

Unilateral Orbital Compartment Syndrome Unmasking Dengue Haemorrhagic Fever: An Unusual Presentation

Ankita Singh¹, Aatish Saraswat²

¹Dept of Ophthalmology, Armed Forces Medical College, India, ²Dept of Pathology, Armed Forces Medical College, Pune

Abstract

Dengue fever may be a potentially life-threatening condition owing to massive capillary leakage and severe bleeding, causing dengue shock syndrome. The earlier literature did not classically describe ophthalmic manifestations in dengue fever. We report a case of a 43-year-old male who presented with sudden onset swelling and pain in the left eye for 2 days along with a history of fever with myalgia for 5 days, for which he did not take any treatment. After a clinical evaluation, a provisional diagnosis of orbital compartment syndrome was made, which required urgent decompression. On investigation, the patient had thrombocytopenia (platelet count of 10,000/ μ L). A positive dengue non-structural protein-1 (NS-1) antigen, along with thrombocytopenia, confirmed the diagnosis of dengue haemorrhagic fever. Dengue IgM antibodies were also found to be positive. The patient was given supportive therapy and urgent platelet transfusions. Lateral canthotomy for orbital decompression, performed after platelet transfusion. However, retropulsion was not possible despite the surgical intervention. During the follow-up period, the left eye chemosis was slightly reduced. However, the visual acuity did not show improvement in the left eye throughout the follow-up. It is recommended that ophthalmologists be aware of such ocular manifestations as the likely initial presentation of dengue fever. Prompt and appropriate treatment through a multidisciplinary approach should prevent sight-threatening complications.

Keywords: Dengue haemorrhagic fever, lateral canthotomy, orbital compartment syndrome, proptosis thrombocytopenia

INTRODUCTION

Dengue fever, which is transmitted by the *Aedes aegypti* mosquito, is a common viral disease in humans.^[1] The infection is more prevalent in Southeast Asian and tropical American regions.^[2] The typical presentation of the illness is an abrupt onset of high-grade fever with chills and rigors associated with a body rash, myalgia, retro-orbital pain, rhinitis, and lumbosacral pain. The clinical manifestations are primarily due to thrombocytopenia leading to bleeding diathesis.^[3] The condition may be potentially life-threatening owing to profuse capillary leakage, severe bleeding causing dengue shock syndrome, and eventually involving the vital organs. Ocular manifestations in dengue fever were not elicited classically earlier in the literature. However, recent years have seen a surge in the frequency of ocular association.^[4,5]

Patients diagnosed with the dengue viral illness reporting sudden diminution of vision need prompt evaluation to prevent any severe complications. Ophthalmic manifestations may possibly be an early indication of

imminent thrombocytopenia.^[5,6] The existing data suggest that most diagnosed cases of dengue fever present with ocular manifestations. However, there is a shortage of literature reporting ocular involvement as the initial presentation of dengue fever. Orbital compartment syndrome is a rare, potentially sight-threatening condition that warrants early identification and prompt and aggressive treatment. Owing to the sight threatening nature of the condition, early diagnosis plays a critical role in the management, which is often caused by the retrobulbar haemorrhage due to various etiologies. The association of orbital compartment syndrome with viral fever has not been reported. Here, we present a case of a patient who presented with orbital compartment

Address for correspondence: Dr Ankita Singh, Dept of Ophthalmology, Armed Forces Medical College, India.
E-mail: ankita.afmc2013@gmail.com

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syndrome and was later diagnosed with dengue haemorrhagic fever.

CASE REPORT

A 43-year-old male patient reported to our hospital with complaints of sudden onset pain and swelling in the left eye for 2 days and a history of fever with myalgia for 5 days, for which he did not seek any treatment. There was no antecedent history of head or eye trauma, altered sensorium, seizures, persistent headache, neck stiffness, diplopia, or deviation of the angle of the mouth. There had been no previous medical or surgical comorbidity. The patient was non-alcoholic and a non-smoker. There was no history of bleeding from any other body parts. On initial examination, the patient had tachycardia (pulse rate of 140 beats per minute) and a blood pressure of 140/96 mmHg in the sitting position. His mental status was E4 V5 M6 (E for eye, V for verbal, M for motor) on the Glasgow coma scale. On ocular examination, his right eye had an uncorrected vision of 6/6, with a normal anterior and posterior segment, whereas his left eye had visual acuity of no light perception. Clinical evaluation of the anterior segment of the left eye revealed severe haemorrhagic axial proptosis measuring more than 30 mm, lagophthalmos, bloody discharge from the eye, periorbital oedema, chemosis, subconjunctival haemorrhage, a hazy cornea and total hyphema with no further view. On palpation, the left eyelid was tender and had a raised temperature with overlying erythema and induration. The intraocular pressure (IOP) was raised, and extraocular movements were limited in all directions of gaze. The presentation and clinical findings suggested a provisional diagnosis of orbital compartment syndrome, which required urgent decompression [Figure 1a]. The right eye's ocular evaluation was essentially normal, with preserved light and pupillary reflex. A general fundus examination of the right eye was also normal. Systemic examination of the abdomen and respiratory systems was within normal limits. Given the sudden onset of proptosis and loss of vision, an urgent CT scan of the brain and orbit was done, which revealed no brain abnormality but left an intraocular haemorrhage. There was

no evidence of retro orbital haemorrhage in the left eye on the CT scan. [Figure 2a and b].

Preliminary haematological investigations were within normal limits, except for thrombocytopenia having counts of 10,000/ μL (normal 150,000–450,000/ μL). The peripheral smear was negative for malaria and any other hemo-parasite. A positive dengue non-structural protein-1 (NS-1) antigen confirmed the diagnosis of dengue haemorrhagic fever with thrombocytopenia. Dengue IgM antibodies were also found to be positive. The patient was managed with urgent platelet transfusions and supportive therapy. The platelet count after five single donor platelets (SDPs) improved to 50,000/ μL . The patient was managed conservatively with IV mannitol and topical carbonic anhydrase inhibitors for raised IOP, along with topical antibiotics and lubricants, and was then advised to obtain a physician's clearance owing to existing thrombocytopenia before lateral canthotomy for orbital decompression. The surgical procedure was performed after a platelet transfusion. Cantholysis was also done, and the excessive fluid was let out following the canthotomy. However, retropulsion was not possible despite the surgical intervention. The patient started bleeding from the incision site, and haemostasis was achieved using interrupted silk sutures [Figure 1b]. He was managed with a pressure bandage and antibiotic eye ointment. The silk sutures were removed 2 weeks after surgery. During the course of follow-up, the left eye chemosis was slightly reduced without any improvement in the visual acuity.

DISCUSSION

The involvement of the eye in this dengue fever, ranging from subconjunctival haemorrhage to optic neuritis, suggests either an underlying infective etiology or an immune-mediated process.^[2] The possible explanations for haemorrhage could be platelet dysfunction, thrombocytopenia with bleeding diathesis, consumptive coagulopathy, and capillary fragility.^[3,4] The ocular complications are generally self-limiting with the remitting course of systemic disease.



Figure 1: (a) Photograph showing severe haemorrhagic proptosis, lagophthalmos, bloody discharge from the left eye, periorbital oedema, chemosis, subconjunctival haemorrhage, hazy cornea, shallow anterior chamber, and total hyphema. (b) Postoperative photograph showing lateral canthotomy incision site with minimal reduction of haemorrhagic proptosis.

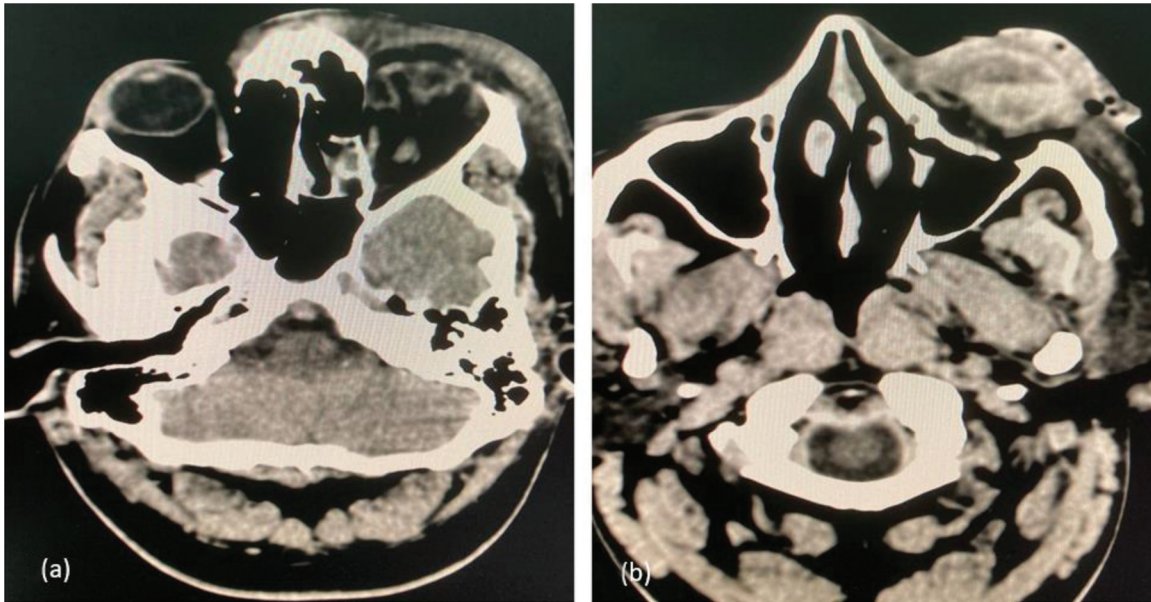


Figure 2: (a) and (b) CT scan axial sections of the brain and orbit showing left expanded globe with displaced eyeball infero-laterally. Intraocular haemorrhage is noted on the left side with no evidence of carotid-cavernous fistula.

As reported in the literature, ophthalmic manifestations of dengue fever are primarily due to the involvement of the posterior segment, like vitreous haemorrhage, macular oedema, vascular occlusion, vasculitis with retinal haemorrhages, chorioretinitis and optic neuropathy.^[4-7] Anterior segment manifestations mostly present as subconjunctival haemorrhages and anterior uveitis, but have not been frequently reported.^[6,7] Sporadic presentations such as orbital compartment syndrome, periorbital ecchymosis, ptosis and globe rupture have been noted.^[8-10] Orbital compartment syndrome is an unusual ocular emergency characterized by a sudden increase in orbital pressure commonly seen in acute onset orbital haemorrhage due to trauma, surgery, local injections, fulminant orbital cellulitis, orbital emphysema, inflammation, intraorbital abscess, tumours and other preexisting medical conditions.^[9-11] Rarely, orbital compartment syndrome has been reported in cases of dengue fever. Damage to ocular and other intraorbital structures can occur due to raised intraorbital pressure, leading to irreversible blindness if not treated promptly. Orbital compartment syndrome is a clinical diagnosis that warrants prompt recognition and urgent orbital decompression to prevent permanent vision loss. Surgical intervention in the form of lateral canthotomy along with inferior cantholysis remains the mainstay of management to save an eye with vision or the potential for visual recovery.^[10,11]

Literature shows previously reported cases of proptosis secondary to retrobulbar haemorrhage and panophthalmitis in dengue patients.^[8,9] In this patient, the proptosis was possibly due to the intraorbital haemorrhage, which was confirmed on a CT scan and could have probably occurred abruptly due to thrombocytopenia leading to orbital

compartment syndrome. However, a literature search reveals this is probably the first clinical case presentation of sudden onset haemorrhagic proptosis, as documented in this patient. In this case, the probable underlying mechanisms for compartment syndrome could be an increase in intraorbital pressure due to intraocular haemorrhage owing to severe thrombocytopenia with capillary fragility, coagulation defects, and platelet dysfunction. Our patient's onset of this ophthalmic emergency corroborated with the fever and severe thrombocytopenia, as also noted in other studies.^[2-4] Lateral canthotomy was challenging as thrombocytopenia posed an imminent risk for inadvertent perioperative bleeding during surgery. The surgical procedure was performed after a platelet transfusion. However, retropulsion was not possible despite the surgical intervention. The patient started bleeding from the incision site, and to avoid haemostasis interrupted silk sutures were placed. Therefore, it is mandatory to evaluate patients with dengue fever who present with ocular symptoms to prevent sight-threatening complications. Such cases may adopt conservative approaches such as IOP-lowering drugs and pressure bandages to control bleeding in such cases.

To conclude, we report an unusual orbital compartment syndrome case diagnosed with dengue haemorrhagic fever. It is recommended that ophthalmologists be aware of such ocular manifestations as the likely initial presentation of dengue fever. Prompt and appropriate treatment by a multidisciplinary approach should prevent such dreaded vision-threatening complications. Written and informed consent was taken from the patient to publish the clinical data.

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Conflicts of interest

There are no conflicts of interest.

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