Case Report

Isolated Unilateral Third Cranial Nerve Palsy : A Rare Presentation of Dengue Fever

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We herein report a case of unilateral 3rd cranial nerve palsy in a 15 years old boy. It can be due to numerous aetiologies like infectious, inflammatory, malignant, metabolic or vascular. In our case the nerve palsy was preceded by history of high grade fever of 5 days. Involvement of 3rd cranial nerve started 9 days after fever onset, insidiously, presenting as Ptosis and Diplopia. No history of altered sensorium, limb weakness, diurnal variation. Routine investigation was normal. Integrated Counselling and Testing Centre (ICTC) was negative. Cerebrospinal Fluid (CSF) study revealed viral picture but was negative for neurotropic viral panel. MRI brain was essentially normal except for presence of small Lipoma over prepontine cistern. Antinuclear Antibody (ANA) and Antineutrophil Cytoplasmic Antibodies (ANCA) were negative. Serology for Dengue was sent considering the history of high grade fever associated with blanchable rash. Dengue IgM report came out to be reactive. CSF Dengue IgM also came out to be reactive. Patient was put on short course of oral steroid therapy and cranial nerve palsy improved gradually. Neurological complications of dengue is uncommon. Few cases of Cranial Nerve Involvement associated with Dengue have been reported in the literature, most of them are associated with encephalitis. But in our case Cranial Nerve involvement was not associated with Encephalitis, it was probably due to immune reactions secondary to Dengue, making this case atypical.

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Key words **: Extra axial occulomotor nerve involvement, Immune reaction, Dengue fever, Convalescent stage.**

Dengue virus is one of the most important Flavivirus. There are 4 serotypes 1-4, which can cause infections with severity ranging from prodrome of constitutional symptoms and signs to severe Hemorrhagic Fever with or without Shock Syndrome. Recently it has been known to involve CNS as well. Neurovirulence is mediated by direct viral invasion and subsequent metabolic changes or from indirect mechanisms. Neurological involvement mainly occur in the form of Encephalopathy. Rarely, it can manifest as Neuritis, secondary to humoral immune response. To establish the diagnosis, serum and CSF reactivity for IgM antibody to be tested. Differential diagnosis to be excluded by appropriate investigations- blood test (to rule out other infectious diseases, inflammatory diseases), CSF examination (cell type and cell count, protein, sugar, RT PCR for neurotropic viral panel), Brain imaging (MRI with contrast). In this article, we are reporting a case of Dengue Fever with Oculomotor Nerve Palsy, extra axial lesion.

Case Report

15 years old aged male patient presented to us with chief complain of

- High graded fever, continuous in nature with chills and rigor, associated with malaise and headache for 12 Days. A febrile for last 7 days.

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Editor's Comment :

- Understanding the pathogenesis of cranial nerve involvement in dengue is of utmost therapeutical importance.
- Short course of oral steroid therapy is needed for early recovery if the underlying mechanism is immune reaction, a rare possibility.

- Drooping of left upper eye lid for 3 days; insidious onset, gradually progressive, associated with Diplopia that disappears on closing either eye and was maximal on looking upwards and right lateral gaze. No diurnal variation present. No history of altered sensorium, convulsion, neck pain, no history suggestive of other Cranial Nerve Palsy or Limb weakness.

Differential diagnosis :

Infectious diseases - Bacterial infections; Viral infections: CMV, EBV, HIV; lyme disease.

Inflammatory - SLE, Vasculitis

Malignancy - Acute Leukaemia

On examination patient was alert and conscious.

Pulse - 88/min, BP - 108/72,

Lymph node - Non palpable

No skin rash, no evidence of Arthritis

CNS Examination - Absent meningeal sign, normal Ophthalmoscopy, left sided complete ptosis, Anisocoria, left pupil fully dilated, not reacting to light, on pursuit movement, other than abduction and intorsions, other movement were impaired in left eye, no limb weakness, plantar- B/L flexor

CVS examination- WNL

Respiratory system- WNL

Gastrointestinal system- No hepatosplenomegaly

Differential diagnosis -

Infectious diseases - Bacterial, viral infections : CMV, EBV, HIV; Lyme disease.

Inflammatory - SLE, Vasculitis (Fig 1).

Investigation :

CBC- Hb 13.7, TLC- 4300 (N24L62), PLT- 1.2 lacs/cumm, ESR-46

LFT- T.Bil 0.5, SGPT/SGOT/AlkPhos- 24/62/121 total protein/Alb- 5.8/3.6

Ur/Cr- 24/0.7, Na/K- 138/4.3

MP, MPDA- Negative

HBsAg/ anti HCV/ **ICTC**- negative

ANA/ anti MPO/anti PR3- non reactive

CSF study -

- cell count & count - 12 cells, all are lymphocytes
- Protein - 60mg/dl,
- Glucose - 46mg/dl
- CSF Neurotropic viral panel - Negative

MRI brain - small area of hyperintensity noted in T1, T2WI suggestive of small lipoma over prepontine cistern (Fig 2).

We re-evaluate the history that fever was associated with blanchable rashes over generalised body, extreme body ache and myalgia. Considering the possibility of dengue infection, we sent for Dengue IgM antibody-

Dengue IgM - 37.8 (reactive).

CSF dengue IgM antibody sent- reactive

Provisional diagnosis - A case of dengue fever (convalescent stage) with extra axial 3rd cranial nerve palsy with lipoma in prepontine cistern region.

Treatment and follow up - He was given supportive treatment with a short duration of intravenous fluids and short course oral steroids and was discharged 7 days after admission with almost full resolution of diplopia and partial improvement of ptosis. The ptosis recovered fully 2 weeks later on follow up.

Discussion

- Viral and host factor play important role in disease pathogenesis.
- Neuropathogenesis involves following mechanism-

metabolic disturbances- cerebral anoxia, edema, hepatic encephalopathy, hemorrhages (secondary to thrombocytopenia)

viral invasion - (a) Passive crossing of the blood brain barrier: dengue induced cytokine immune response

Fig 1 — Dropping of left upper eyelid-weakness of levator palpabrae superioris, supplied by 3rd cranial nerve

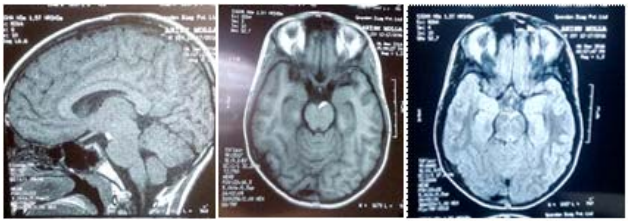

Fig 2 — MRI Angiography of cerebral blood vessel - normal

⟶ disruption of blood brain barrier⟶ invasion of CNS

(b) Actively invading CNS

autoimmune reactions- (a) immunoallergic mechanism in post infectious period

(b) Cell mediated immunologic reactions- activated T cells cross blood brain barrier ⟶ recognise an antigen in endoneural compartment ⟶ produce cytokines ⟶ disruption of blood brain barrier allowing cross reacting antibodies to enter and attack Schwan cells

- Host immune response, both inmate and adaptive, results in effective clearance of pathogenic organism. But dysfunction of immune system may lead to damage to host tissue as well. The immune response in brain secondary to Dengue infection may manifest as Cranial Neuritis.
- Diagnosis is usually made by demonstrating reactive Dengue IgM in CSF.
- Careful exclusion of other differential diagnosis to be done using detailed history, clinical examination, necessary investigations.
- Recovery is spontaneous or may need short course of Steroid Therapy in some cases.

Conclusions

- Neurological complications In Dengue fever can manifest as a form of isolated Cranial Nerve Palsy without other features of encephalopathy.
- Para infectious or post infectious Cranial Nerve Palsy may be due to immune reaction secondary to Dengue infection in CNS.
- Understanding the disease mechanism is important for appropriate treatment.
- A short course of steroid therapy prompt recovery, avoiding longer hospitalisation.

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