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Cutaneous Leuckocytoclastic Vasculitis Revealing Acute Brucellosis: A Case Report

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Abstract The clinical presentations of human brucellosis are very polymorphic, representing a real challenge for clinicians. Among the unusual manifestations of this infection, vasculitis is particularly uncommon with only few sporadic cases in the world literature. We report an original observation of isolated cutaneous leuckocytoclastic vasculitis revealing acute brucellosis in a 44-year-old Tunisian man who was admitted for a rash with fever that had been evolving for two days. Somatic examination noted fever at 38°C, red-purple infiltrated and tender petechial lesions on feet, legs, and hands, small periungual necrotic lesions, and subungual splinter hemorrhages. The basic biological tests showed a biological inflammatory syndrome with an erythrocyte sedimentation rate of 82mmH1 and a C-reactive protein at 62mg/l. The skin biopsy concluded in leukocytoclastic vasculitis, without signs of malignancy and with a negative direct immunofluorescence. The specific etiological assessment of this vasculitis was negative. Wright's serology was positive at 1/1,280 confirming the diagnosis of acute septicemic brucellosis with brucellian vasculitis. The patient was treated with the combination: Doxycycline 200mg/d and Rifampicin 600mg/d with a rapidly favorable evolution. Healthcare professionals should be aware of the possibility of brucellosis in front of any unproven vasculitis, especially in endemic countries. This better knowledge will make it possible to avoid diagnostic delay and improve the prognosis of this infection.

Keywords: *brucellosis, cutaneous vasculitis, leuckocytoclastic vasculitis, angiitis*

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1. Introduction

Brucellosis is the most common bacterial zoonosis [1,2]. Also human brucellosis is frequent and still endemic in many countries with an incidence of more than 500,000 cases annually worldwide [1,2,3]. It represents a real major public health problem with significant economic repercussions and heavy morbidity [4].

It is characterized, moreover, by a great clinical polymorphism making its diagnosis a real challenge for clinicians even in endemic areas [1,2,3,5,6].

Certain clinical presentations are exceptional and uncommon during this infection, particularly in the acute forms; they are thus qualified as "unusual" by several authors [5,6,7,8]. Among these unusual manifestations of brucellosis were: uveitis, neurobrucellosis, peritonitis, orchitis, epididymitis, pericarditis, pancytopenia, myocarditis, and vasculitis [5,6,7,8,9].

We report the original observation of isolated cutaneous vasculitis revealing acute brucellosis.

2. Case Presentation

44-year-old Tunisian man, with no notable pathological history, was admitted to our department for a rash with fever that had been evolving for two days. Somatic examination noted fever at 38°C, red-purple infiltrated and tender petechial lesions on feet, legs, and hands (Figure 1), small periungual necrotic lesions (Figure 2), and subungual splinter hemorrhages (Figure 3). The remainder of the somatic examination was without abnormalities. The basic biological tests showed a biological inflammatory syndrome with an erythrocyte sedimentation rate of 82mmH1 and a C-reactive protein at 62mg/l. The other tests were within normal limits (leukocytes, hemoglobin, platelets, creatinine, serum calcium, transaminases, muscle enzymes, serum protein electrophoresis, fast glycemia, plasma ionogram, thyroid hormones, and urinalysis). Electrocardiogram and chest X-ray were without abnormalities. The skin biopsy concluded in leukocytoclastic vasculitis, without signs of

malignancy, and direct immunofluorescence was negative. The specific etiological assessment of this vasculitis was negative: specialized ENT examination with sinus X-ray, trans-thoracic cardiac ultrasound, abdominal ultrasound, thoraco-abdomino-pelvic tomodensitometry, anti-nuclear antibodies, anti-soluble antigen antibodies, anti-neutrophil cytoplasmic antibodies (ANCA, anti-PR3 and anti-MPO), cryoglobulins, immunoglobulin subclass levels, blood cultures, HLA B51 typing, quantiferon, and myelogram. Likewise, no recent drug intake, toxic exposure, or infection was noted. Considering the notion of consumption of unpasteurized milk revealed in the interrogation and the endemic character of our country for brucellosis, Wright's serology was requested and was positive at 1/1,280.

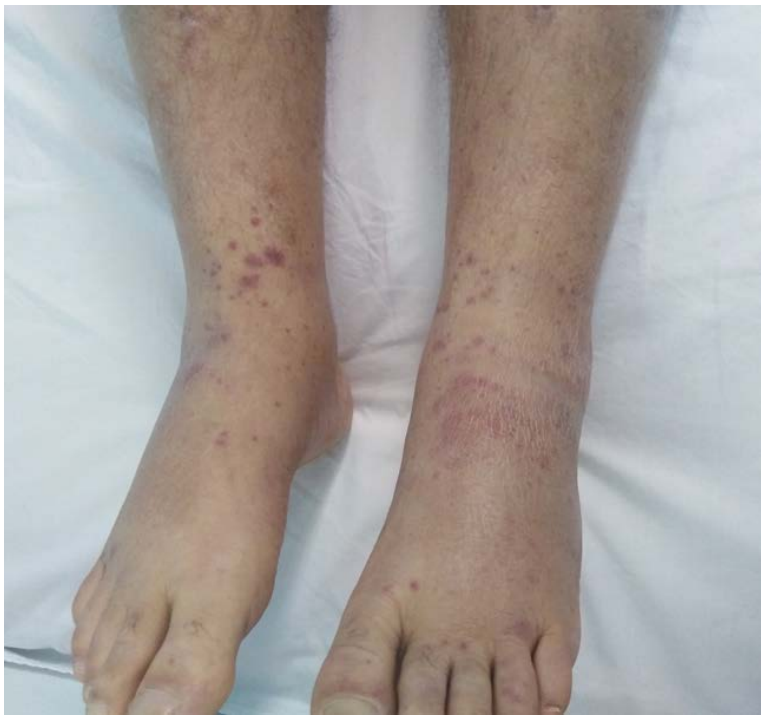

Figure 1. Petechial rash on feet and legs

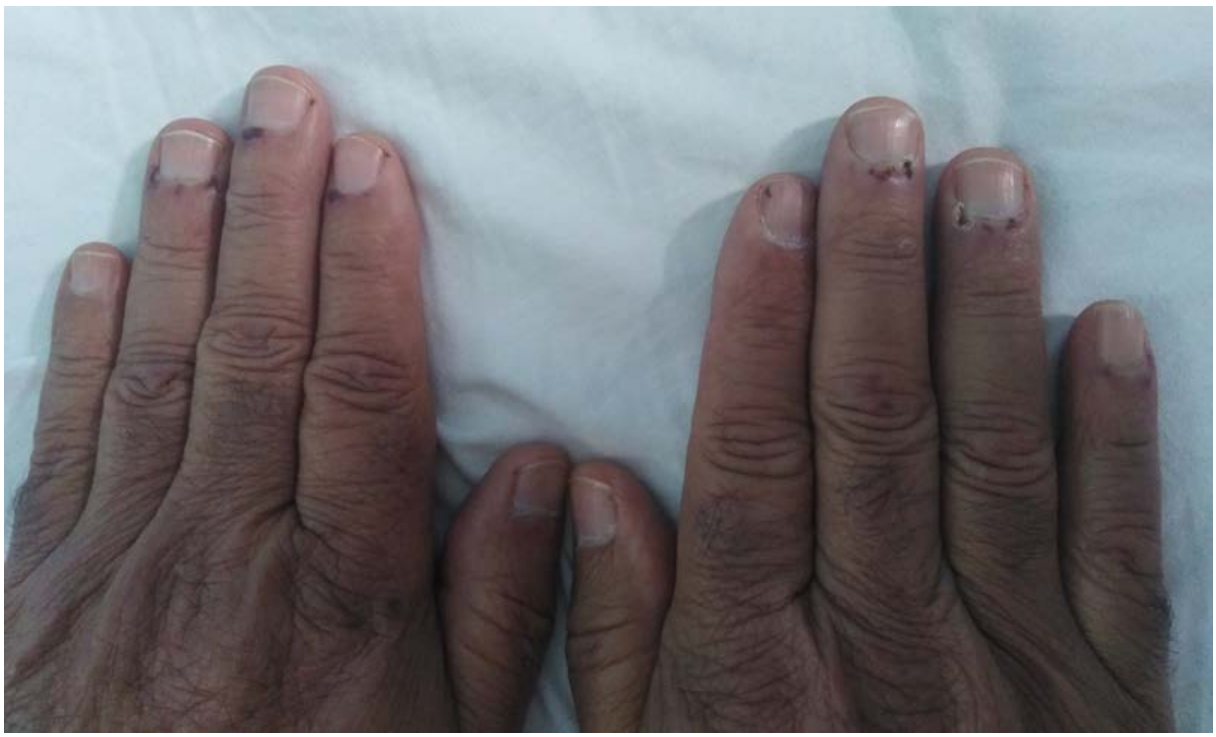

Figure 2. Periungual necrotic lesions

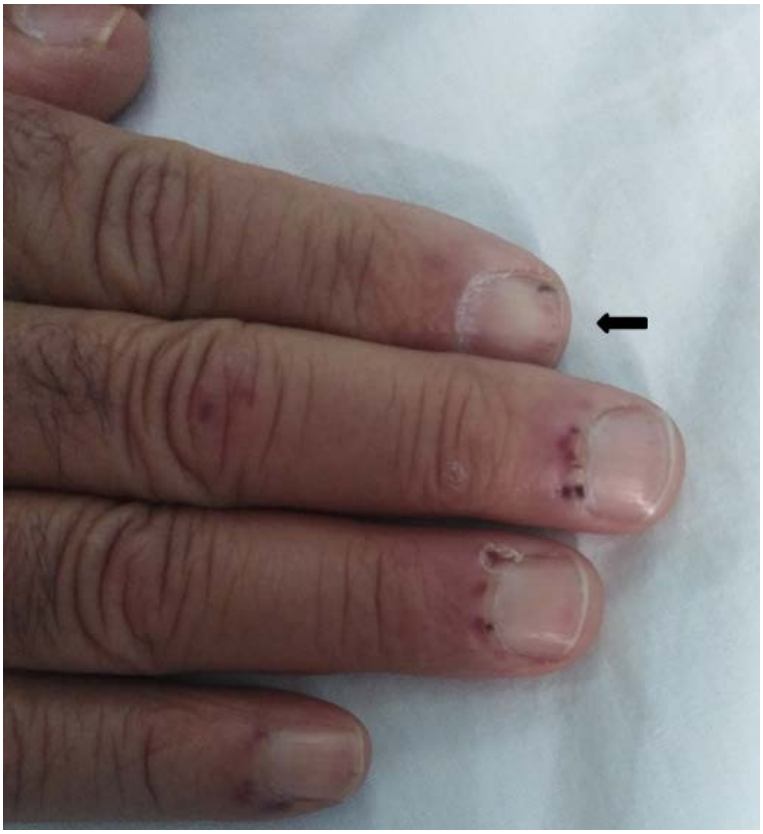

Figure 3. Subungual splinter hemorrhages

Thus the diagnosis of acute septicemic brucellosis with brucellian vasculitis was retained. The patient was treated according to the protocol of the World Health Organization with the combination: Doxycycline 200mg/d and Rifampicin 600mg/d with a rapidly favorable evolution. The patient was afebrile on the second day of treatment. The lesions of cutaneous vasculitis disappear completely after one week of antibiotic therapy. The C-reactive protein was 5mg/l on the tenth day of treatment. Wright's serology was negative at the third month control, and no recurrence of vasculitis has been noted for a year now.

3. Discussion

Due to the large clinical polymorphism and misleading unusual presentations, the diagnosis of brucellosis is often missed, and it is estimated that fewer than 10% of cases of human brucellosis may be clinically recognized [11].

The overall frequency of these unusual clinical presentations was estimated at less than 5% [7,10]. Among these presentations, cardiovascular damage remains exceptional, occurring in less than 1% of cases [1,9].

The vascular involvement itself (vascular brucellosis) is very uncommon [1,12]. This involvement can be such as: endarteritis, arterial ulcers, aortitis, arterial and venous thrombosis, arterial aneurysms, and vasculitis [1,9,12,13,14,15].

Brucellian vasculitis is particularly unusual with only a few sporadic cases reported in the world literature [6,16,17,18,19,20].

This vasculitis can be localized: cutaneous, renal, and cerebral [1,16,17,18,19,21,22,23,24] or systemic [6,20]. Histologically, leukocytoclastic vasculitis is the most frequent [16,17,18]; more rarely, cases of granulomatous vasculitis [19] or Henoch-Schönlein purpura [20] have been reported.

The pathogenesis of brucellosis-associated vascular involvement is still unclear; two mechanisms can be discussed:

- Direct mechanism: the invasion of endothelial cells by the bacteria causing a sustained pro-inflammatory response (infectious vasculitis) [1,12,18],
- Indirect mechanism: induction by the bacteria of an immunological dysregulation with production of circulating immune complexes, immunoglobulins, cryoglobulins or autoantibodies causing immunological vasculitis [16,18,22,25].

Usually this vasculitis has a good prognosis with a rapidly favorable outcome under appropriate systemic antibiotic therapy [26]; more rarely, relapsing forms [3,6,7,8] as well as exceptional forms with fatal outcome [16] have been reported.

4. Conclusion

Healthcare professionals should be aware of the possibility of brucellosis in front of any unproven vasculitis, especially in endemic countries. In our observation, the concordance over time, the negativity of the etiological investigation, as well as the good response to specific antibiotic therapy, and the absence of subsequent recurrences make it possible to confirm the direct causal link between leukocytoclastic vasculitis and brucellosis. This better knowledge will make it possible to avoid diagnostic delay and improve the prognosis of this very simulating infection.

References

[1] Herrick JA, Lederman RJ, Sullivan B, Powers JH, Palmore TN. Brucella arteritis: clinical manifestations, treatment, and prognosis. Lancet Infect Dis. 2014; 14(6): 520-6.

[2] Zheng R, Xie S, Lu X, Sun L, Zhou Y, Zhang Y, et al. A Systematic Review and Meta-Analysis of Epidemiology and Clinical Manifestations of Human Brucellosis in China. Biomed Res Int. 2018; 2018: 5712920.

[3] Franco MP, Mulder M, Gilman RH, Smits HL. Human brucellosis. Lancet Infect Dis. 2007; 7(12): 775-86.

[4] Ulu-Kilic A, Metan G, Alp E. Clinical presentations and diagnosis of brucellosis. Recent Pat Antiinfect Drug Discov. 2013; 8(1): 34-41.

[5] Bouomrani S, Belgacem N, Ben Hamad M, Regaïeg N, Baïli H, Lassoued N, et al. Bilateral Panuveitis Revealing Acute Septicemic Brucellosis. EC Ophthalmology. 2018; 9. 6: 437-441.

[6] Bouomrani S, Dey M, Ahmed A. Acute Renal Failure Complicating Septicemic Brucellosis. J Pathol Infect Dis. 2019; 2(2): 1-2.

[7] Hatipoglu CA, Yetkin A, Ertem GT, Tulek N. Unusual clinical presentations of brucellosis. Scand J Infect Dis. 2004; 36(9): 694-7.

[8] Türksoy O, Tokgöz H, Toparli S. Unusual clinical presentations of brucellosis. Scand J Infect Dis. 2005; 37(10): 784.

[9] Gatselis NK, Makaritsis KP, Gabranis I, Stefos A, Karanikas K, Dalekos GN. Unusual cardiovascular complications of brucellosis presenting in two men: two case reports and a review of the literature. J Med Case Rep. 2011; 5: 22.

[10] Starakis I, Mazokopakis EE, Bassaris H. Unusual manifestations of brucellosis: a retrospective case series in a tertiary care Greek university hospital. East Mediterr Health J. 2010; 16(4): 365-70.

[11] Mantur BG, Amarnath SK, Shinde RS. Review of clinical and laboratory features of human brucellosis. Indian J Med Microbiol. 2007; 25(3): 188-202.

[12] Ferrero MC, Bregante J, Delpino MV, Barrionuevo P, Fossati CA, Giambartolomei GHet al. Proinflammatory response of human endothelial cells to Brucella infection. Microbes Infect. 2011; 13(10): 852-61.

[13] Tosatto V, Boattini M, Nascimento P, Barata Moura R. Lymphadenitis and aortitis due to Brucella melitensis infection. Infection. 2020; 48(2): 313-314.

[14] Anantha T, Chittaragi VB, Karthik MK, Kumar J, Kumar BS, Sai BS. A fatal, rare case of bilateral, upper, lower limbs and abdominal gangrene associated with brucellosis. J Lab Physicians. 2019; 11(4): 388-390.

[15] Wang M, Zhu Q, Yang Q, Li W, Wang X, Liu W, et al. Intestinal brucellosis associated with celiac artery and superior mesenteric artery stenosis and with ileum mucosa and submucosa thickening: A case report. Medicine (Baltimore). 2017; 96(2): e5893.

[16] Dizbay M, Hizel K, Kilic S, Mutluay R, Ozkan Y, Karakan T. Brucella peritonitis and leucocytoclastic vasculitis due to Brucella melitensis. Braz J Infect Dis. 2007; 11(4): 443-4.

[17] Massasso D, Gibson K. Brucellosis mimicking Henoch-Schönlein purpura. Med J Aust. 2007; 186(11): 602-3.

[18] Karaali Z, Baysal B, Poturoglu S, Kendir M. Cutaneous manifestations in brucellosis. Indian J Dermatol. 2011; 56(3): 339-40.

[19] Franco Vicario R, Balparda J, Santamaria JM, Alvaro C, Arizaga C, de la Villa FM, et al. Cutaneous vasculitis in a patient with acute brucellosis. Dermatologica. 1985; 171(2): 126-8.

[20] Akgun C, Akbayram S, Guner S, Aktar F, Temel H, Basaranoglu M. Brucellosis as a trigger agent for Henoch-Schönlein purpura. Bratisl Lek Listy. 2012; 113(8): 506-7.

[21] Elzouki AY, Akthar M, Mirza K. Brucella endocarditis associated with glomerulonephritis and renal vasculitis. Pediatr Nephrol. 1996; 10(6): 748-51.

[22] Yrivarren JL, Lopez LR. Cryoglobulinemia and cutaneous vasculitis in human brucellosis. J Clin Immunol. 1987; 7(6): 471-4.

[23] Catakli T, Arikan FI, Acar B, Dallar Y. Cutaneous vasculitis in a patient with acute brucellosis. Clin Exp Dermatol. 2009; 34(7): e387-8.

[24] Kim EJ, Lee SJ, Ahn EY, Ryu DG, Choi YH, Kim TH. Relapsed Brucellosis Presenting as Neurobrucellosis with Cerebral Vasculitis in a Patient Previously Diagnosed with Brucellar Spondylitis: A Case Report. Infect Chemother. 2015; 47(4): 268-71.

[25] Hermida Lazcano I, Sáez Méndez L, Solera Santos J. Mixed cryoglobulinemia with renal failure, cutaneous vasculitis and peritonitis due to Brucella melitensis. J Infect. 2005; 51(5): e257-9.

[26] Nagore E, Sánchez-Motilla JM, Navarro V, Febrer MI, Aliaga A. Leukocytoclastic vasculitis as a cutaneous manifestation of systemic infection caused by Brucella melitensis. Cutis. 1999; 63(1): 25-7.

[27] Perez C, Hernandez R, Murie M, Vives R, Guarch R. Relapsing leucocytoclastic vasculitis as the initial manifestation of acute brucellosis. Br J Dermatol. 1999; 140(6): 1177-8.