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A Rare Case of ADEM after Japanese Encephalitis

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Abstract: Acute disseminated encephalomyelitis (ADEM) is a monophasic demyelinating disease of central nervous system (CNS) which is most frequently associated with an antecedent infection (identified in ~ 50-77%). 5% of ADEM cases follow immunization. Post infectious autoimmune events associated with Japanese encephalitis (JE) have been limited to Guillian Barre Syndrome (GBS) and JE virus vaccine related ADEM. We hereby report a case of 18 year boy who presented to us with fever, urinary retention, bilateral diminution of vision and acute onset paraparesis. Japanese encephalitis was diagnosed by elevated IgM titres against JE virus in cerebrospinal fluid (CSF). ADEM was confirmed by MRI brain and spinal cord. Our patient also developed bilateral eye optic neuritis presenting clinically as sudden onset blurring of vision in both eye one day after admission and confirmed by visual evoked potential (VEP) study. His symptoms improved after giving high dose intravenous methylprednisolone.

Keywords: ADEM; Japanese Encephalitis.

1. Introduction

Acute disseminated encephalomyelitis (ADEM) is an acute monophasic demyelinating disorder in CNS, typically occurring after infection or immunization [1, 2]. It occurs most commonly after measles. Other antecedent infections include varicella, mumps, parainfluenza, herpes simplex, rubella, influenza, EBV, HIV and mycoplasma pneumonia [1]. Children are predominantly affected. The clinical diagnosis of ADEM is suggested by a close temporal relationship between an infection or an immunization and clinical presentation with multifocal, subcortical white matter abnormalities on an MRI brain. The CSF shows modestly elevated protein (50-150 mg/dl), lymphocytic pleocytosis and elevation of albumin levels.

Japanese encephalitis virus (JEV) is a single stranded positive sense ribonucleic acid (RNA) belonging to family Flaviviridae, transmitted by mosquito Culex and has an incubation period of 5 to 15 days. It is prevalent in South-East Asia [3]. It has 5 genotypes. Genotype III is most common in India. Most patient are asymptomatic with 1 in 250 developing severe disease (encephalitis) [4]. The course of disease is divided into 3 stages: a prodromal stage characterized by fever, myalgia, vomiting, diarrhea, an encephalitic stage and a late stage characterized by recovery or persistence of signs of CNS injury. Thalamic involvement on MRI is highly suggestive of Japanese encephalitis [5]. Some atypical presentation of JE have been reported which include acute transverse myelitis [6]. Post-infectious autoimmune events associated with Japanese encephalitis have been limited to case reports of JEV vaccine associated ADEM [7]. We herein report a possible case of ADEM after Japanese encephalitis.

2. Case Report

A 18 year old boy, resident of Hyderabad, presented to us with fever (acute in onset, mild to moderate grade) for one week, followed by urinary retention and constipation 3 days after onset of fever, followed by bilateral weakness of lower limbs such that patient was not able to stand from sitting position and not able to walk. Patient did not complaint of any sensory loss, alteration in behavior, seizures. On examination, patient was irritable and drowsy. There were decreased power in bilateral lower limb with decreased sensation to touch, pain, temperature. Plantar response was flexor bilaterally and tone of the muscle, deep tendon reflexes were normal. On next day of admission, patient developed blurring of vision in both eyes. On further eye examination only perception of light (PL) in left eye and finger counting at 1 meter was present in right eye. No significant past and family history of similar episodes was noted. All routine investigations were within normal limits. Cerebrospinal fluid (CSF) examination showed lymphocytic pleocytosis with total leucocyte count (TLC) of 50 cells/mm³. MRI spine showed T2 hyperintense signal in the entire cervical spinal cord and upper dorsal cord sparing the periphery of spinal cord suggestive of longitudinal extensive transverse myelitis (Figure 1). MRI brain was subsequently done to differentiate multiple sclerosis (MS), neuromyelitis optica (NMO) and ADEM which showed T2 hyperintensity in bilateral thalami (Figure 2) along with bilateral internal capsule, centrum semiovale and brachium pontis suggestive of Japanese encephalitis.

Figure-1. Sagittal T2- weighted magnetic resonance image (MRI) showing abnormal T2 hyperintense signal intensities in the central region of the cord parenchyma (white arrowhead) from D5 to D12.

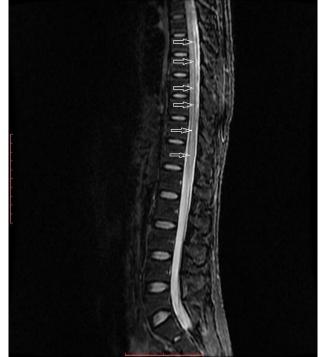
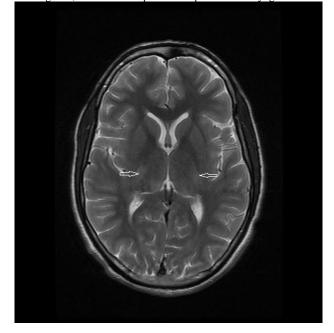


Figure-2. Axial fast spin echo (FSE) MRI showing subtle hyperintensities in bilateral thalamus(white arrowhead). Visual evoked potential showed delayed latencies. On further investigation, CSF showed Japanese encephalitis virus by IgM ELISA. CSF NMO antibodies were negative.



Therefore, ADEM was suspected and high dose methylprednisolone (1g/day x 5 days) was initiated. Two days after starting of therapy, patient had regained the vision completely with gradual improvement in weakness of lower limbs. The patient was discharged on oral prednisolone.

3. Discussion

In our patient, diagnosis was made on the basis of clinical symptoms, MRI findings, CSF report. There is generally no accepted criteria available for diagnosis of ADEM [8]. Schwartz's concept was used in most studies for adult patients. This concept describes ADEM as the first episode of neurological dysfunction with the evidence of demyelination on brain MRI after virus infection or vaccination without emphasis on the consciousness disturbance [9]. There is no definite criteria for MR diagnosis of ADEM. Previous studies showed ADEM usually presented as widespread, bilateral, asymmetric white matter lesions, large confluent demyelination lesion or even multiple small scattered lesions [10]. In our patient subcortical white matter showed hyperintensities on FLAIR and T2WI. There is no widely accepted rule for the latent periods between infection and onset of ADEM. In contrast, a maximal period of 3 months between vaccination and onset of ADEM is suggested. In our patient, the latent period was of few days (3-4 days) [1].

The possible mechanism of Japanese encephalitis-induced ADEM has been explained in an animal study, in which severe inflammation with numerous demyelinating axons, elevated anti-myelin basic protein (MBP) antibody titre, and proliferation of MBP-specific T-lymphocytes were found in a mouse model of Japanese encephalitis [11]. The anti-MBP may cause severe CNS inflammation with subsequent demyelination [1].

JE is diagnosed by detection of antibodies in serum and CSF by IgM capture ELISA [12]. There is no specific treatment for Japanese encephalitis and treatment is mainly supportive. High dose steroid therapy is recommended to treat ADEM patients according to previous review article and 50-70% ADEM patients have full recovery after treatment. We started our patient on high dose of methylprednisolone for 5 days followed by oral steroid which resulted in improvement of symptoms and signs.

4. Conclusion

ADEM is one of the rare presentations of JE and should be considered in patients from endemic area. Early introduction of steroid fastens recovery and diminish the neurological sequelae.

References

- [1] Menge, T., Hemmer, B., and Nessler, S., 2005. "Acute disseminated encephalomyelitis: An update." *Arch Neurol*, vol. 62, pp. 1673-1680.
- [2] Tenembaum, S., Chitnis, T., and Ness, J., 2007. "Acute disseminated encephalomyelitis." *Neurology*, vol. 68, pp. 23-36.
- [3] Umenai, T., Krzysko, R., Bektimirov, T. A., and Assaad, F. A., 1985. "Japanese encephalitis: current worldwide status." *Bulletin of the World Health Organization*, vol. 63, pp. 625–631.
- [4] Mathur, A., Arora, K. L., Rawat, S., and Chaturvedi, U. C., 1986. "Persistence, latency and reactivation of Japanese encephalitis virus infection in mice." *Journal of General Virology*, vol. 67, pp. 381–385.
- [5] Kalita, J. and Misra, U. K., 2000. "Comparison of ct scan and mri findings in the diagnosis of japanese encephalitis." *J. Neurol Sci.*, vol. 174, pp. 3-8.
- [6] Dickerson, R. B., Newton, J. R., and Hansen, J. E., 1952. "Diagnosis and immediate prognosis of Japanese B encephalitis. Observations based on more than 200 patients with detailed analysis of 65 serologically confirmed cases." *The American Journal of Medicine*, vol. 12, pp. 277–288.
- [7] Plesner, A. M., Arlien-Soborg, P., and Herning, M., 1998. "Neurological complications to vaccination against Japanese encephalitis." *Eur. J.Nurol.*, vol. 5, pp. 479-485.
- [8] Liao, M. F., Huang, C. C., and Lyu, R. K., 2011. "Acute disseminated encephalomyelitis that meets modified mcdonald criteria for dissemination in space is associated with a high probability of conversion to multiple sclerosis in taiwanese patients." *Eur. J. Neurol*, vol. 18, pp. 252-259.
- [9] Schwarz, S., Mohr, A., and Knauth, M., 2001. "Acute disseminated encephalomyelitis: A follow-up study of 40 adult patients." *Neurology*, vol. 56, pp. 1313-1318.
- [10] Singh, S., Alexander, M., and Korah, I. P., 1999. "Acute disseminated encephalomyelitis: Mr imaging features." *AJR Am J Roentgenol*, vol. 173, pp. 1101-1107.
- [11] Tseng, Y. F., Wang, C. C., Liao, S. K., Chuang, C., and Chen, W. J., 2011. "Autoimmunity-related demyelination in infection by japanese encephalitis virus." *J. Biomed Sci.*, vol. 18, p. 20.
- [12] Shrivastva, N., Tripathi, M., and Parida, 2008. "Comparison of a dipstick enzyme-linked immunosorbent assay with commercial assays for detection of Japanese encephalitis virus-specific IgM antibodies." *Journal of Postgraduate Medicine*, vol. 54, pp. 181–185.