Unusual presentation of more common disease/injury

Bilateral facial palsy as a manifestation of Japanese encephalitis

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Correspondence to Professor Rajesh Verma, drrajeshverma32@yahoo.com**Summary**

Japanese encephalitis is a cause of substantial morbidity and mortality, prevalent mainly in South East nations. It is caused by group B arbovirus and transmitted with bite of infected culex mosquitoes. The clinical features described are: headache, vomiting, altered sensorium, convulsions and both hypokinetic and hyperkinetic movement disorders. In this submission, we described a 68-year-old man suffering from Japanese encephalitis, who presented with bilateral facial palsy with encephalitic features. The bilateral facial palsy has not been reported previously in association with Japanese encephalitis.

BACKGROUND

Japanese encephalitis has been emerged as significant health issue in India. Every year seasonal epidemic of Japanese encephalitis takes away many lives and caused significant morbidity.¹ Japanese encephalitis is caused by a group B arbovirus and transmitted through bite of infected culex mosquitoes. The disease is characterised by acute headache, vomiting, seizures, altered sensorium and movement disorders.² In this presentation, we described a patient of Japanese encephalitis, who had bilateral facial palsy with encephalitic manifestation. Bilateral facial palsy has not been reported previously and this case report may impress physicians to consider Japanese encephalitis in endemic zone, if a patient presents with bilateral facial palsy and encephalitic syndrome.

CASE PRESENTATION

A 68-year-old man presented with low-to-moderate-grade fever, headache, vomiting and bilateral facial palsy for previous 5 days. He had abnormal behaviour since 1 day prior to admission. The history was negative for seizures, arthralgia, rash, trauma, cough, chest complaints, radicular pain, glandular swellings and weight loss. The patient was non-smoker, non-diabetic and not had tuberculosis. The examination revealed drowsy state. The Glasgow coma scale was E4, V3 and M5. Lymphadenopathy was not present. The cranial nerves assessment showed bilateral lower motor neuron facial palsy. Motor system evaluation revealed generalised lead pipe rigidity. Meningeal and cerebellar signs were negative. The peripheral nerves were not thickened on examination.

INVESTIGATIONS

The haematological and biochemical parameters including liver function test, renal evaluation, serum electrolytes and laboratory assessment for thyroid functions did not reveal any abnormality. The study for falciparum malaria and typhoid fever was negative. The autoantibodies for anti-nuclear antibody, antineutrophilic cytoplasmic antibody

and antiphospholipid lipid antibodies were non-reactive. Serum ACE level was normal. Chest x-ray did not reveal hilar lymphadenopathy. The study for Lyme disease, leptospirosis and Wegner granulomatosis depicted negative results. EEG displayed generalised slowing. The sera for herpes simplex virus, Epstein-Barr virus, varicella zoster virus, cytomegalovirus, HIV and dengue virus revealed negative study. The cerebrospinal fluid (CSF) analysis showed 10 cells (all lymphocytes), proteins –51 mg% and sugar value of 68mg/dl. The sera as well as CSF revealed elevated immunoglobulin M titres for Japanese encephalitis with COMBO ELISA (Panbio, Australia) (Serum-22.27pbu: panbio units, CSF-44.94 panbio units, less than 9-negative). The CSF was negative for malignant cells. MRI, on T2 fluid attenuated inversion recovery axial image showed bilateral symmetrical hyperintense lesions in the pontine area (figure 1).

TREATMENT

The clinical evaluation and investigative workup suggested the diagnosis of Japanese encephalitis with predominant presentation of bilateral facial palsy. Our patient was treated conservatively with dopamine agonist (ropinirole—0.25 mg thrice a day and other supportive measures taking into account the presence of generalised rigidity.

OUTCOME AND FOLLOW-UP

In the following weeks, his facial palsy was completely improved. The clinical outcome was satisfactory and he regained activities of daily life after 3 months.

DISCUSSION

Our patient had bilateral facial palsy as the predominant manifestation of Japanese encephalitis, substantiated by bilateral pontine lesions on MRI. The manifestation of bilateral facial palsy is quite uncommon and often due to secondary cause as compared with unilateral facial palsy which is mainly idiopathic in nature. The incidence of

Figure 1 MRI, T2 fluid attenuated inversion recovery axial image demonstrated bilateral hyperintense lesions in pons (arrows).

bilateral facial palsy has been reported to be one per five million per year.³

The aetiology of facial palsy ranged from congenital, traumatic, infectious, metabolic, toxic, vascular, neoplastic and idiopathic category⁴ (box 1). The infectious aetiology of bilateral facial palsy commonly reported are infectious mononucleosis, HIV infection, Lyme disease, syphilis, brain stem encephalitis and human T-lymphotropic virus-1 infection.⁵

Recently, a case of bilateral facial palsy due to scrub typhus meningitis has been reported which occurred during convalescent phase of illness. The author emphasised to include typhus in the differential diagnosis of

bilateral facial palsy.⁶ Japanese encephalitis has never been reported to be the cause of bilateral facial palsy.

The bilateral facial palsy in our patient could be explained on the basis of bilateral facial nucleus involvement in pons. This is just equivalent to anterior horn cell involvement of spinal cord, well reported in Japanese encephalitis. The polio like illness, due to involvement of anterior horn cells described in literature are: rapidly evolving acute asymmetrical flaccid weakness, weakness starting from lower limbs, severe myalgia, reduced or absent deep tendon jerks, intact sensation and persistent weakness and atrophy even after 2 months.⁷

The isolated bilateral pontine facial nucleus involvement causing bilateral facial palsy is rarely reported in the literature. A case was described of bilateral facial palsy and ageusia due to bilateral nucleus lesions in pons associated with *Mycoplasma pneumoniae* pulmonary infection. The author tried to explain bilateral facial diplegia with autoimmune hypothesis.⁸

The classical neuroimaging features of Japanese encephalitis are T2 and T2 flair hyperintense signals in thalamus and midbrain, although signal changes have also been reported in cerebral cortex, brainstem and basal ganglia structures.⁹ In our patient, MRI of cranium revealed bilateral pontine lesions with positive serology for Japanese encephalitis.

Learning points

- ▶ Bilateral facial palsy is a rare clinical finding of varied aetiology.
- ▶ The bilateral pontine facial nucleus involvement causing facial diplegia is rarely observed.
- ▶ The bilateral facial palsy associated with Japanese encephalitis has not been previously reported in the literature.
- ▶ In the endemic zone of Japanese encephalitis, presentation of facial diplegia should raise suspicion of this lethal clinical disorder.

Competing interests None.

Patient consent Obtained.

Box 1 Causes of bilateral facial palsy

- A Upper Motor Neuron Type
 - ▶ Multi-infarct state
 - ▶ Motor neuron disease
- B Lower Motor Neuron Type
 - ▶ Infectious—HIV, leprosy, lyme disease, syphilis, poliomyelitis, infectious mononucleosis and brain stem encephalitis
 - ▶ Autoimmune—Guillain Barre syndrome, sarcoidosis, vasculitis and multiple sclerosis
 - ▶ Metabolic—diabetes mellitus, porphyria
 - ▶ Congenital—Mobius syndrome, Melkersson Rosenthal syndrome and osteopetrosis
 - ▶ Neoplastic—acute leukaemias, meningeal metastasis and bilateral neurofibromas
 - ▶ Idiopathic polyneuritis cranialis

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