



Case Report

Acute mesenteric lymphadenitis revealing systemic lupus erythematosus

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ARTICLE INFO

Article history:

Received 5 March 2020

Received in revised form 17 March 2020

Accepted 18 March 2020

Keywords:

Acute mesenteric lymphadenitis

Systemic lupus erythematosus

Lymphadenitis

Mesenteric adenitis

ABSTRACT

Acute mesenteric lymphadenitis (AML) remains unusual during systemic lupus erythematosus (SLE), and only a few sporadic observations have been found in the medical literature. It may exceptionally be the first manifestation revealing SLE representing a real diagnostic and therapeutic challenge for clinicians (specific lupus manifestation or infectious complication?).

We report an original observation of isolated AML revealing SLE in a 29-year-old Tunisian woman without pathological medical history. She was explored for acute and febrile abdominal pain evolving for a week and not improved by symptomatic treatment. The somatic examination noted a fever at 38.5°C and a discrete diffuse abdominal tenderness. Biology showed a marked biological inflammatory syndrome, leukopenia at 3400/mm³, neutropenia at 1900/mm³, and lymphopenia at 1140/mm³ with no other abnormalities. The abdominal ultrasound and computed tomography objectified multiple large, hypoechoic, and richly vascularized on Doppler mesenteric lymph node compatible with the diagnosis of AML. Subsequent investigations concluded to SLE.

The patient was treated with systemic glucocorticoids and hydroxychloroquine with favorable outcome. No recurrence has been noted for two years.

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1. INTRODUCTION

The digestive manifestations of systemic lupus erythematosus (SLE) are frequent and very polymorphic. They remain by far dominated by intestinal pseudo-obstructions, peritonitis, ascites, and lupus enteritis [1, 2]. Other rarer complications have been reported: pancreatitis, mesenteric vasculitis, gastrointestinal necrosis and/or

perforation, hepatitis, splenic infarction, mesenteric vessel thrombosis, and mesenteric panniculitis [3]. Abdominal symptoms can be the primary presenting signs of lupus disease and thus represent a real diagnostic challenge for clinicians [2-4].

Acute mesenteric lymphadenitis (AML) remains a rare and unusual manifestation during SLE; forms revealing the disease are exceptional [5, 6].

We report the original observation of isolated AML as the

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<http://doi.org/10.5281/zenodo.3714992>

first manifestation of SLE in a 29-year-old woman.

2. CASE REPORT

A 29-year-old Tunisian woman, without pathological medical history, was explored for acute and febrile abdominal pain evolving for a week and not improved by symptomatic treatment. There have been no reports of associated vomiting or bowel transit disturbances.

The somatic examination noted a fever at 38.5°C and a discrete diffuse abdominal tenderness with peri-umbilical maximum. No peripheral lymphadenopathy, palpable masses, or visceromegalies have been noted.

Biology showed a marked biological inflammatory syndrome with erythrocyte sedimentation rate at 87 mm/H1, C-reactive protein (CRP) at 56 mg/l, and polyclonal hypergammaglobulinemia at 19.3 g/l. The total blood count showed leukopenia at 3400/mm³, neutropenia at 1900/mm³, and lymphopenia at 1140/mm³ with no other abnormalities. The other basic laboratory tests were within normal limits: hemoglobin, platelets, creatinine, blood sugar, serum ionogram, transaminases, muscle enzymes, calcemia, lipid parameters, and urine tests.

The abdominal ultrasound and computed tomography (CT) objectified multiple large, hypoechoic, and richly vascularized on Doppler mesenteric lymph node compatible with the diagnosis of AML (Figure 1). There has been no evidence of tumor, intraperitoneal effusion, or thrombosis of the mesenteric vessels.

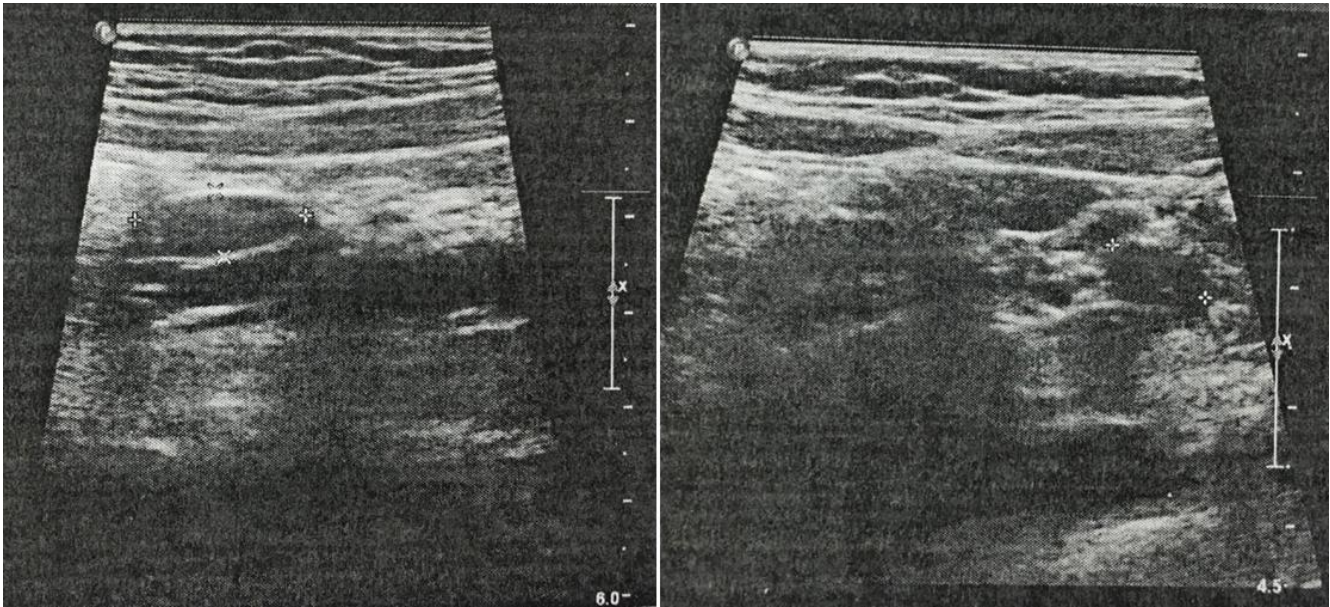


Figure 1: Abdominal ultrasound: multiple large and hypoechoic mesenteric lymph node.

Subsequent investigations made it possible to eliminate an infectious or cancerous cause: blood cultures, urine direct examination and culture, different bacteriological and viral serodiagnostics, tumor markers, chest radiography,

thoraco-abdomino-pelvic CT scan, gastroscopy, colonoscopy, transthoracic echocardiography, and tuberculosis tests.

The immunological assessment revealed positive anti-nuclear, anti-double-stranded DNA, and anti-phospholipid antibodies. anti-neutrophil cytoplasmic antibodies (ANCA) and cryoglobulins were negative.

The diagnosis of SLE with revealing acute mesenteric lymphadenitis was retained. Investigations for other specific visceral disorders of SLE (renal, cardiac, and neurological) were negative.

The patient was treated with hydroxychloroquine (400 mg/day), salicylic acid (100 mg/day), and systemic glucocorticoids (0.5 mg/kg/day for four weeks followed by progressive decrease).

The outcome was favorable with disappearance of clinical complaints during the first week, normalization of the hemogram and CRP after two weeks of treatment, and normal abdominal ultrasound at one month of treatment. No recurrence has been noted for two years.

3. DISCUSSION

Lymph node involvement is far from uncommon during SLE [6-9]. Its prevalence is estimated at 12-59%, and the most common sites are: cervical (43%), mesenteric (21%), axillary (18%), and inguinal (17%). Other sites are more unusual (hilar, mediastinal, and retroperitoneal) [9]. Lymphatic involvement is only exceptionally reported as the primary presenting manifestation of the lupus disease

[6-9].

Acute mesenteric lymphadenitis (AML) remains unusual during lupus, and only a few sporadic observations have been reported [5,6].

SLE is also an exceptional and unusual etiology of acute mesenteric lymphadenitis; indeed only one case in the series of Macari M et al, of 60 patients with AML had SLE as underlying cause (1.6%) [10].

As in our case, AML may exceptionally be the first manifestation revealing SLE [5, 6]. It therefore represents a real diagnostic and therapeutic challenge for clinicians (specific lupus manifestation or infectious complication?) [11].

The differential diagnosis of lupus AML includes the infectious causes which are particularly frequent in lupus patients (tuberculosis and cryptococcosis) [5, 11], metastatic disease (squamous cell carcinoma and germ cell-tumor) [5], and histiocytic necrotizing lymphadenitis (Kikuchi-Fujimoto disease) which is also frequently associated with SLE [5, 12].

Lupus mesenteric lymphadenitis can be isolated or associated with other lupus manifestations such as: ascites, peritonitis, peritoneal panniculitis, and pancreatitis [5,12-14]. It can also be associated with lymphadenopathies in other lymphatic sites [6]. It can even precede the diagnosis of lupus disease by several years [13].

SLE-specific mesenteric lymphadenitis usually has a good prognosis and responds quickly to systemic corticosteroid therapy [5].

4. CONCLUSION

As exceptional and unusual as it is, this abdominal manifestation of SLE deserves to be known by healthcare professionals to avoid diagnostic and therapeutic delays. Our observation is distinguished by its isolated and inaugural character.

It thus seems useful to screen for SLE in front of any AML that does not prove itself, in particular in young women.

5. REFERENCES

1. Fawzy M, Edrees A, Okasha H, El Ashmaui A, Ragab G. Gastrointestinal manifestations in systemic lupus erythematosus. *Lupus*. 2016;25(13):1456-62. doi: 10.1177/0961203316642308.
2. Sran S, Sran M, Patel N, Anand P. Lupus enteritis as an initial presentation of systemic lupus erythematosus. *Case Rep Gastrointest Med*. 2014;2014:962735. doi: 10.1155/2014/962735.
3. Stoddard CJ, Kay PH, Simms JM, Kennedy A, Hughes P. Acute abdominal complications of systemic lupus erythematosus. *Br J Surg*. 1978;65(9):625-8. doi: 10.1002/bjs.1800650911.
4. Henc-Mitis J. Peritoneal symptoms in the initial stage of systemic lupus erythematosus. *Wiad Lek*. 1987;40(16):1131-3.
5. Radin R. Acute mesenteric and retroperitoneal lymphadenitis in systemic lupus erythematosus: case report. *Abdom Imaging*. 2001;26(4):411-3. doi: 10.1007/s002610000182.
6. Kitsanou M, Andreopoulou E, Bai MK, Elisaf M, Drosos AA. Extensive lymphadenopathy as the first clinical manifestation in systemic lupus erythematosus. *Lupus*. 2000;9(2):140-3. doi: 10.1191/096120300678828037.
7. Smith LW, Gelber AC, Petri M. Diffuse lymphadenopathy as the presenting manifestation of systemic lupus erythematosus. *J Clin Rheumatol*. 2013;19(7):397-9. doi: 10.1097/RHU.0b013e3182a6a924.
8. Al-Hashimi H, Bhowmik A. Generalised lymphadenopathy as the first manifestation of lupus nephritis. *BMJ Case Rep*. 2010;2010. doi: 10.1136/bcr.08.2009.2221.
9. Eisner MD, Amory J, Mullaney B, Tierney L Jr, Browner WS. Necrotizing lymphadenitis associated with systemic lupus erythematosus. *Semin Arthritis Rheum*. 1996;26(1):477-82. doi: 10.1016/s0049-0172(96)80028-x.
10. Macari M, Hines J, Balthazar E, Megibow A. Mesenteric adenitis: CT diagnosis of primary versus secondary causes, incidence, and clinical significance in pediatric and adult patients. *AJR Am J Roentgenol*. 2002;178(4):853-8. doi: 10.2214/ajr.178.4.1780853.
11. Kim SH, Kim SD, Kim HR, Yoon CH, Lee SH, Kim HY, et al. Intraabdominal cryptococcal lymphadenitis in a patient with systemic lupus erythematosus. *J Korean Med Sci*. 2005;20(6):1059-61. doi: 10.3346/jkms.2005.20.6.1059.
12. Fernandez SH. Kikuchi's lymphadenitis (necrotizing lymphadenitis) and systemic lupus erythematosus: a case report. *Malays J Pathol*. 2000;22(1):25-9.
13. Hammoudeh M, Siam AR. Recurrent peritonitis with ascites as the predominant manifestation of systemic lupus erythematosus. *Clin Rheumatol*. 1995;14(3):352-4. doi: 10.1007/bf02208353.
14. Bairy I, Berwal A, Prabhu M, Budania LS. Systemic Lupus Erythematosus with Acute Pancreatitis as an Initial Presentation. *J Clin Diagn Res*. 2018;12(9):OR01-OR03. doi: 10.7860/JCDR/2018/35404.12019.