

Bilateral Panuveitis Revealing Acute Septicemic Brucellosis

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Abstract

Introduction: Brucellosis is a highly contagious infection that is still endemic in several countries. The septicemic forms are characterized by a polymorphic clinical presentation and a systemic damage presenting sometimes a real diagnostic challenge. The revealing ocular manifestations are unusual. We report one.

Case Report: A 43-year-old woman with no medical history, hospitalized for a sudden and bilateral decrease of visual acuity with eye pain and fever. Ophthalmological examination concluded to a bilateral non-granulomatous panuveitis. The somatic examination noted a fever at 39°C and a moderate and painless hepatosplenomegaly. Biology showed a marked inflammatory syndrome with neutrophilic leukocytosis at 18 000/mm³. Screening for immunological diseases, tuberculosis, connective tissue diseases, systemic granulomatosis and hematological malignancies was negative. Wright's serologic test was positive at 1/320. The Rose Bengal test was also positive confirming the diagnosis of brucellosis. With Rifampicin-Doxycycline, the evolution was favorable with recovery of the visual acuity and normalization of ophthalmologic check-up at two months.

Conclusion: Ocular lesions during brucellosis are rare: 3.35% and the revealing forms remain exceptional: 0.83%. Panuveitis are associated with the worst visual prognosis. Brucellosis should therefore be mentioned in the presence of recurrent uveitis or uveitis having a poor response to corticosteroid therapy, particularly in an endemic country.

Keywords: Panuveitis; Brucellosis; Uveitis; Infection

Introduction

Brucellosis is a highly contagious anthroponosis that is still endemic in the Middle East, Latin America, and the countries around the Mediterranean. It is a systemic infection that can occur in two clinical forms: acute or chronic [1-4].

This infection is characterized by polymorphic and sometimes unusual clinical manifestations, thus representing a real diagnostic challenge for the clinician [2,4,5]. Among the "unusual" clinical manifestations that may reveal brucellosis, the authors recognize neuro-brucellosis, peritonitis, pericarditis, pancytopenia, uveitis and orchiepididymitis [4]. The brucellar uveitis remains exceptional; indeed in the series of 240 patients with brucellosis of Hatipoglu CA., *et al.* uveitis was found as a revealing manifestation of the infection only in two patients thus representing 0.83% of all cases and 4.58% of the unusual presentations [4].

On the other hand, brucellosis remains an exceptional cause of uveitis [6]; indeed it accounted for only 0.5% of the etiologies in the series of 122 cases of uveitis of Cernea P., *et al* [7].

The ocular complications of brucellosis remain, therefore, very little known despite their potential severity that can lead to irreversible blindness [3].

Here we report the observation of bilateral panuveitis revealing an acute septicemic brucellosis in a 43-year-old woman.

Case Report

This is a 43-year-old woman with no notable pathological history who was hospitalized for acute decline in visual acuity of both eyes associated with eye pain and fever. Ophthalmological examination showed visual acuity at 6/10 on the right and 5/10 on the left, and concluded with non-granulomatous bilateral panuveitis. The somatic examination noted fever at 39 °C and mild hepatic overflow and moderate splenomegaly. In biology there was a marked inflammatory syndrome with hyperleucocytosis and hepatic cytolysis (Table 1). The abdominal ultrasonography confirmed the moderate hepatosplenomegaly and objectified in addition some mesenteric and celiac lymphadenopathies. Subsequent investigations had eliminated sarcoidosis, tuberculosis, connective tissue disease, neoplasia, hematological malignancy, systemic granulomatosis, or underlying systemic vasculitis. The serodiagnostics of viral hepatitis A, B and C as well as screening for HLA haplotypes A29, B51, and B27 were negative.

Tests	Results
ESR	120 mmH1
CRP	68 mg/l
Gammaglobulins	18 g/l
WBC	18000/mm ³
PNN	14400/mm ³ (80%)
Platelet	450000/mm ³
ASAT	52 IU/l
ALAT	68 IU/l
PT (P/C)	100%

Table 1: Biological data of our patient.

Abbreviations: ECR: Erythrocyte Sedimentation Rate; CRP: C-Reactive Protein; WBC: White Blood Cell; PNN: Polynuclear Neutrophils; ASAT: Aspartate Aminotransferase; ALAT: Alanine Aminotransferase; PT: Prothrombin Time; P/C: Patient/Control.

Motivated by the notion of eating unpasteurized fresh milk at the anamnesis, Wright’s serology was requested and returned positive at 1/320. The Rose Bengal test was also positive confirming the diagnosis of acute septicemic brucellosis. Treatment with Rifampicin 600 mg/day with Doxycycline 200 mg/day was initiated. The evolution was quickly favorable with apyrexia from the second day and progressive disappearance of eye complaints. Complete blood cell count and C-reactive protein were normalized after ten days of treatment. Recovery of visual acuity was complete after two weeks and ophthalmologic examination was strictly normal at two months.

Discussion

The first description of human ocular involvement in brucellosis dates back to 1924 by Lemaire, who reported the observation of bilateral optic neuritis with oculomotor nerve palsy associated with brucellar meningitis. Since then, several other ocular lesions have been described during this infection: uveitis, keratitis, conjunctivitis [2,8,9] and more rarely: episcleritis, dacryocystitis [8], choroiditis, retinal hemorrhages, papillary edema [9], optic neuritis and retinal detachment [3,10].

Schematically, these damages can be classified into two main types [2]:

- Direct ocular lesions: uveitis, keratitis, conjunctivitis, episcleritis, choroiditis,
- Neuro-ophthalmic disorders: papillitis, papillary edema, optic neuritis, oculomotor nerve palsy.

Neuro-ophthalmic disorders are most often associated with a neurological complication of brucellosis: meningitis or more parenchymal brain damage (neurobrucellosis). This classification refers to the two pathophysiological mechanisms explaining ocular lesions during brucellosis [2,3,11]:

- The direct invasion of ocular tissues by the pathogenic microorganisms via septic emboli,
- The immunological mechanism secondary to the production of immunoglobulins and circulating immune complex forming deposits in the different structures of the eye. This has been proven by animal experimentation [11].

The frequency of ocular lesions during brucellosis appears to be variable depending on countries and ethnicity, the type of study, and the size of the sample. In fact, in the small Turkish series (132 patients of Sungur GK, *et al.* and 147 patients of Güngür K., *et al.*) the prevalence was 21 to 26% [8,9] whereas in the large Peruvian series of Rolando I., *et al.* of 1551 patients with brucellosis collected over 26 years, this prevalence was only 3.35% [3].

Among ocular lesions, anterior uveitis and conjunctivitis are the most common; posterior and intermediate uveitis are rare, whereas total uveitis or panuveitis remains exceptional [2,3,8,9]; their frequency was only 0.58% in the large series of Rolando I., *et al.* (9 cases/1551 patients with brucellosis and 9 cases/43 brucellar uveitis) [3].

Ocular complications of brucellosis appear to be more frequent in adults [2,3,5], women [3,12], and in chronic forms of brucellosis compared to acute forms: 7,9% Vs 0, 7%, $p < 0.001$ [3].

The damage may be uni- or bilateral [3,5,8,9], episodic or recurrent [13], and single or multiple with the possibility of having several ocular lesions concomitantly in the same patient, like illustrated Mohammadi Z., *et al.* in his observation (episcleritis, panuveitis, and chorioretinitis in the same patient) [2].

The clinical manifestation of ocular involvement during brucellosis is not specific. It can combine in variable degrees: visual blur, redness, pain, sensation of intraocular foreign bodies, decreased visual acuity, scotomas, and tearing [3,14]. It should be noted, however, that in 20% of cases, these lesions may remain completely asymptomatic and may be discovered by routine ophthalmological examination in a patient with brucellosis [3,14].

The diagnosis of ocular brucellosis is most often based on clinical criteria with a positive *Brucella* serology [3]. Intraocular tests are rarely used. They can be of great help in questionable forms [3,8,9,15]. These are mainly serological tests specific to brucellosis performed in intraocular fluids, the bacterial culture of these fluids and the calculation of the Goldmann-Witmer coefficient (ratio of specific antibody levels in the ocular fluids and plasma). Biopsies with histological study of ocular tissues are only exceptionally used [3,8,9,15]. Specific agglutination in ocular fluids is characterized by a sensitivity of 66.7% and a specificity of 100% [3,15].

Treatment is based on appropriate systemic antibiotic therapy [13]; topical (ocular) and/or systemic corticosteroid therapy may be necessary in combination with this antibiotic therapy, especially in severe forms and forms having immunological mechanisms [3,6,7,8].

Untreated, poorly treated, or late diagnosed; ocular involvement of brucellosis can progress to severe sequelae in 36% of cases: cataract, maculopathy, glaucoma, retinal neovascularization, optic atrophy, retinal detachment... [3].

Brucellar uveitis is associated with the most serious clinical forms of this infection and the most unfavourable prognosis:

- In fact, subjects with brucellar uveitis are more exposed to the osteoarticular manifestations of this infection [8],
- It should also be noted that in the world literature, subjects with brucellar uveitis are more likely to have an associated spondyloarthropathy; in particular ankylosing spondylitis [1,8,16].

Panuveitis are associated with the worst prognosis among the brucellar ocular lesions: cause blindness in 89% (8 patients/9 in the series of Rolando I., *et al.*) [3].

Conclusion

As rare as they are, the ocular manifestations of brucellosis deserve to be known by practitioners, especially in endemic countries. Only the early diagnosis and the rapid and appropriate treatment will allow the preservation of the visual functional prognosis of these disorders. Thus, several authors recommend a routine ophthalmological examination in any patient with confirmed brucellosis. Similarly, the diagnosis of brucellosis should be systematically evoked in the presence of recurrent uveitis or uveitis not responding to well-conducted corticosteroid therapy.

Conflicts of Interest

No conflicts.

Bibliography

1. Garip Y, *et al.* "Brucellosis in spondyloarthritis mimicking an exacerbation". *Acta Reumatológica Portuguesa* 39.4 (2014): 351-352.
2. Mohammadi Z, *et al.* "Ocular manifestations in a child with systemic brucellosis". *Journal of Research in Medical Sciences* 19.7 (2014): 677-679.
3. Rolando I, *et al.* "Ocular manifestations associated with brucellosis: a 26-year experience in Peru". *Clinical Infectious Diseases* 46.9 (2008): 1338-1345.
4. Hatipoglu CA, *et al.* "Unusual clinical presentations of brucellosis". *Scandinavian Journal of Infectious Diseases* 36.9 (2004): 694-697.
5. Karahocagil MK, *et al.* "Brucellosis and uveitis". *Annals of Ophthalmology* 40.1 (2008): 48-50.
6. Akduman L, *et al.* "A case of ocular brucellosis: importance of vitreous specimen". *Acta Ophthalmologica* 71.1 (1993): 130-132.
7. Cernea P, *et al.* "Aetiological aspects of uveitis". *Archives D'Ophthalmologie* 37.11 (1977): 715-722.
8. Güngür K, *et al.* "Ocular complications associated with brucellosis in an endemic area". *European Journal of Ophthalmology* 12.3 (2002): 232-237.
9. Sungur GK, *et al.* "Ocular involvement in brucellosis". *Canadian Journal of Ophthalmology* 44.5 (2009): 598-601.
10. Rabinowitz R, *et al.* "Bilateral multifocal choroiditis with serous retinal detachment in a patient with Brucella infection: case report and review of the literature". *Archives of Ophthalmology* 123.1 (2005): 116-118.
11. Woods A. "Experimental brucellar uveitis". *American Journal of Ophthalmology* 36 (1953): 1025-1042.
12. Puig Solanes M, *et al.* "Ocular complications in brucellosis". *American Journal of Ophthalmology* 36.5 (1953): 675-689.
13. Tabbara KF and al-Kassimi H. "Ocular brucellosis". *British Journal of Ophthalmology* 74.4 (1990): 249-250.
14. Ghasemi Barghi R, *et al.* "A review on ophthalmic manifestations of brucellosis and reporting a case of ophthalmic brucellosis". *Iranian Red Crescent Medical Journal* 13.5 (2011): 352-353.

15. Rolando I., *et al.* "Brucellar uveitis: intraocular fluids and biopsy studies". *International Journal of Infectious Diseases* 13.5 (2009): e206-e211.
16. Akyol L., *et al.* "Bilateral sacroiliitis and uveitis comorbidity: brucellosis? Ankylosing spondylitis?" *BMJ Case Reports* (2015): bcr2015211461.

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