

Case Report

Brucella-related multiple cerebral aneurysms: Report of a case and review of the literature

Rouzbeh Shams Amiri, Hamed Hanif¹, Ali Ahmadi², Abbas Amirjamshidi¹

Department of Neurosurgery, Imam Hospital, Mazandaran University of Medical Sciences, Sari, Departments of ¹Neurosurgery and ²Pathology, Sina Hospital, Tehran University of Medical Science, Tehran, Iran

E-mail: Rouzbeh Shams Amiri - rshamsa@gmail.com; Hamed Hanif - h-hanif@razi.tums.ac.ir; Ali Ahmadi - ahmadise@sina.tums.ac.ir;

*Abbas Amirjamshidi - abamirjamshidi@yahoo.com

*Corresponding author

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Abstract

Background: Mycotic cerebral aneurysms are uncommon. We intend to report the first case of multiple mycotic cerebral aneurysms due to *Brucella* infection that were treated surgically.

Case Description: A 34-year-old man with neurobrucellosis presented with intracerebral haemorrhage (ICH). Three mycotic aneurysms were detected in the vicinity of middle cerebral artery (MCA). Medical treatment failed to treat them and aneurysms had to be managed surgically.

Conclusion: *Brucella*-related cerebral mycotic aneurysm has rarely been reported. This is the first report of three mycotic aneurysms occurring in a young man with neurobrucellosis treated surgically.

Key Words: *Brucella*, endovascular treatment, meningitis, mycotic aneurysm

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INTRODUCTION

Brucellosis is one of the most common zoonotic infections around the globe.^[9] It is a multi-systemic disease which can present with different clinical pictures. Involvement of the nervous system includes the features of meningitis, meningoencephalitis, myelopathy, polyradiculitis, mononeuritis, and also vascular insults and angiopathies.^[12,16]

The inflammatory processes involving intracranial vessels may lead to lacunar infarcts, venous thrombosis, small hemorrhages, or aneurysm formation.^[1,12]

This is the first case reported in the English literature of multiple cerebral mycotic aneurysms secondary to *Brucella* infection that were treated surgically. The other five cases of single *Brucella*-related aneurysm reported are

summarized in Table 1, among which only one case was amenable to surgery.

CASE REPORT

A 34-year-old male, a computer engineer by profession, was found drowsy at home and was referred to a nearby medical center. There was history of a recent business trip to Greece which took almost 6 weeks. He was suffering from headache and fever and was drowsy when admitted in the hospital [modified Rankin scale (MRS) =2]. Preliminary workups were all negative and brain computed tomography scan (CTS) showed a small left frontal opercular intracerebral hemorrhage (ICH) and intraventricular hemorrhage (IVH) accompanied with mild hydrocephalus [Figure 1]. He refused to stay in the hospital for further investigations

after improvement of his mental state. Headache, fever, and intermittent changes in his state of consciousness recurred after a month and he was admitted in our center after another attack of severe headache. At the time of admission, he was stuporous with mild right hemiparesis and pure motor dysphasia (MRS = 4). Brain CTS showed a left frontal opercular ICH [Figure 2].

Table 1: Previously reported cases (according to the reference numbers)

Case	Aneurysm	Angiography	Management
Hansmann and Schenken 1932 ^[10]	Proximal Basilar artery	-	Autopsy finding
McLean <i>et al.</i> 1992 ^[14]	presumed	-	Medical therapy but expired
Jabbour R. 2003 ^[11]	Fusiform, dissecting mid basilar trunk	+	Medical with good outcome
Kaya S <i>et al.</i> 2008 ^[12]	Totally thrombosed basilar	+	Medical with good outcome
Erdogan <i>et al.</i> 2005 ^[6]	Callosopericallosal + distal ACA	+	One was clipped and second one was wrapped
Our case	Multiple distal MCA aneurysms	+	Surgical with good outcome

ACA: Anterior communicating artery

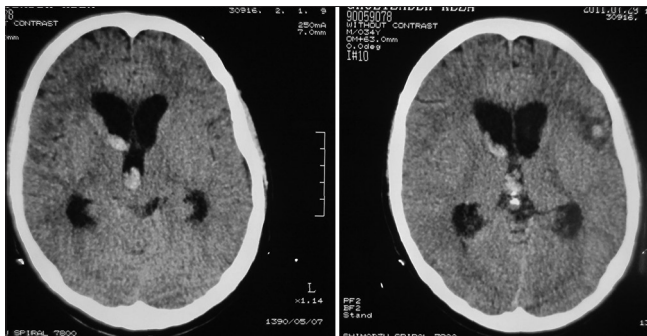


Figure 1: Small hemorrhage within the Sylvian fissure and intraventricular hemorrhage in the right lateral and third ventricles associated with mild hydrocephalus

Four-vessel digital subtraction angiography (DSA) showed a left distal middle cerebral artery (MCA) aneurysm that was 12 × 9 mm in diameter and without an identifiable neck [Figure 3]. This aneurysm was visible in both anteroposterior (AP) and lateral views even though there were two other ill-defined aneurysm blebs only seen in the lateral view of the angiogram. These two blebs were located along the more proximal branches of M3 segment of left MCA [Figure 4].

Considering the past history of fever, headache, detectable neck rigidity, and ICH, the first impression was rupture of a mycotic aneurysm. None of the aneurysms had well-identifiable neck in the angiogram. It was planned to treat the presumable mycotic lesions with a trial of wide-spectrum antibiotic therapy including Vancomycin 1 g twice/day + Rifampin 600 mg three times/day and Tazocin 4.5 g three times/day. All of the preclinical studies were negative except for the following: erythrocyte sedimentation rate (ESR) =28, C-reactive protein (CRP) =3 plus, and positive serology

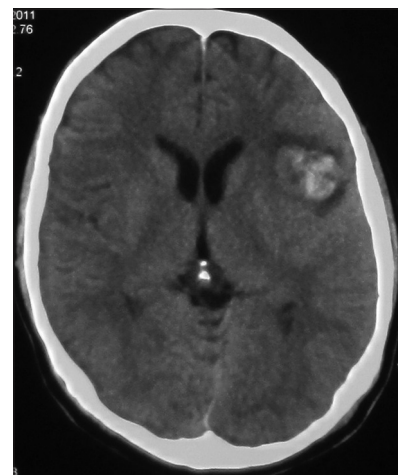


Figure 2: Second CT scan showing normal sized ventricles, but expanded left opercular ICH

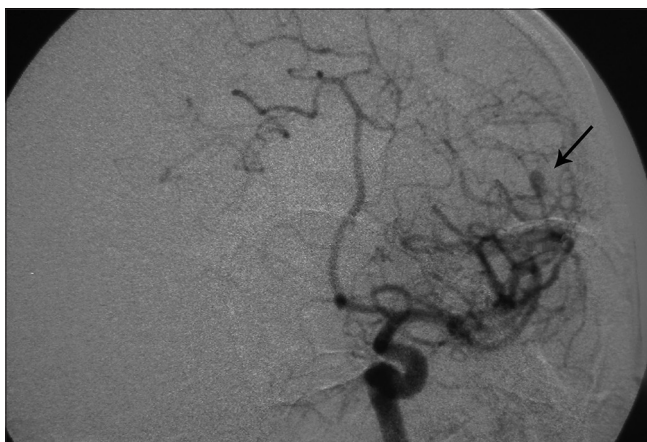


Figure 3: AP view of the DSA showing distal MCA aneurysm



Figure 4: Lateral view DSA showing the larger distal MCA aneurysm (black arrow) and two other smaller aneurysms located more proximally on the Sylvian arterial branches (white arrows)

for brucellosis (direct Coomb's test/*Brucella* micro agglutination test) both in his plasma and CSF with the agglutinin titer of 1/160 and 1/80, respectively. CSF analysis was compatible with partially treated meningitis: WBC = 500/mm³, polymorphoneuclears 60%, lymphocytes = 40%, protein = 55.3mg/dl, and sugar = 56 mg/dl.

His general condition improved moderately after 3 weeks of antibiotic therapy (MRS 3), but intermittent fever persisted and the CSF picture did not change notably. A brain computed tomographic angiography (CTA) did not show any regression in the size of the distal MCA aneurysm. The other two ill-defined aneurysms noted in the previous DSA were well elucidated in the new CTA [Figure 5].

Considering the available facilities, it was decided to treat the aneurysms surgically and it was planned for either clipping-only or excision with reconstruction bypass surgery of the aneurysms according to the findings during surgery. A left posterior temporoparietal craniotomy was done after 30 days of antibiotic therapy. The dura was opened, taking care of the arachnoidal adhesions visible beneath the dura, and attached to one of the cortically exposed aneurysms. A small aneurysm was located along one of the cortical branches of M4, along the superior

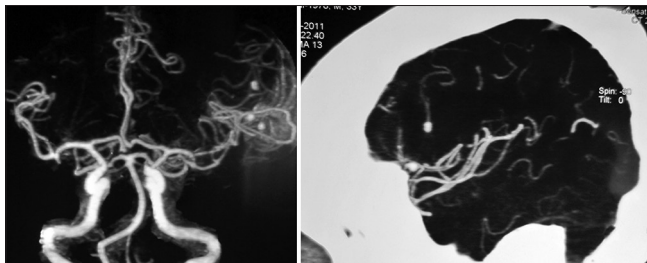


Figure 5: The good-quality CTA demonstrating all three aneurysms in AP and lateral views

temporal gyrus with adhesions in the surrounding subarachnoid space. The arachnoid could be dissected sharply. The aneurysm was a small one located on a distal M4 branch, without a clippable neck. It was coagulated and excised. The second aneurysm was located along M3 segment of the frontal branch of MCA. It was dissected, clipped, and resected. The clip could be removed after micro-coagulation of the parent artery [Figure 6a and b]. The more distal dissection of Sylvian fissure revealed a third small aneurysm located more distally along the M4 segment of MCA, which was dissected, clipped, coagulated, and excised. The histopathological examination of the resected aneurysms was compatible with "mycotic aneurysm" [Figure 7a-c].

The patient's postoperative course was uneventful. Considering the positive serology for brucellosis both in plasma and CSF, the anti-brucella antibiotic regimen was continued. The patient could be discharged after 1 month with improvement of fever and stuttering (MRS 2). The anti-brucella medications were continued for another 6 weeks and negative serology tests

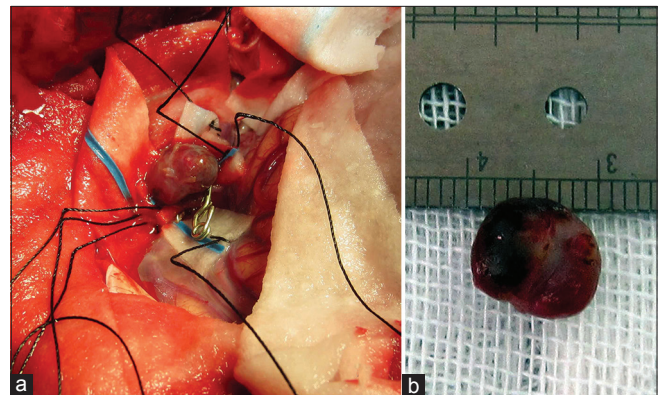


Figure 6: (a) Intraoperative image of second MCA aneurysm clipped and (b) the second mid-size aneurysm resected

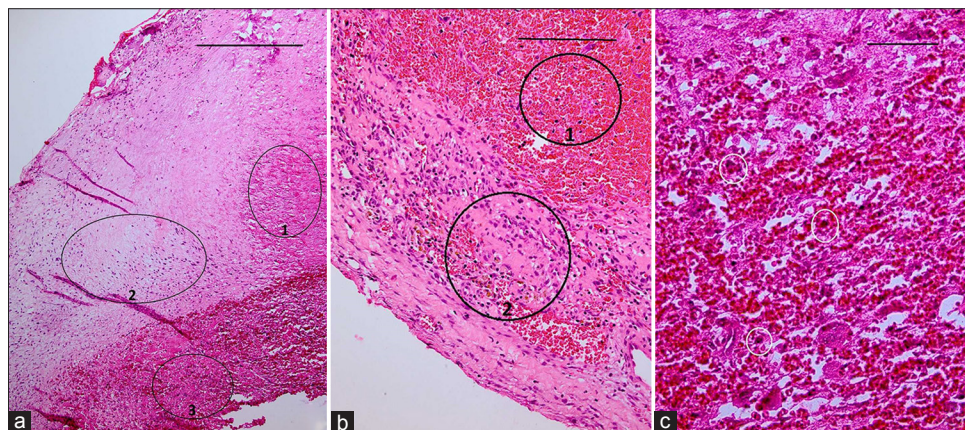


Figure 7: (a) H&E staining $\times 40$ showing portions of vessel wall with necrotic (ring 1), fibrotic (ring 2), and thrombotic (ring 3) changes. Scale bar is 2 mm. (b) H and E, $\times 100$ fibro-necrotic wall containing hemosiderin-laden macrophages (ring 2) admixed with many RBCs (ring 1). Scale bar is 1 mm. (c) H and E, $\times 400$ showing white small rings encircling few foreign body type giant cells and inflammatory cells, interspersed by thrombotic material. Periodic acid Schiff (PAS) staining (for fungi) and acid-fast bacillus (AFB) staining for mycobacteria show no specific microorganism. All these findings are compatible with "necrotizing vasculitis." Scale bar is 100 μm

achieved. The postoperative CTA performed after 1 year confirmed no aneurysmal dilatation along the course of MCA [Figure 8].

DISCUSSION

Neurobrucellosis occurs in about 5% of systemic *Brucella* affections, mostly manifesting as meningitis or encephalomeningitis.^[4] The other major complications in neurobrucellosis are cranial nerve (CN) involvement, polyneuropathy, radiculopathy, depression, paraplegia, stroke, osteomyelitis, and discitis and abscess formation.^[9]

The mechanisms for involvement of CNS in brucellosis may be intracellular involvement by the organism or stimulation of the immune system.^[2]

According to Gul *et al.*, in their pooled analysis of 187 cases of neurobrucellosis, fever, headache, sweating, and weight loss were the predominant complaints, followed by neurological symptoms, just as what happened in our case.^[9] In fact, the diagnosis of neurobrucellosis is by exclusion, i.e. the neurological manifestation not explained by other conditions and findings compatible with systemic brucellosis accompanied by CSF changes.

Although positive culture of *Brucella* organism is the gold standard test for diagnosis, only 28% of neurobrucellosis cases have positive blood cultures and 14% develop positive CSF culture.^[1] We did not obtain any of these positive cultures in our case, but the positive serologic/agglutination tests were diagnostic.

Only five cases of *Brucella*-related cerebral aneurysms have been reported, among which only one has been managed surgically [Table 1].^[6,10-12,14]

The optimum treatment strategy for infectious aneurysm is still a matter of debate. According to several authors,

antibiotic therapy for 4-6 weeks and close follow-up imaging looking for regression of the aneurysm can be the prior step in management of such aneurysms.^[3] In case of increase in the size of the aneurysm and/or hemorrhages, obliteration of the aneurysm by either open surgical intervention or endovascular therapy is another choice.

Endovascular obliteration has been reported as a treatment modality for mycotic aneurysms, but with limited evidences in the literature and the possibility of complications such as rebleeding during the procedure because of the fragility of the infected vessel and also the probability of late abscess formation. This last choice has been considered only in special situations such as mycotic aneurysm occurring in the setting of severe endocarditis which could not be managed by surgical intervention.^[1,3,5-8,12,13,15] The most important factors in choosing between the two approaches are: the morphology and location of the aneurysm, whether it is possible or not to sacrifice the parent artery, whether the patient needs or has received valve replacement surgery, the degree of vasospasm, the amount and location of ICH, and lastly, the patient's overall health status. There is no randomized controlled trial (RCT) comparing endovascular and open surgery in treating mycotic aneurysms, and they are being treated in either way considering the available facilities and feasibilities as in our case.

That was the exact algorithm undertaken in this reported case. Surgical intervention was attempted when the modality of wide-spectrum antibiotic therapy failed to manage the disease. Not only the aneurysm did not regress in size after 4 weeks of antibiotic therapy, but also two other aneurysms not well detectable in the first DSA became well distinguishable in the control imaging.

CONCLUSION

To the best of our knowledge, this is the first report in the literature on multiple mycotic cerebral aneurysms occurring in a patient with brucellosis and treated surgically.

The case reported by Hansmann *et al.*^[10] was an autopsy finding; the cases reported by McLean *et al.*^[14] presented with SAH and ICH, but angiography was negative in both cases and autopsy not performed; the proximally located basilar artery aneurysm reported by Jabbour *et al.*^[11] vanished in control angiography after successful medical treatment; Kaya *et al.*^[12] reported a thrombosed distal dissecting aneurysm of the basilar artery without surgical intervention; and unsuccessful medical management in neurobrucellosis in a 61-year-old man led to exploration and clipping of an aneurysm located on the callosomarginal artery and wrapping of an adjacent aneurysmal dilatation reported by Erdogan *et al.*^[6]

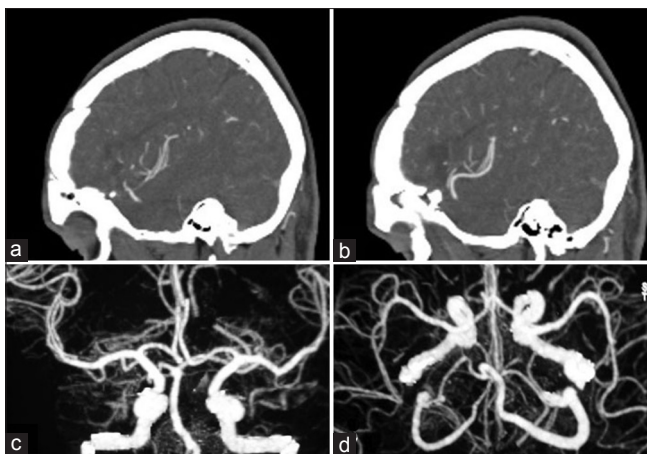


Figure 8: (a-d) The CTA performed after 14 months showing the course of MCA along the Sylvian fissure without aneurysmal dilatation

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