Bilateral Intracerebral Haemorrhages: An Atypical Presentation of Japanese Encephalitis

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Abstract
Japanese encephalitis is common human endemic encephalitis seen over various parts of the world. Usual presenting features include an encephalitic syndrome, symptoms of frontal lobe, basal ganglia and thalamic involvement. Characteristic radiological picture is bilateral thalamic and basal ganglia hypo density in the CT scan and hypo-intensity in T1 and hyperintensity in T2 weighted image in MRI. Very rarely occurrence of bilateral hemorrhage may be seen in these regions. This radiological change may be early indicator of the disease before serological confirmation by the available diagnostic modalities. In this communication, we have reported a case of Japanese encephalitis presented with bilateral basal ganglia hemorrhages.

INTRODUCTION
Japanese encephalitis is a common human endemic encephalitis occurring in various parts of the world including India. Usual clinical presentation is an acute febrile illness associated with behavioral abnormalities, altered sensorium, convulsion, abnormal movements, meningeal sign and focal neurodeficits in various combinations. It has a high rate of mortality and morbidity accounting for 20-24% and 20-30% of affected persons respectively. Usually the cortex and the deeper gray matter such as, thalamus, basal ganglia and brainstem nuclei including anterior horn cells of cervical cord are affected. The common CT finding is bilateral hypodensity in the thalamus and basal ganglia with occasional hemorrhage, though bilateral hemorrhage is rare. MRI picture shows bilateral thalamic and basal ganglia hypointensity in T1-weighted images, which becomes hyper-intense in T2. Diagnosis of Japanese encephalitis depends on the clinical, imaging abnormalities and serological evidence of at least four-fold rise of antibody titer. As serological report may be delayed, early radiological evidence is very much helpful for suspecting JEV infection, especially in a setting of atypical presentation.

We are presenting a case of Japanese encephalitis presenting with acute onset unconsciousness and CT scan evidence of bilateral basal ganglia hemorrhage in which JEV encephalitis was suspected because of imaging abnormality.

CASE HISTORY
A young male aged 40 years from the district of Bankura in West Bengal, was admitted in neurosurgical ward in an altered sensorium with suspected head injury one day before admission. His CT scans of head showed bilateral basal ganglia hemorrhages (Fig. 1). The possibility of head injury was reasonably excluded after a careful neurosurgical evaluation and subsequently patient was referred to our neuromedicine unit for opinion. On reviewing, it was revealed that he had been suffering from low-grade fever, malaise and headache for 3 days preceding his ictus. But there was no history of abnormal behavior, involuntary movement or....
convulsion during that period. His recorded blood pressure within 30 min of ictus was 180/90 mm Hg. He was smoker but, non-alcoholic, non-diabetic and non-hypertensive. He had no significant past illness. On examination we found him drowsy and his blood pressure was 160/90 mm Hg. He had only mild external evidence of injury in the form of bruises over arm and chest. He was febrile (100°F). There was no neck rigidity or Kernig sign. Fundus examination was normal. Only neurodeficit was left sided complete hemiplegia. Within next 72 hrs he gradually became conscious with vacant expressionless look, hypophonia and there was hypokinesia on his right side with left hemiplegia. His blood pressure became normal. His routine blood examination including Hb, total count and differential count was normal. ESR was 14mm/1 st hr. Blood biochemistry including, sugar, urea, creatinine and LFT were normal and his serum for ANF, APLA and ANCA (p and c) were negative. His blood coagulation profile was within normal limits. ECG and ECHO-cardiography were normal. USG of abdomen was normal. MRI (Fig. 2) and MRA of brain had substantiated the CT findings and excluded any possibility of surgical cause for the hemorrhages. His CSF showed mild pleocytosis (385 / c mm. with 80 % polymorph), increased protein (109 mg/dl) and normal sugar (68 mg/dl). CSF for culture, AFB and fungal stains were negative. Blood culture ware also negative for consecutive three samples and no virus was isolated. Serum titre for anti-JEV antibody in 1st week was positive in 1: 20 and in 3rd week it was positive in 1:80. Serum was non-reactive for dengue and West Nile viral antibody as well. EEG showed diffuse theta rhythm. CT head after six weeks of onset showed complete resolution of hematomas. Patient made a rapid recovery and had started walking without support within 2 months of his initial illness. Presently he has mild emotional abnormality with clinical signs of mild emotional lability. He is independent in his activity of daily living.

**DISCUSSION**

Diagnosis of Japanese encephalitis in a patient of endemic region depends on clinical features and radiological changes in the thalamus and basal ganglia, supported by CSF abnormalities and increased titre of antiviral antibody in the convalescent serum. As the initial viremia is very short lasting, a positive viral culture is less likely and at the same time dependence on serological evidence of the JE unduly delays the diagnosis also. In these circumstances, radiological changes in CT/MRI of brain may be very much helpful as an important diagnostic criterion even in early stage of the JE and could guide physician in the right direction.

Patient was resident in the district of Bankura, West Bengal, an endemic zone of Japanese encephalitis. His illness started with sudden onset loss of consciousness with left hemiplegia in a setting of suspected head injury. However in nearly all cases of traumatic intracerebral hemorrhages, there are evidences of other extra or intracranial injury as well. Traumatic intra-cerebral hemorrhage (ICH) occurs usually at the tip of frontal and temporal poles because of closeness to bony parts. Traumatic basal ganglia hemorrhage is itself rare and simultaneous symmetrical, bilateral hemorrhages are also extremely uncommon.

Other causes for bilateral basal ganglia haemorrhage considered were stroke, methanol intoxication, coagulopathies, vasculitis and infection. Primary hypertensive basal ganglia haemorrhage is usually unilateral and occurs in a setting of long standing hypertension or previously undetected hypertension. Spontaneous multiple ICH can occur occasionally. In a series of 600 consecutive cases of ICH diagnosed by CT head, Weishberg found multiple hemorrhages in 2% of cases. Incidence of hypertension was unusually low (2 out of 12) in this series suggesting that the cause of multiple spontaneous ICH may frequently be due to other causative factors. Moreover, absence of past history of hypertension, lack of evidence of target organ involvement such as normal fundoscopy, normal ECG and echocardiography and subsequent conversion into normal blood pressure during hospitalization without any anti hypertensives spoke against the possibility of hypertensive ICH and transient hypertension noted after ictus is most likely reactionary.

Absence of past history of addiction, circumstantial evidences, lack of visual impairment and absence of
acidosis with stable haemodynamic status along with clinical findings together had ruled out the possibility of methanol intoxication as a cause of ICH.

In the presence of normal blood count including platelet count, normal result of coagulation profile and absence of any obvious cause of bleeding disorder, possibility of any coagulopathy or hematological disorder as a cause for this hemorrhage was less likely.

The course of the illness, normal ESR with negative serum ANF results and a normal MR angiogram of brain together made vasculitis less likely. History of preceding febrile illness, clinical setting as well as bilateral location of hemorrhage in the basal ganglia in this patient from endemic area of JE virus aided the possibility of infection in the form of Japanese encephalitis as a cause for bilateral basal ganglia hemorrhage. Other arboviral infections may cause similar type of illness. Of those, West Nile virus and dengue hemorrhagic fever are important. Absence of maculopapular rash, lymphadenopathy and hepatic involvement went against the possibilities of both the diseases. More over, absent bleeding diathesis, clinical shock, leukopenia and thrombocytopenia together made dengue unlikely. Serological study for viral antibodies by haemagglutination inhibition titer showed four-fold rise of antibody titre of JEV in convalescent serum and low titer for the dengue and West Nile virus, which together with clinicoangiological finding in a patient of endemic zone have supported the diagnosis of Japanese encephalitis.

Results of the studies by Kalita J et al\textsuperscript{2} have shown that the cause of hemorrhage in acute stage of encephalitis is due to increased blood flow in thalamus and basal ganglia. Therefore, it may be assumed that increased blood flow to the thalamus and basal ganglia secondary to inflammation could be the cause of hemorrhage.

In conclusion, CT scan and MRI of brain are very much helpful to suspect the possibility of Japanese encephalitis, especially in early course of the atypical presentation as it was in our case with bilateral hemorrhages in basal ganglia and indeed may give clue to the physician to go for viral study for JEV.

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REFERENCES


Erratum

The case report entitled "Polyglandular Autoimmune Endocrinopathy in Type 2 Diabetes" published in December 2004 (J Assoc Physician India 2004;52:999-1000) issue of JAPI should be read as “Polyglandular Autoimmune Endocrinopathy Type 2”

Sd/-
Hon. Editor