



Ruptured Infective Intracranial Aneurysm with Intracerebral Hemorrhage in an Infant

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Intracranial aneurysms are rare in children. Less than 10% of pediatric intracranial aneurysms are infective aneurysms, and few cases of such aneurysms in infants have been reported in the literature. In this study, we report the case of a 1-month-old preterm infant presenting with recurrent intracerebral hemorrhage due to a ruptured intracranial infective aneurysm. Blood culture yielded *Candida* species. Brain computed tomography and cerebral angiography revealed a large intracerebral hematoma in the left frontal lobe and a 2.7-mm sized aneurysm in a distal branch of the middle cerebral artery. The patient underwent craniotomy for hematoma evacuation and aneurysm excision. After the surgical intervention, antifungal therapy, and rehabilitation treatment, the patient recovered with mild neurological sequelae. This case demonstrated that the aggressive treatment for infective intracranial aneurysm is key to a favorable outcome, even in a young infant.

Key words: Infective intracranial aneurysm, intracerebral hemorrhage, infant, *Candida*

INTRODUCTION

Intracranial aneurysms are cerebrovascular lesions that are uncommon in the pediatric population, and they are rarely seen in infancy. Moreover, very few intracranial aneurysms are known to have resulted from infection, with <20 infant cases of infective intracranial aneurysm (IIA) having been reported in the literature. Once ruptured, these lesions require a surgical or endovascular intervention to prevent further bleeding. Here, we report the case of a 1-month-old preterm infant who suffered from recurrent cerebral hemorrhage due to a ruptured IIA. The course of treatment is presented, and the clinical characteristics and contemporary management strategy for IIA are reviewed.

CASE REPORT

A preterm baby was delivered by Cesarean section at a gestational age of 34 weeks due to fetal distress. The birth body weight was 1245 g. The baby developed hypotension

and tachycardia at 1 week of age. A laboratory study showed leukopenia and thrombocytopenia, and a blood culture grew *Candida parapsilosis*. Echocardiography revealed neither valvular vegetation nor cardiac anomaly. The neonate's condition improved after antifungal therapy, but then worsened suddenly at 33 days of age. Transcranial ultrasound showed hyper-echogenicity in the left frontal lobe. Brain magnetic resonance imaging (MRI) demonstrated subacute hemorrhage in the left frontotemporal lobe with mild mass effect, and a nodular lesion near the Sylvian fissure [Figure 1a], yet contrast study showed no abnormal enhancement. The child was then transferred to our hospital. Intensive medical care continued and her condition improved.

At the age of 49 days, sudden onset of the conscious disturbance, right hemiparesis, and left pupil dilatation occurred. Brain computed tomography (CT) showed enlarging cerebral hematoma and brain herniation [Figure 1b]. Emergency craniotomy was performed to remove the hematoma. Postoperative CT angiography (CTA) [Figure 1c and d] and cerebral angiography [Figure 1e and f] showed a small aneurysm arising from a frontal branch of the left middle cerebral artery. Trans-arterial embolization was not done for fear of ischemic stroke and infection. Therefore, the patient underwent reoperation for obliteration of the aneurysm, which was 10 mm in diameter with partial thrombosis. The aneurysmal wall was friable, compatible with a pseudo-aneurysm [Figure 2a and b]. The lesion was excised along with the thrombosed distal parent artery. Pathology reported an organizing thrombus without visible microorganisms.

Received: August 3, 2014; Revised: August 25, 2014;
Accepted: September 9, 2014

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Infective intracranial aneurysm in an infant

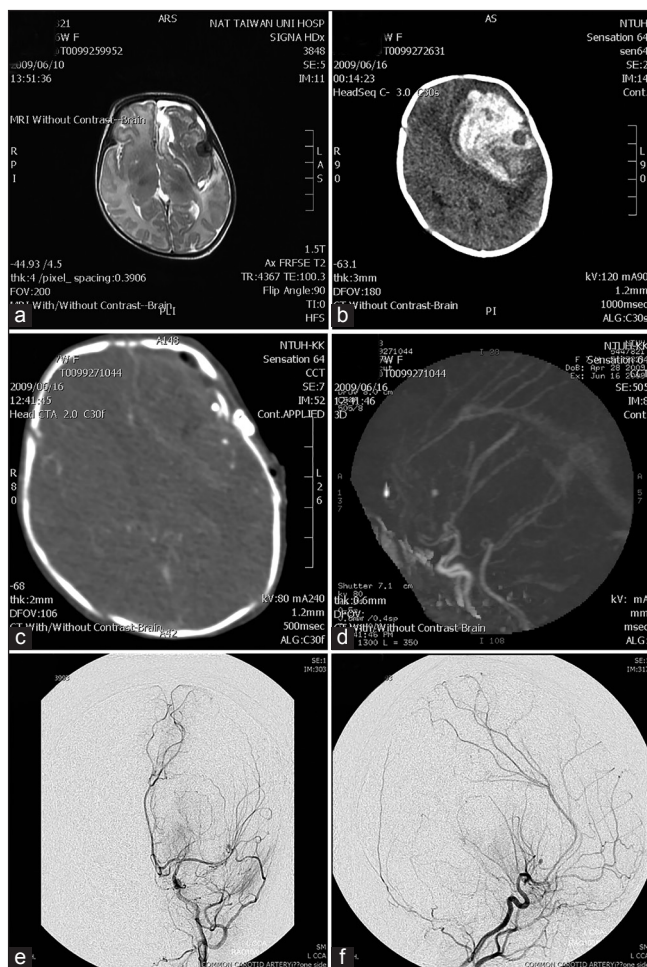


Figure 1. (a) T2-weighted magnetic resonance imaging (MRI) showed subacute hematoma in left frontotemporal lobe. There was a hypointense nodule at sylvian fissure, which did not enhance after contrast medium injection. (b) Computed tomography (CT) revealed enlarging intracerebral hemorrhage with midline shift. There was a hypodense nodule at the same position as in MRI. (c-f) After hematoma evacuation, CT and conventional angiography demonstrated an aneurysm at a distal branch of left middle cerebral artery. The position of the aneurysm was compatible with the nodule shown in the previous MRI and CT scan, indicating a partially thrombosed aneurysm

The infant recovered gradually and was discharged 5 weeks after surgery. There were neurological sequelae of right hemiparesis and epilepsy, which improved after rehabilitation therapy and anticonvulsant treatment.

DISCUSSION

Intracranial aneurysms are rare in the pediatric population, accounting for only 3.5-5% of all such lesions. There are different pathophysiological types of aneurysms: saccular, dissecting, traumatic, and infectious aneurysms. Although IIAs tend to occur more frequently in children than in adults,

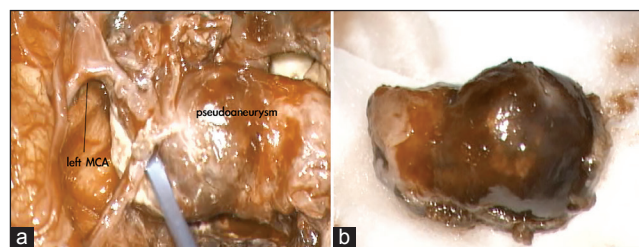


Figure 2. (a) During the second operation, we found the aneurysm arose from the wall of the frontal branch of M2 segment. The aneurysm appeared larger than it was shown in conventional angiography. The aneurysm wall was fragile and adherent with surrounding tissue tightly, indicating a pseudoaneurysmal nature. (b) Due to thrombosis of its distal parent artery, the pseudoaneurysm was excised. The final pathology revealed an inflamed and destructed vessel wall with thrombus inside the lumen

<10% of pediatric intracranial aneurysms are infectious in nature. IIAs are extremely rare in children <1 year of age. A systematic review of the literature from 1966 to 2005 identified 13 infant cases of IIA.¹ Two additional cases were identified after 2006.^{2,3} However, the true incidence is probably underestimated because many IIAs remain asymptomatic and resolve following antibiotic treatment.⁴

The pathogens responsible for IIAs include bacteria, fungi, and in rare cases, viruses. Two pathophysiological mechanisms lead to the formation of an IIA. First, infectious emboli from endocarditis or septicemia may lodge on the cerebral arterial walls. Second, a local infection, e.g., cavernous sinus thrombophlebitis, osteomyelitis, orbital cellulitis, or meningitis, can extend to the intracranial vessels. In the past, the most common etiology was infectious emboli from bacterial endocarditis, but direct extensions from meningitis or other local infections play an equally important role nowadays.^{5,6} Among the published cases of infant IIA, a definitive etiology was found in 10 cases: Four were caused by meningitis, four were caused by septicemia, one was caused by osteomyelitis, and one was caused by infective endocarditis. Our patient's IIA was likely related to bloodstream infection by *Candida* species, which also accounted for two other cases of infant IIA.^{2,4}

The diagnosis of an infant IIA is frequently delayed due to complicated clinical scenarios with concomitant infections, e.g., septicemia and meningitis. The typical course, as in our case, is that the infection improves initially after antibiotic treatment, but later deteriorates due to IIA rupture. Bleeding from a ruptured IIA usually results in subarachnoid or intracerebral hemorrhage. For infants, the clinician could use transcranial ultrasonography to screen for intracranial bleeding, and then confirm the results with CT or MRI. To delineate the culprit lesion requires angiographic study. While digital subtraction angiography (DSA) is the gold standard for diagnosis, CTA has comparable sensitivity and specificity in depicting IIA. MR angiography (MRA) is another diagnostic

alternative, but the longer acquisition time required is less desirable for infant patients. In our case, the contrast MRI done after first bleeding did not reveal any vascular lesion in the local hospital, and no further angiography was performed until secondary bleeding from the IIA occurred. To avoid missing a tiny aneurysm, angiography by MRA, CTA, or DSA must be performed even if the result of contrast MRI or CT study was negative.

Mortality from ruptured IIAs has dropped from 80% in the 1970s to 32% in the modern era.^{5,6} Current treatment guidelines recommend a multimodality management algorithm.⁷ Patients with unruptured IIA are treated medically with antibiotics and serial angiography. Patients with ruptured IIA not associated with mass effect from hematoma or involving the eloquent area are treated endovascularly. Patients with ruptured IIA are treated surgically if the hematoma is large or if the aneurysm is in the eloquent vascular territory. Ideally, the aneurysm is secured by clipping, with the parent artery preserved. However, the wall of the pseudoaneurysm is often fragile, and excision of the lesion is an alternative.^{2,8} In our case, the IIA ruptured twice because the lesion was not found by contrast MRI after the first bleeding. To prevent recurrent bleeding, neurovascular intervention to control the lesion is mandatory once an IIA bleeds. Our patient received surgery instead of endovascular embolization due to concerns regarding potential ischemic and infectious complications. In the future, endovascular therapy may become the mainstay treatment as this technique has demonstrated a low complication rate in recently reported studies.^{9,10}

CONCLUSION

Infant IIAs are critical cerebrovascular lesions that require a multi-disciplinary approach for management. In such cases, the young age and frail condition of the patient pose a major challenge for the clinician. Timely treatment through a neurosurgical or endovascular intervention for a ruptured IIA will prevent further bleeding, and improve the patient's outcome.

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