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

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ABSTRACT

تلخص هذه الدراسة مدى التحدي الذي يواجه عملية معالجة تمزق أم الدم الخمجي داخل تجويف القحف وذلك في المرضى الذين خضعوا لعملية استبدال الصمام في القلب. وبالرغم من إزدياد خطر الإصابة بتمزق أم الدم بعد إجراء عملية القلب إلا أن عدد الحالات التي تم الإبلاغ عنها والتي تثبت حدوث تمزق أم الدم في الفترة المحيطة بجراحة استبدال الصمام كان قليلاً. نستعرض في هذا المقال حالة مُصاب بنزيف في الدماغ إثر تمزق أم الدم داخل تجويف القحف وذلك بعد مرور أسبوعين من عملية استبدال الصمام المترالي. ولقد كانت أم الدم تمد بالدم من قبل فرعين من الشرايين في الجزء المتوسط الأيسر من الدماغ. وتمت معالجة التمزق بإجراء عملية إرقاء مباشرة للأوعية النازفة (التشبيك) وذلك من خلال استخدام مشبكين مع الحفاظ على الشرايين الأم. تثبت هذه الدراسة أنه بالرغم من ندرة حدوث مثل هذه الحالات إلا أن تمزق أم الدم داخل القحف والذي لم يتم علاجه قد يحدث في الفترة المحيطة بجراحة القلب، ولهذا ومن أجل سلامة المرضى ننصح بوضع خطة علاجية محددة للتغلب على هذا التمزق قبل حدوثه وذلك قبل جراحة القلب وخصوصاً إذا كان هناك حاجة لإعطاء مضادات تخثر الدم بعد الجراحة.

Management of patients with infectious intracranial aneurysms (IIAs) who require valve replacement remains a challenge. Although there is potential risk of aneurysmal rupture associated with cardiac surgery, there have been few reported ruptures of IIAs during the perioperative period of valve replacement. We present a unique patient who suffered intracerebral hemorrhage due to rupture of an IIA 2 weeks after mitral valve replacement. This unique aneurysm is fed by 2 adjacent branches of the left middle cerebral arteries. Direct clipping of the aneurysm was successfully performed using 2 clips with preservation of the parent arteries. This case demonstrates that although it is rare, rupture of an untreated IIA might occur during the perioperative period of cardiac surgery. For patient safety, definitive treatment of unruptured IIAs is recommended before cardiac surgery, especially when long-term anticoagulation is needed after surgery.

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Infectious intracranial aneurysms (IIAs) are uncommon and potentially devastating neurological complications of infectious 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 of the left distal middle cerebral arteries (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

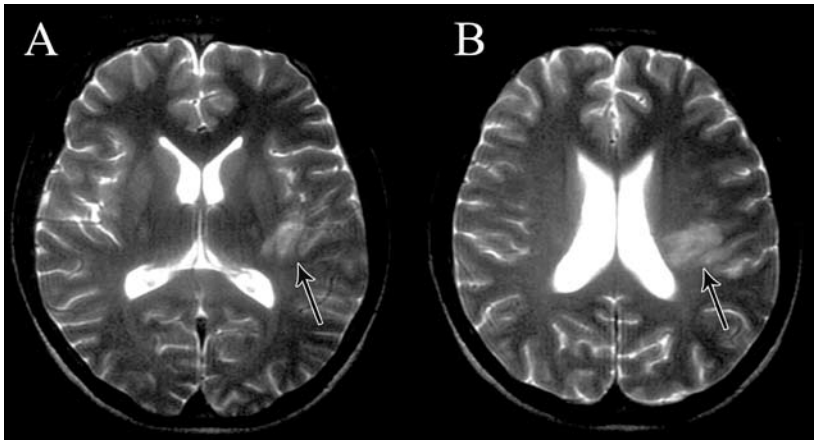

Figure 1 - Noncontrast T2-weighted magnetic resonance imaging shows a high-signal-intensity lesion in the left temporoparietal lobe (arrows) (**A & B**), suggesting an ischemic lesion involving the territory of the left middle cerebral artery.

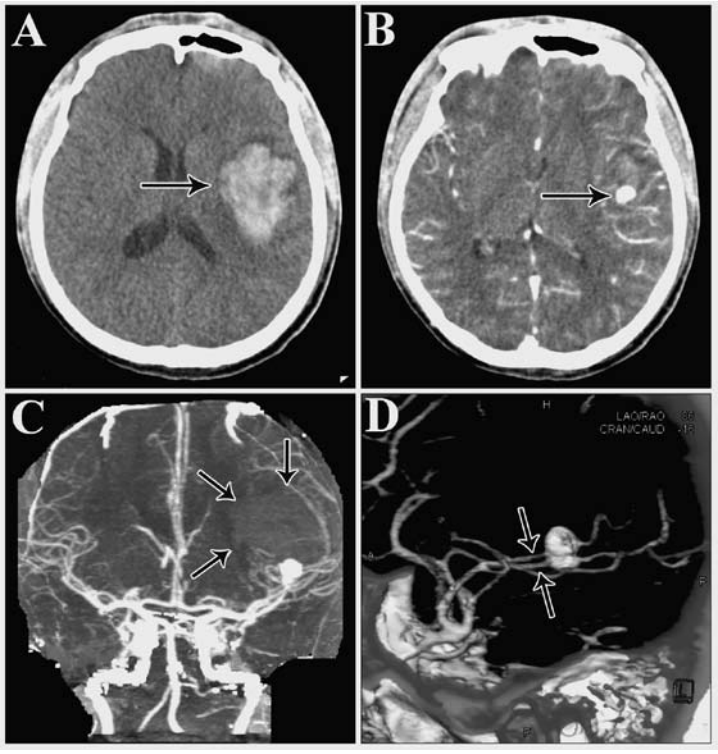

Figure 2 - Preoperative computed tomography (CT) and CT angiography showing **A**) Non-contrast CT shows a temporoparietal hematoma (arrow). **B**) Contrast CT shows a well-defined lesion (arrow) with homogenous contrast-enhancement in the temporal lobe. **C**) CT angiography reveals a distal middle 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.

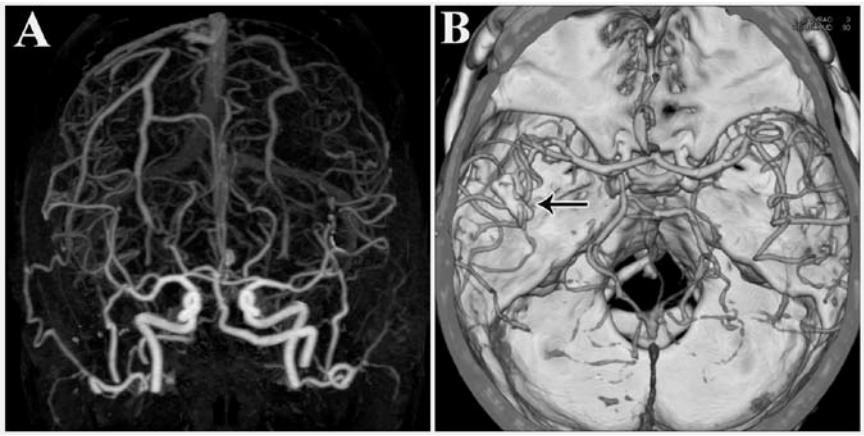

Figure 3 - Postoperative CT angiography shows the occlusion of the aneurysms and the patency of the branches of the left middle cerebral artery (**A & B**). Two clips could also be seen (arrow).

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.[1] 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 neurological complications.[3] In a previous report,[4] 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.[3] 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.[4] 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.[4,5]

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.[2,3] 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.[3]

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.[2,3] 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.[2] Aneurysms

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

1. Gillinov AM, Shah RV, Curtis WE, Stuart RS, Cameron DE, Baumgartner WA, et al. Valve replacement in patients with endocarditis and acute neurologic deficit. ***Ann Thorac Surg*** 1996; 61: 1125-1129.
2. Kannoth S, Thomas SV. Intracranial microbial aneurysm (infectious aneurysm): current options for diagnosis and management. ***Neurocrit Care*** 2009; 11: 120-129.
3. Peters PJ, Harrison T, Lennox JL. A dangerous dilemma: management of infectious intracranial aneurysms complicating endocarditis. ***Lancet Infect Dis*** 2006; 6: 742-748.
4. Salgado AV, Furlan AJ, Keys TF. Mycotic aneurysm, subarachnoid hemorrhage, and indications for cerebral angiography in infective endocarditis. ***Stroke*** 1987; 18: 1057-1060.
5. Eddleman C, Nikas D, Shaibani A, Khan P, Dipatri AJ Jr., Tomita T. HydroCoil embolization of a ruptured infectious aneurysm in a pediatric patient: case report and review of the literature. ***Childs Nerv Syst*** 2007; 23: 707-712.
6. van de Beek D, Rabinstein AA, Peters SG, Cloft H, Wijdicks EF. *Staphylococcus endocarditis* associated with infectious vasculitis and recurrent cerebral hemorrhages. ***Neurocrit Care*** 2008; 8: 48-52.
7. Derex L, Bonnefoy E, Delahaye F. Impact of stroke on therapeutic decision making in infective endocarditis. ***J Neurol*** 2010; 257: 315-321.
8. Wang H, Rammos S, Fraser K, Elwood P. Successful endovascular treatment of a ruptured mycotic intracavernous carotid artery aneurysm in an AIDS patient. ***Neurocrit Care*** 2007; 7: 156-159.
9. Eddleman CS, Surdell D, DiPatri A, Jr., Tomita T, Shaibani A. Infectious intracranial aneurysms in the pediatric population: endovascular treatment with Onyx. ***Childs Nerv Syst*** 2008; 24: 909-915.
10. Nakahara I, Taha MM, Higashi T, Iwamuro Y, Iwaasa M, Watanabe Y, et al. Different modalities of treatment of intracranial mycotic aneurysms: Report of 4 cases. ***Surg Neurol*** 2006; 66: 405-409.

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Farag A, Al-Yamany M. Surgical clipping of ruptured anterior circulation cerebral aneurysms: KFMC experience. ***Neurosciences (Riyadh)*** 2008; 13 (Suppl): 32.

Jabbour RMD, Khalifeh RMD, El-Kutoubi AMD, Atweh S. Dissecting aneurysm of the basilar trunk in a young man with CNS brucellosis. ***Neurosciences (Riyadh)*** 2003; 8 (Suppl): 59.

Sami AS. Aneurysm coiling in sub-arachnoid hemorrhage. Two years Egyptian experience and follow-up. ***Neurosciences (Riyadh)*** 2003; 8 (Suppl): 45.