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KIKUCHI-FUJIMOTO DISEASE: A CASE REPORT FROM KASHMIR

Otolaryngology

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KEYWORDS

INTRODUCTION

Cervical lymphadenopathy persisting for greater than three weeks should be referred to a head and neck clinic as a suspected case of cancer according NICE guidance.In this article, we examined a case of persistent tender cervical lymphadenopathy which had confounded diagnosis until a histological specimen was obtained from the lymph tissue.Kikuchi-Fujimoto disease (KFD) or histiocytic necrotizing lymphadenitis is an uncommon, idiopathic, generally self-limited cause of lymphadenitis.[1, 2]The cause of Kikuchi-Fujimoto disease is unknown.Some kind of viral or post viral etiology has been proposed.There have also been reports of a possible link between KFD and systemic lupus erythematosus (SLE).Kikuchi-Fujimoto disease is an extremely rare disease.Its incidence has been reported worldwide with a higher prevalence among Japanese and other Asiatic individuals.KFD is more common in females compared to males with a male to female ratio of 1:4.People under 30 years of age are more affected by this disease than any other age group.[3]

Case History:

A 18 year old girl presented with history of fever for 4 weeks which was intermittent,mild to moderate degree,associated with mild headache and mild pain in the right side of the neck with swelling at the same site which is gradually progressive in size over the last 10 days.No history of anorexia,weight loss,nausea,rashes,arthralgia,oral ulcerations,respiratory or genitourinary symptoms or contact with animals or recent travel.There was no previous history of tuberculosis or contact with tuberculosis.She did not have history of any drug intake or atopy.She did not have any other significant medical problems.On examination patient was afebrile,2 cervical lymph nodes were palpable on the right side of the neck mainly posterior triangle of neck,discrete,mobile, minimally tender and varying sizes, largest being 2×3 cm, with normal overlying skin.Rest of the general physical and systemic examination were normal.The patient was referred from department of ENT GMC SRINAGAR to the department of Radiational oncology GMC Srinagar where patient was further evaluated and followed.

On investigation: hemoglobin,10gm%;total leukocyte count, 6000cells/ cumm; ESR, 50mm/1st hr; platelet count, 1,55,000/cumm; chest x ray was normal; Mantoux test, Widal test, HBsAg, HCV ab, HIV by ELISA were negative.Bone marrow examination was normal,ANA and anti dsDNA was negative,ultrasound abdomen showed mild hepatomegaly of 156mm. Histopathological examination of the lymph node was showing large mottled area suggestive of necrosis and foamy macrophages in sheets along with abundant karyorrhectic debris and cresentic histiocytes with eccentrically placed nucleus and engulfed debris consistent with Kikuchi's disease.Gram stain and Ziehl-Neelsen stain for acid fast bacilli were negative.Fine needle aspiration cytology (FNAC) of the right posterior triangle node showed features suggestive of reactive lymphadenitis and the patient was started on oral antibiotics. Since the patient continued to have fever and persistent lymphadenopathy, in spite of one week of antibiotics,lymph node biopsy was done and the histological features suggested the diagnosis of Kikuchi-Fujimoto disease.Immunohistochemistry custom panel (CD 3,5,10,20,30,23,BCL-2,CYCLIN D1,Ki-67,CD-68) was also done to rule out any malignancy. The Patient was treated symptomatically with nonsteroidal anti-inflammatory drugs and the lymph nodes regressed in four weeks.

Discussion

Kikuchi-Fujimoto disease was first reported in 1972 by Kikuchi[1]and Fujimoto[2]et al, as lymphadenitis with focal proliferation of reticular cells associated with numerous histiocytes and extensive nuclear debris. The etiology and pathogenesis of Kikuchi's disease is not known but a viral[4,5] bacterial like yersinia and protozoa infections are suggested[6]. A genetic predisposition has been noted and there is female preponderance with a mean age of 30yrs[7,8]. It is a benign and self limiting condition with a low recurrence rate of 3-4%.[9]It usually presents as fever of low grade which may persist for about one week , rarely up to a month along with lymphnode involvement usually localized (cervical), painful, moderately enlarged (1-2cm), occasionally much larger but usually not beyond 7cm and sometimes as cutaneous manifestations like rashes, malar rash, erythematous papules. Other symptoms, like myalgia, arthralgia, chest pain and abdominal pain may also be present.

Histopathological examination of lymph node is diagnostic and shows irregular paracortical areas of coagulative necrosis with abundant karyorrhectic debris distorting nodal architecture. Large number of different types of histiocytes at the margin of the necrotic areas.The karyorrhectic foci are formed predominantly by histiocytes and plasmacytoid monocytes, medium to large sized transformed lymphocytes (immunoblasts). Neutrophils are characteristically absent and plasma cells are either absent or scarce.CT scan of affected lymph node shows hypodense centre with peripheral ring enhancement corresponding to central necrosis.[10]

Because of lack of signs and symptoms and serological markers, diagnosis of Kikuchi's disease is based on histopathological findings. So early recognition of this disease helps in avoiding unnecessary investigations and treatment.

Conclusion

Kikuchi disease is benign and self limiting disorder and treatment is mainly symptomatic. Its association with systemic lupus erythematosus makes these patients to be followed up. Clinicians and pathologists should be aware of this disorder to prevent misdiagnosis and inappropriate treatment.

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