Case Studies

Cerebral Infarcts by Nonbacterial Thrombotic Endocarditis Associated with Adenomyosis: A Case Report

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We report a case of multiple embolic cerebral infarcts associated with nonbacterial thrombotic endocarditis (NBTE) in a patient with adenomyosis. The patient presented with dysarthria, left perioral sensory change, and left-hand weakness. Magnetic resonance imaging revealed multiple vascular territory infarctions involving the bilateral cerebellum and the right precentral gyrus. Magnetic resonance angiography was normal. D-Dimer, carbohydrate antigen (CA) 19-9, and CA125 levels were elevated. Abdominal and pelvic computed tomography with iodine contrast enhancement revealed a huge adenomyosis with left ovarian cyst. Transesophageal echocardiography (TEE) with agitated saline injection test demonstrated shaggy vegetation at the coapting edge of both mitral leaflets and mitral regurgitation. A diagnosis of NBTE was established and treatment with anticoagulation was initiated. Two weeks later, the thrombi reduced significantly on follow-up TEE and transthoracic echocardiography. The patient underwent an abdominal hysterectomy with bilateral salpingo-oophorectomy, and pathological specimens confirmed adenomyosis. The possibility that adenomyosis can be associated with NBTE suggests one of the underlying thromboembolic mechanisms in adenomyosis. Clinicians should be aware of the potential thromboembolic risk of adenomyosis. Further reporting of similar cases is needed to confirm the thromboembolic mechanism. **Key Words:** Adenomyosis—ischemic stroke—nonbacterial thrombotic endocarditis—anticoagulation.

Introduction

Adenomyosis is a condition in which heterotopic endometrial glands and stroma are present in the myometrium accompanied by adjacent smooth muscle hyperplasia. Arterial thromboembolisms, including cerebral arterial infarcts and thrombi in the brachiocephalic trunk and the subclavian artery have been reported in adenomyosis patients.[1] In addition to arterial thromboembolism, venous thrombosis manifesting in the cerebral dural venous sinus has also been identified in a patient with adenomyosis.[2] Given the widespread systemic thromboembolism with consistent negative results of echocardiography in previous reports, the authors speculate that the underlying mechanism of thromboembolism in adenomyosis is hypercoagulability rather than cardiogenic embolism.[1,2]

Nonbacterial thrombotic endocarditis (NBTE) is characterized by sterile vegetations composed of platelets and fibrin that adhere to the heart valves and are susceptible to embolization.[3,4] Patients with advanced malignancy and patients with systemic lupus erythematosus are the most common populations affected by NBTE,[5] but an association with adenomyosis has been reported very rarely.[6]

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Received July 18, 2017; revision received September 29, 2017; accepted October 8, 2017.

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1052-3057/$ - see front matter

https://doi.org/10.1016/j.jstrokecerebrovasdis.2017.10.005

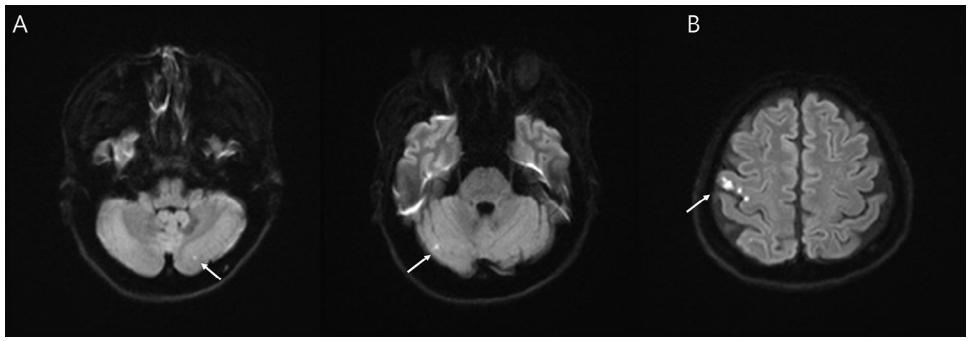

Figure 1. *Multiple vascular territory infarctions involving the bilateral cerebellum (A, arrows) and the right precentral gyrus (B, arrow) on diffusion-weighted brain magnetic resonance imaging.*

We report a rare case of multiple embolic cerebral infarcts caused by NBTE in a patient with adenomyosis.

Case Presentation

A 49-year-old woman presented to our hospital with a sudden onset of dysarthria, left perioral sensory change, and left-hand weakness. The woman's medical history included hypertension currently treated with a calcium channel blocker. She had a history of severe menorrhagia and dysmenorrhea 10 days before admission, which lasted for 4 days. At admission, the patient was not actively menstruating. On admission, the patient's vital signs were stable without fever. Brain diffusion-weighted magnetic resonance image revealed multiple vascular territory infarctions involving the bilateral cerebellum and the right precentral gyrus (Fig 1, A,B). Magnetic resonance angiography of intracranial and extracranial vessels from the common carotid artery to the circle of Willis demonstrated unremarkable findings. Chest computed tomography angiography with iodine contrast enhancement revealed no abnormal findings such as cancer, pulmonary artery fistula, or thrombus on the aortic arch, the subclavian artery, or the brachiocephalic artery. Abdominal and pelvic computed tomography with contrast enhancement showed a huge adenomyosis with left ovarian cyst (Fig 2), without any filling defect lesion reflecting a venous thrombus in the inferior vena cava or common iliac veins.

Hemoglobin level was 9.9 g/dL and the hematocrit level was 30.2%. The D-dimer level was 3.99 μg/mL (normal <1.0 μg/mL). Carbohydrate antigen (CA) 19-9 level was 69.2 U/mL (normal 0-35 U/mL) and CA125 level was 379 U/mL (normal 0-35 U/mL). Other laboratory studies were all normal, including platelet count, prothrombin time, partial thrombin time, antithrombin III level, fluorescent antinuclear antibody (FANA), rheumatoid factor, lupus anticoagulant antibody, anticardiolipin antibody, antineutrophil cytoplasmic antibodies, anti-Sjogren syndrome-related antigen type A or B, human leukocyte antigen (HLA) B27 and HLA B51, complement 3 and C4, C-reactive protein, factors V, VII, and VIII levels, factor V, Leiden gene, protein C activity, protein S activity, peripheral blood morphology, and blood culture.

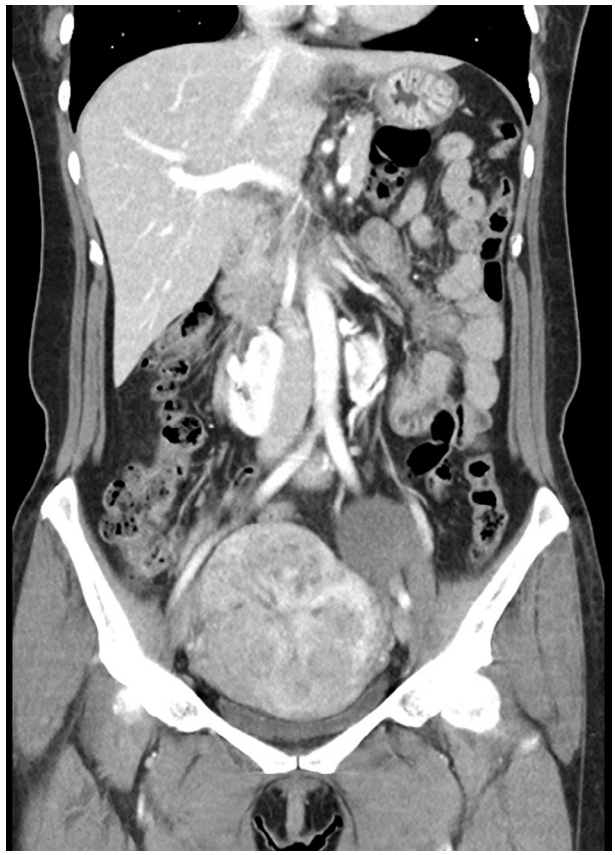

Figure 2. *Abdominal and pelvic computed tomography with contrast enhancement showed a huge adenomyosis with left ovarian cyst.*

Continuous 24-hour electrocardiography Holter monitoring showed unremarkable findings. Transthoracic echocardiography (TTE) and transesophageal echocardiography (TEE) with agitated saline injection test demonstrated shaggy vegetation at the coapting edge of both mitral leaflets and mitral regurgitation with no shunts, or patent foramen ovale (PFO) (Fig 3). TEE revealed no thrombus in the left atrium, left atrial appendage, right atrium, or descending thoracic aorta. Based on vegetation