An unusual occurrence of stromal keratitis in dengue fever

Pritam Bawankar, Tatyarao Lahane[1], Ragini Parekh[1], Sayalee Lahane, Sumeet Lahane[1], Pooja Pathak[1], Sayli Sonawane[1]

Access this article online

Quick Response Code:

Website: www.ijo.in

DOI: 10.4103/ijo.IJO_387_18

Departments of Vitreo-Retina Surgery, [1]Ophthalmology, Grant Medical College and Sir J.J. Group of Hospital, Mumbai, Maharashtra, India

Correspondence to: Dr. Pritam Bawankar, Department of Vitreo-Retina Surgery Services, Grant Medical College and Sir J.J. Group of Hospital, Mumbai - 400 008, Maharashtra, India. E-mail: dr.pritambawankar@gmail.com

Manuscript received: 27.03.18; Revision accepted: 14.06.18

Dengue is a mosquito-borne infection endemic in the tropical and subtropical regions of the world. Classic dengue fever is a self-limiting, influenza-like illness transmitted by *Aedes aegypti* mosquito. Ophthalmic manifestations though rare can involve both the anterior and posterior segments and are usually associated with the thrombocytopenic state. However, ophthalmic complications such as anterior uveitis and vasculitis suggest immune-mediated pathogenesis. Herein, we report a rare case of stromal keratitis and an unusual occurrence of simultaneous bilateral blindness following dengue fever in a young girl.

Key words: Anterior uveitis, dengue fever, retinal detachment, stromal keratitis, vitreous hemorrhage

Dengue is a mosquito-borne infection endemic in the tropics and warm temperate regions of the world. The highest

Cite this article as: Bawankar P, Lahane T, Parekh R, Lahane S, Lahane S, Pathak P, *et al*. An unusual occurrence of stromal keratitis in dengue fever. Indian J Ophthalmol 2018;66:1631-3.

incidence occurs in areas of Central and South America, Southeast Asia, the western Pacific, Africa, and eastern Mediterranean.[1] Four closely related but serologically distinct viruses that can cause dengue fever (DF) have been identified. *Aedes aegypti* is the principal vector for human transmission in urban areas, which is found worldwide between latitudes 35°N and 35°S.[2] Classic DF is a self-limiting, influenza-like illness, whereas dengue hemorrhagic fever (DHF) is a severe and potentially fatal form of the disease, characterized by multisystem hemorrhagic manifestations, thrombocytopenia, plasma leakage, and increased vascular permeability.

Ophthalmic complications associated with DF and DHF are mostly posterior segment manifestations such as macular edema, vascular occlusions, vasculitis with related retinal hemorrhages, choroidal effusion, or exudative retinal detachment.[3] Anterior segment manifestation has mostly been reported in the form ofsubconjunctival hemorrhage and anterior uveitis.[3]

Herein, we describe an unusual occurrence of simultaneous bilateral blindness in a case with history of DF. We also report a rare corneal complication of DF that has not been previously described in literature.

Case Report

A 25-year-old girl presented to our outpatient department with sudden and severe loss of vision in both the eyes since 3 days. She was admitted in the medicine department 15 days before with a history of fever, severe malaise, headache, abdominal pain, and vomiting of 5 days duration. She had also developed maculopapular rash over the trunk, limbs, and face on the sixth day of fever. She denied any history of visual disturbance during the course of her systemic illness. On investigation, the peripheral smear was negative for malarial parasite, and all other routine investigations were within normal limits except for thrombocytopenia with platelet counts of 9,000/μL at the time of admission. A diagnosis of DF was made after she was found positive for IgM, IgG, and dengue nonstructural protein 1 (NS-1) antigen. She was started on supportive therapy and platelet transfusions, following which there was an improvement in platelet count. She was recovered and discharged from the hospital after a week.

On ophthalmic evaluation, the vision in her both eyes was no perception of light. External ocular examination revealed bilaterally symmetrical normally placed eyelids. Ocular movements were full and free on both sides. Anterior segment evaluation of the right eye revealed total yellowish white opaque cornea mimicking corneal abscess, thinned out inferiorly near the limbus. Left eye showed ciliary flush, mild corneal edema, medium to large sized keratic precipitates, and 360° posterior synechiae with complicated cataract precluding fundus view. The intraocular pressure (IOP) was high in the left eye. Penetrating keratoplasty was planned in the right eye (RE) to preserve corneal integrity and for the removal of inflamed corneal tissue. Intraoperative findings revealed edematous opaque cornea [Fig 1a], inflamatory pupillary membrane [Fig 1b], and complicated cataract [Fig 1c]. Corneal graft was sutured with twelve interrupted bites using 10-0 nylon [Fig 1d]. The specimen was sent for histopathology and microbiological examination. Preoperative B-scan ultrasonography of both eyes showed vitreous hemorrhage, total retinal detachment, and choroidal effusions [Fig. 2a and b]. The patient was started on medical therapy for raised IOP along with topical corticosteroids and lubricants in both the eyes. Histopathological examination of corneal tissue revealed stromal breakdown with myxoid change and neovascularization. Stroma also showed mixed inflammatory infiltrate comprising neutrophils and lymphocytes [Fig. 3]. There was no evidence of neutrophilic abscess or micro-organism. Overlying epithelium was unremarkable. Microbiological examination was unremarkable. The vision remained no perception of light in both the eyes after about 6 months of follow-up [Fig. 2c and d].

Discussion

DF is one of the most common arthropod-borne viral diseases in humans, characterized by an abrupt onset of fever after an incubation period of 2–7 days. Globally, 2.5 billion people live in areas where dengue viruses can be transmitted and approximately 100 million cases of illness are estimated to occur annually.[3] Dengue infection is known to cause fever, headaches, myalgia, thrombocytopenia-related hemorrhagic complications, and also hypotension, especially in DHF/dengue shock syndrome causing high morbidity and mortality.

In recent times, ophthalmic complications due to dengue infection are being reported more frequently in medical literature. Chan *et al.*[4] in their published study demonstrated various ophthalmic manifestations of DF which included involvement of both the anterior and posterior segments of eye suggesting widespread inflammatory processes. DF can impair the vision ranging from mild blurring of vision to catastrophic and severe blindness due to choroidal effusion, optic neuritis, exudative maculopathy, or retinal thickening in the macula.

The pathophysiologic mechanisms responsible for various ocular complications in dengue are not known. The first and most obvious mechanism would be the thrombocytopenic state, which gives rise to increased incidence of hemorrhage. The resultant bleeding tendency manifests as retinal blot hemorrhages in the macula and periphery. However, a hypocoagulable state alone would not account for the entire range of complications such as vasculitis, anterior uveitis, and macular edema which indicates a hyperpermeable and inflammatory process. Immune-mediated pathogenesis has been suggested for these manifestations, and it involves T-cell proliferation and recognition of dengue viral antigens on infected monocytes by sensitized CD4+ and CD8+ cytotoxic T cells.[4] This results in the release of cytokines with vasoactive and procoagulant properties in response to immunological activation.

Ocular findings in our patients primarily included anterior uveitis, vitreous hemorrhage, exudative retinal detachment, and choroidal effusion, which could be due to generalized increased capillary leakage, breakdown of the blood-aqueous barrier and hemorrhagic diathesis associated with platelet destruction, and consumptive coagulopathy.[5] The possible mechanism of corneal complication in our case is believed to be a result of complex immune-mediated process which might have caused autoantibody formation against corneal endothelial cells.[5] Histopathological examination also revealed stromal keratitis more likely secondary to viral etiology.

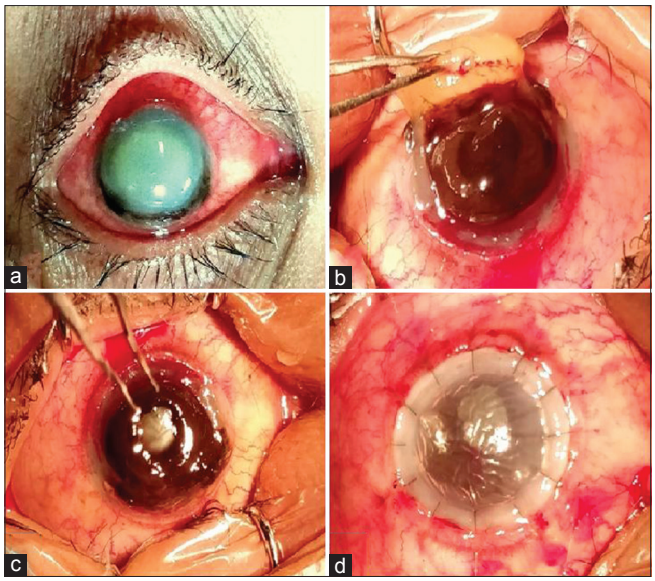

Figure 1: Colored anterior segment images of the right eye showing (a) total edematous opaque cornea thinned out inferiorly, (b) inflammatory membrane at pupillary area, (c) complicated cataract, and (d) postoperative image of penetrating keratoplasty

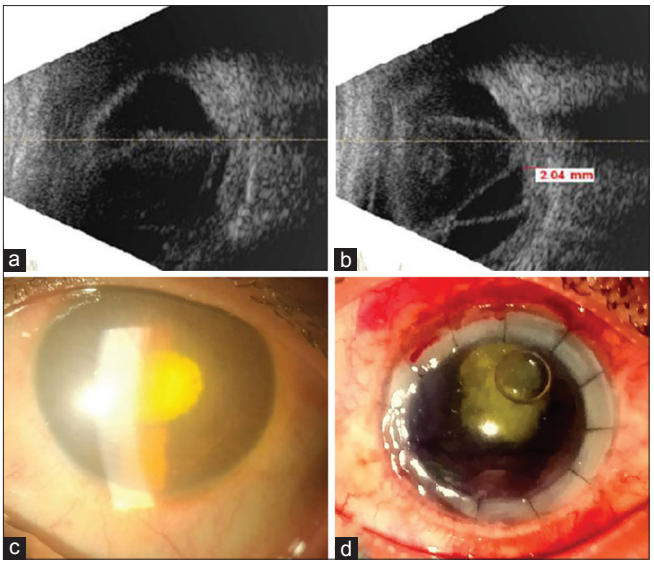

Figure 2: B-acan ultrasound images showing vitreous hemorrhage, retinal detachment, and choroidal effusion in right (a) and left (b) eyes. Posttreatment anterior segement pictures at 1 week follow-up showing (c) complicated cataract with 360° posterior synechiae in the left eye and (d) clear corneal graft with complicated cataract in the right eye

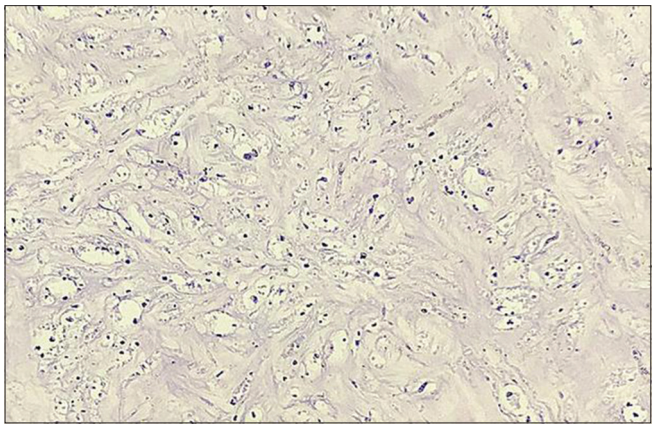

Figure 3: Histopathological examination of right eye corneal tissue showing stromal breakdown with myxoid change and neovascularization. Stromal inflammatory infiltrate comprising neutrophils and lymphocytes

The onset of ophthalmic complications in our patient does not relate with low platelet count. Thus, a complex immune-mediated pathogenesis has been suggested for these manifestations. There is no established treatment for ocular manifestations of DF. Aggressive and effective treatment with topical and systemic corticosteroids permitted control of anterior uveitis. Unfortunately, vision remained no perception of light in both the eyes throughout the follow-up due to severe posterior segment manifestations. Corneal complication was well managed with penetrating keratoplasty.

In conclusion, we present a rare case of stromal keratitis in a DF which was not previously well-described in medical literature. We also report an unusual occurrence of simultaneous bilateral blindness in DF that does not coincide with the thrombocytopenic state. This current case provides an insight to primary care physicians, general ophthalmologists, and other eye care professionals of all the possible ophthalmic complications in patients with severe dengue so that early diagnosis and referral for appropriate supportive therapy can considerably reduce mortality related to this potentially fatal disease.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

References

1. Ramirez-Ronda CH, Garcia CD. Dengue in the Western Hemisphere. Infect Dis Clin North Am 1994;8:107-28.
2. Gibbons RV, Vaughn DW. Dengue: An escalating problem. BMJ 2002;324:1563-6.
3. Gupta A, Srinivasan R, Setia S, Soundravally R, Pandian DG. Uveitis following dengue fever. Eye (Lond) 2009;23:873-6.
4. Chan DP, Teoh SC, Tan CS, Nah GK, Rajagopalan R, Prabhakaragupta MK, *et al*. Ophthalmic complications of dengue. Emerg Infect Dis 2006;12:285-9.
5. Kapoor HK, Bhai S, John M, Xavier J. Ocular manifestations of dengue fever in an East Indian epidemic. Can J Ophthalmol 2006;41:741-6.