Rupture of an infectious intracranial aneurysm involving two parent arteries after surgical treatment of infective endocarditis

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Infectious intracranial aneurysms (IIAs) are uncommon and potentially devastating neurological complications of infective endocarditis (IE). The natural history of IIAs remains unclear. Although rupture of IIAs could occur as the initial manifestation of IE, or even weeks to months later after adequate treatment of IE, there have been few reported ruptures of IIAs during the perioperative period in patients who underwent valve replacement.1,2 We present an unusual patient who had IE complicated by rupture of an IIA 2 weeks after mitral valve replacement. The unique IIA aneurysm was fed by 2 distal branches of the MCA after mitral valve replacement. The unique IIA aneurysm was fed by 2 distal branches of the MCA, which were both clipped successfully. Our objective in presenting this particular case is to emphasize the importance of recognizing the risk of IIAs following embolic events, and the risk of aneurysmal rupture during the perioperative period of cardiac surgery in patients with IE.

Case Report. A 23-year-old male with a one-month history of cough and intermittent mild fever presented with the sudden onset of right hemiparesis. He was sent to a local hospital, where MRI scans revealed a temporoparietal lesion consistent with cerebral ischemia (Figure 1). Contrast MRI did not show abnormal enhancement. Echocardiography showed a vegetation on the mitral valve as well as a ruptured chordae tendineae and mitral regurgitation. Chest CT showed bilateral moderate pleural effusion and cardiac enlargement.
Staphylococcus aureus was subsequently isolated from the blood samples. The diagnosis of IE was made, and he was treated with antibiotics. Repeated echocardiograms showed the worsening of mitral regurgitation during 2 weeks of medical treatment. He was then transferred to our hospital, and he underwent urgent mitral valve replacement after admission. The postoperative course in hospital was uneventful. His hemiparesis improved rapidly without any new neurological signs, and subsequent blood cultures were negative. He was discharged 10 days after surgery on continuous oral anticoagulant and antibiotics. Unfortunately, he suffered the sudden onset of right hemiparesis with dysarthria 4 days after release. He returned to our hospital, and CT revealed a large temporoparietal hematoma (Figure 2a). On his admission, neurologic examination revealed

Figure 1 - Noncontrast T2-weighted magnetic resonance imaging shows a high-signal-intensity lesion in the left temporoparietal lobe (arrows) (A & B), suggesting an ischemic lesion involving the territory of the left middle cerebral artery.

Figure 2 - Preoperative computed tomography (CT) and CT angiography showing A) Non-contrast CT shows a temporoparietal hematoma (arrow). B) Contrast CT shows a well-defined lesion (arrow) with homogenous contrast-enhancement in the temporal lobe. C) CT angiography reveals a distal middle cerebral artery (MCA) aneurysm inferior to the hematoma (arrows). D) Oblique view of the CT angiography shows 2 adherent aneurysmal sacs respectively originating from 2 adjacent distal branches (arrows) of the left MCA.
dysarthria and complete hemiplegia in his right limbs (grade 0 in upper and lower limbs). Contrast CT revealed a homogeneously enhanced lesion with well-defined margins adjacent to the hematoma (Figure 2b), which was shown to be an aneurysm originating from 2 adjacent distal branches of the left MCA (Figures 2c & d). The patient and his family did not accept the conventional angiogram. Coagulation studies at that time revealed a prolonged prothrombin time and an activated partial thromboplastin time. Repeated CT showed no enlargement of the hematoma, so delayed surgical procedure was chosen considering the benefit of evacuation of the hematoma. He was treated with fresh frozen plasma and vitamin K. Three days later, he underwent temporoparietal craniotomy. After the distal sylvian fissure was opened, dissection was performed proximal to distal to expose the aneurysm. The aneurysm seemed to have 2 sacs adherent closely to one another. Two parent arteries were identified and the aneurysmal sacs were confirmed to be communicating. Direct clipping was successfully performed using 2 clips. Subsequent CT angiography (Figure 3) revealed complete disappearance of the aneurysm and patency of the 2 parent arteries. He recovered well after surgery, and was subsequently transferred to a rehabilitation hospital. At the latest follow-up (15 months), his right hemiplegia resolved completely in the lower limbs, and power in the upper limbs recovered to grade 3. Repeated CT angiography revealed complete occlusion of the aneurysm.

**Discussion.** Neurologic complications occur in up to 40% of patients with IE, which most commonly result from septic embolization. The IIAs tend to occur secondary to septic embolization. Although IIAs were reported in only 2-4% of patients with IE, they produce potentially devastating neurologic complications. In a previous report, a focal deficit consistent with embolism was found to be the most common prodrome of the IIAs. Therefore, any neurological symptom should raise the suspicion for an intracranial aneurysm and may lead to further diagnostic evaluation. The time course for development of IIAs following embolism is uncertain. An experimental study suggested that IIAs usually form within 48 hours, so angiography was suggested 48 hours after the initial neurological symptoms. In the present case, the location of the aneurysm was consistent with the involved vessels from the initial ischemic distribution. Because there were no diagnostic tests, the IIA in our patient was not detected until it ruptured, as in some reported cases.

Compared with saccular aneurysms, aneurysms associated with IIAs are more often multiple, and a peripheral location, poorly-defined neck, irregular outline, and fusiform shape are more frequently found in IIAs. It is very unusual that 2 adjacent branches of the MCA were involved in this aneurysm. Though the exact pathophysiology of the special aneurysm is not known, we presume multiple vessel involvement is associated with extravascular spread of infection among adjacent vessels.

Management of unruptured IIAs remains controversial because there have been no randomized controlled trials to guide clinical decision-making. When considering treatment, the most important factor is whether the aneurysm has ruptured. Unruptured IIAs might be initially managed with medical therapy and serial follow-up angiography. A high proportion of such aneurysms were reported to respond to antibiotic therapy and resolve completely without the necessity of a surgical or endovascular procedure. The greatest risk of medical treatment is aneurysmal rupture during therapy. However, it is impossible to predict the response of these aneurysms to medical treatment. Aneurysms
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that enlarge or exhibit dynamic morphological features during follow-up may require surgical and endovascular procedures.\(^2,3\)

When cardiac surgery, such as valve replacement, is indicated in patients with unruptured IIAs, the safety of intraoperative cardiopulmonary bypass and long-term anticoagulation after surgery must be considered. Although there have been few cases reported of aneurysmal rupture during the perioperative period of valve replacement, most authors recommended definitive treatment of the aneurysms before cardiac surgery.\(^1,6,7\) In our patient, intracerebral hemorrhage due to aneurysmal rupture occurred 2 weeks after successful valve replacement, which might be related to intraoperative and postoperative anticoagulation therapy. Our case supports the notion that aneurysmal rupture is a risk during the perioperative period of cardiac surgery. Definitive neurosurgical or endovascular therapy has generally been used for treatment of ruptured IIAs. Recently, great advances have been achieved in endovascular strategies and tools, which make treatment of IIAs safe and effective using endovascular treatment.\(^2,8,9\) However, one of the major limitations of endovascular treatment for ruptured IIAs is the inability of surgery to evacuate an adjacent hematoma and to prevent increased intracranial pressure. Moreover, for IIAs that involve parent arteries for eloquent regions of the brain, surgical management has a better rate of parent artery preservation or revascularization than does endovascular management.\(^10\) Considering the hematoma and the potential loss of multiple parent arteries in our patient, the decision to perform surgery was preferred and direct clipping was successfully carried out.

In summary, our case illustrates a rare rupture of an untreated IIA during the perioperative period of cardiac surgery. This case is even more unusual in that the aneurysm was fed by 2 adjacent distal branches of the left MCA. Our experience with this patient emphasizes the risk of IIAs following embolic events and neurological symptoms, as well as risk of aneurysmal rupture during the perioperative period of cardiac surgery in patients with IE. Therefore, we recommend definitive treatment of unruptured IIAs before cardiac surgery, especially in patients who require long-term anticoagulation.

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**References**


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