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LABORATORY DIAGNOSIS OF MACROPHAGEAL ACTIVATION SYNDROME IN VISCERAL LEISHMANIASIS IN AN INFANT: A CASE REPORT

Zineb AL Youssoufi[1,2]*, Mehdi Aziki[1,3], Hicham Yahyaoui[1] Mohamed Chakour[1]

[1]Hematology Laboratory, Avicenne Military Hospital, Marrakech, Morocco.
[2]Faculty of Medicine, Mohammed VI University of Health Sciences, Casablanca, Morocco.
[3]Faculty of Medicine and Pharmacy, Cadi Ayyad University, Marrakech, Morocco.

***Corresponding Author: Zineb AL Youssoufi**
Hematology Laboratory, Avicenne Military Hospital, Marrakech, Morocco.

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ABSTRACT
Visceral leishmaniasis (VL) is a severe parasitic disease that can lead to macrophage activation syndrome (MAS), a serious hematological complication that can be fatal without early management. The diagnosis of MAS is complex due to the clinical similarity with other inflammatory or infectious pathologies. This study reports a case of VL complicated by MAS in an infant and highlights the importance of biological diagnosis. The main objective is to describe the diagnostic process, highlighting the role of the laboratory in identifying this potentially fatal complication. The study concerns an 18-month-old infant, hospitalized for prolonged fever, pancytopenia and hepatosplenomegaly in a region endemic for leishmaniasis. A biological assessment was carried out including a blood count, a blood smear, the dosage of inflammatory markers, a Leishmania ELISA serology and an analysis of the bone marrow smear. Despite a first negative myelogram, the persistence of biological and clinical abnormalities motivated the performance of a second myelogram, confirming the presence of hemophagocytosis.

KEYWORDS: Visceral leishmaniasis, Macrophage activation syndrome, Hemophagocytic lymphohistiocytosis.

INTRODUCTION

Visceral leishmaniasis (VL) is a common cause of secondary macrophage activation syndrome (MAS) in children in Morocco. The latter can be a real threat to life and warrant specific treatment.

This article presents a case study on the diagnosis of SAM on a VL in an infant and its revelation in the hematology laboratory.

METHODS

The study was conducted on an 18-month-old infant born to 1st degree consanguineous parents, originally from the Taroudant region (leishmaniasis endemic area), hospitalized for prolonged fever, incomplete bone marrow failure syndrome (anemic syndrome, infectious syndrome, no hemorrhagic syndrome), partial tonic - clonic convulsions of the left hemibody and a tumor syndrome consisting of hepatosplenomegaly.

Based on these clinical data we suspected visceral leishmaniasis, a biological assessment was carried out in this direction.

We performed a complete blood count, blood smear, inflammatory marker assay, leishmaniasis serology, and bone marrow smear analysis.

The results were as follows

- Pancytopenia: Normochromic normocytic aregenerative anemia Hb = 9.8 g/ dL, Neutropenia at 552/mm3 and Thrombocytopenia at 63,000/mm3
- Increases in inflammatory markers, namely an ESR of 58 mm, ferritin = 3261 ng / mL and LDH of 1151 IU / L.
- Leishmaniasis serology by the ELISA technique positive with an index of anti-Leishmania sp. type IgG antibodies at 31 NTU (for a positivity threshold > 11)
- Analysis of the bone marrow smear stained with May- Grünwald Giemsa noted an absence of Leishmania bodies.
- Correct liver and kidney function

Based on these clinical -biological data, the diagnosis of VL was made and etiological treatment based on meglumine antimoniate (Glucantime) at 60 mg/kg/day was started.

However, we noted a persistence of cytopenia and fever

despite probabilistic antibiotic therapy (based on ceftriaxone and gentamicin) started in parallel with the etiological treatment, with no improvement in the tumor syndrome. Which leads us to suspect a complication of VL by a macrophage activation syndrome.

A second myelogram was requested to confirm this diagnostic hypothesis. The result was positive and demonstrated images of hemophagocytosis (figures 1),

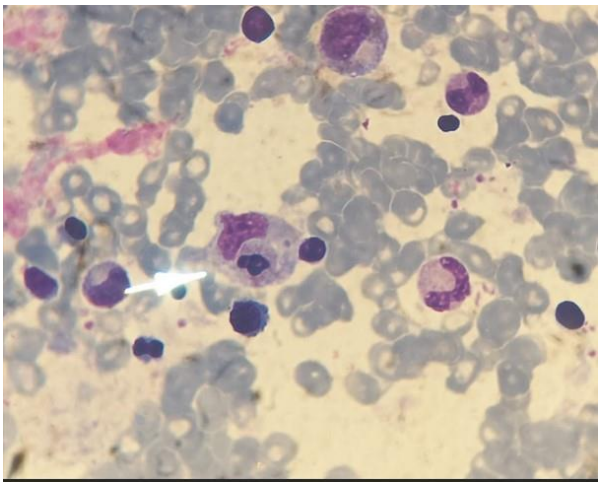

Figure 1: Image of hemophagocytosis on a bone marrow smear stained with MGG.

We then used, in addition to the etiological treatment, steroids in a short course with intravenous immunoglobulins.

The evolution was favorable with improvement of the general condition and apyrexia after day 3 of treatment with an increase in neutrophils and platelets

However, a febrile recovery was noted on day 10 with an increase in triglycerides, ASAT, ALAT and metabolic acidosis with PNN hyperleukocytosis and thrombocytopenia. The outcome was unfortunately fatal and death occurred in a setting of sepsis.

DISCUSSION

Visceral leishmaniasis (VL) is caused by flagellates of the genus Leishmania, transmitted by the bite of female sandflies.[1,3]

Macrophage activation syndrome (MAS) is related to uncontrolled activation and proliferation of T lymphocytes and macrophages in the bone marrow and lymphoid system, it can be primary or secondary to various hematological, autoimmune or infectious diseases.[1,3]

These two entities have similar presentations, the diagnosis of SAM having no clinical specificity can be difficult in case of association. The latter is not rare in our context. Indeed, in a series of 12 cases of SAM followed at the University Hospital of Fez between 2005 and 2009, VL was incriminated in eight cases.[4] Similarly, during a study conducted in Algeria over 10 years, 13 cases of VL collected among 276 were associated with SAM, i.e. a prevalence of 4.7%, with a median age of 30.7 months.[5]

The clinical picture of SAM being not very specific, it is the association of biological signs, very evocative, which leads to the diagnostic presumption. The appearance of the medullogram is characteristic, but inconstant, with histiocytic infiltration, erythroblastosis and image of hemophagocytosis.[1,2] According to the HLH-2004 criteria (hemophagocytic lymphohistiocytis)[2], the diagnosis of SAM was retained in our patient on the basis of clinical signs not resolving after etiological treatment of VL and biological signs including the medullogram showing images of hemophagocytosis , neutropenia and thrombopenia, marked hypertriglyceridemia and hyperferritinemia.

Although our patient's first medullogram came back negative, it was necessary given the high endemicity in our context to do the serology which came back positive. Indeed, in a literature review of 56 cases of SAM-LV association, the first marrow failed to reveal Leishmania bodies in 64.7% of cases.[6]

The treatment of secondary SAM, which is not consensual, involves etiological treatment, associated or not with intravenous immunoglobulins, short-term steroids or immunosuppressants[2,6], which was the case for our patient.

When it is at the origin, VL can be treated, ideally, by liposomal amphotericin B according to a schedule of 20 mg/kg spread over two days, or by antimonial derivatives.[1] Indeed, the survey of Rajagopala et al. revealed a success of 100% of cases treated by liposomal amphotericin B, and of 65.4% in the case of recourse to stibial derivatives.[6]

In our case, treatment with meglumine antimoniate has a possible promoting effect on SAM[6] diagnosed and treated late, a factor that could explain the clinical - biological deterioration observed in our patient and the occurrence of death in a context of sepsis not controlled by probabilistic antibiotic therapy.

CONCLUSIONS

The severity of the prognosis of the SAM-LV association in infants, not negligible in our context, should lead to the repetition of the medullogram in order to make the diagnosis of SAM complicating VL as early as possible and to the early adoption of an immediately aggressive therapeutic approach.

Declaration of interests

The authors declare that they have no competing interest.

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