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MORPHOLOGY UPDATE

Unexpected *Leishmania* in a bone marrow aspirate

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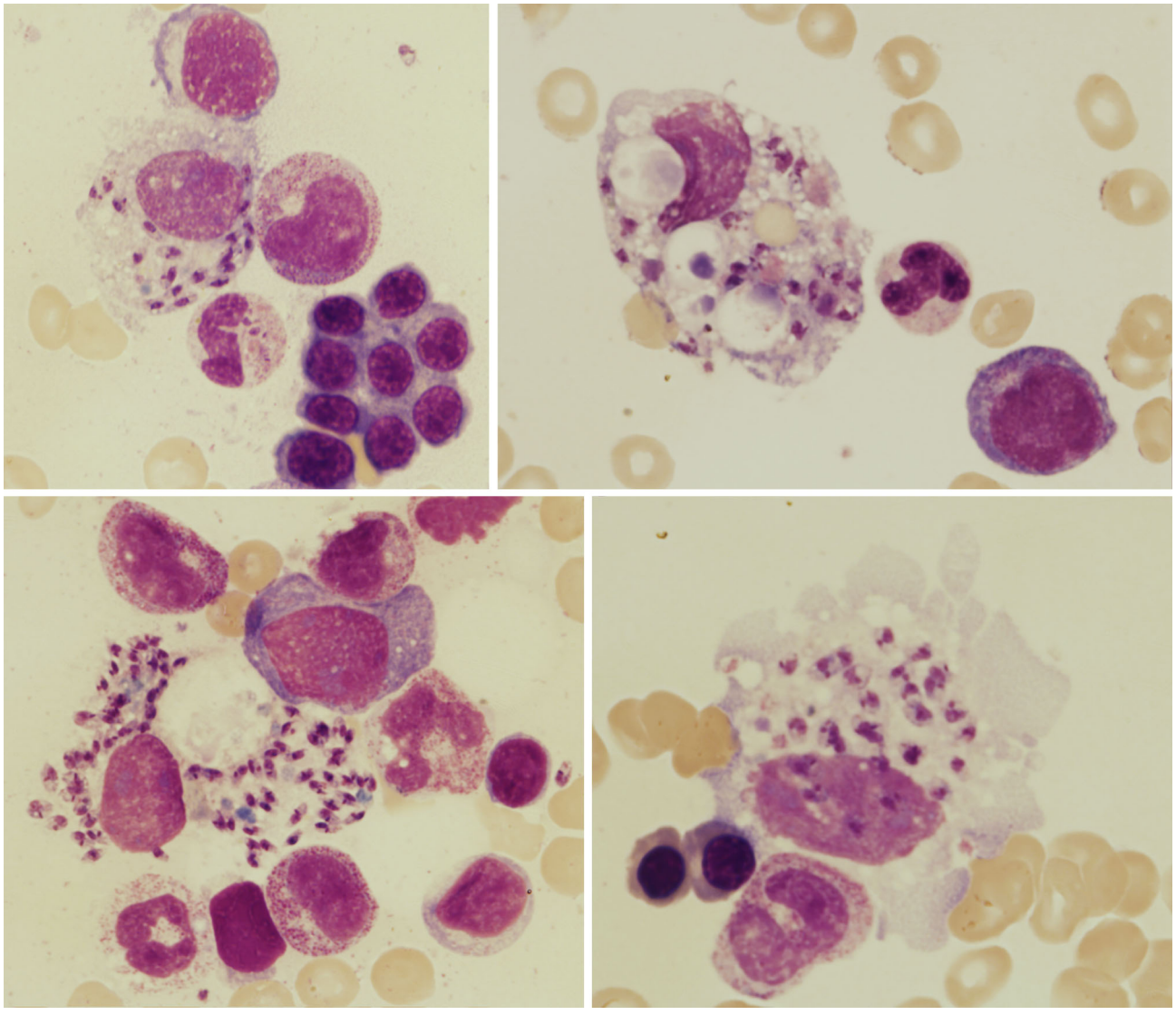

A 57-year-old man, a Belgian resident, was referred for evaluation of refractory hemophagocytic lymphohistiocytosis (HLH), thought to be due to Epstein–Barr virus (EBV) infection. The patient had been treated with high dose corticosteroids, etoposide and rituximab. On admission, he had persistent fever with profuse sweating and splenomegaly. Laboratory tests showed: hemoglobin concentration 93 g/L, platelet count 39×10^9/L, neutrophils 1.9×10^9/L, ferritin >40 000 μg/L, fibrinogen 1.97 g/L and triglycerides 3.66 mmol/L. Based upon these parameters, the HLH probability (Hscore) score was calculated and indicated a 99.8% probability of HLH.[1]

Because of the patient's ongoing illness, a bone marrow aspiration was performed. Cytomorphology showed, as expected, hemophagocytosis (top right, ×100 objective) but importantly also large numbers of intra-cellular and extracellular parasites with the morphological features of *Leishmania* amastigotes (all images, ×100). Polymerase chain reaction for *Leishmania* was positive for *Leishmania donovani-complex* but it was not possible, on the basis of PCR analysis, to differentiate between *L. donovani* and *L. infantum*. However, the patient's previous travels in southern Italy argued in favor of *L. infantum* infection.[2]

Hemophagocytic lymphohistiocytosis is a life-threatening condition of excessive immune activation, a severe hyperinflammatory syndrome induced by aberrantly activated macrophages and cytotoxic T cells. Among the underlying causes of secondary HLH, infections and neoplasms, particularly lymphoid neoplasms, are the most frequently encountered, but autoinflammatory/autoimmune disorders can also be causative and the condition is then sometimes designated macrophage activation syndrome. Visceral leishmaniasis is among the many infections that may cause secondary HLH.[1,3] Leishmaniasis is endemic in Southern but not Northern Europe. However travel is now so common that occasional cases are seen in Northern Europe.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

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REFERENCES

1. La Rosée P, Horne A, Hines M, et al. Recommendations for the management of hemophagocytic lymphohistiocytosis in adults. *Blood*. 2019;133:2465-2477.
2. Cunze S, Kochmann J, Koch LK, et al. Leishmaniasis in Eurasia and Africa: geographical distribution of vector species and pathogens. *R Soc Open Sci*. 2019;6:190334.
3. Diamantadis M, Palioura A, Ioannou M, et al. Hemophagocytic lymphohistiocytosis as a manifestation of underlying visceral leishmaniasis. *Cureus*. 2020;12:e11911.

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