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Case Report

Kikuchi Fujimoto: A Forgotten Etiology of Cervical Lymphadenopathy: Case Report and Literature Review

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Abstract

Kikuchi-Fujimoto's disease is a rare disease, little known by clinicians. It is a necrotizing lymphadenitis, affecting young subjects, with a female predominance. Lymphadenitis are localized, especially in the cervical region, but sometimes also diffuse. The lymph node biopsy allows the diagnosis by finding a necrotizing lymphadenitis with the presence of plasmacytoid monocytes and numerous cells in apoptosis (CD8+ T lymphocytes). The course is usually spontaneously favorable within a few weeks to a few months, only occasionally requiring brief corticosteroid therapy. We report the case of a young patient, who consulted for cervical lymphadenitis in whom the diagnosis of Kikuchi Fujimoto was retained.

Kikuchi's disease may reveal or evolve towards an autoimmune disease, in particular lupus, requiring a long-term clinicobiological follow-up.

Keywords: case report; histology; kikuchi fujimoto; lymphadenopathy

Introduction

Kikuchi-Fujimoto's disease or necrotizing histiocytic lymphadenitis is a benign pathology affecting essentially young women, of unknown etiology. Its diagnosis is primarily histological. It is an often unrecognized cause of cervical adenopathy associated with various clinical symptoms. Kikuchi's disease may reveal or evolve towards an autoimmune disease, in particular lupus, requiring a long-term clinical and biological follow-up.

Case report:

We report the case of a 29-year-old female patient, without any notion of tuberculosis contagion or any past medical history of note, who consulted for the appearance of painful cervical lymphadenitis progressively increasing in size without any other associated signs in a context of fever and conservation of the general state.

Blood tests including immunological tests, chest x-ray and tuberculin test were all normal.
Ultrasound revealed bilateral necrotic lymphadenitis, the cavum was free at nasal endoscopy, a cervicotomy was performed and the histolopathologic evaluation completed by an immunohistochemical study allowed the diagnosis of nonsuppurative histiocytic and necrotizing lymphadenitis refering to Kikuchi Fujimoto disease.

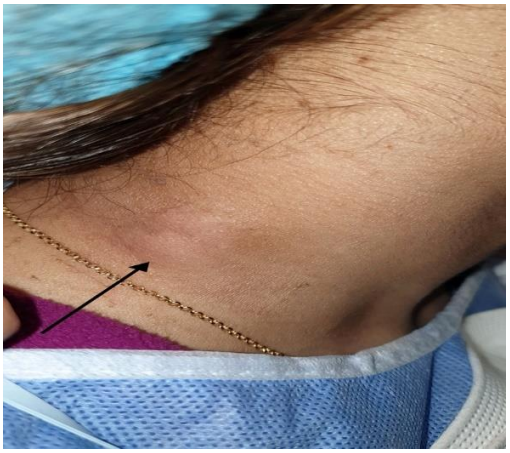

Figure 1: Image of the patient showing the cervical lymphadenitis**.**

Discussion:

Kikuchi-Fujimoto disease (KF) was first described in 1972. It is generally admitted to be a non-neoplastic disease of lymph node tropism, reactive lymphoid hyperplasia type. KF has been found mainly in young people with a very low recurrence rate (about 3%) [1]. May affect both sexes with female predominance [2]. Mostly observed in East Asia, sporadic cases have been found outside.

A viral or autoimmune cause of KF has been suggested. Some initial reports hinted at Yersinia enterocolitica and Toxoplasma gondii as possible causative agents of KF, mainly on the basis of positive serologic test results. But subsequent studies failed to support these hypotheses. In addition with these microorganisms, the histologic features of lymphadenitis associated clearly differ from those of KF [3,4,5].

To diagnose the disease quickly and accurately, clinical radiological, biological and histological analysis is essential. The clinical manifestations of KF are mainly fever, varying between 38 and 41 °C, lasting about 4 to 6 weeks, superficial lymphadenitis mainly in the neck, 0.5 to 3 cm in diameter, congestive maculo-papular rash, usually on the trunk, limbs and cheeks, sometimes may be associated with mild hepatosplenomegaly with liver enlargement of about 0.5 to 2 cm. Because of the non-specificity of the symptomatology, other more specific analyses must be performed to avoid misdiagnosis and mistreatment.

An anatomo-pathological analysis of the lymphadenopathy biopsy should be performed. Showing extensive coagulative necrosis and histiocytosis. The histological features, such as clusters of plasma-like mononuclear cells with nuclear debris and crescent-shaped tissue cells, are indistinguishable from those of lymphoma, hence the value of complementing with an immunohistochemical study that can reveal MPO-positive and CD68-positive cells [6], such a pathological tool would be useful for diagnosis and rule out the differential diagnosis.

KF is a subacute disease, usually lasting 1-3 months, but can persist for up to 1 year [2]. There is no universally accepted treatment plan, as each case may be different. The primary treatment for KFD is to manage the disease by supporting the patient mentally and physically to accelerate relief of symptoms. Antibiotics are not effective, but their use in this case [7] prevents potential bacterial infections.

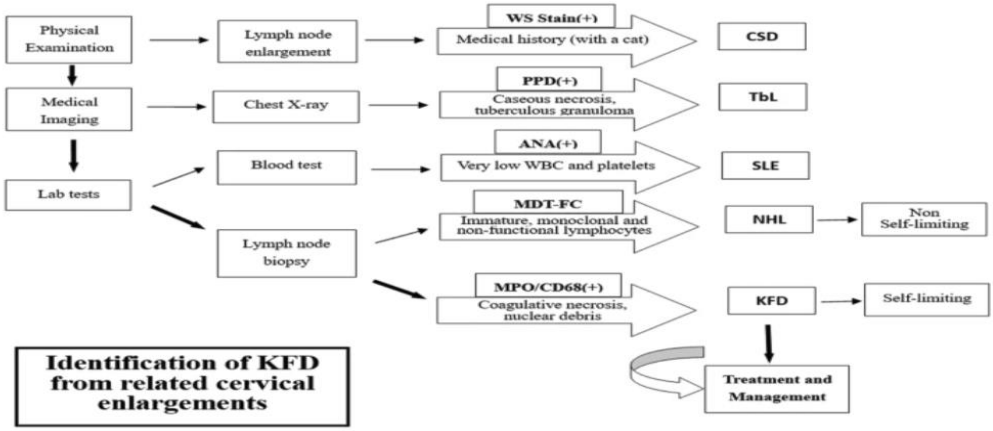

Figure 2: Decision tree of Kikuchi-Fujimoto disease [8].

Conclusion:

Cervical lymphadenitis are a very frequent reason for consultation, requiring a wide range of paraclinical examinations in search of the etiology. Kikuchi-Fujimoto's disease is a rare cause and of elimination, confirmed by histology, an autoimmune work-up is initiated and a long-term clinical-biological monitoring is imposed because of the risk of secondary appearance of autoimmune disease, especially lupus.

Informed Consent: Free and informed consent has been given by the patient.

Conflicts of Interest: The authors declare no conflicts of interest.

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