



Unilateral lateral rectus palsy following dengue: a case report

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Introduction and importance: Dengue fever is a mosquito-borne viral infection presenting with high-grade fever and other constitutional symptoms.

Case presentation: This case report details a rare occurrence of lateral rectus palsy in an 18-year-old male subsequent to dengue infection. The patient initially presented with fever, a generalized tonic-clonic seizure (GTCS), and symptomatic hypoglycemia, leading to multiple organ dysfunction syndrome (MODS) necessitating intensive care. Remarkably, no haemorrhagic manifestations were observed. The MODS gradually resolved by the 12th day of admission, coinciding with the emergence of complaints about uniocular diplopia and right esotropia. Systemic examination, including a normal computed tomography (CT) head scan, did not reveal any abnormalities. Additionally, potential causes contributing to esotropia and diplopia were ruled out. The patient was subsequently managed expectantly for lateral rectus palsy following severe dengue. Follow-up assessments indicated a gradual improvement in esotropia and diplopia, and the patient was advised to continue the prescribed medications.

Conclusion: This is the first documented case report of paralytic squint post-severe dengue in Nepal, emphasizing the importance of considering it as a differential diagnosis in tropical regions with endemic dengue infections. The case report advocates early identification and treatment of ophthalmic issues, notably with prednisolone, to achieve favourable outcomes, as evidenced by improvements in visual acuity, esotropia, and diplopia during follow-ups. Further research is essential to determine optimal treatment strategies for such neurological complications associated with dengue fever.

Keywords: Dengue, Diplopia, Esotropia, Lateral rectus palsy, MODS

Introduction

Dengue, a viral illness caused by four serotypes (DEN-1, DEN-2, DEN-3, and DEN-4) within the Flavivirus genus, is transmitted through the bite of infected female *Aedes* mosquitoes^[1]. Approximately 390 million dengue infections occur annually across 128 countries, with 75% of this burden concentrated in Southeast Asia and the Western Pacific region. Notably, the Southeast Asia region reports a 1% fatality rate^[2].

In Nepal, the case fatality rate (CFR) for dengue was recorded at 0.04%. Spatial and temporal shifts in dengue patterns were observed in 2022 and persisted into 2023, with Nepal witnessing earlier spikes in case numbers than usual^[3]. Despite historically circulating all four dengue serotypes, DENV-1 and DENV-3 were predominant in 2022, with no evidence of DENV-4. This year,

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HIGHLIGHTS

- In this case report, we present the unique case of an 18-year-old male diagnosed with dengue who experienced multiple organ dysfunction syndrome (MODS) and right lateral rectus palsy during the convalescence period, highlighting the rare occurrence of the palsy as a consequence of dengue infection.
- Prompt identification and treatment of ophthalmic complications with prednisolone led to significant improvement in visual acuity and resolution of diplopia, highlighting the importance of early intervention.
- This case emphasizes the critical need for clinicians to remain vigilant for neurological complications in dengue fever cases and highlights the role of timely intervention in optimizing patient outcomes.

Nepal also reported its highest-ever recorded number of dengue cases, reaching 54 784 cases and resulting in 88 deaths^[4]. After recovering from the primary infection, individuals acquire life-long serotype-specific immunity^[5]. However, antibodies generated from the initial infections often cross-react, and subsequent infection with different serotypes can result in severe and potentially fatal dengue due to antibody-dependent enhancement (ADE)^[6].

Clinically, dengue manifestations vary from self-limiting mild febrile illness to potentially life-threatening dengue haemorrhagic fever (DHF) and dengue shock syndrome (DSS). Additionally, dengue is known to affect multiple organ systems, including the eyes. Nevertheless, isolated abducens nerve palsy, presenting as a

paralytic squint following dengue, is exceptionally rare, with only a few cases reported earlier in the literature^[7-9]. In this case report, we present the unique case of an 18-year-old male diagnosed with dengue who developed multiple organ dysfunction syndrome (MODS) and right lateral rectus paralysis during the convalescent period, highlighting the rare occurrence of the palsy as a consequence of dengue infection.

Case presentation

An 18-year-old male patient was referred to our centre with a three-day history of fever and headaches. One day prior to the referral, the patient experienced three episodes of generalized tonic-clonic seizures, each lasting 3–5 min. These seizures were characterized by tongue biting, up-rolling of the eyes, frothing from the mouth, and urinary and faecal incontinence. The post-ictal period was eventful for the absence of consciousness for ~45 min, prompting immediate hospital transport. His Glasgow Coma Scale (GCS) was E1VTM1, with a score of 1 for eye-opening (E), 1 for verbal response (V), and 1 for motor response (M), SpO₂ 75% and was intubated and referred to our centre.

Upon admission, the patient had a respiratory rate of 24 breaths/min, a Pulse of 130 beats/min, a temperature of 99.1°F, SpO₂ of 97% with FiO₂ at 60%, and GCS was E1VTM1 with unrecordable blood pressure. He was severely hypoglycaemia at presentation, with a glucose level of 13 mg/dl. Investigations on the same day revealed the following values: Haemoglobin (Hb) at 16.8g/dl (normal range: 12–16 gm/dl), packed cell volume (PCV) at 54.9% (normal range: 36–54%), white blood cell (WBC) count at 17 420/mm³ (normal range: 4000–11 000/mm³), platelet count at 65 000/mm³ (normal range: 150 000–400 000/mm³), Serum creatinine at 3.20 mg/dl (normal range: 0.4–1.4 mg/dl), Serum urea at 43 mg/dl (normal range: 15–45 mg/dl), sodium at 140.1 meq/dl (normal range: 135–150 mmol/l), potassium at 2.67 mmol/l (normal range: 3.5–5.5 mmol/l), alanine transaminase (ALT) at 49 IU/l (normal range: <45IU/l), aspartate transaminase (AST) at 261 IU/l (normal range: <40IU/l), prothrombin time (PT) exceeding 90 sec (normal range: 10–14 sec), international normalized ratio (INR) greater than 7sec, and the patient tested positive for IgM dengue antibodies.

All systemic examinations yielded normal results except for the central nervous system (CNS), which could not be evaluated. However, normal muscle tone and downgoing plantar reflexes were observed.

He had no significant past medical or surgical history.

He was managed with Intravenous (IV) fluid bolus, injection of dextrose, IV antibiotics, sedation with Midazolam and fentanyl, and a potassium chloride (KCl) infusion for hypokalemia. The patient was transferred to the Medical ICU (MICU) for further management.

The patient was drowsy the next day with persistently elevated AST/ALT and deranged PT INR. Serum urea was increased to 60 mg/dl, while serum creatinine reached 1.22 mg/dl.

Subsequently, the patient was managed based on the protocol for severe dengue with MODS.

The platelet count reached its lowest point at 34 000 on the sixth day of fever, surprisingly without any observed haemorrhagic manifestations. The patient gradually became afebrile on the 11th day. Additionally, both acute kidney injury (AKI) and Acute Liver Failure subsided over the course of the 12-day admission period. However, the patient complained of unocular

diplopia on the 12th day of admission, and right eye esotropia was noticed. His visual acuity was 6/36, and pinholes were 6/12p on both eyes. The anterior and posterior segments, including the fundus and intraocular pressure (12 mm Hg on both eyes) were normal. The computed tomography (CT) head was normal, liver function test, kidney function test, serum electrolytes (sodium, potassium, calcium) were normal, viral markers were non-reactive, and potential causes leading to right esotropia with diplopia were ruled out.

A normal CT head scan ruled out the presence of intracranial haemorrhage, intracranial space-occupying lesions, and skull fracture in this case.

The absence of compressive mass lesions confirmed through imaging, along with clinical and serological exclusion of other infectious, inflammatory, and vasculitic causes, and the clear temporal association with dengue fever, establishes the connection between abducens nerve palsy and dengue fever in our patient.

Therefore, he was expectantly managed for the right lateral rectus palsy, including treatment with ketorolac tromethamine, prednisolone, lubricating eye drops, multivitamins, and advised extraocular muscle physiotherapy. The patient was kept on follow-up at 1 month intervals, and the status of esotropia and diplopia during two subsequent follow-ups is depicted in Figures 1, (Fig: 1.1, 1.2, 1.3) and 2, respectively.

The esotropia and diplopia were gradually improving, and his visual acuity improved to 6/12 on the right eye and 6/36 on left eye. With pinhole correction, visual acuity improved to 6/6p in the right eye and 6/9 in the left eye. The phenomenon of overshooting is observed in the contralateral eye, as illustrated in Figs. 1.2 and 1.3, are governed by Hering's law of equal innervation, representing a physiological principle. He was advised to maintain the current medication and physiotherapy regimen. The extraocular motility on different version movements is demonstrated below in Fig. 3.

Discussion

Dengue, an ssRNA virus, comprises three structural (capsid 'C,' precursor membrane prM, and envelope 'E') and seven non-structural proteins (NS)^[10]. Typically, dengue fever manifests as an acute febrile illness characterized by retro-orbital pain, severe headache, myalgia, arthralgia, and rash. Haemorrhagic manifestations are present in both dengue fever and dengue haemorrhagic fever, with plasma leakage being the primary feature distinguishing the two conditions. The progression of a decreasing platelet count and increasing haematocrit serves as an indication of impending shock^[11-13]. Moreover, the lowest platelet count during the course of the illness has been observed to correlate with the onset of ophthalmic symptoms, typically occurring approximately one week after the fever onset. Among the common visual complaints associated with dengue are blurred vision (51.2%) and central scotoma (34.1%). Dengue-associated ocular inflammation is a rising ophthalmic concern, frequently affecting the posterior segment. The self-limiting course of the disease usually favors good restoration of vision even without treatment. However, patients may experience mild, persistent relative central scotoma for several months^[14,15].

Ocular findings in dengue fever were considered rare previously. Now, a broad spectrum of ocular manifestations ranging

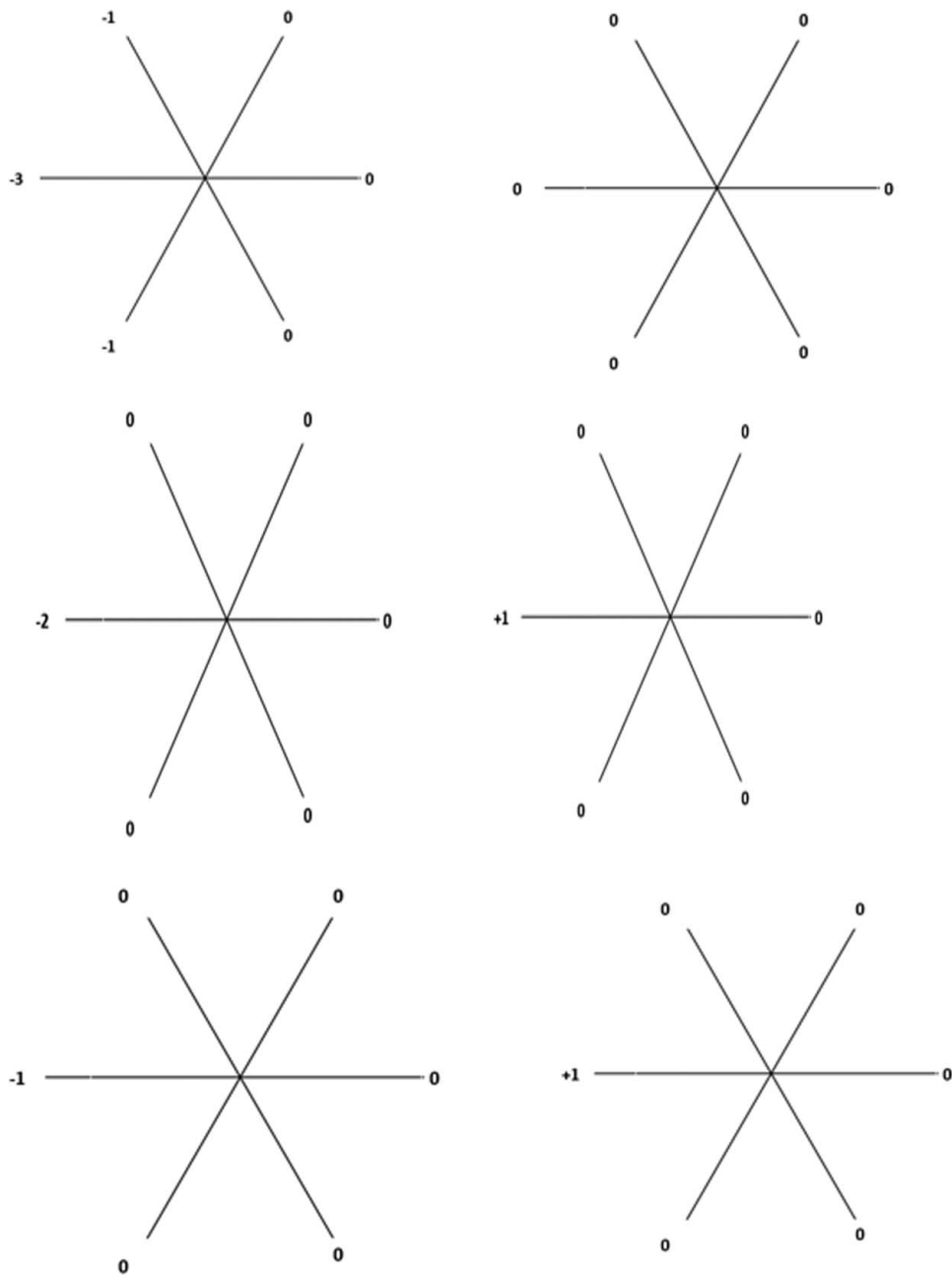


Figure 1. 1.1: Status of esotropia during diagnosis (Right eye esotropia with normal left eye). 1.2: Status of esotropia one month later (Improving right eye esotropia with left eye overshoot). 1.3: Status of esotropia two months later (Improving right eye with left eye overshoot).

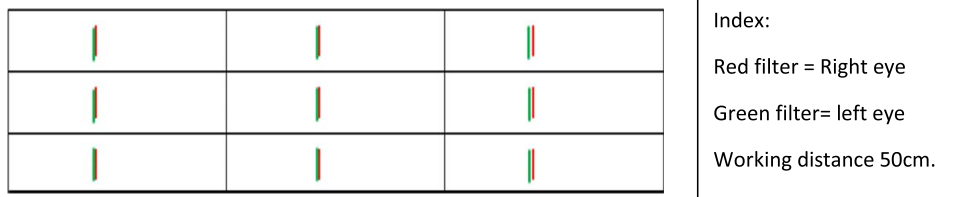


Figure 2. Diplopia charting showing uncrossed diplopia in right gaze.

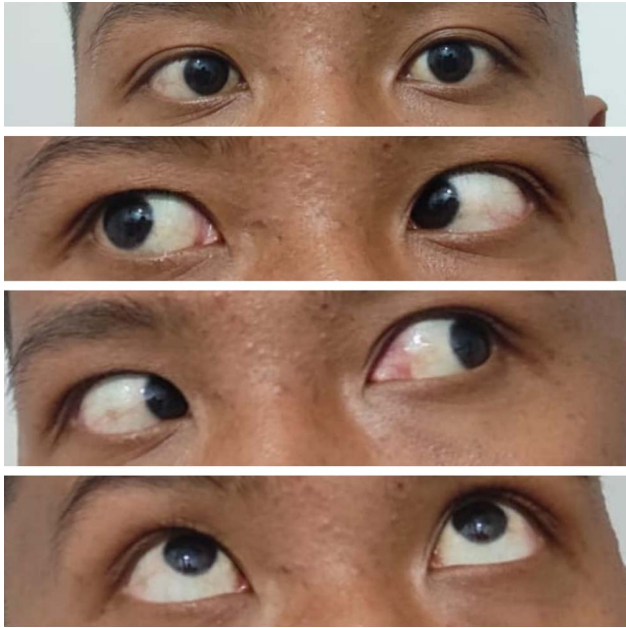


Figure 3. Ocular examination showing right esotropia and limitation of movement in right lateral gaze.

from mild non-specific symptoms to severe retinal haemorrhages are being commonly reported. Previously uncommon neurological complications such as encephalopathy, Guillain–Barre syndrome, radiculopathies, plexopathies, and mononeuropathies have now been recognized. Additionally, isolated cranial nerve involvement, such as optic neuropathy and oculomotor nerve palsy, has become more frequently reported in recent studies^[16]. However, abducens nerve palsy, manifesting as lateral rectus palsy, remains a rare occurrence, with only a few documented cases reported thus far^[6,7,15]. In this report, we presented a case of an 18-year-old male with severe dengue requiring intensive organ care who had undergone lateral rectus palsy manifested as right esotropia and uniocular diplopia.

The precise mechanism behind neurological complications is not fully comprehended. The three main pathogenic mechanisms for the neurological complications of dengue infection were identified: (1) direct invasion of the CNS by the virus, leading to neurotropic effects such as meningitis, encephalitis, and myositis; (2) metabolic abnormalities causing CNS complications like encephalopathy, stroke, and hypokalemic paralysis; and (3) autoimmune complications, including encephalomyelitis, Guillain–Barré syndrome (GBS), cranial nerve palsy, and optic neuritis. All four serotypes of dengue virus (DenV-1 to DenV-4) have been associated with neurological manifestations in dengue fever^[2].

In a retrospective study involving thirteen patients with ophthalmic complications following dengue, there is a close correlation between the onset of visual symptoms and the lowest recorded platelet counts. Among the nine patients monitored daily for platelet levels, all experienced visual symptoms within one day of reaching their lowest platelet counts. Specifically, 55.6% of patients reported visual symptoms on the same day as their lowest platelet count, while others experienced symptoms either one day before or after^[13]. However, in our case, visual

symptoms were evident only 6 days after the patient's platelet count reached its lowest level.

Conclusion

This is the first documented case report of paralytic squint post-severe dengue in Nepal. In regions with endemic dengue infections like Nepal, where the likelihood of encountering unusual complications of this very common disease increases, healthcare professionals should consider post-dengue infection abducens neuropathy as a potential cause of lateral rectus palsy, especially in cases presenting with acquired convergent squint and diplopia within compatible clinical contexts.

It emphasizes early identification and appropriate treatment of ophthalmic complications, particularly with prednisolone, for favourable outcomes. The observed improvement in visual acuity, esotropia, and diplopia during follow-ups emphasizes the potential for recovery with timely interventions. It also highlights the importance of considering neurological complications in dengue cases and the need for ongoing vigilance for ocular manifestations, even without haemorrhagic manifestations. Further research and advanced imaging modalities are essential to enhance our understanding and refine management strategies for such cases.

Informed consent

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

Limitation

Although prior head imaging, CT head, revealed no abnormalities, MRI could have provided further insights into potential underlying causes of the lateral rectus palsy following dengue infection. However, conducting an MRI to investigate possible pathologies along the abducent nerve pathway was not feasible due to financial constraints, as the patient could not bear the additional expenses associated with MRI imaging. Nevertheless, our case report highlights the rare occurrence of paralytic squint as a neurological complication after severe dengue. It sheds light on the positive impact of prednisolone in enhancing visual acuity, correcting esotropia, and alleviating diplopia, highlighting the potential for recovery through timely interventions.

Ethical approval

Since this case report has not involved clinical trials upon patient, we have not acquired ethical approval, informed written consent from the patient is taken

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We do not have any funding for research.

Author contribution

B.N.: conceptualization, validation, visualization, writing an original draft, review and editing. B.A.: conceptualization, validation, visualization, writing an original draft, review and

editing. A.S.: writing and review. S.K.G.: writing and review. D.G.: writing and review

Conflicts of interest disclosure

There is no conflicts of interest.

Research registration unique identifying number (UIN)

Not applicable.

Guarantor

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Data availability statement

Data are publically available.

Provenance and peer review

Not applicable.

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