Reversible splenial lesion syndrome caused by rubella infection

1Pahn Kyu Choi, 2Eun Ju Yoon, 3Sang Woo Ha, 1Hyun Goo Kang

1Department of Neurology, 2Department of Radiology, and 3Department of Neurosurgery, Chosun University Hospital, Gwang-ju, South Korea

Abstract

Reversible splenial lesion syndrome can be caused by viral infection. Rubella generally occurs in childhood, and it is rarely accompanied by neurological complications in adulthood. A 35-year-old man visited our hospital due to conjunctival injection, mild fever, and headache 3 days after experiencing skin rash. Brain magnetic resonance imaging (MRI) revealed distinct lesions involving white matter in the splenium of the corpus callosum approximately 3 days following the onset of symptoms. Enzyme immunoassay performed on serum and CSF samples was positive for rubella virus IgM. A follow-up brain MRI was performed 24 days after the onset of symptoms, and reduced lesion size with decreased signals were observed on diffusion weighted image. This case showed that rubella infection can result in reversible splenial lesion accompanied by only mild neurological symptoms.

Keywords: Corpus callosum; reversible splenial lesion syndrome; rubella

INTRODUCTION

Reversible splenial lesion syndrome (RESLES) which involves the splenium of the corpus callosum, can be caused by infection, high-altitude cerebral edema, seizure attacks, antiepileptic drugs, and metabolic disorders.1 The typical clinical symptoms of RESLES include mildly altered states of consciousness, delirium, and seizure. When RESLES occurs due to infection, most of the cases were accompanied by encephalitis or seizure.

The known causative viruses are mainly influenza A and B, Epstein-Barr virus, rotavirus, herpesvirus-6, mumps, varicella-zoster virus, and adenovirus.1 Rubella is a viral eruption disease characterized by fever, rash, and lymph node swelling in the neck. It shows a range of symptoms from inapparent infection to severe complications. However, it generally occurs in childhood and is rarely accompanied by neurological complications in adulthood.2

We report here an adult patient who had RESLES with mild symptoms after rubella infection, and performed a literature review.

CASE REPORT

A 35-year-old man, who was in good general health, presented with conjunctival injection, mild fever, and headache 3 days before having a skin rash (Figure 1). He had severe holocephalic headache (numerical rating scale 8/10), which was worse while sitting or walking. His vital signs on
admission were body temperature 37.8°C, blood pressure 120/78 mmHg, pulse rate 78 beats/min, and respiratory rate 20 breaths/min. Physical examination revealed skin rash reportedly started from the face and spread to the entire body. Multiple macular rashes (1-2 mm in diameter) accompanied by discoloration were scattered on the face, body, and both arms and swelling of lymph nodes at the back of the ears and the head (Figure 1). The patient recalled being vaccinated against various viruses but could not remember specific information regarding these vaccinations.

Laboratory test showed that white blood cell count, erythrocyte sedimentation rate, and C-reactive protein level were 5,050 cells/mm³, 10 mm/h, and 8.78 mg/dL. Urinalysis, hepatic and renal function indicators, serum concentrations of electrolytes, and glucose were within normal ranges. Brain magnetic resonance imaging (MRI) revealed distinct lesions involving white matter in the splenium of the corpus callosum approximately 3 days following the onset of symptoms. These lesions showed restricted diffusion with hyperintense signal on diffusion-weighted imaging (DWI) and hypointense signal on apparent diffusion coefficient (ADC) sequence (Figure 2). Fluid attenuated inversion recovery (FLAIR) and T2-weighted images showed similar hyperintense signal on the splenium (C and D).

Consequently, the patient was diagnosed with rubella infection. The patient gradually recovered only with fluid therapy and was discharged 10 days after symptoms onset. A follow-up brain MRI showed no contrast enhancement. Cerebrospinal fluid (CSF) examination was conducted to test for viral meningitis and intracranial hypotension; however, white blood cell count (0/mm³), protein (40 mg/dL), glucose (45 mg/dL) were within normal ranges. An electroencephalography performed was normal. An enzyme immunoassay on serum and CSF showed that rubella virus IgM was positive and other viral markers with mumps virus, measles virus, Epstein-Barr virus, herpes simplex virus, and cytomegalovirus were negative. Consequently, the patient was diagnosed with rubella infection. The patient gradually recovered only with fluid therapy and was discharged 10 days after symptoms onset. A follow-up brain MRI showed no contrast enhancement. 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The patient gradually recovered only with fluid therapy and was discharged 10 days after symptoms onset.
performed 24 days after symptoms onset showed reduced size of lesions with decreased signals in fluid attenuated inversion recovery (FLAIR) and T2 weighted image (Figure 3). The patient was asymptomatic during follow up.

**DISCUSSION**

Clinical manifestations of rubella can be divided into natural rubella and congenital rubella syndrome depending on the infection pathway. Acute rubella encephalitis, encephalomyelitis, or progressive rubella panencephalopathy may occur when rubella invades the central nervous system. A previous study reported that radiological findings in adults rubella infection include edematous change and abnormal signals in the temporal lobe. Pediatric autopsy cases showed extensive edema in the cerebral white matter, brainstem, and cerebellum. Histopathology showed infiltration of lymphocyte without demyelination.

Common prodromal neurological signs in RESLES include delirium, disturbance of consciousness, and seizure, with RESLES occurring 1-3 days after symptoms onset. Influenza viruses A and B are the most common causative viruses. We reported here an uncommon case of RESLES caused by rubella virus infection. The neurological signs were not severe despite having RESLES.

The pathogenesis of RESLES has not been clearly elucidated. It was shown that splenial lesion was not caused by neurotoxicity from inflammatory reactions since inflammatory markers such as interleukin-6 in the CSF and tumor necrosis factor were normal. The most probable hypothesis to date is intramyelinc edema and influx of inflammatory cells, with separation of the myelin layers, leading to transient decreased diffusion. The reversibility of the lesion suggests a distinctive pathophysiology from the cytotoxic edema evident in cellular failure.

In conclusion, we present a patient with novel manifestation of rubella virus infection associated with RESLES, who recovered completely without any interventions. This case showed that rubella virus infection could cause RESLES, with mild clinical symptoms only.

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**DISCLOSURE**

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Conflicts of interest: None.

**REFERENCES**


