

Pregnancy, Japanese encephalitis and superior sagittal sinus thrombosis: coincidence or causal?

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Japanese encephalitis is a common central nervous system (CNS) infection and affects all group of population. It has been seen in pregnant women too. Pregnancy also predisposes to cerebral venous sinus thrombosis.^{4,7} However the combination of pregnancy, Japanese encephalitis and superior sagittal sinus thrombosis has not been described previously. The authors describe the diagnosis and management of a 19-year-old pregnant woman who had both Japanese encephalitis and SSS thrombosis successfully managed in our institution. We describe this unique case in detail and review the literature.

Japanese encephalitis is a common infectious condition in Nepal. Cerebral venous thrombosis is an uncommon clinical condition affecting persons with certain predisposing factors. But the simultaneous occurrence of these two conditions in extremely rare and to the best of our knowledge not described previously in the literature. In this paper we describe the clinical features and management of a 19-year old pregnant woman who suffered from Japanese encephalitis and superior sagittal sinus (SSS) thrombosis in our institution. In addition we review the literature.

Key Words: Japanese encephalitis, pregnancy, sagittal sinus thrombosis

Case Report

History

This 19-year-old previously healthy woman presented to us with two weeks' history of headache & fever, one-week history of altered sensorium, and four days' history of urinary incontinence. She was amenorrhic for 3 months, her previous cycles being regular. Rest of her history was unremarkable.

Examination

Upon arrival, she was irritable but conscious with normal vital signs. There was no nuchal rigidity. Motor examination revealed left sided hemiparesis with upgoing planter reflex bilaterally. Rest of her neurological and systemic examination was normal.

Laboratory and Imaging studies

Routine laboratory tests revealed leucocytosis with neutrophilia (total count 13,300/cubic millimeters; neutrophils 89%). Pelvic ultrasonography (USG) revealed a viable intrauterine fetus of 12 weeks gestation.

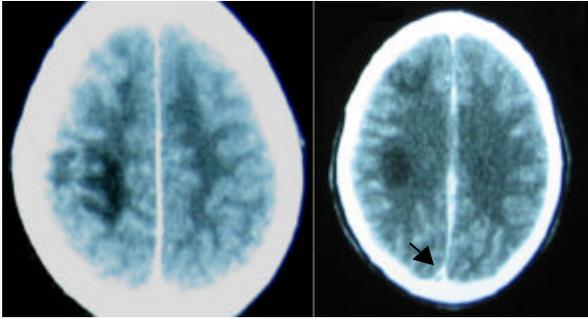


Figure 1. Noncontrast CT scan of head at admission demonstrating a lesion in the right parafalcine region suggestive of a venous infarction (left) and enhancement of adjacent meninges (right) giving rise to Delta sign (arrow).

Cerebrospinal fluid (CSF) obtained from lumbar puncture was as follows: Total count-200/high power field (hpf); neutrophils 80%, and lymphocytes 20; Sugar 2.3 millimols/liter (blood sugar 4millimols/liter) and protein 0.27gram/ liter. Culture of the CSF was sterile. Coagulation profile was within normal limits (though we were unable to perform the estimation of Protein C & S as the test was not available in our laboratory) Anti Japanese encephalitis antibody test was positive. Computerized tomography scan of the head (Figure 1) showed features suggestive of sagittal sinus thrombosis. EEG showed generalized slow wave, suggestive of encephalitis.

Patient was managed conservatively, and she recovered completely without any neurological deficits in 3weeks.

Discussion

Japanese Encephalitis virus, a flavi virus is found throughout Asia including far eastern Russia, Japan, China, India, Pakistan and South East Asia spreading upto Malaysia and Australia. In Nepal it is one of the leading causes of encephalitis spreading in 26 districts of Terai and inner Terai.⁹ Most of the infections are asymptomatic and among the people who develop a clinical illness, the case fatality rate may be as high as 30%.⁹ Japanese Encephalitis usually starts as a flu like illness, 6 to 8 days following bite of a mosquito (incubation Period 5-15 days). The illness is characterized by prodrome of non-specific constitutional symptoms. This may be rapidly followed by variety of neurological signs and symptoms including focal deficits and convulsions. The acute illness usually lasts from few days to as long as 2-3weeks and recovery may take weeks to months.

Diagnosis is made on the basis of clinical features, CSF analysis (which usually shows lymphocytic pleocytosis) and demonstration of Anti JE IgM antibody in CSF or serum. Treatment of this condition is essentially supportive, as no effective antiviral therapy has been found till date.⁹ There are limited studies of Japanese Encephalitis in pregnant women. Chaturvedi et al. reported 5 cases of Japanese

Encephalitis in pregnant women and have concluded that JE virus infection in pregnancy is capable of producing transplacental infection and fetal wastage.³ However, whether JE virus causes fetal malformation was undetermined.³ In our case, USG study showed no abnormalities in fetus.

Thrombosis of cerebral venous sinuses usually develops in relation to infection of the ear, paranasal sinuses, meninges or to various hypercoagulable states like cancer, cyanotic congenital heart diseases, sickle cell disease, antiphospholipid antibody syndrome, protein C or S deficiency, antithrombin III deficiency, polycythemia, thrombocytopenia etc. It also occurs in certain settings such as consumption of birth control pills, pregnancy, postpartum and postoperative states which are often characterized by thrombocytosis and hyperfibrinogenemia. Occlusion of cortical vein that are tributaries of dural sinus take the form of venous infarctive stroke. Thus a patient with stroke and above mentioned conditions should have a suspicion for cerebral venous thrombosis.^{1,7} Thrombosis of superior sagittal sinus presents as headache, nausea, vomiting, confusion and focal or generalized seizures. There may be rapid development of stupor and coma. Weakness of lower extremities with bilateral Babinski sign or hemiparesis is often present. If sagittal sinus thrombosis occurs as a complication of bacterial meningitis, meningeal signs may be present. The diagnosis of venous sinus thrombosis is suggested by an absent flow void within the affected venous sinus on MRI and confirmed by MR venography or venous phase of cerebral angiography.² CT scan shows “empty triangle” so called Delta sign, secondary to enhancement of meninges around the area of thrombus.¹⁰ Anticoagulant therapy beginning with heparin for several days, followed by warfarin, combined with antibiotics if the venous occlusion.

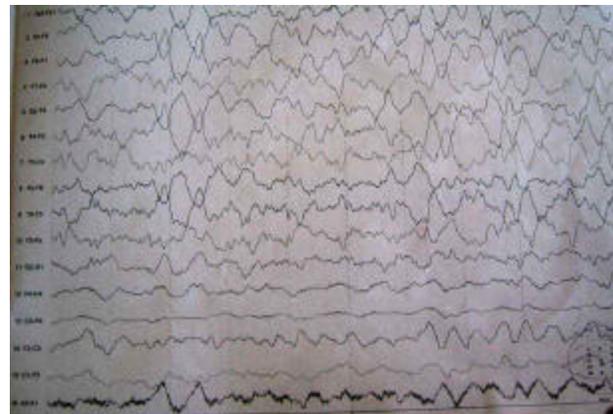


Figure 2. Awake EEG tracing of the patient, showing generalized slow wave suggestive of encephalitis.

is inflammatory, has been life saving in some cases but overall mortality rate remains high at 10 to 20%, in part due to large hemorrhagic venous infarctions.¹

There are many studies and case reports of cerebral venous sinus thrombosis in pregnancy and postpartum states. Gokcil et al. have reported a woman with 28 weeks pregnancy who develop superior sagittal sinus thrombosis associated with decreased free protein S level.⁶ Similarly Eon et al. have reported a case of cerebral venous thrombosis associated with protein C deficiency in a 25 year old woman with six months pregnancy.⁵ However in our set up protein C and protein S level assay were not available. McDonnell et al. have reported a case of sagittal sinus thrombosis occurring even during an ectopic pregnancy.⁸

Conclusions

A patient with Japanese Encephalitis can present with diversity of neurological signs and symptoms, as it can be true with a case of cerebral venous sinus thrombosis. If we come across a patient with CNS infection with neurological deficit along with hypercoagulable states, a possibility of cerebral venous sinus thrombosis should be also kept in mind.

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