Case Report
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Does Systemic Lupus Erythematosus Increase the Risk of Procedure-Related Complication in Endovascular Treatment of Intracranial Aneurysm?

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Cerebral aneurysms associated with systemic lupus erythematosus (SLE) are more likely to grow rapidly and rupture, compared to those found in the general population. The main underlying pathology of intracranial aneurysm and its rupture is presumed to be SLE-related intracranial vasculitis and fragility of blood vessels due to prolonged use of steroid. For these reasons, both surgical and endovascular options are challenging. On the other hand, given the possibility that SLE may predispose to growth and rupture of intracranial aneurysm, early intervention for cerebral aneurysms associated with SLE may be more necessary and beneficial than other cerebral aneurysms in the general population. Here we would like to report on the unexpected complications that occurred during or after endovascular treatment of an SLE patient with multiple aneurysms. The complications include intraprocedural rupture of unruptured aneurysm, coil stretching, contrast-induced encephalopathy, and delayed ipsilateral intraparenchymal hemorrhage after stent-assisted coiling. Our unique case highlights that the SLE patient with multiple intracranial aneurysms had a higher risk of endovascular procedure-related complications, which might be due to the increased bleeding tendency and fragility of blood vessels.

Key Words: Cerebral aneurysm, endovascular treatment, systemic lupus erythematosus

INTRODUCTION

Systemic lupus erythematosus (SLE) is an autoimmune chronic multisystemic inflammatory disease of unknown etiology. The main underlying pathology of intracranial aneurysm and its rupture is presumed to be SLE-related vasculitis and fragility of blood vessels due to prolonged use of steroid. Arterial inflammation causes lumen vessel narrowing and cerebral flow reduction, leading to ischemia and hemodynamic stress, which are cofactors for aneurysmal genesis.[1,2] Therefore, patients who have undergone successful clipping of intracranial aneurysms often experience complications of SLE. Here we would like to report on the unexpected complications that occurred during or after endovascular treatment (EVT) of an SLE patient with multiple aneurysms.

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CASE REPORT

The patient's informed consent was obtained for this case report. A 44-year-old woman with hypertension and a 17-year history of SLE was referred to our department with fluctuant left ptosis, chemosis, and ocular pain for 2 months. At the time of hospitalization, she met four criteria of the 2012 Systemic Lupus Collaborating Clinics criteria for SLE, including chronic cutaneous lupus erythematosus, positive ANA titer, positive anti-dsDNA Ab on two occasions, and positive antiphospholipid Ab.[3] Recent SLE activity was mild, and the patient had been taking low-dose oral prednisone as maintenance therapy. Brain magnetic resonance angiography revealed a 7.0 mm-sized an-

eurysm of the left anterior choroidal artery. Relapse and remission of Tolosa-Hunt syndrome resulting from that aneurysm was assumed to be the cause of the present symptoms. After loading 300 mg of clopidogrel and 300 mg of acetylsalicylic acid at 12 hours before the procedure, EVT was performed under intravenous sedation. Intraprocedural aneurysmal rupture was identified by the angiographic visualization of contrast agent extravasation without a rapid change in her vital signs (Fig. 1). However, the angiographic evidence of coil perforation was not observed. To prevent continuous leaking of contrast material, protamine sulfate was given for heparin reversal and additional coils were deployed for dome protection. After the entire procedure, both the condition and computed tomography (CT) finding of the patient were intact, indicating that intraprocedural aneurysmal rupture led to minimal extravasation of blood, fortunately. The postoperative course was uneventful, and the patient was maintained on dual antiplatelet therapy.

A month after the previous procedure, the patient was readmitted to our hospital for coiling of another aneurysm located on the right paraclinoid internal carotid artery (ICA). During

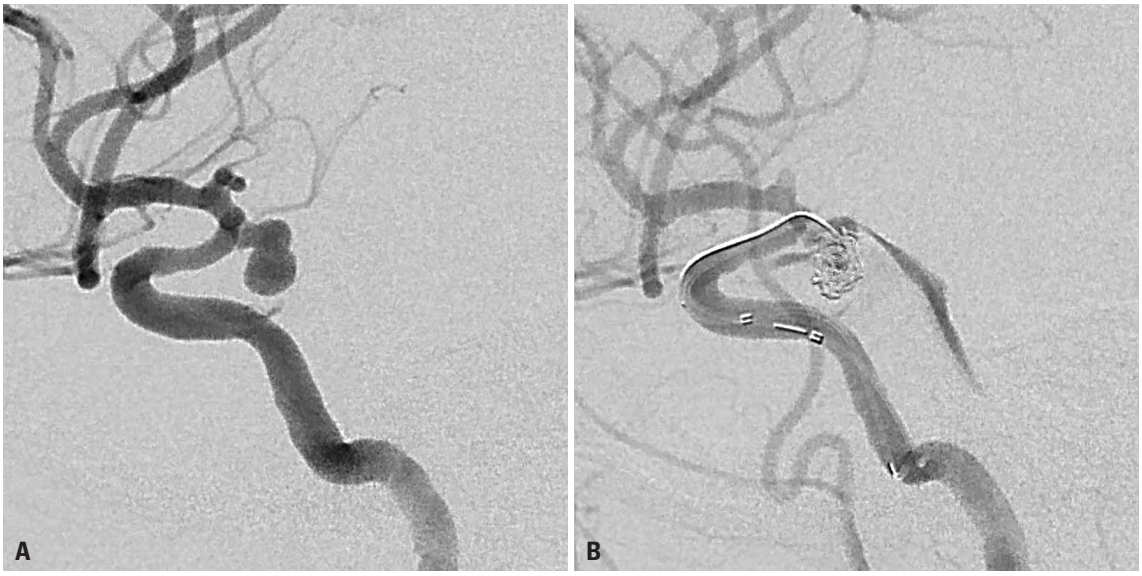

Fig. 1. (A) Left internal carotid artery angiography showing a symptomatic unruptured aneurysm at the origin of the left anterior choroidal artery. (B) Leak of contrast agent surrounding aneurysm neck indicates intraprocedural aneurysm rupture.

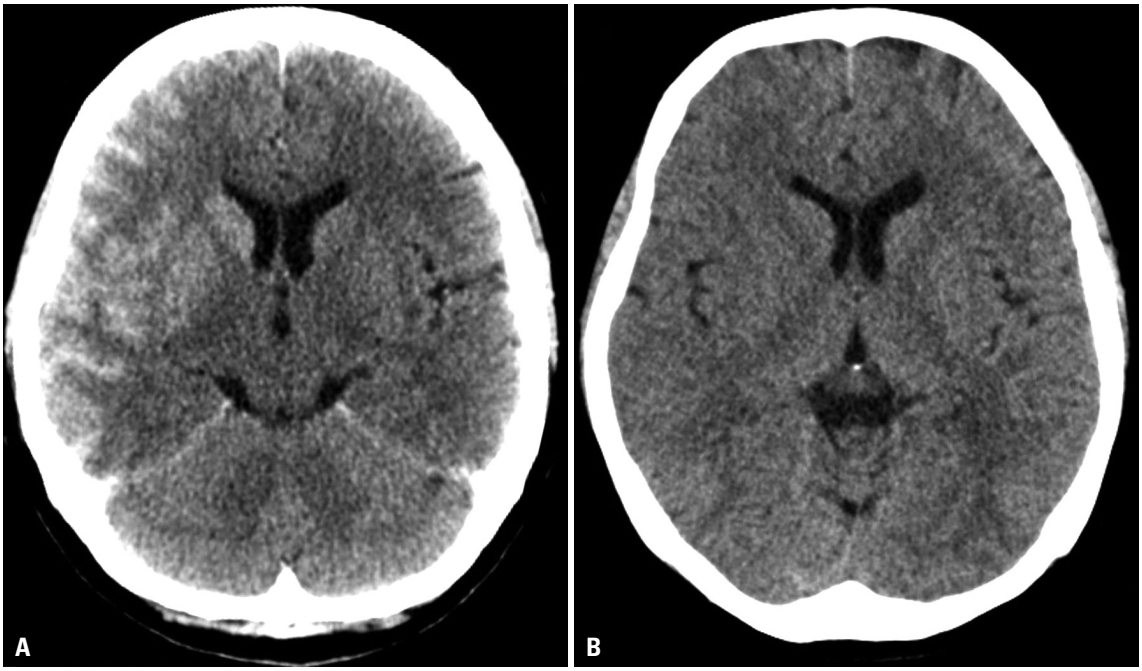

Fig. 2. (A) Postangiographic brain computed tomography showing marked enhancement throughout the right cerebral cortex and right basal ganglia, indicative of contrast-induced encephalopathy. (B) Repeat brain computed tomography obtained after 24 hours demonstrating no enhancement on cerebral cortex and basal ganglia.

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right ICA angiography, just before coiling, the patient developed rapidly worsening confusion and left hemiparesis on the table. Right ICA angiography revealed no abnormal findings. A total of 40 mL of Iopromide (Ultravist 300, Bayer Healthcare Pharmaceuticals, Wayne, NJ, USA), a low osmolar non-ionic contrast agent, was administered and the procedure was abandoned. Non-contrast CT showed marked enhancement throughout the right cerebral cortex and right basal ganglia with concordant diffuse swelling of the right cerebral hemisphere, which are indicative of contrast-induced encephalopathy (CIE) (Fig. 2A). Magnetic resonance imaging was then performed to exclude the possibility of other mimicking conditions, including an acute ischemic complication, and it showed no abnormal findings. Twenty-four hours later, repeat brain CT scan revealed no enhancement on cerebral cortex and basal ganglia (Fig. 2B). The patient gradually recovered after supportive care, and was discharged after 3 days with a modified Rankin scale score of 0.

Despite concerns about the side effect of contrast material, EVT was retried for the treatment of the aneurysm 3 months later. To prevent contrast-induced adverse reaction, another type of contrast medium (iobitridol, Xenetix® 300, Guerbet, Sulzbach, Germany) was selected, and adequate hydration was performed with intra-arterial saline infusion and intravenous fluids during and before the procedure. The procedure using stent-assisted coiling was uneventful, and the patient was well-recovered; however, remote intracerebral hemorrhage (ICH) occurred after 2 days with a generalized tonic-clonic seizure (Fig. 3). Hematoma was well-absorbed after conservative treatment. However, the patient experienced two more seizure attacks despite taking anti-epileptic medication, and she is currently on anti-epileptic medication without any neurological deficits.

DISCUSSION

Cerebral aneurysms associated with SLE are more likely to grow rapidly and consequently rupture, compared to cerebral aneurysms in the general population.[4] Therefore, when an aneurysm is present and requires occlusion in patients with SLE, it should be considered for more aggressive treatment, especially if a growth or related symptom of aneurysm is observed, as in our case.

Therapeutic management of intracranial aneurysm and subarachnoid hemorrhage in these patients is not well-established and depends highly on the patient's clinical condition. Successful surgical clipping and EVT of aneurysm have been previously reported in patients with SLE.[4-7] However, unexpectedly fragile intracranial arterial walls due to multifocal disease spread make them difficult to handle, and could prompt a rupture during both surgical and endovascular procedures. In the case of this study, intraprocedural aneurysmal rupture and delayed ipsilateral ICH that occurred during or after aneurysm coiling are assumed to be related to lupus vasculitis and fragility of blood vessels due to prolonged use of steroid, although the patient was found to have neither hematologic abnormalities nor angiographically proven vasculitis at the time. The incidence of remote ICH after EVT of unruptured intracranial aneurysms has been reported as 0.46%.[8] Another study reported that the incidence of remote ICH after stent-assisted coiling of intracranial aneurysms was 2.2%.[9] This event occurred mostly in patients with stents, hypertension, and unruptured intracranial aneurysms on the ICA.[8] Our patient had all of the risk factors mentioned above. This case suggests that SLE may also predispose to this complication after aneurysm coiling. Therefore, in order to avoid these hemorrhagic complications, it seems necessary to limit the use of stents and antiplatelet medication in EVT for SLE patients.

Iodinated CIE is a rare complication of angiography. Iodinated contrast media are reported to temporarily disrupt the blood-brain barrier, causing an encephalopathy that is usually self-limiting. Based on current knowledge, this complication appears to be an idiosyncratic reaction to contrast.[10] This makes avoidance of CIE challenging. Although it is difficult to establish a

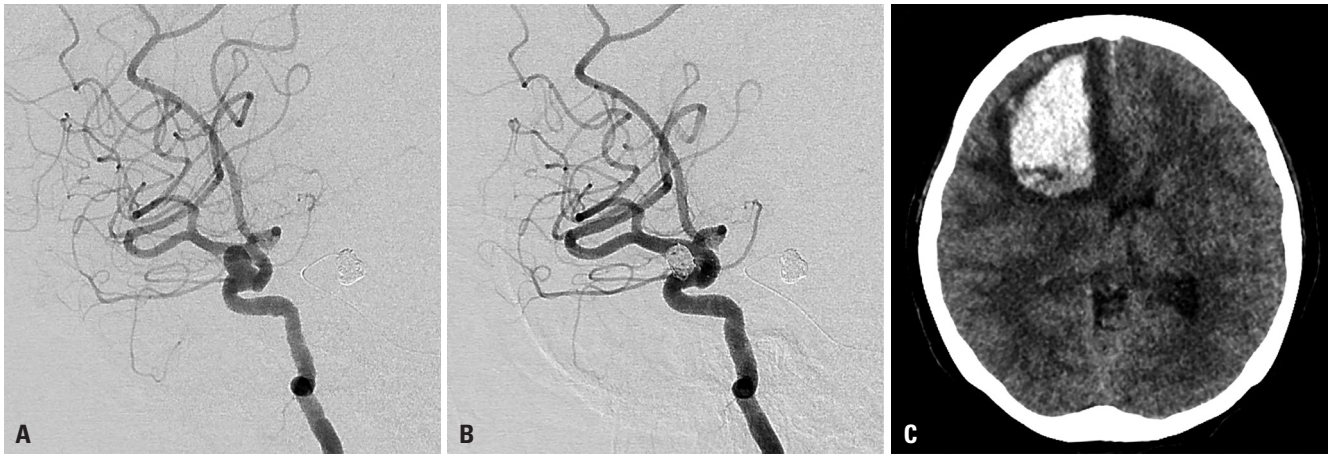

Fig. 3. (A and B) Right ICA angiographies before and after endovascular treatment showing another aneurysm located on the right paraclinoid ICA and uneventful stent-assisted coiling. (C) Non-contrast computed tomography showing a right frontal hematoma. ICA: internal carotid artery.

direct correlation between SLE and CIE, SLE pathology may have contributed in part to the occurrence of this complication, which should be always considered and prepared for.

Based on our experience, when a cerebral aneurysm associated with SLE requires endovascular occlusion, increased risk of procedural complication should be noted and high degree of individual tailoring will be demanded.

AUTHOR CONTRIBUTIONS

Conceptualization: Chang Hwa Choi. Data curation: Jun Kyeung Ko. Formal analysis: Sang Weon Lee. Funding acquisition: Jun Kyeung Ko. Investigation: Chang Hwa Choi. Methodology: Chang Hwa Choi. Project administration: Jun Kyeung Ko. Resources: Jun Kyeung Ko. Software: Jung Hwan Lee. Supervision: Sang Weon Lee. Validation: Sang Weon Lee. Visualization: Jung Hwan Lee. Writing—original draft: Jun Kyeung Ko. Writing—review & editing: Jung Hwan Lee. Approval of final manuscript: all authors.

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