Case Studies

Nonbacterial Thrombotic Endocarditis Complicated by Cerebral Infarction in a Patient with Adenomyosis with High Serum CA125 Level; A Case Report

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We report a case of a 48-year-old woman with multiple cerebral infarctions caused by nonbacterial thrombotic endocarditis (NBTE) because of adenomyosis with high serum carbohydrate antigen (CA)125 level. Transesophageal echocardiography (TEE) showed a vegetation, 4 mm in diameter, adjacent to the anterior leaflet of the mitral valve on day 2. Soluble CA125 level was elevated to 901 U/mL. Intravenous infusion of unfractionated heparin sodium was started. On day 35, TEE revealed reduction of the vegetation in size, 2 mm in diameter. On day 38, she was transferred to the hospital for further rehabilitation. CA125 is a transmembrane mucin that contributes to the progression of epithelial ovarian cancer. It is important to keep in mind that adenomyosis with abnormally high serum CA125 level may be at high risk of NBTE. **Key Words:** Cerebral infarction—nonbacterial thrombotic endocarditis—adenomyosis—CA-125—mucin.

Introduction

Nonbacterial thrombotic endocarditis (NBTE) is a well-known form of thromboembolism in which endothelial cells are damaged by immune complexes and hypercoagulability because of malignant tumors and autoimmune disease.[1] NBTE is often associated with malignant tumors such as ovarian, pancreatic, and lung adenocarcinomas, but may on very rare occasions arise as a complication of benign tumor.[2] Here, we report the case of a patient with cerebral infarction because of NBTE caused by adenomyosis with high serum concentrations of carbohydrate antigen (CA)125.

Case Presentation

A 48-year-old woman was admitted to our hospital with right hemiplegia and aphasia. She was a previously healthy woman with no history of vascular risk factors. About 1 month earlier, she had experienced abnormal vaginal bleeding and presented to another hospital, where iron deficiency anemia and adenomyosis were diagnosed.

On presentation, level of consciousness according to the Glasgow Coma Scale was E4V1M5. Body temperature was 37.4°C, blood pressure was 118/67 mm Hg, and heart rate was 80 beats/min. She showed motor aphasia and right hemiplegia, and National Institutes of Health Stroke Scale score was 9.

Initial laboratory examination indicated a hemoglobin level of 8.5 g/dL, and a platelet count of $135 \times 10^3/\mu L$

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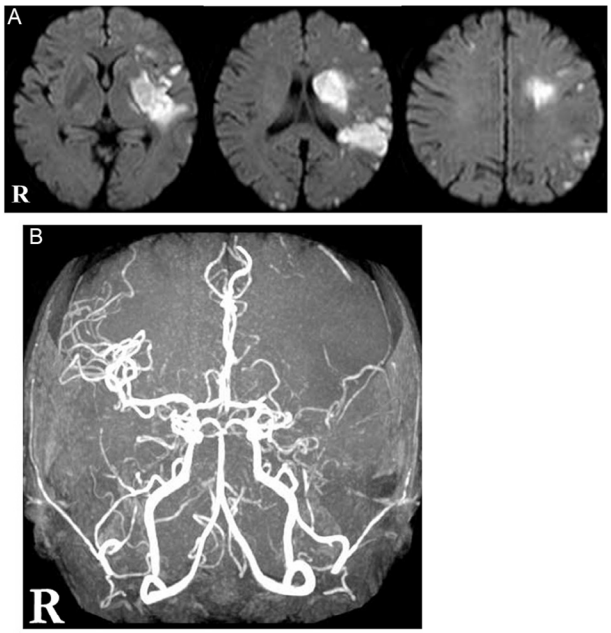

Figure 1. *Head MRI DWI and MRA: (A) Diverse embolic infarction is observed in the left MCA area. Cerebral infarction is also scattered in other areas. (B) Left MCA admits occlusion at M1 origin. Abbreviations: DWI, diffusion-weighted imaging; MCA, middle cerebral artery; MRA, magnetic resonance angiography; MRI, magnetic resonance imaging.*

and slightly elevated C-reactive protein (1.09 mg/dL), D-dimer (1.9 μg/mL), and markedly elevated serum CA125 concentration (901 units/mL; normal range, 0-35 units/mL), and CA19-9 level (1791 units/mL; normal range, 0-37 units/mL). Diffusion-weighted magnetic resonance imaging (MRI) showed high-intensity lesions in the territories of the right and left middle cerebral arteries (Fig 1, A). Magnetic resonance angiography revealed an occluded M1 segment of the left middle cerebral artery (Fig 1, B). Transesophageal echocardiography (TEE) revealed a mobile vegetation on the mitral valve (Fig 2). MRI of the pelvis revealed giant adenomyosis (10 cm × 10 cm) with multiple hemorrhages in the myometrium (Fig 3).

Post-Hospitalization Course

The patient had no history of heart disease or tooth extraction, and 2 sets of blood culture collected on the day of admission remained sterile. Based on the medical history, head MRI or magnetic resonance angiography, and TEE findings, cardiogenic cerebral embolism caused by NBTE was suspected. Continuous infusion of unfractionated heparin at 10,000 units/d was started from the first day of illness. We suspected NBTE associated with malignant tumor, and examined tumor markers,

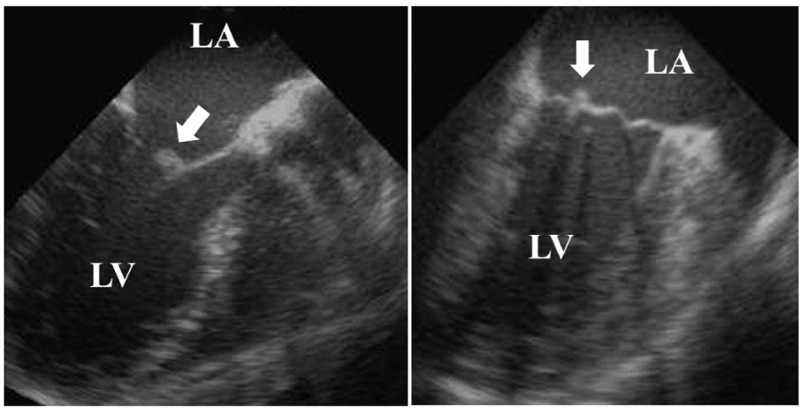

Figure 2. *TEE (day 1): A high echo structure (➡) with a size of about 4 mm movable is attached to the anterior cusp of the mitral valve. Abbreviations; LA, left atrium; LV, left ventricle; TEE, transesophageal echocardiography.*

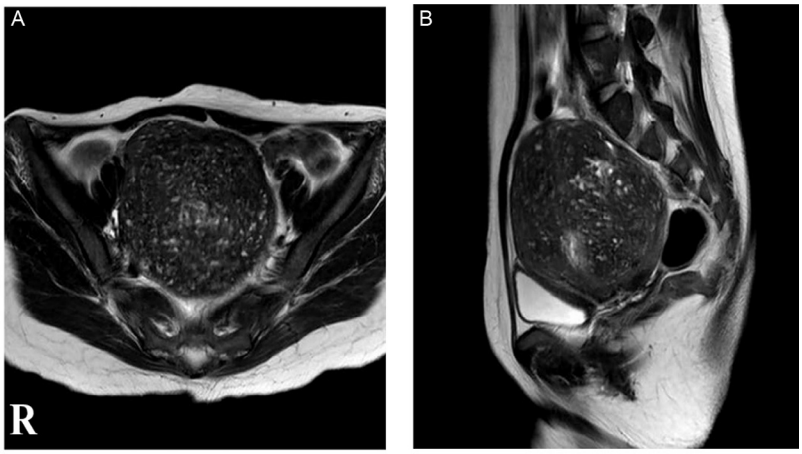

Figure 3. *Lower abdomen MRI T2 highlight image (day 2). 10 × 10 cm uterine adenomyosis with numerous small bleeding inside (A, horizontal cut; B, arrowhead). Abbreviation: MRI, magnetic resonance imaging.*