



## **Oculogyric Crisis – Atypical Presentation in Patient with Dengue Fever**

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### **Authors' contributions**

*This work was carried out in collaboration among all authors. All authors read and approved the final manuscript.*

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**Case Study**

### **ABSTRACT**

Here we are presenting a case of dengue fever presented with an atypical symptom of “oculogyric crisis” with features of multi organ dysfunction syndrome. Increased vascular permeability, plasma leakage, haemorrhagic manifestations, and thrombocytopenia are characteristic of dengue fever. A 19 year old male patient, with no major co-morbidities came to Dhiraj hospital with chief complaints of fever which was high grade and intermittent; associated with chills & rigors for 5 days. He also complained of abdominal pain over epigastric region which was dull aching, non radiating associated with decreased appetite since 5 days. Dengue infection has a wide range of clinical features. Neurological complications may occur in any spectrum of dengue infection. The diagnosis of oculogyric crisis associated with dengue infection has not been reported in any Indian literature so far. Oculogyric crisis is a clinical diagnosis supported with detailed clinical history and physical examination requiring exclusion of the other possible etiologies.

**Keywords:** *Oculogyric crisis; multi organ dysfunction syndrome; dengue fever; dengue hepatitis.*

## ABBREVIATIONS

RR	: Respiratory rate
WBC	: White blood cell
ALF	: Acute liver failure
GBS	: Guillain barre syndrome
LVEF	: Left ventricular ejection fraction.

## 1. INTRODUCTION

Dengue is one of the most widespread arthropod- borne viral disease and is transmitted in humans by *Aedes aegypti* mosquitoes. DEN-1, DEN-2, DEN-3 & DEN-4 are the four subtypes of Dengue [1].

Increased vascular permeability, plasma leakage, haemorrhagic manifestations, and thrombocytopenia are the characteristic features of dengue. Disease exists in a wide range of spectrum ranging from asymptomatic infection to dengue shock syndrome [2].

Dengue infection can cause multi organ dysfunction syndrome, involving cardiovascular system in the form of myocarditis, central nervous system features, hematological involvement & other various manifestations. It is an important cause for multi organ dysfunction syndrome among febrile illness [3].

Neurological involvement in dengue infection is a well known feature. The extrapyramidal involvement in the form of oculogyric crisis has not been described in Indian literature so far. The most common complication of dengue is Encephalopathy. Encephalitis is considered a clinical presentation of dengue infection in endemic areas. Serological types 2 & 3 of dengue infection are commonly associated with severe neurological complications. For other

possible neurological diagnosis like myelitis, myositis, GBS etc., one should always keep the possibility of dengue infection in mind [4].

Here we present a peculiar case of dengue fever with atypical “oculogyric crisis” associated with features of multi organ dysfunction syndrome.

## 2. CASE REPORT

A 19 years old male patient, with no major co-morbidities came to Dhiraj hospital with chief complaints of high grade intermittent fever, associated with chills & rigors for 5 days. He also complained of abdominal pain over epigastric region which was dull aching, non radiating, associated with decreased appetite for five days. Two days prior to admission, he developed involuntary upward deviation of eyeballs, with difficulty in downward gaze; with this the patient came to outpatient department of medicine, Dhiraj hospital following which we admitted the patient in ICU. After detailed history and clinical examination, we found that the patient was having “Oculogyric crisis” with recurrent episodes during the hospital stay. Patient denied intake/abuse of any medications which could explain the atypical phenomena or extra pyramidal symptoms. However, it was remitted with medical management and supportive care.

On examination patient had bradycardia with pulse rate of 56 bpm, regular rhythm, other vital data were stable, petechiae were found on right arm with skin blanching (+) with delayed capillary refill time. Upon Investigating, Hb was found to be 15.3 gm/dl, WBC- 9600 and Platelets- 20,000 per microliter. His Dengue NS1 Antigen was positive. Liver function test showed LDH -1200, SGOT - 110 and SGPT - 127units per liter.



Fig. 1. Oculogyric crisis

S. creatinine and S. electrolytes were within normal range. USG showed mild volume of Ascites. ECG & chest x-ray were normal. 2D echo was performed in view of bradycardia, which showed mild LV global hypokinesia with LVEF 45% and suggestive of myocarditis. A neuro imaging was undertaken which was normal. On further investigations concomitant infections like malaria, other viral diseases with similar clinical pathology were excluded. A probable diagnosis of dengue fever with viral myocarditis with atypical oculogyric crisis was considered and the patient was treated with IV fluids, symptomatic & supportive management and patient recovered.

### 3. DISCUSSION

A young gentleman with paroxysms of “oculogyric crisis” in a case of dengue fever, with no other identifiable cause to account for the atypical abnormal eye movements is presented here.

The word "oculogyric" alludes to the bilateral elevation of the visual gaze. The “Oculogyric crisis” is a dystonic response to specific medications or medical ailment, characterized by prolonged involuntary upward deviation of the eyes.

Dengue infection has a wide range of clinical scenarios. Neurological complications can emerge in any spectrum, for example, in dengue fever, or in dengue haemorrhagic fever. Encephalopathy and encephalitis are the most commonly found neurological complications[4].

Neurological complications in dengue infection on the basis of pathophysiology are secondary to systemic complications like electrolyte abnormalities, microcapillary hemorrhage, release of toxic products, and liver and kidney failure; secondarily a post-infectious immune mediated mechanism such as GBS and neuralgic amyotrophy, and a direct neurotropic impact of the infection as encephalitis, myelitis or myositis have been documented [4].

In a review from India, Misra U.K. *et al.* published series of 17 patients of which 11 had febrile encephalopathy. Real case scenario of these eleven patients showed that six had acute motor weakness without sensory involvement of which five had raised CPK levels and one having biopsy proven myositis. Three patients had seizure and one had myoclonus. There was globus pallidus involvement and thoracic spinal myelopathy in one case each as shown by MRI

studies [5]. MRI study showed hyperintense foci in globus pallidus in one of the nine patients with dengue encephalitis in clinical case series reported Thai medical clinic [6]. Dengue encephalitis patients can have normal MRI like our patient or may have scattered focal abnormalities like haemorrhage and oedema [5,6]

In a recent study of neuroimaging (20 MRI + 1 CT) done by Bhoi S. K. *et al.* in 21 confirmed cases of dengue fever with altered sensorium, the abnormalities were seen in 10 patients. The imaging features in the study were cerebellar bleed, subdural haematoma, signal changes in the thalamus, basal ganglia, white matter, and cortical area and meningeal enhancement. The imaging pattern did not affect the outcome [7].

A study done by Fragoso Y. D. *et al.* (2016) [8] “Encephalomyelitis Associated With Dengue Fever” reported neurological symptoms like diplopia, optic neuritis, para paresis, paraesthesia, lower limb hyper reflexia, unusual proprioception, urinary incontinence, dizziness, and gait ataxia. Magnetic resonance imaging of his brain and spinal cord confirmed the presence of demyelination.

Our patient had bradycardia with global hypokinesia, features suggestive of heart-involvement in the form of “viral myocarditis.” A study by Lia Yingying *et al.* (2014) “Characterization of the Myocarditis during the worst outbreak of dengue infection in China” documented myocarditis as a common complication. In this study from china, total 1782 confirmed dengue patients were admitted from August to October, 2014 were studied of which 201 cases had myocarditis. Prevalence of myocarditis in hospitalized dengue was 11.28% which ranged from 9.72% in dengue cases without warning signs and 46.7% in severe dengue as well as in non-severe dengue with warning signs. Myocarditis is an especially common & important cause leading to death in patients with severe dengue. However, review of myocarditis in dengue is still incredibly inadequate [9].

Our patient had slightly raised enzymes of liver function test suggestive of “dengue hepatitis”.

A review done by Devarbhavi Harshad *et al.* (2014) on “Dengue hepatitis with acute liver failure:

Clinical, biochemical, histopathological characteristics and predictors of outcome” reported 36 cases of acute liver failure (ALF) of 10,108 patients diagnosed to have dengue. Among them, around 72.2% and 27.8% patients satisfied criteria for hyper-ALF and ALF, respectively. Dengue hepatitis advanced to ALF in 0.35%. Development of ALF was related with a high mortality (>50%). Lactate level, pH, and MELD score at the time of admission were significant determinants of result [10].

A study done by Ishak S. H. et al. [11] “Severe Dengue with Hemophagocytosis Syndrome” present a case report of patient diagnosed to have multi-organ dysfunction and highly elevated liver enzymes. There has been one case series done by Tan et al. [12] of eight dengue cases related with hemophagocytosis syndrome reported from Malaysia.

The diagnosis of oculogyric crisis is to a great extent clinical and includes taking a focused history and physical examination to recognize potential triggers for the crisis and to exclude other causes of abnormal eye movements.

Medications that can cause an oculogyric crisis include neuroleptics (such as haloperidol, chlorpromazine, olanzapine) [13]. also carbamazepine, chloroquine, cisplatin, diazoxide, levodopa [14], lithium, metoclopramide, domperidone, nifedipine, phencyclidine [15] reserpine, and cetirizine, an antihistaminic are responsible for an oculogyric crisis. High-potency neuroleptics are the most widely recognized reason.

Different causes of oculogyric crisis can include aromatic L-amino acid decarboxylase deficiency [16], postencephalitic Parkinson's, Tourette's syndrome, multiple sclerosis, neurosyphilis, head injury, infarction of bilateral thalami, fourth ventricle lesions, cystic glioma of the third ventricle, encephalitis due to herpes virus, kernicterus and juvenile Parkinson's disease. Concomitant malaria and dengue may coexist [17] and neurological features can be due to neuromalaria also.

#### 4. CONCLUSION

In our study, 19 years old male patient of dengue hemorrhagic fever with multi system involvement had atypical oculogyric crisis with the exclusion of other possible etiologies was considered a

peculiar finding associated with dengue fever syndrome.

#### ETHICAL APPROVAL

As per international standard or university standard written ethical approval has been collected and preserved by the author(s).

#### CONSENT

As per international standard or university standard, patients' written consent has been collected and preserved by the author(s).

#### COMPETING INTERESTS

Authors have declared that no competing interests exist.

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