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Case Report

Increased levels of anti-phosphatidylcholine and anti-phosphatidylethanolamine antibodies in pediatric patients with cerebral infarction

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Abstract

Cerebral infarction in children is rare and often occurs secondary to moyamoya disease, hereditary coagulopathies, vasculitis, antiphospholipid antibody syndrome, heart disease, mitochondrial disease. However, in some cases, the causes of cerebral infarction is unknown.

In this study, we detected increased levels of serum anti-phosphatidylcholine and anti-phosphatidylethanolamine IgG antibodies in three pediatric patients with cerebral infarction whose primary disorders are unknown by routine examination. For the five disease control patients of cerebral infarction due to other primary disorders, there was no such increase in these antibodies levels.

Phosphatidylcholine and phosphatidylethanolamine are major components of the phospholipids of vascular endothelial cells, while cardiolipin is a minor component. Anti-phosphatidylcholine and anti-phosphatidylethanolamine antibodies, as well as anti-cardiolipin antibody, might also be risk factors with cerebral infarction.

Keywords: Antiphosphatidylcholine antibody; Antiphosphatidylethanolamine antibody; Cerebral infarction

1. Introduction

Cerebral infarction in children is rare and often occurs secondary to moyamoya disease, hereditary coagulopathies, vasculitis, antiphospholipid antibody syndrome, heart disease, mitochondrial disease [1]. However, in some cases, their causes of cerebral infarction and primary disorders is unknown.

Among causes of cerebral infarction, antiphospholipid antibody syndrome is characterized by venous and/or arterial thrombosis, and its diagnosis requires the presence of both clinical and laboratory findings, such as positive anti-cardiolipin (CL) and anti-β2 glycoprotein I antibodies and lupus anticoagulant [1]. However, CL is a minor component of the phospholipids of the vascular endothelial cells in human, while phosphatidylcholine (PC) and phosphatidylethanolamine (PE) are major components [2].

In this study, we detected increased levels of anti-PC and anti-PE antibodies in three pediatric patients with cerebral infarction whose primary disorders are unknown by routine examination. These antibodies, as well as anti-CL antibody, might also be risk factors for cerebral infarction.

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2. Case reports

Patient 1 was an 1-month-old female infant. There was no family history of cerebral infarction, coagulopathies or autoimmune disease. Her mother was negative for anti-CL antibody. Preceded by fever, she suddenly developed convulsion and impaired consciousness. Her head CT revealed right thalamic hemorrhage and intraventricular rupture (Fig. 1 left), while head MRA did not show obvious rupture of artery (Fig. 1 right). She was examined for cerebrovascular malformation, hereditary coagulopathies, heart disease, and congenital metabolic disorder, but no abnormalities were identified. Finally her pathology was suspected to be thrombosis of internal cerebral veins. Following intensive treatments, warfarin treatment was started. Thereafter, she has not relapsed.

Patient 2 was an 1-year and 1-month old female. There was no family history of cerebral infarction, coagulopathies or autoimmune disease. There were no remarkable physical problems in her past history. She suddenly developed frequent vomiting and diarrhea associated with adenoviral enterocolitis. Moreover, she was complicated with right ataxia, nystagmus and Horner's syndrome. She was examined for cerebrovascular malformation, hereditary coagulopathies, heart disease, and congenital metabolic disorder, but no abnormalities were identified. Her head MRI T2-weighted imaging and FLAIR imaging revealed high intensity of right cerebellar hemisphere and right lateral medulla oblongata (Fig. 2 left) and her head MRA could not detect right vertebral artery and posterior inferior cerebellar artery (Fig. 2 right). Finally she was diagnosed with Wallenberg syndrome caused by infarction of territory irrigated by right vertebral artery and posterior inferior cerebellar artery. Following intensive treatments, warfarin and aspirin treatment were started. Thereafter, she has not relapsed.

Patient 3 was 5-year and 8-month old female. There was no family history of cerebral infarction, coagulopathies or autoimmune disease. There were no remarkable physical problems in her history. At the age of 3 years, she suddenly developed febrile status convulsivus resulting in severe motor intellectual disabilities. At the age of 5 years, she was referred to Oita University Hospital. Her head MRI revealed brain atrophy, and T1-weighted imaging low and T2-weighted imaging high intensity area of left posterior cranial fossa (Fig. 3 left). Moreover, her enhanced abdominal CT revealed multiple low density areas of bilateral renal parenchyma and enlargement of left renal pelvis (Fig. 3 right). Finally her pathology at the age of 3 years was suspected to be infarction of cerebral territory irrigated by left posterior cerebral artery and bilateral renal infarction. As a primary disorder of multiple infarction, she was examined for hereditary coagulopathies, heart disease, and congenital metabolic disorder, but no abnormalities were identified. Following dipyridamole treatment was started, she has not relapsed.

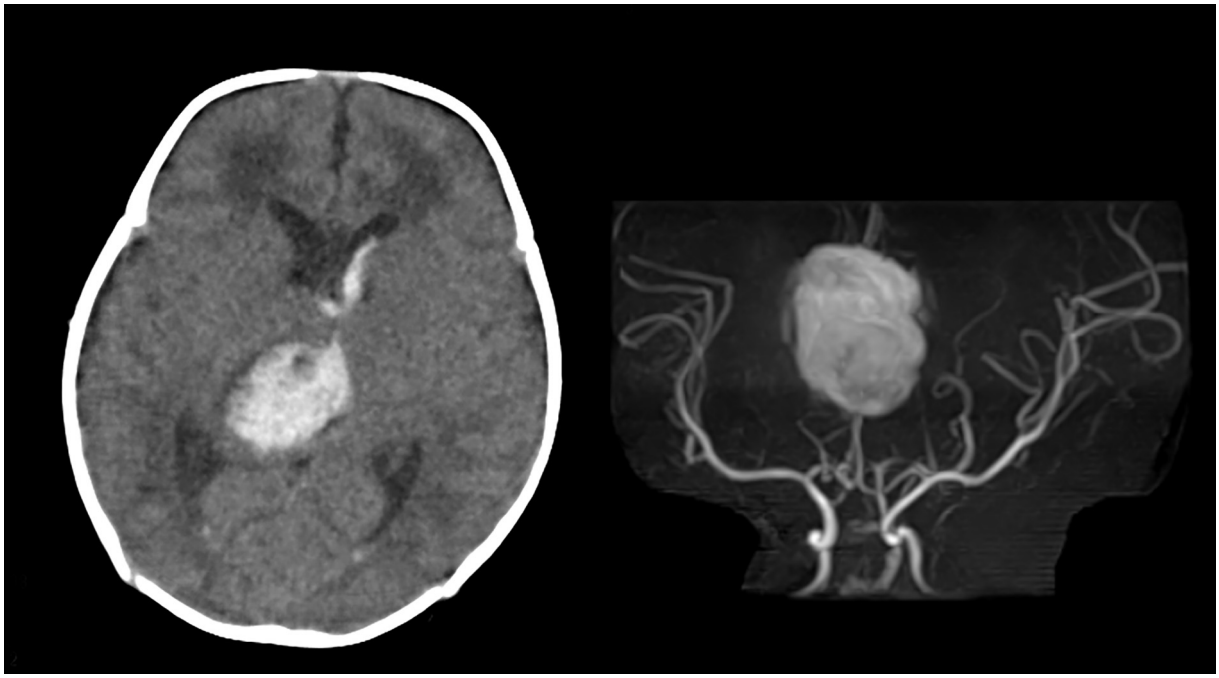

Fig. 1. Head MRI T2-weighted imaging and MRA imaging of patient 1. *left:* Her head CT revealed right thalamic hemorrhage and intraventricular rupture. *right:* Her head MRA did not show obvious rupture of artery.