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Case Report

Brainstem infarction associated with HHV-6 infection in an infant

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Abstract

Introduction: The relevant literature includes several case reports on cerebral infarction in children with HHV-6 infection; however, there is no report of brain stem infarction.

Case: An 11-month-old girl was hospitalized because of fever. She was unable to stand up and meet her mother's gaze. Magnetic resonance imaging (MRI) indicated a right pons and mid-brain lesion; a diagnosis of brainstem infarction was made. After her fever subsided, a rash developed on her trunk and limbs; blood examination results indicated a primary HHV-6 infection. She was treated with aspirin, edaravone, and mannitol to prevent further complications. At the age of 18 months, the auditory brainstem response (ABR) was unremarkable and she is developing well.

Discussion and conclusion: A limited number of studies have reported HHV-6 infection-associated infarction, and no cases of brainstem infarction have been reported. One possible cause of cerebral infarction is antiphospholipid antibody syndrome (APS) triggered by the infection. HHV-6 may also directly infect vascular endothelial cells and cause angiopathy. However, the real mechanism of infarction remains unclear. Our patient had a favorable prognosis despite brainstem infarction. © 2017 The Japanese Society of Child Neurology. Published by Elsevier B.V. All rights reserved.

Keywords: HHV-6; Exanthema subitum; Brain infarction; Antiphospholipid antibody syndrome (APS); Magnetic resonance imaging (MRI)

1. Introduction

Population-based studies of cerebral infarction in children estimate an annual incidence of 7.8 per 100,000 persons [1]. In contrast to adults where the main risk factor is arteriosclerosis, various risk factors are involved in pediatric infarction including congenital heart disease, cardiac disorder, metabolic disease, hyper-coagulable states, and infection [2]. Human herpes virus

6 (HHV-6) is known to cause exanthema subitum in infancy. Recently, several case reports on cerebral infarction in children with HHV-6 infection have been published; however, there are no guidelines for its treatment or long-term surveillance studies. We report a pediatric case of brainstem infarction associated with HHV-6 infection, and review the relevant literature.

2. Case report

An 11-month-old girl with no history of growth or developmental disorder was hospitalized because of

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fever. She was unable to stand up and meet her mother's gaze. All vitals were normal with the exception of a fever of 39.4 °C. She was not dehydrated. Glasgow Coma Scale score was E4V5M5. She had spontaneous eye opening, and her pupils were equal and reactive. Brainstem function was intact and she had spontaneous limb movements. Deep tendon reflex was not examined. Her chest examination was clear except for decreased breath sounds on the right side.

2.1. Investigations

C-reactive protein, alanine aminotransferase, aspartate aminotransferase, ferritin, fibrin degradation product, and D-dimer levels were mildly elevated on blood analysis (Table S1). Blood urea nitrogen, creatinine, and cerebrospinal fluid (CSF) protein were also high, and venous blood gas analysis showed mild metabolic acidosis from circulatory failure likely due to fever and infection. Urinalysis analysis were unremarkable. Norovirus was slightly positive by rapid antigen examination, but we believed that this was a false positive because she had no digestive symptoms. Cranial computed tomography showed no lesions. Chest radiography revealed right upper consolidation with an effusion, suggesting pneumonia.

2.2. Clinical course

Although ampicillin (100 mg/kg/day) was administered and her fever had reduced the next day, her unusual condition continued, which we believe resulted from a consciousness disturbance (Fig. 1). Thus, MRI was performed on day 2; a diagnosis of brainstem infarction was made (Fig. 2). Magnetic resonance angiography showed no abnormality. Blood samples were analyzed for the presence of hypercoagulable states. Only protein C activity was abnormal at 50%; however, re-examination several days later showed an increase to 78%. There were no abnormal findings on the electrocardiogram or echocardiogram.

On day 2, after her fever subsided, a rash developed on her trunk and limbs. Antiviral antibodies were measured and the results were compatible with primary HHV-6 infection. A CSF sample was negative for HHV-6 by polymerase chain reaction.

She was treated for brain infarction with aspirin (5 mg/kg/day), edaravone (10 mg/kg/day), and mannitol (7.5 mL/kg/day) to prevent further complications and gradually returned to a normal state of health. On day 9, a rash appeared all over her body. It was thought to be caused by cefotaxime administration; the rash vanished immediately following the ceasing of administra-



CTX, cefotaxime; ABPC, amylase-binding protein C;



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