CASE REPORT AND REVIEW OF LITERATURE

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Facial nerve palsy: An atypical presentation of dengue fever

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ABSTRACT

Dengue fever is a grave public health concern in tropical countries. The disease spectrum of dengue fever is multi-systemic, although neurologic complications of dengue fever is relatively uncommon. The pathogenesis of neurologic manifestations in dengue fever is not well understood and may be due to the neurotrophic effect of the virus, or related to the systemic effects of dengue infection, and immune mediated. Here in, we discuss the occurrence of isolated 7th cranial nerve lower motor neuron type palsy in a 23-year-old lady suffering from dengue fever. To date, there are very few case reports citing the involvement of facial nerve in dengue fever.

Key words: Tropical fever; Seventh cranial nerve; Corticosteroid; Mononeuropathy; Bell's palsy

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INTRODUCTION

Dengue fever is caused by dengue virus, belonging to the *Flaviviridae* family of viruses and is an important arthropod-borne infection in tropical countries. Disease spectrum of dengue is diverse with multisystemic involvement. Nervous system involvement is associated with serotypes 2 and 3.[1] Dengue infections are usually asymptomatic but can present with classic dengue fever, dengue hemorrhagic fever, or dengue shock syndrome. Some unusual manifestations of dengue infection that have been reported as a part of expanded dengue syndrome include liver failure, renal failure and neurological complications such as polyradiculoneuropathy, myelitis, encephalitis, and cranial nerve palsies. There are very few cases of isolated Bell's palsy occurring with dengue infection that have been documented in literature.

CASE REPORT

A 23-year-old unmarried non-pregnant female, was admitted to MR Bangur Hospital, Kolkata, India, with the complaint of fever for 5 days (high grade, intermittent in nature) along with headache and deviation of angle of mouth to the left for the past 1 day. The patient did not have any associated cough, diarrhoea, hoarseness, shortness of breath, dysuria, abdominal pain, bleeding manifestations, ear ache, ear discharge, skin rash, difficulty of vision, photo-phobia, lesions over ears, seizures at the time of admission. She did not have any prior history of seizure disorder, heart disease, diabetes, or hypertension. The patient has a history of extra-pulmonary tuberculosis (TB meningitis) for which she had completed anti-tubercular therapy 4 years ago, and subsequently developed a communicating hydrocephalus as a sequalae; however,

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she did not face any functional debilitation over the course of time.

Examination

At the time of admission, the general consciousness of the patient was good, and she was alert, coherent, and cooperative, febrile (102F), hydrated, pulse rate - 116/min, regular; blood pressure - 110/70 mmHg, SpO_2 - 98% in room air, a respiratory rate of 23/min, random capillary blood glucose – 118 mg/dL. No cervical/axillary/inguinal lymph nodes were palpable. No rashes, or lesions were visible on the skin/mucosa. Cardiac auscultation revealed tachycardia and normal dual heart sounds, whereas bilateral broncho-vesicular breath sounds were heard during the examination of the respiratory system. GI examination revealed no oral ulcers/abdominal tenderness/organomegaly.

Neurological examination revealed the following: right lower motor neuron facial palsy, Bell's phenomenon (+), slight flattening of (right) nasolabial fold, and deviation of angle of mouth to the left (Figure 1). Neurological weakness did not deteriorate with repeated intention to action, and no diurnal variation was cited. Pupils were bilaterally equal, reacting to light. Tone/power was normal and deep tendon reflexes were preserved in all limbs. Plantar response was bilateral flexor. No clinical evidence of bulbar paresis was found.

No other cranial nerve involvement was noted. No spinal tenderness, Kernig's sign was elicited. No objective evidence of sensory system/bladder bowel involvement was noted.

Investigations

At the time of admission, complete blood count revealed a hemoglobin - 11.1 g %, total count - 4200/mm^3 (N50L40E5M5) and thrombocytopenia (95000/mm^3) which deteriorated over the next 2 days (88,000/mm^3 ---> 76000/mm^3) and thereafter showed a rising trend: Haematocrit of 49%, suggestive of haemo concentration. The patient was detected with dengue NS1 antigen positive. MPDAT and COVID-19 reverse transcription polymerase chain reaction (PCR) was negative.

Serum electrolytes (including $Na^+/K^+/Mg^{2+}$), arterial blood gas analysis, liver function tests, creatinine, and lipid profile were within normal limits. Fasting plasma glucose of 102 mg/dL was noted on day 2 of admission. On day 3 of admission, dengue IgM came (+) whereas standardized serological assays for malaria, scrub typhus, leptospira, typhoid, brucella, hepatitis B, hepatitis C, human immunodeficiency virus (HIV)1/2 were negative. Coagulation profile, bleeding time/clotting time were within normal range. Sr. ANA (1:160, Hep2 Method) was negative. Serum homocysteine and serum angiotensin-converting enzyme levels were normal.

Noncontrast computed tomography brain (Figure 2) on day 1 of admission revealed-obstructive hydrocephalus with acqueductal stenosis, which led to conduction of an magnetic resonance imaging (MRI) brain with contrast on day 2 of admission, revealing moderate communicating hydrocephalus, with few gliotic changes in periventricular white matter. The residual communicating hydrocephalus was a sequelae of the previous TB meningitis 4 years ago (Figure 3).

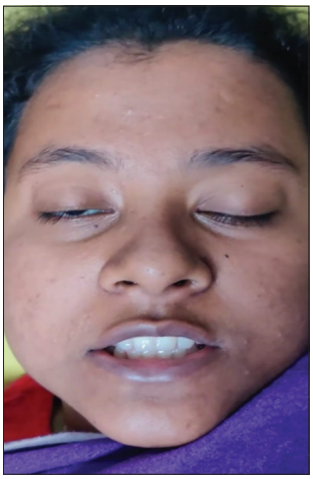


Figure 1: Bell's phenomenon (right eye) and deviation of angle of mouth to left on day 1 of admission

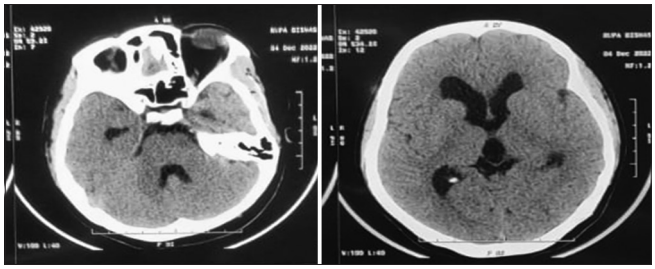


Figure 2: Non-contrast computed tomography brain, showing no parenchymal changes in brainstem, but horns of lateral ventricles are dilated

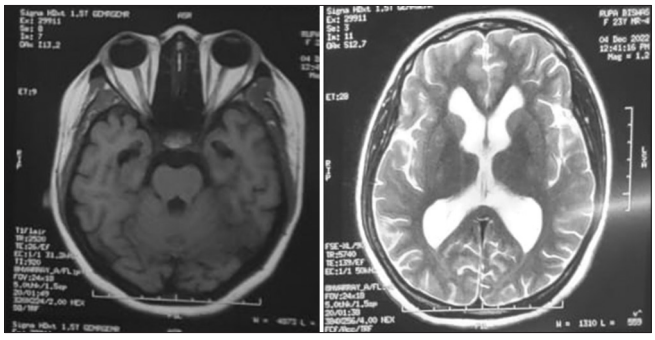


Figure 3: Magnetic resonance imaging brain - T1 image on the left showing no parenchymal signal change in pons. Similar to non contrast computed tomography brain, a communicating hydrocephalus is noted in T2 image with dilatation of horns of lateral ventricle

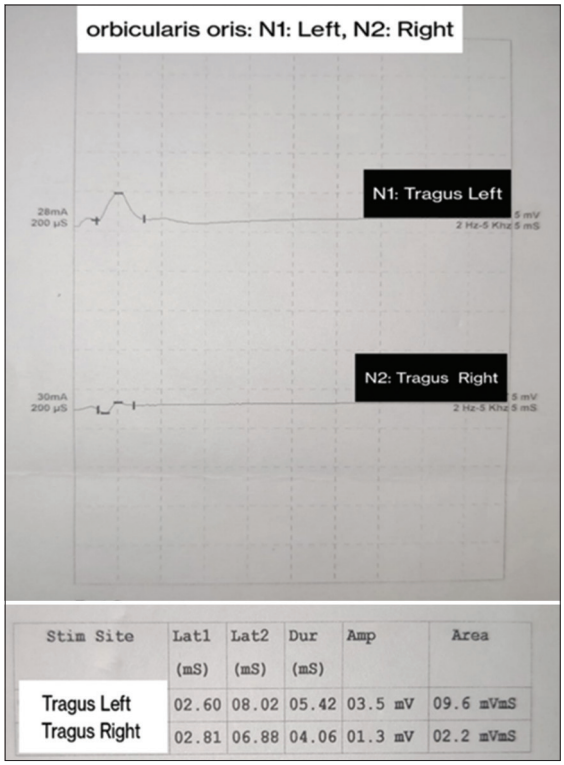
Cerebrospinal fluid (CSF) analysis on day 2 revealed the following - 5 cells (polymorphs - 67% and mononuclear cells 33%), glucose - 77 mg/dL (RBS - 110 mg/dL), protein - 32 mg/dL; CSF adenosine deaminase - 0.3 U/L. Paired serum/CSF sample for JE was negative, and CSF PCR for dengue was positive.

Facial nerve conduction study showed axonal changes in the right facial nerve when orbicularis oris was stimulated (Figure 4).

Echocardiography and echo-cardiogram was normal. Bilateral carotid artery Doppler did not reveal any intima thickening or plaque. Chest X-ray revealed no hilar lymphadenopathy or parenchymal lesions.

Clinical course

The patient remained febrile till day 4 of admission, after which fever subsided, along with an improvement in haemo-concentration. A working diagnosis of dengue fever with lower motor neurons (LMN) type cranial nerve VII palsy was arrived at, and the patient was initiated on a short course oral prednisolone initiated at 1 mg/kg with rapid tapering over 14 days, along with supportive measures for fever and hydration. Physiotherapy was initiated, and at the time of discharge patient's facial muscle weakness improved wherein she could pout and purse her lips to some extent.



Stim Site	Lat1 (mS)	Lat2 (mS)	Dur (mS)	Amp	Area
Tragus Left	02.60	08.02	05.42	03.5 mV	09.6 mVmS
Tragus Right	02.81	06.88	04.06	01.3 mV	02.2 mVmS

Figure 4: Decreased amplitude of right facial nerve in facial nerve conduction study

DISCUSSION

Dengue fever is a known arborviral disease mediated by the dengue virus of the Flaviviridae family, known in the history of medicine since 1780. It was coined as "break bone fever" by Benjamin Rash due to its associated symptomatology.[2] The disease burden in tropical countries is very ominous and it accounts for the second-most common mosquito-borne disease after malaria.[3] The most common presentation of dengue is fever with thrombocytopenia and its variants include dengue hemorrhagic fever and dengue shock syndrome being the grave entities.

Dengue fever leads to systemic protean manifestations. Though it is not a neurotropic virus, the involvement of neurological aspects is a well-known topic in the literature. Neurological outcomes range from neuropathy (long thoracic nerve palsy), radiculoneuropathy (Gullain Barre syndrome [GBS]), demyelination (acute disseminated encephalomyelitis [ADEM]), vascular issues like infarct or hemorrhage or even disastrous encephalitis. The neurological outcomes are attributed pathologically to the following mechanisms such as postinfectious immune-mediated damage which leads to GBS, ADEM, or peripheral neuritis; systemic complications leading to stroke, encephalopathy, or hypokalemic paresis; neurotropism resulting into meningitis, myelitis, myositis, encephalitis.[4]

Mononeuropathies in dengue fever have been documented in case reports, wherein paralytic squint in patient of DF during the critical phase of illness and LMN paresis of hypoglossal nerve involvement resulting in D5 of dengue fever has been reported by authors in India.[5,6]

Here, we present a case of dengue fever with expanded dengue syndrome due to neurological consequences with right LMN facial palsy. The case was initially an uncomplicated dengue fever, but later on, the unique manifestation twisted the scenario. So far as the literature is concerned, LMN facial palsy is associated with various microbes such as herpes zoster, cytomegalovirus, HIV, borrelia, and treponema, but flaviviridae such as dengue virus is not very commonly associated with Bell's palsy. Recent evidence suggest that subtle features of neurotropism is proven by dengue virus beaching blood–brain barrier, dengue viral particles in CSF.[7]

As isolated Bell's palsy is not so common in dengue, we had evaluated the patient with possibilities like brainstem demyelination, but MRI brain didn't show any lesion in the brainstem. There was no clinical and laboratory evidence that this episode was a post-TB meningitis manifestation. We managed this case conventionally with intensive fluid therapy; in view of concurrent facial palsy, oral corticosteroid

was initiated for a short duration. The use of a short course of corticosteroids in dengue fever complicated with facial nerve paralysis has been shown to be effective, as has been demonstrated in another case study from India.[8] Oral prednisolone was started at the dose of 1 mg/kg and tapered over subsequent weeks in a patient from Punjab,India who had developed LMN facial palsy on 4th day of dengue fever.[9]

The patient had recovered well. The clear temporal relationship with dengue infection establishes the relationship between facial nerve palsy and dengue fever in our patient.

CONCLUSION

This case is one of the very rare occurrences of Bell's palsy with dengue fever as described in literature previously. Thus, LMN facial palsy in endemic countries like India should be evaluated for dengue if associated with febrile illness.

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ETHICAL CONCERNS

Written informed consent was obtained from the patient prior to the case report drafting, for publication of treatment data and images of patient.

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RM- Case management, Final draft review. **DD-** Literature review, First draft preparation.

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