generally a self-limiting condition usually affecting the female sex under the age of 30 years.[1] This disease was initially described in Japan (1972) when pathologists Kikuchi and Fujimoto reported a non-malignant, self-limiting lymphadenopathy with necrotizing lymphadenitis and characteristic histologic appearance.[2] KFD cases have been reported all over the world (Germany, United States, Iran, and Italy), although there has been a higher prevalence among people of Japanese and Asiatic descent. The pathogenesis for KFD suggested ranges from viral to autoimmune disease processes, with organisms such as Yersinia, Toxoplasma, Ebstein Barr virus, cytomegalovirus amongst others have been speculated but further studies have been unsuccessful in confirming the causality relationship.[3]

Clinical features

The disease onset is most often acute or subacute with enlarged painful and tender lymph nodes measuring 0.5-4 cm in size, with pain and tenderness being an inconsistent finding. Lymphadenopathy is noted in 59% of patients; majority present in the posterior cervical triangle, although 2% to 40% cases have shown involvement of other anatomic locations and rare generalized lymphadenopathy.[4,5] Extra-nodal involvements of this disease is atypical, but cutaneous, eye and bone marrow affections have been well documented.[4,5]

The cutaneous manifestations are erythematous papules, plaques, indurated lesions, facial malar erythema, or even ulcers usually presenting on the face or upper body.[4,6] The known ocular manifestations reported have been vaso-occlusive retinopathy, bilateral retinal vasculitis, papillary edema, oculomotor palsy, Parinaud oculoglandular syndrome, pre-retinal or vitreous hemorrhage, angiitis, anterior uveitis or panuveitis and papillary conjunctivitis.[7]

KFD has also been documented to occur in conjunction with systemic lupus erythematosus (SLE), with the latter being diagnosed at the same time or after being diagnosed

with KFD.[8-10] The clinical picture seen is not specific, and differential diagnoses should include bacterial infections (tuberculosis, cat-scratch disease), viral infections (infectious mononucleosis), malignant lymphoma, or metastatic cancer.[10]

As pathogenesis remains unclear, it becomes a task to differentiate KFD from various other causes of lymphadenitis. The rate of misdiagnosis with the other forms of lymphadenitis is around 40% and can lead to frequent misdiagnosis and unnecessary treatment.[9] Medical imaging techniques such as ultrasonography and magnetic resonance imaging have been performed. Although these imaging procedures are quicker and less invasive, they only provide information regarding lymph node enlargement and cause confusion.[9,11] Imaging studies using 18 F-FDG on PET/CT are unable to differentiate KFD from malignant lymphoma.[11]

KFD presently is a diagnosis of exclusion and confirmed by histopathological examination of affected lymph nodes. The features of widespread coagulative necrosis with histiocytosis are noted in the cortical and paracortical areas of the affected lymph nodes. An immunohistochemical staining to reveal the presence of MPO-positive and CD68-positive cells can help in differential diagnosis, as features of mononuclear cells with scattered nuclear debris and crescent-shaped tissue cells are indistinguishable from the picture seen in lymphoma.[9,12] An immunohistology analysis conducted by Kuo revealed histiocytes to be the predominant cells along with the plasmacytoid monocytes. CD8(+) T cells were noted, whose number correlated to the disease duration, with absent B cells and insignificant number of OPD4(+) cells. Also, a study of eight cases with flow cytometric analysis of the DNA showed it to be diploid.[13]

Owing to the benign and self-limiting nature of the disease and non-availability of treatment guidelines, management has largely been based on published articles and experts. Patients presenting with symptoms and extra-nodal manifestations have responded to short pulse corticosteroids, nonsteroidal anti-inflammatory drugs and anti-pyretics.[6] Patients can be managed with titrated doses of either single or combined regime of corticosteroids and hydroxychloroquine or a dual immunosuppressive therapy.[14,15] A case reported in a Japanese woman who priorly showed response to high dose prednisolone relapsed on tapering of doses and was subsequently managed with combination of hydroxychloroquine and low dose prednisolone.[15] Rezai and et al reported a case with a history of travel to India to have been treated with hydroxychloroquine on two occasions and shown rapid response.[16]

CONCLUSION

This case report of Kikuchi-Fujimoto disease highlights how a benign condition can closely resemble more serious causes of cervical lymphadenopathy, making diagnosis challenging in clinical practice. It demonstrates the limitations of initial investigations such as FNAC and imaging and underscores the importance of an excisional biopsy for accurate diagnosis. Imaging studies (e.g., PET/CT) are nonspecific, making histopathological lymph node examination the gold standard for diagnosis. The patient's good response to conservative management further supports the self-limiting nature of the disease and the need to avoid unnecessary aggressive treatment. Additionally, the possibility of recurrence and its association with autoimmune conditions like systemic lupus erythematosus emphasize the importance of careful follow-up. Overall, this case aims to build on existing knowledge by reinforcing a practical and patient-centered approach to diagnosis and management, while highlighting the need for greater awareness and further research into this uncommon condition.

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