



Management of Consecutive Development of Ruptured Intracranial Mycotic Aneurysms: Case Report

Arka Arkaya Gelişen Rüptüre İntrakraniyal Mikotik Anevrizmaların Takibi: Olgu Sunumu

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ABSTRACT

Intracranial mycotic aneurysm (MA), also known as intracranial infectious aneurysm, is a rare type of intracranial aneurysm. However, it carries a high mortality rate of 80% when ruptured and 30% even when unruptured. Here we report a 22-year-old male patient who consecutively developed two ruptured intracranial MAs related to infective endocarditis. The time interval between the two attacks was approximately seven months. The second aneurysm formed and ruptured regardless of long-term antibiotic therapy and cardiac surgery to replace the dysfunctional aortic valve. This case indicates that intracranial MAs may develop even after successful treatment of the underlying cardiac disorders, and hence close angiographic follow-ups may be warranted following the cardiac surgery.

KEYWORDS: Intracranial mycotic aneurysm, Intracranial infectious aneurysm, Surgical treatment, Endovascular treatment

ÖZ

İntrakraniyal mikotik anevrizma (MA), intrakraniyal enfeksiyöz anevrizma olarak da bilinir ve nadir bir intrakraniyal anevrizma tipidir. Ancak rüptüre olduğunda %80 ve rüptüre olmadığında bile %30 gibi yüksek mortalite oranına sahiptir. Burada enfektif endokarditle ilgili olarak arka arkaya iki rüptüre intrakraniyal MA gelişen 22 yaşında bir erkek hastayı sunuyoruz. İki atak arasındaki aralık yaklaşık yedi aydı. İkinci anevrizma disfonksiyonel aort kapağını değiştirmek için yapılan kardiyak cerrahi ve uzun dönemli antibiyotik tedavisine rağmen gelişti ve rüptüre oldu. Bu olgu intrakraniyal MA'ların altta yatan kardiyak bozukluğun başarılı tedavisinden sonra bile gelişebileceğini ve bu nedenle kardiyak cerrahi sonrasında anjiyografik takibin faydalı olabileceğini düşündürmektedir.

ANAHTAR SÖZCÜKLER: İntrakraniyal mikotik anevrizma, İntrakraniyal enfeksiyöz anevrizma, Cerrahi tedavi, Endovasküler tedavi

INTRODUCTION

Intracranial mycotic aneurysm (MA), also known as intracranial infectious aneurysm, is a rare type of intracranial aneurysm. It carries a high mortality rate of 80% when ruptured and 30% even when unruptured (1). Treatment for this disease can be divided into medical, surgical and endovascular therapy (2, 3, 7). Here, we report a 22-year-old male patient who consecutively developed two ruptured intracranial MAs and was treated using combined therapeutic approaches.

CASE REPORT

A 22-year-old male patient suddenly presented with vomiting and progressively deteriorated consciousness on Aug 25, 2011. On admission to his local hospital, the Glasgow Coma Scale (GCS) score was 5 with a fixed dilated right pupil and constricted left pupil. The head computerized tomography (CT) scan showed intracerebral hemorrhage in the right temporal-occipital lobe (Figure 1A). An emergent surgery of hematoma evacuation plus decompressive craniectomy was

performed (Figure 1B). His neurological condition gradually improved after the surgery: the GCS score rose to 13 and both pupils regained light reaction. On the 10th day after the surgery, the GCS score unexpectedly decreased to 10. The immediate CT scan showed rebleeding at the surgical site (Figure 1C), and digital subtraction angiography (DSA) disclosed a ruptured aneurysm at the distal branch of the right posterior cerebral artery (Figure 1D). Then he was transmitted to our department for further treatment. Embolization of the ruptured aneurysm and the parent artery using a liquid embolic agent (Onyx) was performed uneventfully (Figure 1E). During hospitalization, he also suffered from a severe incision infection that extended to the subdural space, with high fever and nuchal rigidity. The cerebrospinal fluid panel confirmed the diagnosis of meningitis while the blood culture showed negative results. He underwent wound debridement and lumbar drainage, and received a four-week course of intravenous antibiotics. He was discharged without neurological deficit six weeks after admission.

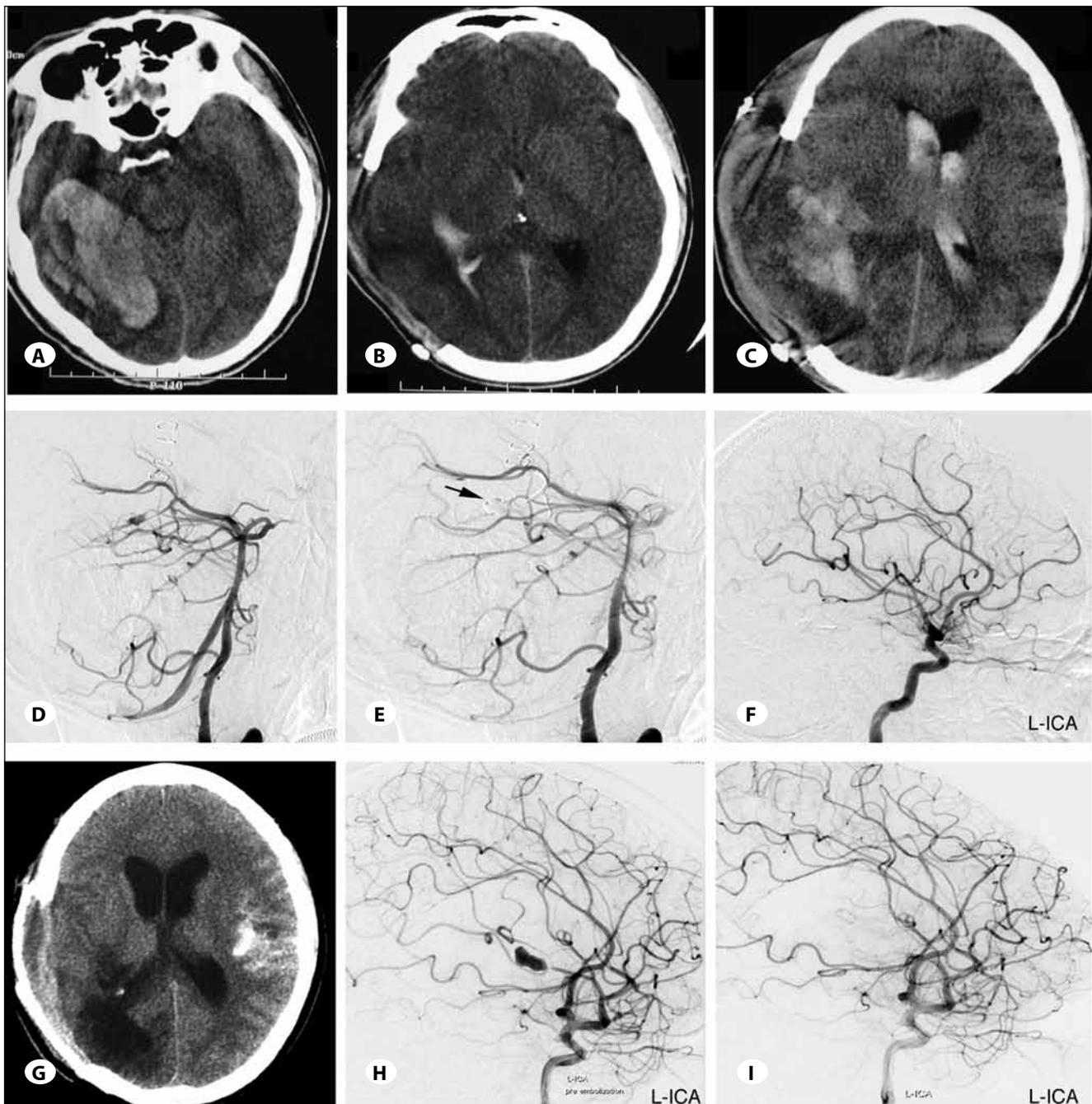


Figure 1: Radiographic findings of the patient during the treatment of two ruptured intracranial aneurysms. The arrows denoted the ruptured aneurysm before and after embolization. **A)** The head computerized tomography (CT) scan showed intracerebral hemorrhage in the right temporal-occipital lobe. **B)** CT scan showing an emergent surgery of hematoma evacuation plus decompressive craniectomy was performed. **C)** The immediate CT scan showed rebleeding at the surgical site, and **D)** digital subtraction angiography (DSA) disclosed a ruptured aneurysm at the distal branch of the right posterior cerebral artery. **E)** Then he was transmitted to our department for further treatment. Embolization of the ruptured aneurysm and the parent artery using a liquid embolic agent (Onyx) was performed uneventfully. **F)** The DSA follow-up at three months after the discharge showed no relapse or new formation of intracranial aneurysms. **G)** The head CT showed subarachnoid hemorrhage on the left parietal-temporal lobe. **H)** The urgent DSA disclosed a ruptured aneurysm at the distal branch of the left middle cerebral artery, which was not present in the latest DSA (**F**). **I)** We embolized the aneurysm and the parent artery using Onyx.

The DSA follow-up at three months after the discharge showed no relapse or new formation of intracranial aneurysms (Figure 1F). One week later, however, he presented with high fever, and the blood culture showed the presence of hemolytic streptococci. An echocardiogram disclosed the regurgitated aortic valve with mobile vegetation on its leaflet. He underwent cardiac surgery to replace the aortic valve at another hospital, and this was followed by six weeks of intravenous antibiotic treatment. He recovered well from the surgery.

On Mar 26, 2012, about three months after the cardiac surgery, he was admitted to our department again for cranioplasty. He was alert and showed no neurological deficit before surgery, but he became sluggish after the surgery. The head CT showed subarachnoid hemorrhage on the left parietal-temporal lobe (Figure 1G). The urgent DSA disclosed a ruptured aneurysm at the distal branch of the left middle cerebral artery (Figure 1H), which was not present in the latest DSA (Figure 1F). We embolized the aneurysm and the parent artery using Onyx (Figure 1I). After these measures, his consciousness status gradually improved, and he underwent four weeks of intravenous antibiotic treatment before discharge. Now he lives a normal life and is under close follow-up.

DISCUSSION

To our knowledge, this case is rare not only because this patient consecutively developed two ruptured intracranial MAs within seven months, but also because the second aneurysm developed and ruptured after long-term antibiotic therapy and cardiac surgery. These findings may support close follow-up for patients with intracranial MAs, even after surgical treatment of the underlying cardiac disorders.

Intracranial MA accounts for 0.7-6.5% of all intracranial aneurysms (4). Sources of infection can be divided into systemic infections such as infective endocarditis (IE), and local infections like meningitis, cavernous sinusitis, or orbital cellulitis (5). The proposed diagnostic criteria for this disease are mainly based on the documentation of the intracranial aneurysm by angiography and a series of predisposing conditions (6). Although the patient developed meningitis following the decompressive craniectomy, both the intracranial MAs are more likely to be associated with IE, which was supported by the positive results from blood culture and echocardiogram.

In patients with IE, septic emboli from cardiac valves may lodge at intracranial vessels at branching points or distal segments. These emboli may also cause focal infection and corrupt the vessel wall, which subsequently gives rise to aneurysm formation (4). Therefore, targeting the underlying cardiac disorders is mandatory in the treatment of IE-related intracranial MAs. However, the current case indicates that even successful treatment of cardiac disorders may not guarantee a cure. In our opinion, the delayed formation of the second intracranial MA may be due to the spread of septic emboli preceding the cardiac surgery with an incubation period

between the emboli lodgment and the aneurysm formation.

Management of a ruptured intracranial MA is an emergency situation, but therapeutic strategy may be optimized in individuals. The endovascular approach may generally be the first choice if possible because of less invasiveness than the surgical approach. However, decompressive surgery is reserved for cases with increased intracranial pressure (4). In our case, the first ruptured aneurysm presented with massive intracerebral hemorrhage, which indicated the necessity for emergent decompressive surgery. Unfortunately, the ruptured aneurysm was not clipped during the surgery and it was embolized by an endovascular approach. The second ruptured aneurysm showed mild subarachnoid hemorrhage, which warranted endovascular treatment.

Long-term intravenous antibiotics for four to six weeks have been recommended for treating intracranial MAs(5). Sometimes mere antibiotic treatment leads to complete resolution of the aneurysm (2). In our case, the second intracranial MA formed and ruptured despite long-term antibiotic treatment between the two attacks. The therapeutic effect of antibiotic treatment appears to vary in individuals, and hence close angiographic follow-up is necessary in patients with intracranial MAs.

In summary, intracranial MAs may develop in a delayed form, even after successful management of the underlying cardiac disorder. Therefore, close angiographic follow-up may be warranted following the cardiac surgery.

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