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Case Report

Gastric perforation and critical illness polyneuropathy after steroid treatment in a patient with encephalitis/encephalopathy with transient splenial lesion

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Abstract

The outcome of mild encephalitis/encephalopathy with reversible splenial lesion (MERS) is favorable whether or not specific treatment is performed. We report a patient with MERS treated with methylprednisolone, complicated by gastric perforation followed by critical illness polyneuropathy. The patient was a 14-year-old male with mildly impaired consciousness and hyponatremia who was treated with methylprednisolone pulse therapy. High fever appeared after methylprednisolone pulse therapy and free air was recognized on an abdomen roentgenogram. Gastric perforation was recognized on emergent endoscopic surgery and omental implantation repair was performed. His consciousness was fully recovered after surgery, whereas he was noted to have motor and sensory impairment of the lower extremities and vesico-rectal disturbance. Nerve conduction studies revealed decreased compound muscle action potentials with preserved motor conduction velocity and decreased sensory nerve action potentials. He was diagnosed as having critical illness polyneuropathy, and bedside physical rehabilitation was initiated. His neurological symptoms resolved within 6 months. Our patient highlighted possible serious adverse events associated with steroid treatment for children with MERS.

Keywords: Splenial lesion; Steroid; Gastric perforation; Critical illness polyneuropathy

1. Introduction

Mild encephalitis/encephalopathy with a reversible splenial lesion (MERS) is a subtype of acute encephalopathy, characterized by a reversible lesion with reduced diffusion in the corpus callosum mainly involving the splenium, sometimes associated with symmetrical white matter lesions [1,2]. Delirious behavior and/or mild to moderate impairment of consciousness are the most common neurological symptoms. The outcome of MERS is favorable, with a large majority of patients achieving a full recovery [3], whether or not specific treatment such as steroid and intravenous immunoglobulin is provided. Thus, it is generally considered that specific treatment is not necessary in children with MERS. However, it is difficult to make early treatment decisions because final diagnosis is only made several days after when many data including clinical information and neuroimaging findings are

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integrated. Even in children with splenial lesion, other types of brain lesions such as cerebellitis [4] and late reduced diffusion in subcortical white matter [5] has been reported.

Among the treatments for acute encephalopathy, methylprednisolone pulse therapy is commonly performed without major adverse events; however, there is the potential risk for various problems, including hypertension, gastrointestinal ulceration, and thrombosis. Here, we report an adolescent with MERS treated with methylprednisolone complicated by gastric perforation followed by critical illness polyneuropathy (CIP). Based on this report, an avoidance of steroid use should be considered for patients with MERS.

2. Patient report

A 14-year-old male was transferred to Juntendo University Hospital due to a 5-day illness with pyrexia, headache, and mildly impaired consciousness associated with hyponatremia. He was the first child of unrelated healthy parents. His past and family history was unremarkable. He visited another hospital because of pyrexia and headache, and lumber tap revealed a cell count of 176/μL and protein level of 86 mg/dL. He was diagnosed as having aseptic meningitis and was admitted to the hospital. Hyponatremia (serum sodium level, 128 mEq/L) was recognized after admission and worsened gradually despite extracellular fluid infusion and fluid restriction. Mild impairment of consciousness was observed, and head magnetic resonance imaging (MRI) showed a high intensity area in the splenium of the corpus callosum on diffusion-weighted images (Fig. 1). He was transferred to our hospital 5 days after the onset of fever.

His clinical course was shown on Fig. 2. Upon arrival at our hospital, his consciousness was E4V4M6; He could respond to verbal questions, whereas his responses were inconsistent. Laboratory data showed hyponatremia of 125 mEq/L. Later, plasma antidiuretic hormone was reported to be 3.64 pg/mL. Otherwise, his vital signs and laboratory data were unremarkable. Head MRI showed persistence of the splenial lesion on diffusion-weighted images. No other lesions were present on head MRI. EEG 2 days after admission demonstrated posterior dominant mild slowing. He was diagnosed as having MERS associated with inappropriate secretion of antidiuretic hormone. He was treated with fluid restriction against hyponatremia and intravenous dexamethasone against encephalopathy. Although hyponatremia was gradually corrected, impaired consciousness persisted. Methylprednisolone pulse therapy was started 5 days after admission to our hospital. No H2-blocker was used during methylprednisolone pulse therapy. Hypertension appeared after the administration of methylprednisolone, and he was treated with nicardipine.

At 10 days after admission, high fever was newly recognized. Laboratory examination showed white blood cell counts of 23,100/μL and a C-reactive protein level of 8.9 mg/dL. Although abdominal pain was not observed, free air was recognized on a routine abdomen roentgenogram (Fig. 1). Intestinal perforation was strongly suspected and emergent endoscopic surgery was performed. Gastric perforation was identified and omental implantation repair and intestinal lavage were performed. Rocuronium was adminisitered as a neuromuscular blocking agent during operation and was not used after operation. Artificial ventilation was discontinued 12 h after surgery. Intravenous tazobactam/piperacillin, meropenem, and immunoglobulin were administered for peritoneal infection.

After surgery, his consciousness was fully recovered and the general condition became stable within a few days. Splenial lesion was not observed on MRI at 2 days after surgery (Fig. 1). However, motor and sensory impairment of the lower extremities and vesico-rectal disturbance were noticed after surgery. Manual muscular test was grade 2 in the lower extremities. Mild numbness, temperature, and vibration sensation was observed in the lower legs. Deep tendon reflexes were normal in the upper extremities but weak in the lower extremities. No pyramidal signs were observed. Cranial nerves were not affected. Muscle pain was not noticed and serum creatine kinase levels were within the normal range. Nerve conduction studies revealed decreased compound muscle action potentials with preserved motor conduction velocity in the bilateral tibial and peroneal nerves, decreased sensory nerve action potentials in the right peroneal nerve, and absence of F-waves in the bilateral tibial nerves (Fig. 1). He was diagnosed as having CIP. Bedside physical rehabilitation and mobilization were initiated since 5 days after the surgery and neurological symptoms gradually recovered. He could walk with support 3 weeks later, walk independently 2 months later, and run 3 months later. No neurological sequelae or cognitive disability were present 6 months later. Nerve conduction studies were not performed after discharge.

3. Discussion

Gastrointestinal perforation followed by CIP was unexpectedly observed in our patient with MERS after steroid treatment. Although the outcome of our patient was favorable, gastrointestinal perforation may have resulted in severe neurological sequelae or even death. After the outbreak of influenza-associated acute encephalopathy in the 1997/98 winter season, methylprednisolone pulse therapy has been widely administered to children with acute encephalopathy, regardless of its subtypes and severity. Although an efficacy of ster-