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RESEARCH ARTICLE

A RARE DISEASE DIAGNOSED AS LYMPH NODE TUBERCULOSIS: KIKUCHI-FUJIMOTO DISEASE

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ABSTRACT

Kikuchi-Fujimoto disease (KD) is a rare and self-limiting disease. It is characterized by benign necrotizing lymphadenitis, which is accompanied by fever and lymphadenopathy, with or without extension to other organs, such as skin, eyes and bone marrow. The histopathological characteristics of this disease are nuclear debris surrounded by a histiocytic infiltrate with different degrees of lymphocytes and immunoblast. Its etiology is unknown, it has been associated with systemic lupus erythematosus and infections by Epstein-Barr virus (EBV), parvovirus B-19, HIV and humanos herpesvirus 6; although the nature of the relationship is unclear. The case of a girl with Kikuchi-Fujimoto disease is presented.

INTRODUCTION

Kikuchi-Fujimoto disease is a rare and benign disease, first described in Japan in 1972, which presents with fever and cervical lymphadenopathy (Altinel Acoglu, 2018); sometimes with other manifestations, which usually increase the misdiagnosis, confusing mainly with infectious causes that are also accompanied by lymphadenopathy, such as tuberculosis, lymphoma, HIV, systemic lupus erythematosus, toxoplasmosis and infectious mononucleosis. Having a very general clinical picture, the diagnosis is made by histopathological examination of a node. Extrapulmonary tuberculosis cover 15-20% of tuberculosis presentations. Peripheral lymphadenitis is the second most common presentation of extrapulmonary tuberculosis in children, and the cervical region is the most common site affected. Lymphadenopathy at the cervical-facial level presents painless, with slow growth. Systemic symptoms may not be present, however, 55% must be taken into account (Neyro, 2018). A histological early0 diagnosis could prevent misdiagnosis and inappropriate use of antibiotics or immunomodulatory medications (Tariq, 2014). The case of a 6-year-old school patient with bilateral cervical lymphadenopathy is presented.

CASE REPORT

A 6-year-old female girl, with bounded bilateral cervical lymphadenopathy, of hard consistency, local temperature increase, non-painful, not adhered to deep planes; of three weeks of evolution, with intermittent fever between 38-40^{O} Celsius, accompanied by diarrhea, diffuse abdominal pain and generalized joint pain, without weight loss or diaphoresis. Initially, lymph node tuberculosis was suspected and treatment with clarithromycin 15 mg/ kg/day orally for 14 days, ceftriaxone 100 mg/kg/day intravenously for three days and ibuprofen 10 mg/ kg/doce, orally for 15 days; without improvement. Laboratory studies were performed; hematic biometrics reported leukocytosis 16400, absolute neutrophils 8850, absolute lymphocytes 5084, monocytes 1200, hemoglobin 12.5 mg/dl, hematocrit 39.9%, platelets 565000, procalcitonin less than 0.20 ng/dl, elevated C-reactive protein 4.6 mg /dl. Serology for Epstein Barr Virus (EBV) with old infection data: EBV positive nuclear antigens, IgG against positive capsular antigen, IgM against negative capsular antigen, early negative antigen (AB ANTI VEB IgG 139, IgM 10, AB ANTI. AG NUCLEAR DE CAPSIDE IgG 1670, IgM 5), antinuclear antibodies, anticardiolipins negatives. Peripheral blood cultures without development, VDRL, ELISA for HIV, IgM serology for Toxoplama gondii, Rubella virus, Cytomegalovirus and Herpes virus 1 and 2 negatives. An ultrasound was performed that was reported with adenomegalies in the neck, right side of 8x6 cm. and left side of 6x7 cm., with data suggestive of lymph node necrosis, with probable inflammatory-infectious process versus infiltrative process, without other findings (Figure 2, 3, 4 and 5). The tomography neck report, with cervical and bilateral lymphadenopathy, in IIB and VII zone, with data of central necrosis; thoracic region unchanged. The size reported was 8x6 cm right and left of 6x7 cm (Figure 6a, 6b). In the chest tomography, with pulmonary window, nodules or

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Figure 1a

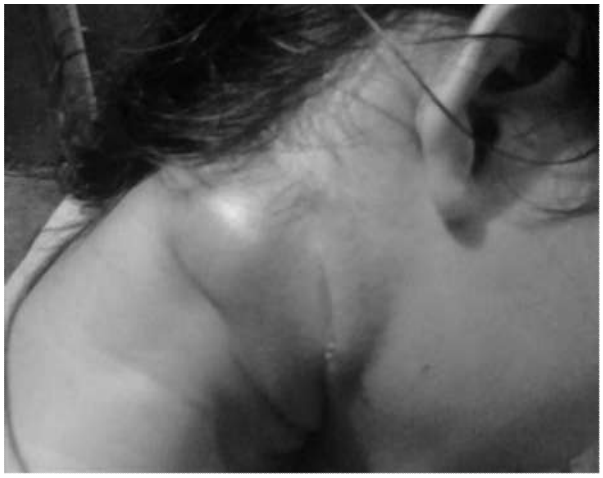
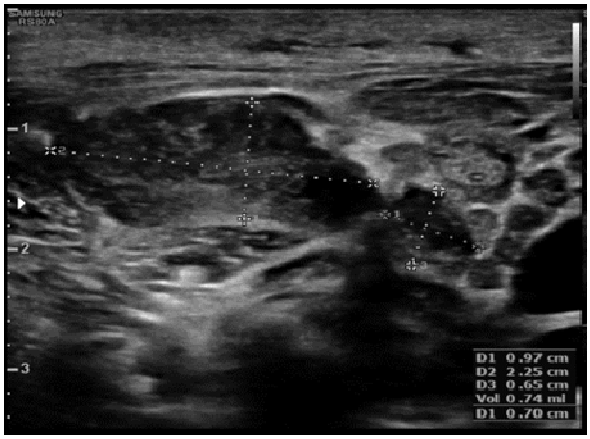
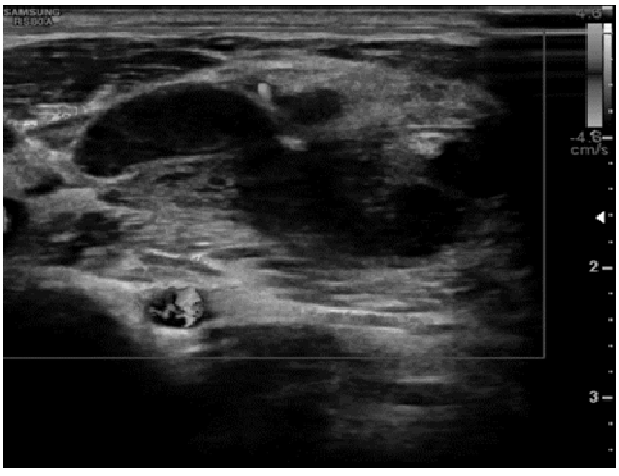
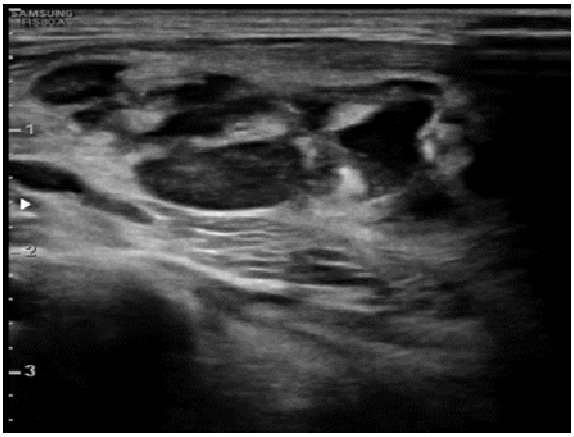


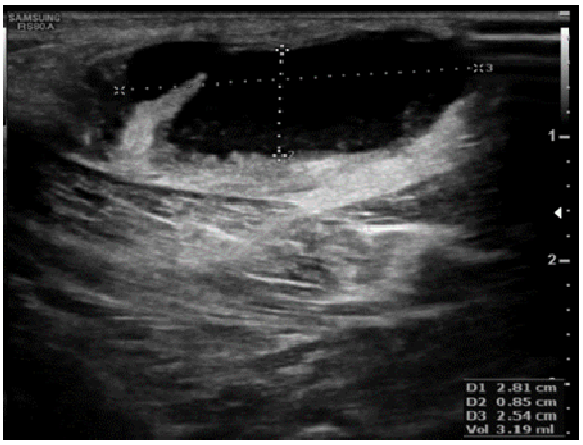
Figure 1b

Figure 1a, 1b: Presence of cervical lymphadenopathy right side (arrow)









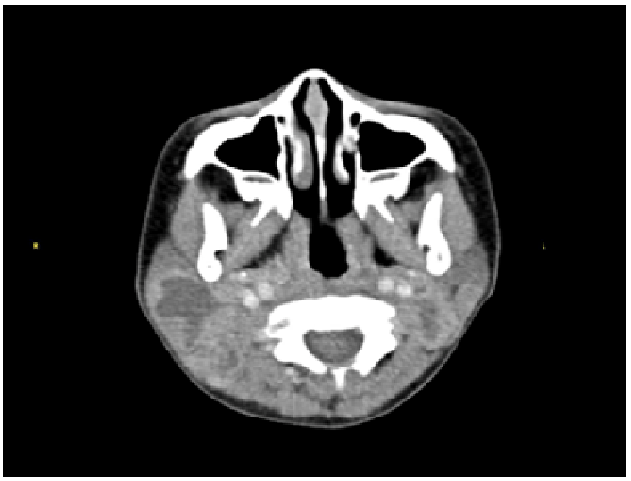


Figure 2, 3, 4 and 5: Neck ultrasound where multiple adenomegalies are identified in bilateral V A and I B levels, predominantly on the right side with asymmetric cortical thickening, lobed contours, associated with aberrant vascularity.

consolidations were not found (Figure 7). A biopsy of the lesion was taken, with reported Ziehl Neelsen staining, no alcohol resistant acid bacilli were observed, Gram staining without bacteria, polymerase chain reaction (PCR) GeneXpert tuberculosis (TB) negative for complex detection Mycobacterium TB, PCR for Bartonella henselae (for history of living with cats) and and culture for Mycobacterium non-tuberculosis, negatives. Histopathological diagnosis: necrotizing granulomatous lymphadenitis (Figure 8). Subsequently, the patient received treatment with a non-steroidal anti-inflammatory drug for one month, which showed improvement and decrease in the size of the lymph nodes, with complete remission of the condition in two months.

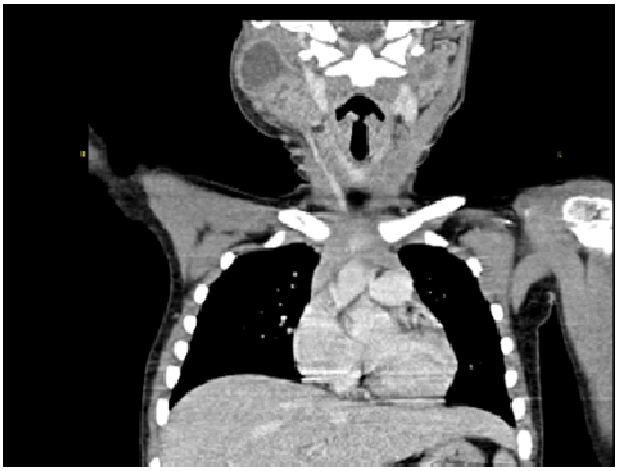


Figure 6a, 6b:Contrast neck tomography showing right retroauricular lesion with peripheral enhancement, associated with hypodense center that suggests necrosis.

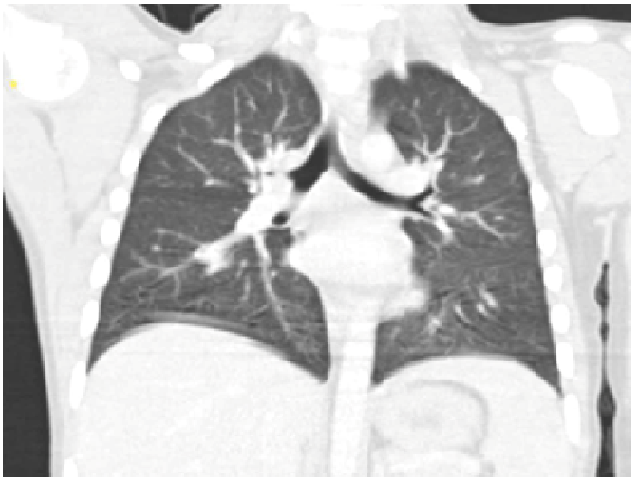


Figure 7: Chest tomography, pulmonary window, with no evidence of nodules or consolidations

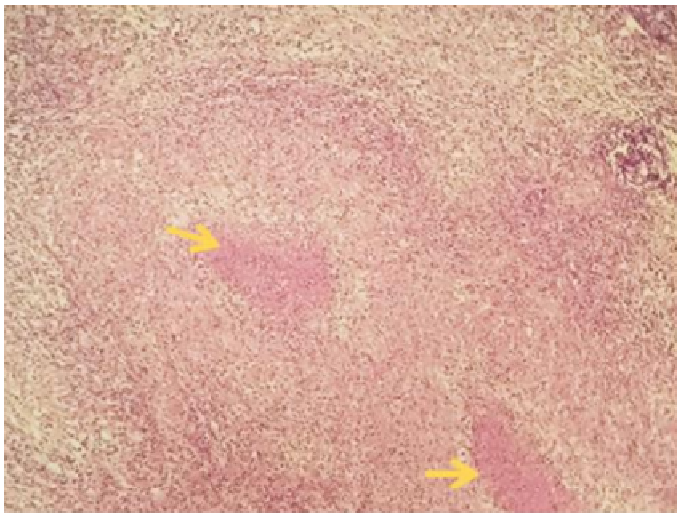


Figure. 8: Histological section of lymph node where granulomas with necrozant center are observed.

DISCUSSION

Kikuchi disease is a pathology that is difficult to diagnose, the quality of being diagnosed in the differential diagnoses of cervical adenopathies, with or without fever; the most frequent is the tuberculosis (Cruz, 2016; Andriamampionona, 2015) and lymphoproliferative diseases, connective tissue and immunological diseases (Williams et al., 2019). Currently, the finding of fever and cervical lymphadenopathy, in young women, lead to an initial diagnosis of tuberculosis or lymphoma, in developing countries, initiating unnecessary treatment in children (El-Wajeh, 2018). As background, in the first studies of Kikuchi-Fujimoto, 21 cases out of 40 studied, were frequently diagnosed as tuberculous lymphadenitis (Kaur et al., 2014). Clinically it occurs more frequently in females, with high fever and cervical lymphadenopathy (Dalugama, 2017), as presented in our case. Regarding imaging studies, some ultrasound features that allow differentiating KD from TB lymphadenopathy have been studied. In a study that included 135 patients with kikuchi disease, the presence of echogenic hilum was found in 90% of them (Ryoo et al., 2015), which is observed in the ultrasound of our patient. Regarding the location, no conclusive results have been found, because the location of lymphadenopathy in KD is cervical, as well as in TB lymphadenitis (Pang et al., 2018; Estomba et al., 2016). The presence of calcifications, the cluster of nodes and the absence of vessels have been found in TB lymphadenitis, and not in KD (Sigg et al., 2018), findings that were not found in the reported case. The ultrasound has a negative predictive value (NPV) of 96% and a positive predictive value (PPV) of 36% to determine the malignancy of a nodule, with a sensitivity and specificity of 77% and 80% respectively (Whitman et al., 2011). In the tomography it has been studied that the presence of bilateral lymphadenopathy, perinodal infiltration, and minimal ganglionic necrosis, are characteristic of KD (15). A study of 96 patients with KD, reported that lymphadenopathy is more frequent in levels II to V (Anikhindi et al., 2017). The definitive diagnosis is pathological, so it is necessary to perform a biopsy, with the characteristic finding necrozant lymphadenitis, with a few or no neutrophils and a macrophage histiocytic infiltrate (Al-Allaf, 2018; Andola, 2016).

CONCLUSION

Kikuchi disease is a pathology that is difficult to diagnose, which should be considered in the differential diagnoses of cervical adenopathies, with or without fever, such as tuberculosis, lymph proliferative diseases, connective tissue and immunological diseases. It is very important to know how to differentiate, because the treatment, clinical evolution and prognosis are different. For this reason, we consider that imaging studies together with the clinic are of great help in determine the need for biopsy or follow-up studies and therefore, avoid unnecessary exposure to the pediatric patient to invasive studies and avoid worries in family members.

Glossary of abbreviations

KD Kikuchi disease
EBV Epstein-Barr virus
HIV Human immunodeficiency virus
TB Tuberculosis
PCR Polymerase chain reaction
NPV Negative predictive value
PPV Positive predictive value

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