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Case Report

CASE REPORT

Meningitis retention syndrome associated with complicated mild encephalitis/encephalopathy with reversible splenial lesion in a young adult patient: a case report

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Abstract

Meningitis retention syndrome (MRS), comprising aseptic meningitis and urinary retention, is a self-limiting disease that resolves within a few weeks. Refractory urinary retention and encephalitic syndromes are rare. A 32-year-old man presented with acute fever and headache followed by acute urinary retention (UT). Neurological examination revealed meningeal irritation, UT, constipation and ataxic gait. The cerebrospinal fluid showed mononuclear leukocytosis, and the etiological examination was negative. We suspected MRS. However, magnetic resonance imaging demonstrated an abnormally intense lesion in the splenium of the corpus callosum (SCC). He also developed delirium on day 4 of hospitalization. We diagnosed the patient with MRS associated with mild encephalitis/encephalopathy with a reversible splenial lesion (MERS). While his delirium and constipation improved, and the SCC lesion disappeared, UT was refractory and required 6 months to complete recovery. Our case suggests that severe MRS can exhibit refractory UT and may associate with MERS.

INTRODUCTION

Meningitis retention syndrome (MRS) is a para-infectious disease comprising aseptic meningitis and urinary retention (UT) and generally resolves within a few weeks [1]. Here, we report a case of severe MRS that showed refractory UT as well as constipation and required 6 months to complete recovery. During the clinical course, he also showed transient mild consciousness disturbance with reversible lesion in the splenium of the corpus callosum (SCC); this condition is known as mild encephalopathy with reversible splenial lesion (MERS) [2].

CASE REPORT

A previously healthy 32-year-old man was admitted to our hospital with acute fever, mild headache, UT and constipation. Seven

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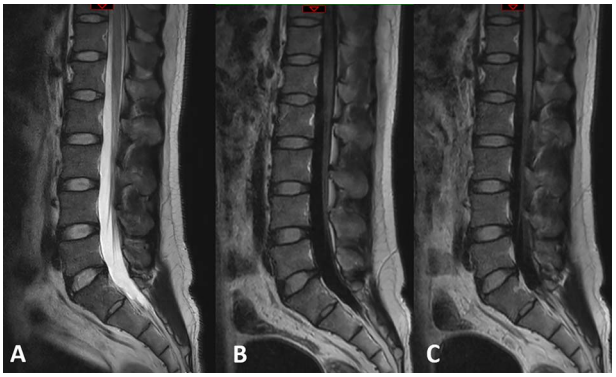

Figure 1: Spinal MRI without enhancement conducted on the fourth day of admission. (**A**) Sagittal T2WI, (**B**) T1WI and (**C**) fluid-attenuated inversion recovery image did not reveal any lesions in the spinal cord.

days before admission, he suddenly developed a fever of 39°C; the following day, he was administered antibiotics at another hospital. Two days prior to admission, he experienced acute UT and was treated with temporary transurethral catheterization at our hospital. He was suspected to have meningitis and was referred to our department.

On admission, he presented with a fever of 38°C and UT. On neurological examination, he was alert (Glasgow coma scale score 15) and revealed nuchal rigidity. The cranial nerve, motor and sensory functions were normal. Deep tendon reflexes were intact, and Babinski reflex was absent. He showed mild ataxic gait. His bladder-filling sensation was preserved; however, he could not urinate. He complained of constipation, and his anal sphincter reflex was depressed. A urodynamic study revealed a non-contractile neurogenic bladder. Laboratory examination revealed normal complete blood count and serum chemistry. Lumbar puncture showed elevated Cerebrospinal fluid (CSF) pressure was elevated (240 mmH_2O). Mononuclear leukocytosis (127 cells/μl) and increased protein level (202 mg/dl) in the CSF were observed. Bacterial smears and cultures were negative. The interleukin (IL)-6 level in the CSF was elevated (18.3 pg/ml). Myelin basic protein levels were elevated (401.5 pg/ml), while the oligoclonal band was negative. Lumbosacral spinal cord magnetic resonance imaging (MRI) did not reveal any lesions (Fig. 1). We started intravenous acyclovir on admission day and discontinued on the ninth day of hospitalization because polymerase chain reaction for Herpes Symplex Virus type-1 in the CSF was negative.

Transient delirium accompanied by automatism, such as finger rubbing, lip smacking or chewing, was observed several times a day from the fourth day of hospitalization; no convulsions were observed. Cranial MRI performed on the same day revealed an abnormal intensity lesion at the SCC (Fig. 2). An electroencephalogram conducted on the 11th day of hospitalization was normal. Follow-up MRI on the same day showed complete disappearance of the SCC lesion with no additional abnormalities (Fig. 3). We diagnosed the patient with MRS associated with MERS. On the 12th day of hospitalization, consciousness disturbance disappeared without any treatment. Although he still had mild ataxic gait, constipation, and UT, he was discharged on the 19th day of hospitalization. Two months later, all symptoms disappeared except for UT, and self-transurethral catheterization was still required. The patient required 6 months to completely recover from the UT after onset. He has had an uneventful course without recurrence for more than 3 years.

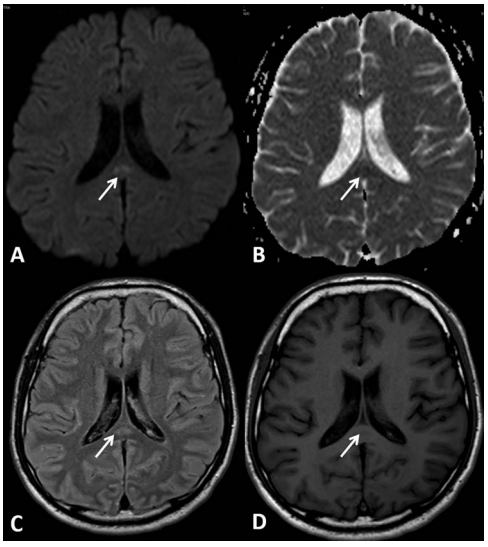

Figure 2: Brain MRI conducted on the fourth day of admission. (**A**) Axial diffusion-weighted image, (**B**) ADC image, (**C**) fluid-attenuated inversion recovery (FLAIR) image and (D) T1WI. A splenial lesion was revealed. The signals in the splenium of the corpus callosum were hyperintense on FLAIR, T2WI and DWI, decreased on ADC and had a slightly low intensity on T1WI (arrow).

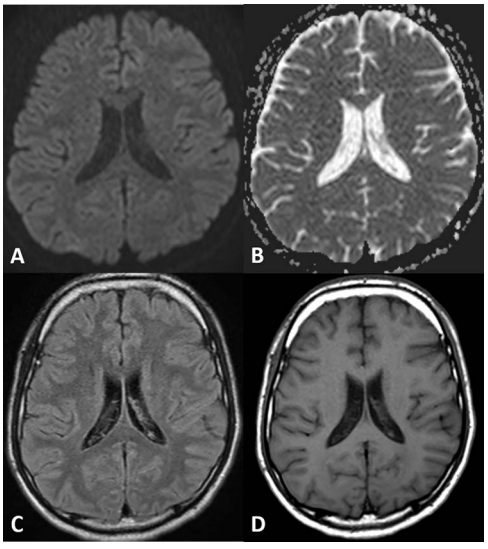

Figure 3: Follow-up brain MRI conducted on the 11th day. (**A**) Axial diffusion-weighted image, (**B**) ADC, (**C**) fluid-attenuated inversion recovery image and (**D**) T1WI. Follow-up MRI showed no lesions on any sequences.

DISCUSSION

We presented a case of MRS associated with MERS. Several differential diagnoses resemble patients with MRS, such as myelitis, Guillain–Barre syndrome (GBS), sacral herpes, multiple sclerosis (MS) and acute disseminated encephalomyelitis (ADEM) [3]. Lack of leg numbness and paresthesia, negative for oligoclonal band in the CSF, no occasional lesion on spinal MRI helped us to rule out myelitis, GBS and MS. Sacral herpes was also unlikely because HSV DNA was not detected in the CSF. Our patient initially presented with acute UT and constipation with aseptic meningitis, which is characteristic of MRS [1]. A urodynamic study of MRS usually shows an atonic or hypotonic neurogenic bladder with intact bladder sensation. Although the exact

Table 1: Reported cases of MERS complicated with UT

Case no.	Author, reference	Age/sex	UT timing	CSF IL-6 (pg/ml)	UT duration
This case	Hidaka *et al.*	32/M	Preceded MERS	18.3	24 weeks
1	Tascilar *et al.* [6]	26/F	4 days after MERS onset	NM	7 weeks
2	Kitami *et al.* [7]	23/F	Simultaneous	NM	4 weeks
3	Yuan *et al.* [8]	37/M	Simultaneous	NM	10 days
4	Pan *et al.* [9]	26/M	NM	NM	NM
5	Pan *et al.* [9]	21/M	NM	NM	NM
6	Suzuki *et al.* [10]	38/M	Simultaneous	NM	12 days
7	Suzuki *et al.* [10]	36/M	Preceded MERS	1260	NM

F, female; M, male; NM, not mentioned.

pathophysiology of MRS is poorly understood, it is speculated that direct viral/bacterial inflammation or post-infection inflammatory demyelination causes sacral myeloradiculopathy. Therefore, MRS might be a spinal form of ADEM restricted to the sacral spine; however, this is unclear because most previous cases of MRS have not reported any abnormalities of the spinal cord MRI. In the present case, insult to the parasympathetic nerves originating from the conus medullaris was suspected based on the neurological manifestation. Thus, his condition does not contradict sacral myeloradiculopathy. The clinical course of MRS is generally self-limiting and resolves within a few weeks [1]; however, our case was unique in that it required 6 months to recover completely from UT and temporal encephalopathy occurred during the clinical course.

Transient encephalopathy with reversible SCC lesions observed in our case appears compatible with MERS proposed by Tada *et al.* [2]. Although both ADEM and MERS are para-infectious disorders, their radiological findings and pathology differ in some aspect. Patients with ADEM typically demonstrate multiple, asymmetric, bilateral and poorly marginated hyperintense lesions on T2-weighted image (T2WI) with increased apparent diffusion coefficient (ADC) values. While patients with MERS generally show only well-defined hyperintense lesions on diffusion-weighted image and T2WI with decreased ADC values, suggesting cytotoxic edema confined to the corpus callosum [4], as seen in our case. Their supposed pathologies are also different. ADEM is caused by acquired autoimmune responses to the central nervous system and subsequent demyelination [5]. While in MERS, cytotoxic release is hypothesized to induce water influx into neurons and astrocytes, resulting in cytotoxic edema in SCC rich in myelinated fibers. As our case showed elevated CSF IL-6 levels, the SCC lesion might have resulted from cytotoxic release.

A combination of MRS and MERS is rare. In the present case, we speculate that strong cytokine release due to MRS might have triggered MERS. In most of the reported cases of MERS complicated by UT (Table 1), MERS preceded the UT onset or occurred simultaneously, except in the case of Tascilar [6–10]. IL-6 levels in CSF were measured in only one case, other than ours, and were highly elevated. Further studies are necessary to elucidate the relationship between these two disorders.

In conclusion, patients with MRS can exhibit refractory UT and associate with MERS. When such patients present with encephalopathic symptoms, MRI of the brain should be conducted to evaluate complication with MERS.

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CONFLICT OF INTEREST STATEMENT

No conflicts of interest.

FUNDING

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ETHICAL APPROVAL

None required.

CONSENT

Informed consent was obtained from the patient.

GUARANTOR

Shuji Arakawa is the guarantor of this article.

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