Fulminant endogenous endophthalmitis caused by Brucella melitensis, a case report

SeyedehMaryam Hosseini[1], Mohammad Baghi[1], Mohammad Reza Ansari Astaneh[1], and Mehrdad Motamed Shariati[2]

[1]Mashhad University of Medical Sciences Khatam-al-Anbia Hospital
[2]Mashhad University of Medical Sciences

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Introduction:

Brucellosis is a zoonotic infectious disease with a wide range of manifestations including malaise, anorexia, fever, and profound muscular weakness, as described by Marston in 1860[1]. It is caused by the Gram-negative coccobacillus, Brucella, and remains endemic in some developing countries, such as Iran. There are 6 types of brucella that 4 of which include Brucella melitensis, Brucella abortus, Brucella canis, and Brucella suis were recognized as pathogens involving humans. Brucella melitensis was described as the most common and virulent pathogen worldwide. The first case of ocular brucellosis in a human being was described by Lemaire in 1924 [2], presented with bilateral optic neuritis and external ophthalmoplegia in a patient with brucella meningitis.

Ocular manifestations of acute and chronic infection include anterior and posterior uveitis, panuveitis, keratitis, conjunctivitis, papillitis, cataract, maculopathies, glaucoma, and ocular muscle paresis. Modern treatments of ocular brucellosis, intraocular as well as systemic antibiotics, have improved the prognosis of the disease[3]. Herein, we present a patient with endogenous endophthalmitis caused by Brucella Melitensis (B.Melitensis), which is very rare and unusual.

Case report:

A 25-year-old woman came to the emergency department of Khatam-Al-Anbia eye hospital (affiliated with Mashhad University of Medical Sciences, Mashhad, Iran) with complaints of acute decreased vision, photophobia, and redness in the right eye from one week ago. She had no history of trauma or eye surgery. The patient had a history of mild fever with right shoulder pain 4 months ago. She did not have a history of night sweats or coughing.

Because of the endemic area where she lived, the physician suspected brucellosis. The wright test and 2-mercaptoethanol test were 1/80 and 1/40 respectively which was positive for the patient. ESR (erythrocyte sedimentation test) was 38 and CRP (C-Reactive Protein) 1+ in the labs' test. Accordingly, she was treated with oral doxycycline and trimethoprim-sulfamethoxazole. The patient had poor compliance with medicine consumption. During these 4 months, the patient experienced some sort of pain in her shoulders and a mild fever with an on-off pattern.

The best-corrected visual acuity with a tumbling E-chart in the right eye at the time of presentation was hand motion with projection and 10/10 in the left eye. Intraocular pressure (IOP) was within the normal limit in both eyes. The anterior segment examination of the right eye showed clear cornea, hypopyon and flare, and 4+ vitreous cells (based on SUN Working Group) [4]. We found no iris nodules and posterior synechia. Fundus examination of the right eye revealed optic disc swelling, diffuse vasculitis, and a retinitis

patch located one disc diameter below the optic nerve head. The left eye was entirely normal.

With the possible diagnosis of vision-threatening endogenous endophthalmitis or infectious retinitis, the patient was admitted for further diagnostic evaluations and therapies. Regarding the positive history of Wright test and symptoms of brucellosis, consultation with an infectious diseases specialist for more systemic evaluations was performed. Systemic work-ups and laboratory tests including blood, urine, throat culture, chest x-ray, complete blood count, platelet count, blood urea nitrogen, creatinine, urine analysis, and cardiologic consult for the possibility of infectious endocarditis were unremarkable. However, the Wright test was still positive.

Vitreous sampling was performed with a 25-gauge needle through pars plana and evaluated for polymerase chain reaction (PCR) to detect the Herpes Simplex virus, Varicella Zoster virus, Cytomegalovirus, Brucella, and smear and culture. Intravitreal vancomycin (1 mg/0.1 ml), and ceftazidime (2.25 mg/0.1 ml) were injected. Regarding the suspicion of herpetic retinitis, we started valacyclovir, 1000mg tablets every 8 hours for the patient.

The PCR test was positive only for B. melitensis. The previous systemic medications for brucellosis were continued and oral prednisolone 50 mg/day was prescribed. Due to severe vitreous inflammation, a three-port 23-gauge pars plana vitrectomy with silicone oil tamponade was performed on the third day of admission. After the removal of all vitreous inflammatory debris and membranes, we found diffuse retinal vasculitis and multiple retinitis patches around the optic disc.

Two months later, because of significant cataracts and near-total rhegmatogenous retinal detachment (RRD) and subretinal fibrotic bands under silicon oil, we performed cataract surgery with intraocular lens implantation, silicone oil removal, 23G-re-vitrectomy, and subretinal band removal and re-injection of silicone oil (5700 centistoke viscosity). Systemic antibiotics were prescribed for six weeks, the systemic corticosteroid was tapered off for the patient, and brucellosis treatment was completed.

At the final follow-up, the visual acuity of the right eye was hand motion with projection. IOP was 4 mmHg. A retinal fold was developed, and the right eye was pre-phthisis bulbi condition.

The left eye was completely normal at the last visit (figure 1).

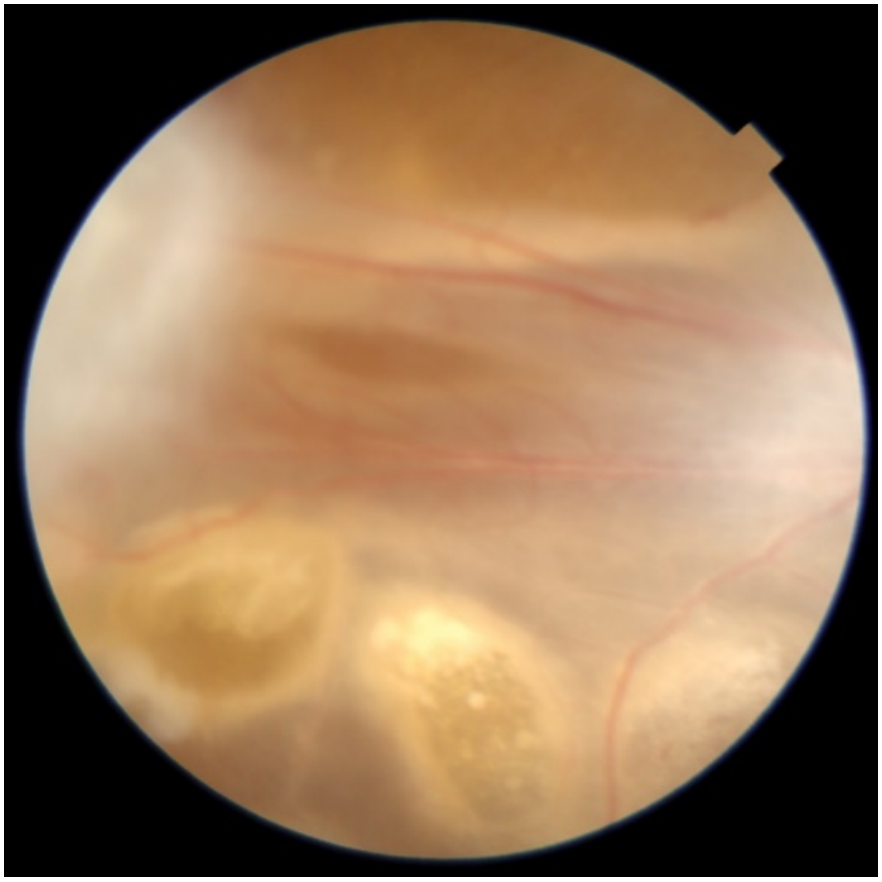

Figure 1: Color fundus photograph of the right eye showing retinal fold under silicon oil.

Discussion:

Brucellosis is a zoonotic disease that can be found worldwide. Although it has been eradicated and is under control in most developed countries, it still represents an important health problem in many parts of the world, including the Middle East, the Mediterranean, Mexico, and Central and South America[5]. In some countries, such as Peru, Kuwait, Saudi Arabia, and Iran brucellosis is endemic[6,7].

Brucellosis presents with a spectrum of clinical manifestations, and diagnosis is based on clinical signs and positive bacteriological and serological tests. Ocular involvement caused by Brucella remains poorly recognized. Some ocular manifestations include dacryoadenitis, episcleritis, chronic sclerouveitis, nummular keratitis, cataract, glaucoma, multifocal choroiditis, exudative retinal detachment, maculopathy, and optic neuritis[8,9].

Cavallaro *et al* . reported a patient with papilledema due to brucellosis treated with sole anti-brucellosis without steroid administration[10]. Lashay *et al* . from Iran reported a case of bilateral optic nerve head swelling following brucellosis, which led to bilateral optic nerve atrophy and visual loss[11].

Endogenous endophthalmitis is an ophthalmic emergency that can have severe sight-threatening complications and still presents a diagnostic and therapeutic challenge even with improvements in therapeutic modalities. The main prognostic factor is the virulence of the causative organism: once the organism enters the eye, it rapidly destroys ocular tissues. However, it should be considered that our patient's poor outcome could also be related to sequelae of endophthalmitis such as RRD and proliferative vitreoretinopathy (PVR) than the high virulence of the organism. Endogenous endophthalmitis is one of the manifestations

of brucellosis which is spreading from ocular blood circulation. The diagnosis of brucella endophthalmitis may be quite challenging and requires a high index of suspicion in the absence of characteristic systemic features. Regarding a 1.3% false positive rate for serology assessment for the diagnosis of brucellosis, we considered other differentials such as fungal, bacterial, and tuberculosis-related endogenous endophthalmitis[12]. However, the patient's systemic work-ups end in brucellosis. The point to notice in this case is the occurrence of endophthalmitis about four months after the patient's systemic symptoms. Orey et al. reported a 26-year-old female with the final diagnosis of brucella endogenous endophthalmitis, which was treated with high-dose systemic corticosteroids and azathioprine with an initial misdiagnosis elsewhere. They concluded that the diagnosis of brucellosis should be considered in any case of panuveitis of unknown origin in endemic areas[7].

While previous studies have shown an appropriate response to treatment in patients with Brucella endophthalmitis[11], in this article, we reported a patient with fulminant endogenous endophthalmitis following brucellosis, which had a poor visual prognosis and is prone to phthisis bulbi despite our therapeutic efforts.

Conclusion

The prevalence of brucellosis has decreased in many developed countries and ophthalmic complications are rare in these regions, but it is suggested that in endemic areas, ophthalmologists consider work up for brucellosis in any case of panuveitis of unknown origin, as it seems that early diagnosis and prompt treatment of the disease could decrease vision-threatening complications.

Declarations

Ethics approval and consent to participate: Not applicable.

Consent for publication: Written informed consent was obtained from the patient for the publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Availability of data and materials: The datasets used during the current study are available from the corresponding author upon reasonable request.

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