Letter to the Editor

Arterial ischemic stroke in a patient with co-existence of antiphospholipid syndrome and ulcerative colitis

Soonchan Park, Chang-Woo Ryu

Department of Radiology, Kyung Hee University Hospital at Gangdong, College of Medicine, Kyung Hee University, Seoul, Republic of Korea

Correspondence to: Soonchan Park. Department of Radiology, Kyung Hee University Hospital at Gangdong, College of Medicine, Kyung Hee University, 892 Dongnam-ro, Gangdong-gu, Seoul 05278, Republic of Korea. Email: mdpark96@naver.com.

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Introduction

Antiphospholipid syndrome (APS) is a systematic autoimmune disorder characterized by vascular thrombosis, and pregnancy morbidity with antibodies against phospholipids or phospholipid-binding protein cofactors (1). Patients with inflammatory bowel disease (IBD) are at a higher risk of thrombosis and APS. We describe a case of arterial ischemic stroke in a patient with co-existence of APS and ulcerative colitis (UC), with varying imaging features during her hospital stay.

Case presentation

A 42-year-old woman presented to the emergency department with left-sided weakness, jerking movement at right arm and leg, and seizure movement 5 hours after symptom onset. She was admitted using intravenous (IV) midazolam, lorazepam due to presumable status epilepticus. Five years prior to presentation, she was diagnosed with moderately active UC and was irregularly taking mesalazine 1 g from once to three times daily. After admission, she had been given vancomycin and acyclovir as an empirical therapy for encephalitis. After cerebrospinal fluid (CSF) infection lab was found normal (protein: 37 mg/dL, glucose: 54 mg/dL), these antibiotics and antiviral agent were discontinued. After that, valproate and levetiracetam was administered as an antiepileptic drug.

Initial magnetic resonance imaging (MRI) revealed right middle cerebral artery (MCA) territorial infarction. Further, magnetic resonance angiography revealed moderate focal stenosis of the right supraclinoid internal carotid artery (ICA) (*Figure 1*). Based on these imaging findings, vasculitis related to UC was suggested as a first diagnosis and digital subtraction angiography (DSA) was planned. Three days later, DSA revealed occlusion of the right supraclinoid ICA, with the right MCA territory filled by the left ICA, through the anterior communicating artery. Contrast-enhanced T1-weighted MRI revealed enhancement of the right supraclinoid ICA wall (*Figure 2*). After all these imaging, IV steroid pulse therapy (methylprednisolone 1 g) was administered as a medication for vasculitis related infarction. One month later, follow-up MRI revealed a new right occipital infarction and recanalized right supraclinoid ICA, with right MCA and anterior cerebral artery stenosis (*Figure 3*).

Due to change of vascular status and a new infarction on imaging, further laboratory examination was done yielding positive results for anti-phospholipid immunoglobulin G (IgG) (15.7 IgG phospholipid units per milliliter (GPL/mL), normal range <10.0 GPL/mL) and anti-cardiolipin IgG (14.7 GPL/mL, normal range <10.0 GPL/mL). The patient showed negative results to systemic lupus erythematosus tests [anti-double stranded deoxyribonucleic acid antibody (Ab), 6.0 units per milliliter (U/mL); anti-Smith Ab, 1.1 U/mL; antinuclear ribonucleoprotein Ab, 1.2 U/mL; anti-Sjogren syndrome A Ab, 0.2 U/mL; Anti-Sjogren syndrome B Ab, 0.6 U/mL]. Based on these clinical findings, she was finally diagnosed with arterial stroke due to co-existence of APS and UC.

All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research committee(s) and with the Helsinki

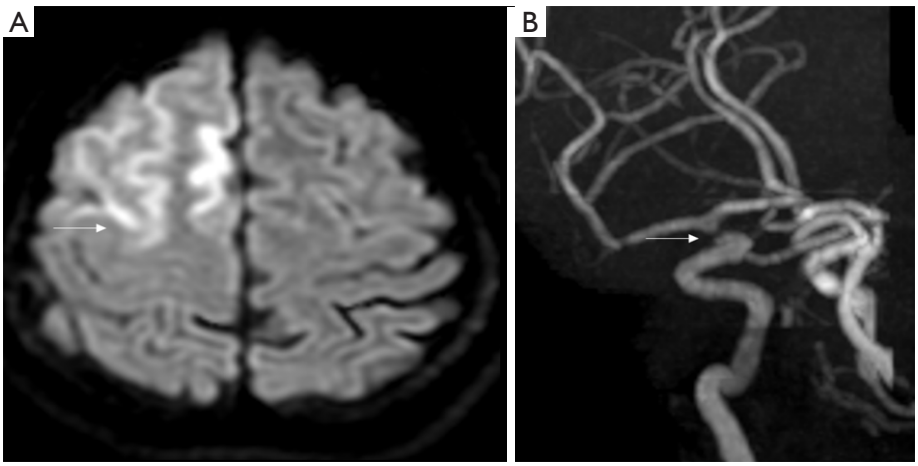

Figure 1 Initial magnetic resonance imaging of the patient. (A) Diffusion-weighted imaging showing a diffusion-restricted lesion in the right frontal lobe (arrow). (B) Magnetic resonance angiography showing severe focal stenosis of the right supraclinoid internal carotid artery (arrow).

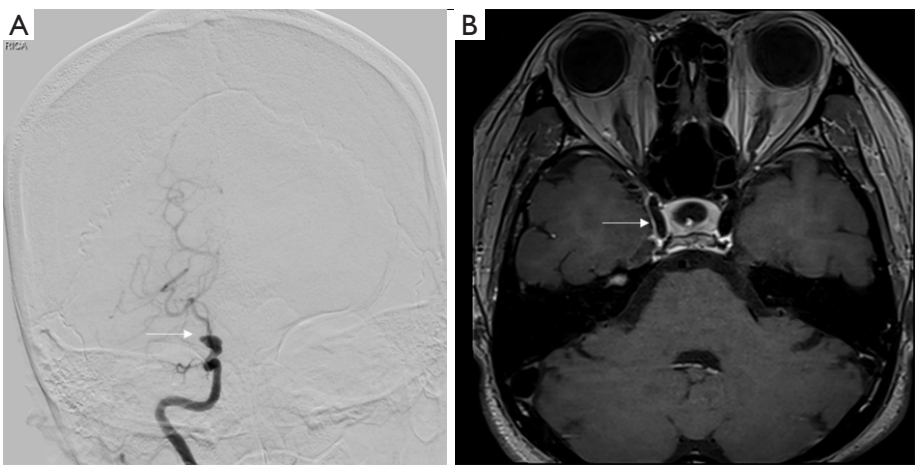

Figure 2 Imaging of the patient three days later. (A) Digital subtraction angiography showing complete occlusion of right supraclinoid internal carotid artery (arrow). (B) High-resolution T1-weighted imaging showing concentric vessel wall enhancement of the right supraclinoid internal carotid artery (arrow).

Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

Discussion

APS is an autoimmune condition characterized by arterial and/or venous thrombosis, and ischemic stroke is the most frequent neurological manifestation in antiphospholipid antibody (APL)-positive patients. In a recent study, IBD was found to be highly correlated with APLs (2). IBD patients have a higher risk of developing thromboembolic complications (1–7.7% of cases), particularly in the active phase of the disease (3). Cerebral arterial thrombosis in patients with UC has been reported (4). There are reports on association between IBD and APS (2). However, there are no reports showing cerebrovascular imaging in a patient with stroke due to co-existence of APS and UC. Patients with IBD have an increased risk of thrombotic events due to the disease per se and/or co-occurrence of other conditions

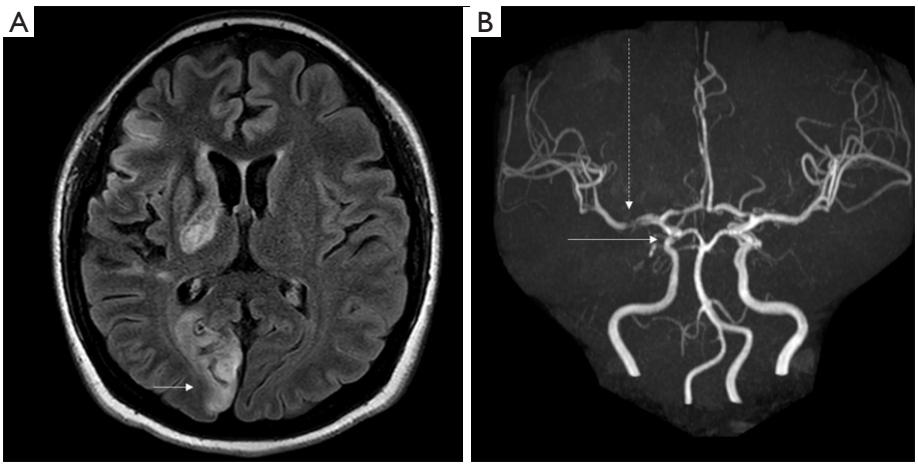

Figure 3 Follow-up imaging of the patient after one month. (A) Fluid-attenuated inversion recovery imaging showing a new right occipital ischemic lesion (arrow). (B) Magnetic resonance angiography showing recanalization of the supraclinoid internal carotid artery (arrow) with focal stenosis of the middle cerebral artery (dashed arrow).

responsible for pro-thrombotic state. Many factors responsible for cerebral thrombotic events are not identified or may not be present at all among IBD patients. Moreover, frequently only discreet symptoms of such conditions may be manifested. So imaging evaluation can be helpful in those patients as in our case (5,6).

APS is usually managed with life-long anticoagulant or antiplatelet medication. However, tailored therapy may be determined on an individual basis (7). There could be some specific type of APS depending on APL. Isolated immunoglobulin M (IgM)-APS is more associated with stroke but relapse rate of Isolated IgM-APS with that of non-isolated IgM-APS was similar (8).

This study uniquely highlights several points. First, our case demonstrates how arterial involvement in the active phase of APS can change and progress over time. Initial imaging revealed a right MCA territorial infarction with supraclinoid ICA stenosis. Three days later, DSA revealed supraclinoid ICA occlusion, which was visualized as a vessel wall enhancement on MRI. This finding may reflect the active phase of the disease. On follow-up 1 month later, the supraclinoid ICA spontaneously was recanalized with a new infarction at right PCA territory. Second, previous reports described cerebral arterial thrombosis in young patients with UC. However, to our knowledge, there were no reports on imaging evaluation in patients with co-existence of APS and UC. This is the first report to highlight a case of an arterial ischemic stroke with imaging findings in a patient with co-existence of APS and UC.

Conclusions

We present a patient with an arterial stroke in which the patient's symptoms changed, supported by concordant changes in imaging findings because of disease progression in co-existence of APS and UC.

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Footnote

Conflicts of Interest: Both authors completed the ICMJE uniform disclosure form (available at https://qims.amegroups.com/article/view/10.21037/qims-22-429/coif). SP reports that this work was supported by a grant from the National Research Foundation of Korea in 2017 (No. 2017R1C1B5076919). The other author has no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work, ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All procedures performed in this study were in accordance with the ethical standards of the institutional and/or national research

committee(s) and with the Helsinki Declaration (as revised in 2013). Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the editorial office of this journal.

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