A case of clinically mild encephalitis with a reversible splenial lesion (MERS) after mumps vaccination

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Abstract

We describe for the first time an 8-year-old male patient who demonstrated clinically mild encephalitis with a reversible splenial lesion after mumps vaccination. He suffered from transient hallucinations, nuchal rigidity, and inappropriate antidiuretic hormone secretion syndrome. On the 5th day of admission, his head MRI showed symmetrical high-signal-intensity lesions on T2, FLAIR, and diffusion-weighted images in the splenium of the corpus callosum and in the periventricular white matter, while an apparent diffusion coefficient map showed reduced diffusion. The images were not enhanced by gadolinium. Follow-up MRI on the 16th day of admission revealed none of these abnormalities. His serum IgM and IgG antibodies against the mumps virus were positive according to an enzyme immunoassay. Mumps Torii vaccine strain was isolated from the patient’s cerebrospinal fluid. Previous reports demonstrated that transient delirious behavior, the syndrome of inappropriate antidiuretic hormone secretion, and good prognosis were the main clinical features of mild encephalitis with a reversible splenial lesion. This case shows that mild encephalitis with a reversible splenial lesion could occur after mumps vaccination.

Keywords: Clinically mild encephalitis; Reversible splenial lesion; Mumps vaccination; MRI

1. Introduction

Clinically mild encephalitis with a reversible splenial lesion (MERS) is a distinct new clinico-radiological entity [1]. Over 30 cases of MERS related to viral infections, bacterial infections, and other conditions have been reported [2–6]. The clinical manifestations consist of relatively mild central nervous system symptoms, most often delirious behavior within a week of fever, and complete recovery within a month [1]. MRI reveals reversible lesions with reduced diffusion in the corpus callosum (at least in the splenium), sometimes related to symmetrical white-matter lesions that completely disappear within a short period [1]. The outcome is favorable in most cases. To our knowledge, this is the first case of MERS after mumps vaccination.

2. Case report

An 8-year-old male was admitted to St. Mary’s Hospital because of fever, headache, vomiting, and eye flickering. He had received eight kinds of immunizations...
before reaching the age of 4 years and a mumps vaccine (Torii strain) 20 days before admission. He had never experienced any adverse events following immunizations before. On admission (day 1), his body temperature was elevated, at 39.3°C. He had not suffered from respiratory infection, parotiditis, pancreatitis, or orchitis. He was conscious and alert. His neurological findings were normal except for photophobia, and he had a positive Kernig’s sign as well as nuchal rigidity. His motor function and deep tendon reflexes were normal. On day 2, he suffered from hallucinations such as that of a stranger stealing his sweets. He showed a score of 12–13 on the Glasgow Coma Scale, and this score continued for 6 days. His delirious behavior and hallucinations persisted for several minutes after the headache, and were repeated for a day. According to laboratory examinations, the patient’s complete blood count, urinalysis, serum concentrations of electrolytes, glucose, and hepatic and renal function indicators were within the normal ranges except for a low sodium level 134 mEq/L (135–147). The cerebrospinal fluid (CSF) on admission showed mild pleocytosis of 17/mm³ (mononuclear cells: polymorphonuclear cells = 5:12), normal levels of glucose and protein. His second CSF sample, taken on day 5, showed an increased cell count of 624/mm³, protein of 52 mg/dl, myelin basic protein levels less than 40 pg/ml (<102 pg/ml), and a negative oligoclonal band. Brain MR imaging on day 5 showed symmetrical high-intensity-signal lesions in the splenium of the corpus callosum (SCC) and in the periventricular white matter on T2-weighted images, FLAIR, and diffusion-weighted images (DWI), and low-intensity-signal lesions on T1-weighted images in the same regions. An apparent diffusion coefficient (ADC) map showed a reduced diffusion. These lesions were not enhanced by gadolinium (Fig. 1A and B). On day 3, the laboratory data demonstrated a further decrease in the levels of serum sodium to 128 mEq/L, and increased urinary sodium of 54 mEq/L, which suggested that he had inappropriate antidiuretic hormone secretion syndrome (SIADH). The fluid volume had been restricted to 40 ml/kg/day since day 1. Steroid pulse therapy was started on day 6 because of the patient’s persistent fever, headache, and delirious consciousness. His serum sodium returned to normal, and his hallucinations and meningeal signs had completely disappeared on day 7. EIA units of serum IgM antibodies against mumps virus were 1.2 on day 5 and increased to 5.7 on day 16. The serum IgG antibodies on day 5 were 3.2 and increased to 23.7 on day 16. Mumps virus was isolated from the patient’s CSF, and sequencing results showed that the partial hemagglutinin-neuraminidase (HN) gene was identical to the Torii vaccine strain. The phylogenetic analysis of the sequence results is shown together with that of the currently circulating wild-type (Fig. 2). The sequence of the mumps virus obtained from the patient was identical to that of the Torri vaccine strain. Genotype G was the currently circulating wild-type [7]. Examinations for serum antibodies against viruses, including influenza A/B, human parvo, echo, coxsackie, and herpes simplex, were negative. ABR and EEG performed on day 7 were showed normal results. On day 16, head MRI revealed normal findings in the SCC and in the periventricular white matter.

3. Discussion

Our case presented with fever, meningeal signs, and delirious consciousness, after mumps vaccination, all of which were completely resolved within a week. A multi-center study performed in Japan investigating the data from 54 cases with MERS showed that the most common neurological symptom is delirious behavior (54%) [1]. In radiological study, SCC lesions

![Fig. 1. Brain MR imaging on the 5th day of admission. (A) Diffusion-weighted images (DWI) showed symmetrical high-intensity-signal lesions on the splenium of the corpus callosum (SCC) [arrow-head] and the periventricular white matter [arrow]. (B) An apparent diffusion coefficient (ADC) map showed reduced diffusion.](image-url)
in MERS patients are symmetrical, not enhanced with gadolinium [6], and initially have reduced ADC values, which indicates cytotoxic edema. These findings completely disappear within a week in most cases. These findings were present in our case. Previous studies have reported a close relationship between MERS and hypo-natremia involving SIADH. Hypotonic hyponatremia causes cerebral edema due to the entry of water into the brain, resulting in headache, nausea, vomiting, confusion, and seizures [5]. Our case had similar symptoms and was also complicated with SIADH.

Although the distinction between MERS and ADEM remains controversial, ADEM patients tend to show asymmetric callosal lesions and lesions enhanced with gadolinium on brain MRI, and their clinical conditions are typically more severe than those of MERS patients. These features indicate that our patient was more likely to have suffered from MERS than ADEM. MERS may be attributed to both intramyelinic edema and inflammatory infiltration [6]. The corpus callosum is a midline white-matter brain region responsible for interhemispheric communication and coordination. The nerve fibers at the posterior section of the corpus callosum innervate the bilateral visual cortex. This implies that the lesions responsible for transient headache and hallucination may be located in the SCC and in the periventricular white matter [8].

The most common pathogens implicated in MERS are influenza virus A/B (19%) and the mumps virus (7%) [1], whereas the major adverse effects after mumps vaccination has generally been considered to be aseptic meningitis caused by mumps vaccine strains [9]. The live-attenuated mumps-virus vaccine used for our patient contained the Torii strain, which was isolated from his CSF. He suffered from MERS 20 days after mumps vaccination. The incubation time for the mumps virus averages 16–18 days within a range of 2–4 weeks [9]. This implies that the Torii strain is strongly associated with MERS in this case.

In conclusion, we herein provide the first reported case of MERS with SIADH that occurred after mumps vaccination.

References