Cutaneous Leukocytoclastic 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 leukocytoclastic 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 hemmorhages. 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, leukocytoclastic vasculitis, angiitis


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
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


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