Clinical Case Report

Medicine®

OPEN

Case report: acute demyelinating encephalomyelitis following viper bite

Anyi Xu, MD[a], Renfei Shan, MD[a], Daochao Huang, MD[a], Jiajia Zhou, NP[a], Anaswasseem Keenoo, MD[b], Jie Qin, MD[a,*]

Abstract

The most serious complications of the central nervous system that occur after venomous snake bite are intracranial hemorrhage and ischemic stroke.

We present a rarely seen central nervous system complication, acute demyelinating encephalomyelitis, after a treated Deinagkistrodon's viper bite.

On April 5, 2015, a 50-year-old male farmer was bitten on his right leg by a Deinagkistrodon's viper. The bite rendered the victim unconscious for 14 days, during which he was treated with tetanus toxoid and polyvalent antisnake venom. Acute demyelinating encephalomyelitis (ADEM) was suspected after magnetic resonance imaging of the brain. After a high dose of methylprednisolone was used as diagnostic treatment, the patient started recovering fast.

ADEM is a rare complication after snake bite, and is triggered by venom or antivenin. Magnetic resonance imaging helps in the early diagnosis of ADEM, and high-dose corticosteroid therapy appears to be effective in ADEM after viper bite or antivenin management.

Abbreviations: ADEM = acute disseminated encephalomyelitis, CNS = central nervous system, CSF = cerebral spinal fluid, MRI = magnetic resonance imaging.

Keywords: antivenin, demyelinating encephalopathy, neuropathy, viper bite

1. Introduction

Acute disseminated encephalomyelitis (ADEM) is an autoimmune-mediated nervous system disease, which occurs mainly following animal or insect bites. However, ADEM after a snake bite has rarely been reported.[1,2] Here, we present a rarely seen central nervous system (CNS) complication, acute demyelinating encephalomyelitis, after a treated Deinagkistrodon's viper bite. The patient recovered after a high dose of methylprednisolone therapy. We discuss the role of magnetic resonance imaging (MRI) and methylprednisolone in the diagnosis and treatment of this rare complication. We obtained the patient's written informed consent for the procedure and subsequent use of data.

2. Case report

A previously healthy 50-year-old man presented to our emergency department after being bitten by a Deinagkistrodon viper on April 5, 2015. He described severe pain and significant swelling on his right leg. One hour later, he was transported to a local hospital and given broad-spectrum antibiotics (amoxycillin sodium and clavulanate potassium 1.2 g intravenously), administered 3 times daily. He was also administered tetanus toxoid, delivered subcutaneously by injection (0.5 mL), and polyvalent antivenin therapy immediately. He received polyvalent antisnake venom (manufactured by Shailun Bio-pharmaceuticals Company, Shanghai, China) with a dose of one vial (6000 units).

Five hours after the treatment, the patient was unconsciousness and showed no response to external stimulation. Emergency endotracheal intubation was performed to maintain an open airway and mechanical ventilation. Meanwhile, the patient was treated with anti-infective and other supportive treatment. Thirteen days later, the patient was transported to our department, still unconscious, and had not responded to treatment.

The patient's medical history was unremarkable. On examination, he was febrile with a temperature of 38.5°C, with normal blood pressure and pulse. There was no skin rash and no lymphadenectasis, but he had mild residual swelling at the bite site. The Glasgow Coma Scale score was 6.

Laboratory findings: blood culture was negative. The patient's blood glucose was normal. Peripheral blood examinations: red blood cell 3.4×10^{12}/L, white blood cell 16.4×10^{9}/L, neutrophilic granulocyte 88.8%, lymphocyte 11.6%, hemoglobin 74 g/L, platelet count 5×10^{9}/L. Renal function: serum creatinine was 211 umol/L, blood urea nitrogen 33.7 mmol/L. Liver function: alanine aminotransferase 105 U/L, aspartate aminotransferase 47 U/L. His erythrocyte sedimentation rate was 98 mm/h and his C-reactive protein was 45 mg/L. Cytology and tuberculosis examination of the cerebral spinal fluid (CSF) were normal.

MRI of the brain was used to confirm whether CNS complications had resulted in long-term unconsciousness. The examination demonstrated multiple nonspecific foci of abnormal signal intensity in the right cerebral hemisphere, basal ganglia,

Editor: Abdelouahab Bellou.

The authors have no funding and conflicts of interest to disclose. Informed consent was given by the patient.

[a] Department of Emergency Medicine, Taizhou Hospital of Zhejiang Province, Wenzhou Medical University, [b] Wenzhou Medical University, Linhai, China.

[*] Correspondence: Jie Qin, Department of Emergency Medicine, Taizhou Hospital of Zhejiang Province, Wenzhou Medical University. No. 150 Ximen Street, Linhai, China (e-mail: QINJIE@enzemed.com).

Medicine (2016) 95:45(e5310)

Received: 6 July 2016 / Received in final form: 9 October 2016 / Accepted: 11 October 2016

http://dx.doi.org/10.1097/MD.0000000000005310

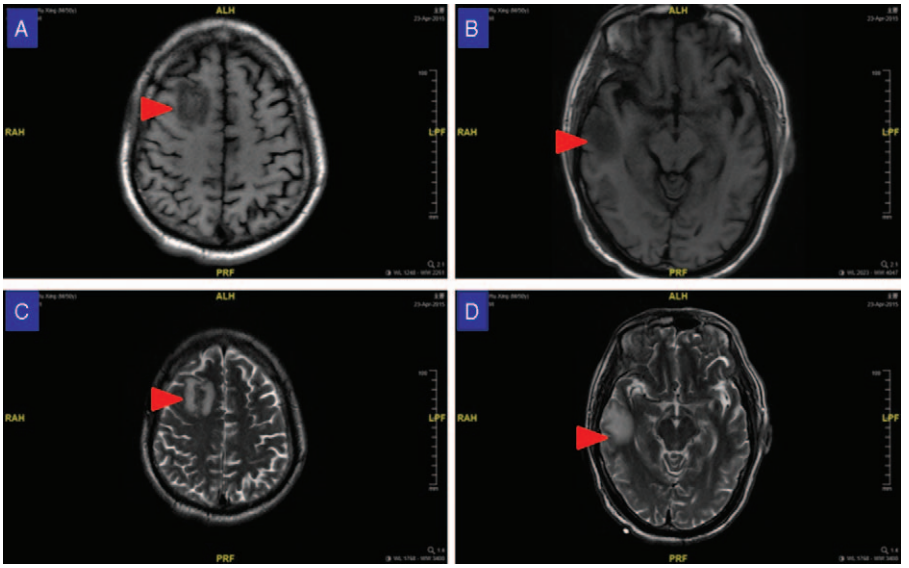

Figure 1. Axial T1-weighted MR image showing decreased signal intensity (A, B). T2-weighted MR image showing increased signal intensity in the lesions. MR = magnetic resonance.

and other small lesions. A T1-weighted MRI showed decreased signal intensity, while a T2-weighted MRI showed increased signal intensity in the lesions (Fig. 1).

Initially, methylprednisolone (500 mg/d) was used as a diagnostic treatment. The patient's body temperature returned to normal within 3 days. Then, prednisone (40 mg/d) was administered for 10 days. A physical examination showed that the patient's Glasgow Coma Scale score was 10. Meanwhile, the patient was given antibiotics, a blood transfusion, protection of liver and kidney function, supportive nutrition, and right leg debridement and dressing to ameliorate his health. An MRI 14 days after admission showed that the space-occupying lesions had significantly reduced, compared with the previous images (Fig. 2).

The final diagnosis was demyelinating encephalopathy. The patient recovered fairly well and was discharged from our hospital. He was advised to take prednisolone (50 mg/d) orally and reduce the doses gradually (minus 2 times a week, each 5 mg). Follow-up 1 month later showed that the lesions revealed by the MRI had almost disappeared (Fig. 3). The patient was able to take full care of himself and participate in house chores.

3. Discussion

Snake bite is a neglected public health issue that affects millions of people worldwide each year. Depending on the species of the offending snake, the clinical manifestations may be different, but include swelling and necrosis in the bitten limb or neurological

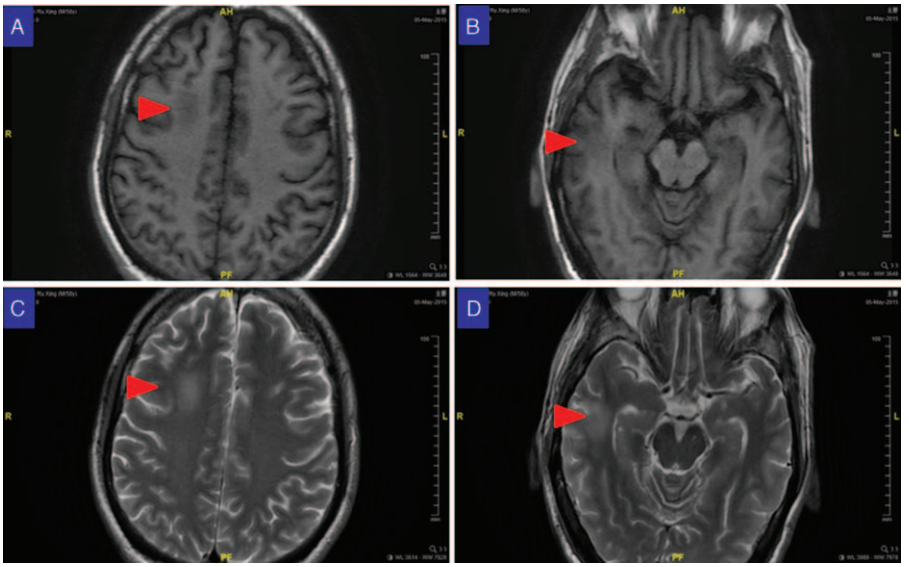

Figure 2. Axial T1 (A, B) and Axial T2 (C,D) weighted MR images on the 14th day after administration of methylprednisolone: the lesions are significantly smaller compared with Fig. 1. MR = magnetic resonance.

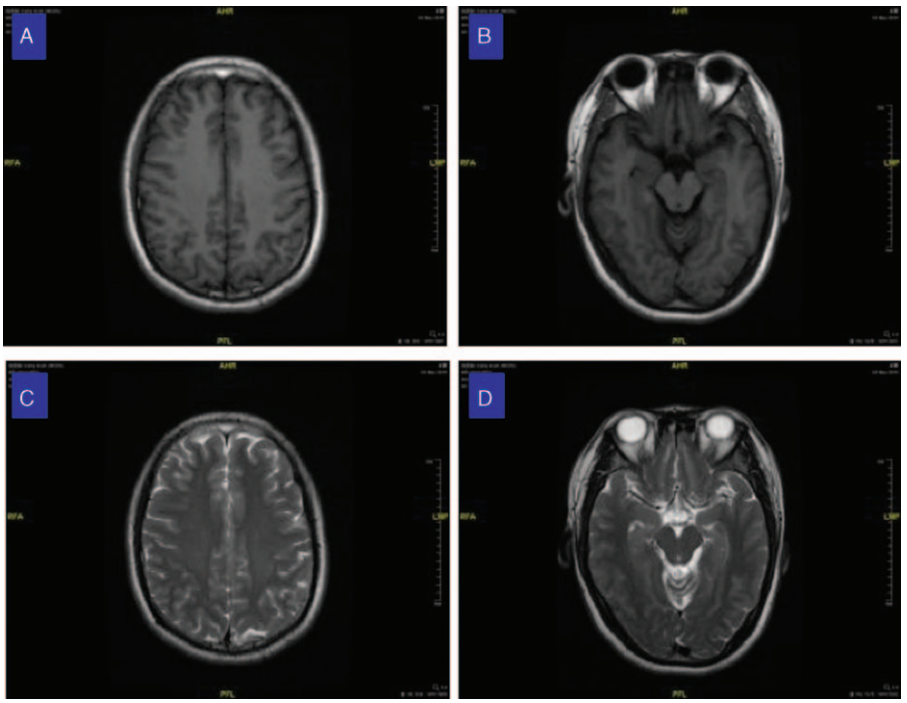

Figure 3. Axial T1 (A, B) and Axial T2 (C, D) weighted MR images 1 month after discharge showing that the lesions have disappeared. MR = magnetic resonance.

symptoms.[1] The most common serious CNS complications following venomous snake bite are intracranial hemorrhage and ischemic stroke. Some patients also experience CNS complications such as delayed cerebellar ataxia or ADEM after a snake bite. These manifestations may relate to the immune-mediated damage triggered by venom or antivenin.

ADEM is an autoimmune demyelinating disease of the nervous system, which often follows viral or bacterial infections, or less often, vaccination for measles, mumps, or rubella.[3] It has rarely been reported in the literature following animal or insect bites, and there is only 1 recorded case that was caused by snake bite. ADEM can be suspected based on clinical features, laboratory features, and magnetic resonance imaging, which is the imaging modality of choice to demonstrate white matter lesion. Among the treatment methods used for ADEM are supportive care, high-dose methylprednisolone, immunomodulation, plasmapheresis, and physical and rehabilitation therapy. More than half of ADEM patients can achieve full recovery after therapy.

In the case of our patient, MRI revealed multiple abnormal signals in both cortical hemispheres, the basal ganglia and the centrum semiovale, and presented several space-occupying lesions. Since the patient did not have any history of CNS disorders, we considered the findings to be more likely due to inflammatory and autoimmune origins than tumor. However, cytology and a tuberculosis examination of the CSF did not show evidence of infectious diseases. After the exclusion of other possible causes of coma, we considered demyelinating encephalopathy. Moreover, the patient's sensitivity to glucocorticoids also supported our diagnosis.

The relationship between cerebral damage and viper bite is unclear based on current information.[4–7] Some researchers have hypothesized that injury to the nerve endings or demyelination may be related to an immune reaction to the venom or antivenin. Since the patient in our case had no history of diseases involving the central nervous system, such as multiple sclerosis, the occurrence and turnover of the nervous system were concomitant with the occurrence of a viper bite. Following snake bite, some patients develop delayed neurological symptoms.

The vast variety of symptoms often makes the diagnosis of ADEM difficult. It is easy to be misdiagnosed as an intracranial infection or tumor. We should pay more attention to the history and take account of the possibility of central nervous system damage caused by systemic diseases. The brain MRI and sensitivity to glucocorticoids helped us to make a provisional diagnosis of ADEM, but eventually biopsy may be needed for definitive diagnosis.

4. Conclusion

ADEM is a rare complication after snake bite, and is triggered by venom or antivenin. MRI helps in the early diagnosis of ADEM, and high-dose corticosteroid therapy appears to be effective in ADEM after viper bite or antivenin management.

References

[1] Kerrigan KR. Venomous snakebite in eastern Ecuador. Am J Trop Med Hyg 1991;44:93–9.
[2] Gawarammana I, Mendis S, Jeganathan K. Acute ischemic strokes due to bites by Daboia russelli in Sri Lanka-First authenticated case series. Toxicon 2009;54:421–8.
[3] Höllinger P1, Sturzenegger M, Mathis J, et al. Acute disseminated encephalomyelitis in adults: a reappraisal of clinical, CSF, EEG, and MRI findings. J Neurol 2002;249:320–9.
[4] Chroni E, Papapetropoulus S, Argyriou AA, et al. A case of fatal progressive neuropathy. Delayed consequence of multiple bites of a non-venomous snake? Clin Neurol Neurosurg 2005;108:45–7.
[5] Chuang TY, Lin SW, Chan RC. Guillaı'n-Barre' syndrome: an unusual complication after snake bite. Arch Phys Med Rehabil 1996;77:729–31.
[6] Ekenback K, Hulting J, Persson H, et al. Unusual neurological symptoms in a case of severe crotalid envenomation. Clin Toxicol 1985;23:357–64.
[7] Srivastava A, Taly AB, Gupta A, et al. Guillaı'n-Barre' syndrome following snake bite. Ann Indian Acad Neurol 2010;13:67–8.