Int J Clin Exp Med 2018;11(5):5222-5226
www.ijcem.com /ISSN:1940-5901/IJCEM0068748

Case Report

Transient spinal cord involvement in reversible splenial lesion syndrome: a case report

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Received November 8, 2017; Accepted April 2, 2018; Epub May 15, 2018; Published May 30, 2018

Abstract: Reversible splenial lesion syndrome (RESLES) has largely been described in encephalitis/encephalopathy. It is typically characterized by transient focal reversible symmetrical lesions, and show T2 hyperintensity with corresponding diffusion restriction and no contrast enhancement in the splenium of the corpus callosum (SCC) on magnetic resonance image (MRI), which can resolve completely or near completely within days or months. Previous cases have revealed the lesions could extend to other areas, such as the basal ganglia region, periventricular white matter, cerebral hemisphere, thalamus, cerebellum, the limbic system and brainstem. In the present case, transient lesions from the 1st to the 10th thoracic spinal cord were first reported in the syndrome in a patient with suspected tuberculous meningitis.

Keywords: Reversible splenial lesion syndrome, splenium of the corpus callosum, reversible, spinal cord

Introduction

Reversible splenial lesion syndrome (RESLES) is a clinical-radiological syndrome, which was first proposed by Tada et al. [1] in 2004 and further expanded by Takanashi et al. [2]. The syndrome has been described to be most commonly associated with seizures, antiepileptic medications and their withdrawal, and systemic and central nervous system infections [3]. It is typically characterized by transient symmetrical focal lesions in the splenium of the corpus callosum (SCC) on magnetic resonance image (MRI), which can resolve completely or near completely within days or months. It has reported that there are RESLES and extended RESLES [4]. This means that other than the SCC, these lesions may extend to other areas such as the basal ganglia region, periventricular white matter, cerebral hemisphere, thalamus, cerebellum, the limbic system and brainstem [5]. The exact mechanism of RESLES remains unknown. In the present case, transient lesion of the spinal cord was first reported in a 14-year-old boy who suffered from suspected tuberculous meningitis. It is possible that more and more new areas that involve this syndrome will be reported in the future.

Case report

A 14-year-old boy presented to our hospital with sudden onset of headache, fever, nausea and vomiting after catching a cold. The patient had no focal neurological symptoms except for nuchal rigidity. MRI scan was normal on T1-weighted images (T1WI) and T2-weighted images (T2WI), while contrast-enhanced MRI scan revealed linear enhancement of the meningeal. Lumbar puncture was performed, which revealed a mildly elevated initial pressure of cerebrospinal fluid (CSF) and level of protein (**Figure 1**), while the acid-fast stain of CSF was positive. Therefore, the patient was diagnosed as suspected tuberculous meningitis, and was given anti-tuberculous treatment. However, his clinical condition worsened five days after hospitalization, and he became drowsy and suffered from a visual disturbance, as well as sensory-motor disorder in his legs. On follow-up MRI of the brain, T2WI and diffusion weight image (DWI) revealed a hyperintense lesion in the SCC with a corresponding hypointensity on apparent diffusion coefficient (ADC) values. Meanwhile, there was a hyperintense signal from the 1st to the 10th thoracic spinal cord on the T2WI image (**Figure 2**). Diffusion tensor

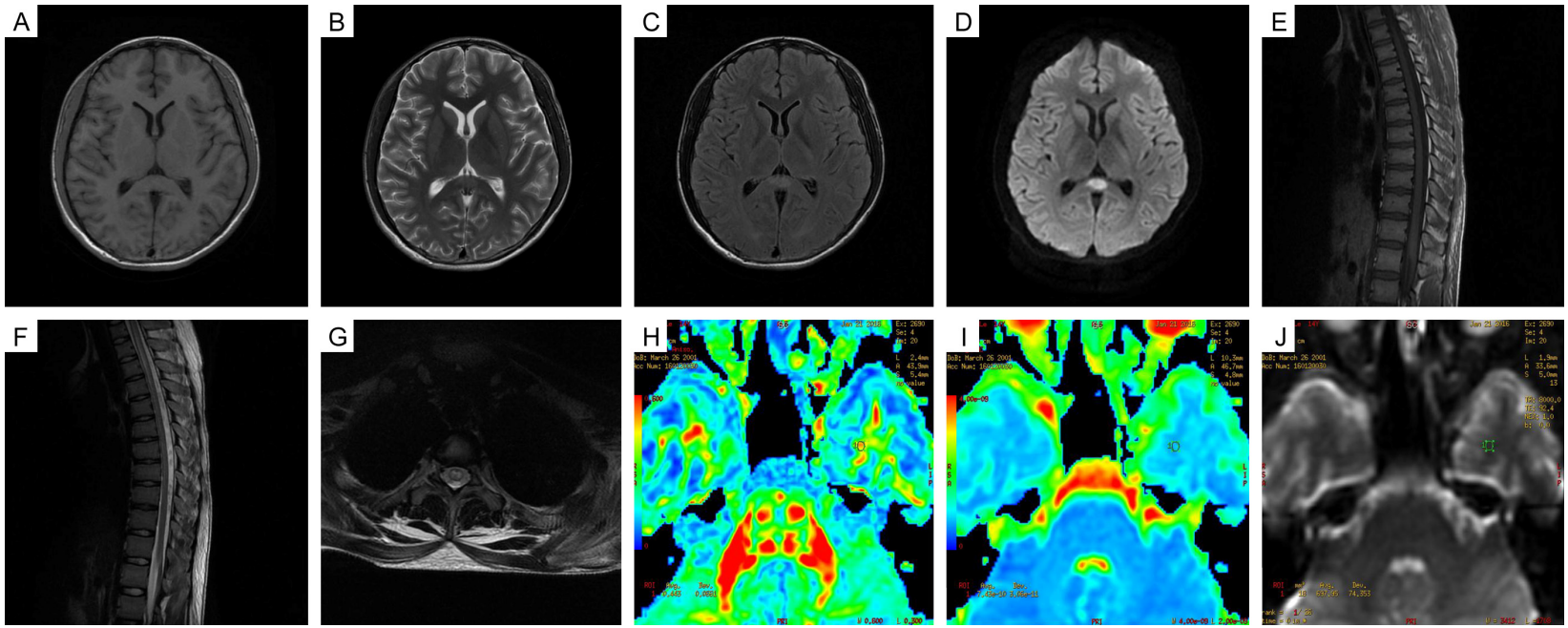

Figure 1. Magnetic Resonance Imaging Scans (3.0T) after his clinical conditions worsened. The lesions of SCC (A-D) and spinal cord (E-G). The DTI showed reduced FA in the SCC (H-J).

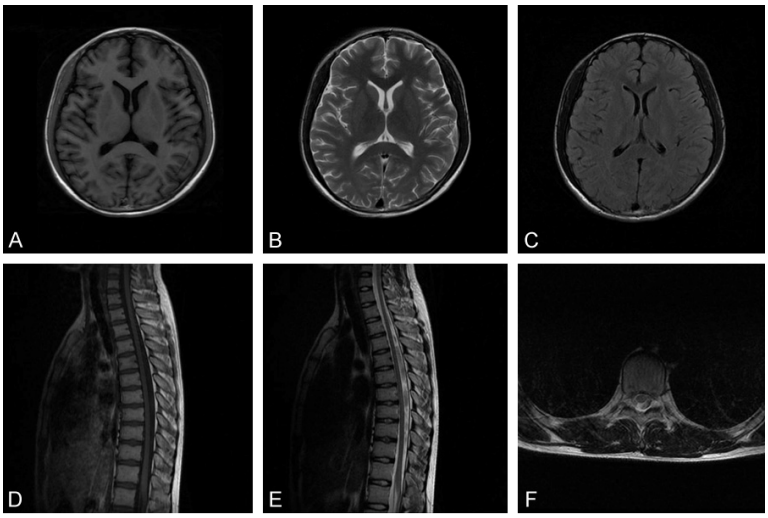

Figure 2. Follow-up MRI of brain (A-C) and spinal cord (D-F) showed radiological conditions were recovered completely.

imaging (DTI) revealed a reduced fractional anisotropy (FA) value in the SCC. Hence, the patient was successively given intravenous immunoglobulin and steroid pulse therapy for two months. The lesions in the spinal cord and SCC totally vanished at 50 days after hospitalization. His clinical conditions were nearly completely recovered (**Figure 3**) except for elevated protein in CSF (**Figure 1**).

Discussion

The patient in the present study revealed a nearly complete clinical and radiological recovery, which supported the diagnosis of RESLES. RESLES is an encephalopathy syndrome involved in the SCC, which was first reported by Tada et al. in 2004. It is characterized by transient symmetrical lesions in the SCC on MRI, which can resolve completely or near completely within days or months. The possible differential diagnoses of RESLES include reversible posterior leukoencephalopathy syndrome, multiple sclerosis, and acute disseminated encephalomyelitis (ADEM). ADEM should be kept as the first differential diagnosis. In ADEM, MRI revealed multiple lesions at T1 and T2 signals in the subcortical white matter, which were typically bilateral, but asymmetrical. Meanwhile the lesions in ADEM would show a variable contrast enhancement different from RESLES [6].

The exact pathophysiology of the splenial lesion is obscure. It has been reported to be associated with seizures, antiepileptic medications and their withdrawal, systemic and central nervous system infections, etc. [3] MRI abnormalities, which are transient and reversible, are characterized by low signals on T1WI, high signals on T2WI, FLAIR and diffuse weighted image, reduced apparent diffusion coefficient and FA values on DTI, and no contrast enhancement [7]. In addition to the SCC, lesions could also extend to other areas. However, the involvement of the spinal cord has not been previously reported in RESLES, as in the present case. We can regard this as extended RESLES. RESLES is associated with a variety of disorders, including blood-brain barrier breakdown, intramyelinic edema due to inflammation and migration of inflammatory cells, extrapontine osmotic myelinolysis due to sodium and glucose imbalance, direct viral invasion, vitamin deficiency, the selective vulnerability of such lesions, and genetic predisposition; and these may explain the MRI features in RESLES patients [8-10]. Why does RESLES selectively occur in the splenium? The vascular supply of splenium differs from other parts of the corpus callosum. The rostrum, genu and trunk, which constitute the anterior part of the corpus callosum, are supplied by branches of the artery that originate from the anterior circulation, whereas the splenium is supplied by both the anterior and posterior branches of the artery. It has been postulated that the splenium has specific vulnerability to excitotoxic injury in metabolic diseases, making it selectively involved in differential pathological events. In the present case, why was the spinal cord also involved? As it is known, the spinal cord, especially the thoracic spinal cord, is vulnerable to ischemia and hypoxia, which would be triggered by all of the above pathological processes. Recently, Ract and Yi have reported cases on spinal cord involvement in posterior reversible encephalopathy syndrome (PRES) [11]. This also suggests that the spinal cord was sensitive to excitotoxic injury [12]. However, further studies are needed. Recently, it has reported that reversible cerebral vaso-

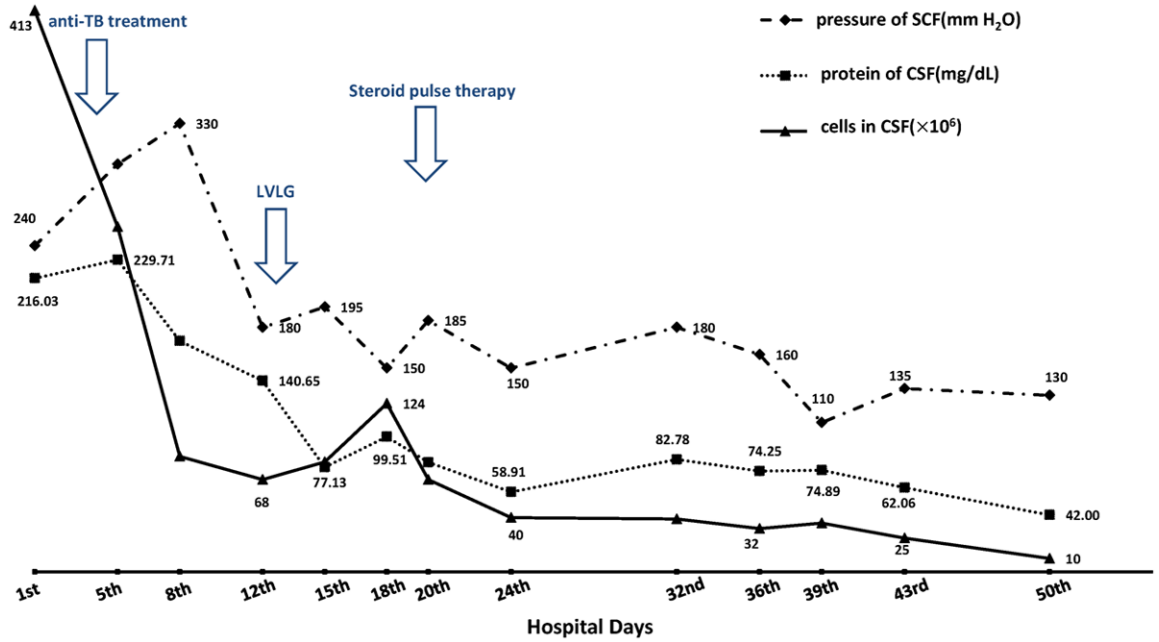

Figure 3. Changes of cells, protein and glucose in the CSF during hospitalization.

constriction syndrome (RCVS) induced transient splenial lesions [13]. Based on the facts above, it should be emphasized that RESLES, RCVS, and PRES may share the same mechanism, which actually needs further research.

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