Unexpected Rupture of a Giant Lobulated Thrombotic Middle Cerebral Artery Aneurysm and Emergency Surgical Treatment With Thrombectomy: A Case Report and Review of the Literature

Vaner Koksal,[1,*] and Selim Kayaci[1]

[1]Department of Neurosurgery, Medical School, Recep Tayip Erdogan University, Rize, Turkey

*Corresponding author: Vaner Koksal, Department of Neurosurgery, Medical School, Recep Tayip Erdogan University, Rize, Turkey. E-mail: vanerkoksal@hotmail.com

Abstract

Introduction: The treatment of giant intracranial aneurysms is one of the most challenging cerebrovascular problems of neurosurgery. We report the rupture of a giant, lobulated, and almost completely thrombosed middle cerebral artery (MCA) aneurysm that is the ninth such report in the literature. We also investigated additional solutions used in the treatment of this patient.

Case Presentation: A 58-year-old man had been admitted with headache 8 years previously (in 2005), and a giant MCA aneurysm was detected. Two separate endovascular interventions were performed, and both failed. The patient began to live with the giant aneurysm. As there was a large thrombosis filling the aneurysm lumen during the previous endovascular procedures, the aneurysm was not expected to rupture. However, a rupture eventually occurred, in 2013. Even if an aneurysm is very large, lobulated, old, and almost completely thrombosed, it can suddenly bleed. During surgery on this patient, we observed severe cerebral vasospasm caused by a giant thrombosed aneurysmal rupture. Despite the complications, surgery is a life-saving treatment for this emergency when other strategies are not possible. Thrombectomy and clipping are approaches that require a great deal of courage for the neurosurgeon, in terms of entering the risky area within the aneurysm.

Conclusions: We believe that it would be more appropriate to plan for combined treatment with surgical and endovascular approaches before the emergency condition could occur.

Keywords: Cerebral Giant Aneurysm, Combined Treatments, Lobulated Aneurysm, Middle Cerebral Artery, Thrombectomy, Thrombosed Aneurysm

1. Introduction

Giant intracranial aneurysms have always been among the most difficult cerebrovascular lesions to treat. Most giant aneurysms, defined as having a greatest diameter of > 2.5 cm, occur in the anterior circulation and the internal carotid artery (ICA). The second most common form of giant aneurysm is reported to occur in the middle cerebral artery (MCA). It is reported that an average of 5% of all intracranial aneurysms are giant in size (1, 2). Intra-aneurysmal thrombosis has been reported to occur in 50% of giant intracranial aneurysms, and 47% of these cases can present with subarachnoid or intraparenchymal hemorrhage (1, 2). A lobular aneurysmal structure is reported very rarely, and only for the ninth time with this case report. Surgical treatment of a giant intracranial aneurysm can be very difficult and is not always successful (3). Several factors may complicate the treatment, including restricted visibility of surrounding vascular and neural structures due to the magnitude of the aneurysm, a widened and calcified aneurysmal neck, a thrombus within the aneurysm, and mass effect with proximity to the cranial nerves and vital neural structures. For these reasons, treatment of giant intracranial aneurysms is one of the most challenging cerebrovascular problems in neurosurgery (3, 4).

There are many reports on the treatment of intracranial giant aneurysms (4-6), including several good outcomes recently reported after combined surgical and endovascular treatment (5). Most of these reports focused on the treatment of aneurysms without thrombus, and there are insufficient data on the causes of failure or reports on the challenges of endovascular treatment in the literature. Our case involved an adult patient who presented with a giant ruptured lobulated thrombotic aneurysm in the MCA. Such a huge, lobulated, thrombosed, and ruptured aneurysm has not previously been reported. This is a unique case of a giant lobulated aneurysm of the MCA treated emergently by thrombectomy and clipping.

2. Case Presentation

In 2013, a 58-year-old man was admitted to the emergency department of Recep Tayyip Erdoğan University, Rize, Turkey, with complaints of severe headache, agitation, and progressive loss of consciousness. His blood pressure was 250/100 mmHg, and his Glasgow coma scale (GCS) score was 12 - 13 in the emergency department. A diagnosis of aneurysm was obtained with computed tomography (CT) scans and magnetic resonance imaging (MRI) of the head. The aneurysm was measured at 56.5 mm with the General Electric Healthcare PACS program (GE; Centricity Universal Viewer Zero Footprint, version 5.0 sp7.1) (Figure 1A and 1B). The aneurysm was seen to have settled in the middle fossa at the base, filled the inferior p ortion of the temporal lobe, and ruptured toward the posterior portion within the temporal lobe. At first, it had been observed on the CT at the orbital level, and the higher axial sections showed lobulation extending up to the third ventricular level. The aneurysmal wall was not calcified, while the thrombosis within the lumen was locally calcified. A midline shift had formed due to the impact of the hemorrhage. In the cerebral angiographic images obtained by MRI, it was observed that the right MCA and all of its branches did not fill up with blood (Figure 2B). It was decided, after the aneurysm's rupture, that the anterior circulation fed by the right ICA was totally occluded due to vasospasm.

The patient's medical history showed that he had been diagnosed with a giant cerebral aneurysm 8 years earlier and that two separate endovascular interventions had been performed, both of which failed. During this process, the patient never had an ischemic cerebral attack. The patient begin living with the giant aneurysm. The patient and his family believed that aneurysmal rupture would not occur because there was a large thrombosis filling the aneurysmal lumen during the previous endovascular procedures. The patient had hypertension, but he did not take any regular anti-hypertensive medication. Although the aneurysm was not expected to rupture, it eventually did.

The patient's clinical condition began to deteriorate after 6 hours in intensive care. He rapidly lost consciousness and began to experience irregular respiration. He was intubated and attached to a ventilator. Upon the occurrence of anisocoria, he was rushed to surgery, both to provide intracranial decompression and to close the aneurysm. All relevant information about the patient is summarized in Table 1.

2.1. Surgical Technique

Based on CT and MRI in the emergency department, the giant-sized aneurysm with a diameter of 56.5 mm, which had developed from the MCA and filled the inside

of the right temporal fossa, was determined to have ruptured. Since the patient's clinical condition was rapidly deteriorating, urgent surgery was decided upon. The actual size of the aneurysm was larger than expected during the surgery. In order to be able to obtain proximal control of the aneurysm more safely and to uncover the aneurysmal neck, the ICA was first closed temporarily. A 6-minute-thrombectomy was performed on the giant aneurysm, and the parent artery was revealed along with the MCA branches. Since the parent artery was the one that directly filled the aneurysm, it was closed with a clip.

Despite vasospasms, the right ICA was prepared because hemorrhage control might be challenging due to the magnitude of the aneurysm. Vascular tape was wrapped around it in a way that would make it possible to close at any time. Following the pterional skin incision, an extensive temporoparietal craniotomy was performed. The dura was opened so that the pedicle base would stay on the temporal base. There was no pulsation in the right cerebral hemisphere. The aneurysm was directly seen in the proximal entry of the sylvian sulcus and on the inferior temporal gyrus (Figure 1A, 1B and 2A). The diameter of the visible surface of the aneurysm was measured at 40 mm. Since it completely filled the proximal part of the Sylvian sulcus, dissection was not performed on the Sylvian sulcus. The aneurysmal walls were not movable, thus the carotid artery could not be reached. The aneurysm was hard and no dissection was likely to be performed around it. At this stage, when it was obvious that the inside of the aneurysm was filled with thrombus, it was decided that the posterior part of the aneurysm would be reached through thrombectomy.

When the outer wall of the aneurysm was opened, the old thrombus wall was encountered (Figure 3A, 3B and 3C). During the thrombectomy, it was noted that the thrombus had became fibrotic and old, while the new thrombus was very small. The aneurysm was filled with thrombus to the parent artery lumen, and it took the shape of the lamina seen within the aneurysm. The thrombus layers were totally rigid, gray, and even calcified due to the fact that the layers close to the external wall, in particular, were rather old. As the center was more closely approached, newly-formed thrombus layers were encountered (Figure 3C). When the center of the aneurysm was reached, it gradually began to bleed. At this point, the right ICA was compressed and the blood flow was stopped. After the contents were discharged, the aneurysmal wall became flexible. The posterior part was examined while being moved, and the aneurysm, along with a separate large lobule, seemed to proceed into the temporal lobe and toward the superior region. The aneurysm was minimized with macro-scissors. After the primary lobule was thoroughly removed, the in-

Figure 1. Giant Cerebral Aneurysm on CT and MRI in the Emergency Department
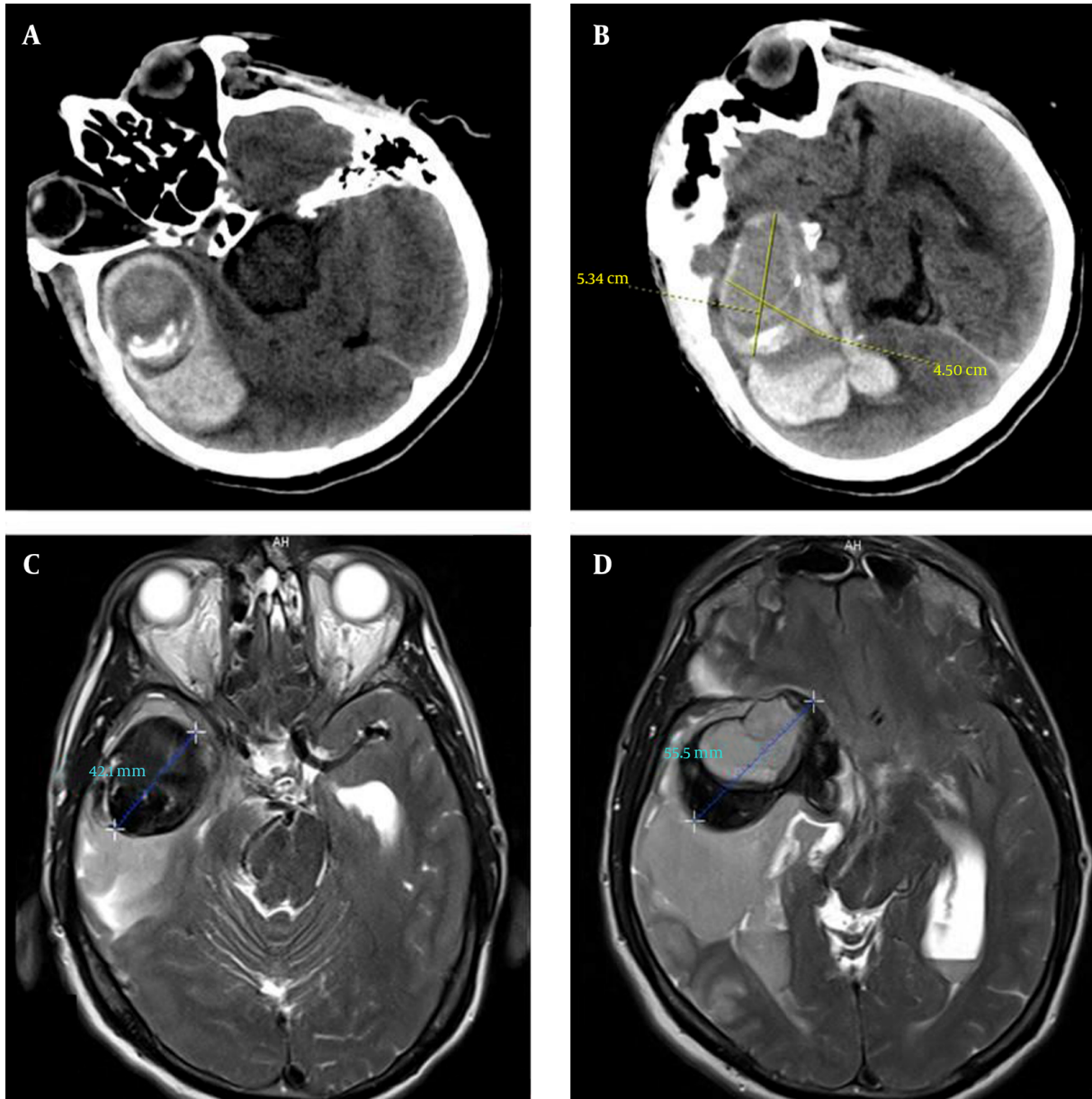
A and B, Ruptured right giant MCA aneurysm on CT; C and D, Giant, lobulated, and thrombotic right MCA aneurysm on T2 axial MRI. Brainstem compression is observed.

terior portion of the secondary lobule was discharged with tumour forceps (Figure 3D and 3E). The M2 branch of the MCA originating from the wall, overlooking the temporal lobe of the giant aneurysm and proceeding toward the distal part of the Sylvian sulcus, could be seen when the walls of the secondary lobule were moved. The M2 originated from the second lobule of the aneurysm. There was no pulsation in the M2 and its diameter was tiny due to

vasospasm (Figure 3D). Perforating branches with rather tiny diameters were separated from the side of the giant aneurysm, which overlooked the temporal lobe and proceeded along it. The proximal part of the aneurysm that was connected to the artery was reached, and the final large thrombus was removed from it. The ICA was then reopened, and the hemorrhage that followed had a higher flow rate than the previous one. It was then understood

Figure 2. Drawing of the Aneurysm and MRI Angiography
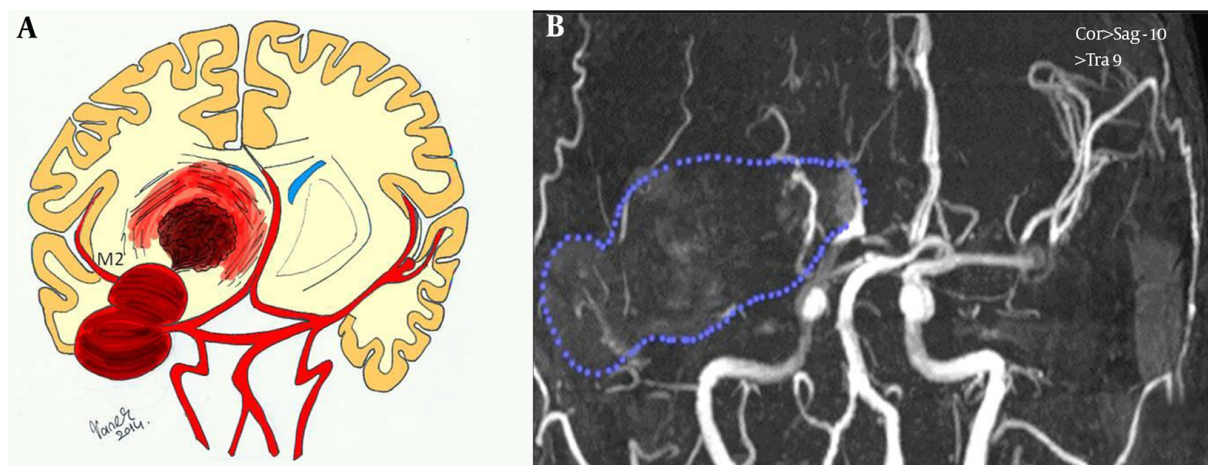
A, Giant lobulated aneurysm on the coronal plane with drawing by the author; B, Cerebral MRI angiography Vessels of the right hemisphere are not observed due to thrombosis. Blood flow of the right distal ICA is not apparent on MRI angiography.

Table 1. Patient Data

| 58-Year-Old Male with 20 Years of Hypertension				
8 Years Earlier(2005)			**2013**	
First complaint: headache		Ischemic stroke and hemorrhage was not observed	**Emergency admission with blood pressure of 250/100 mmHg, severe headache, and agitation.**	Progressive loss of consciousness
GCS: 15	Endovascular treatment was attempted twice	Waiting period for 8 years	GCS: 12 - 13 Major bleeding and large temporal hematoma	Uncal herniation begins
Giant MCA aneurysm was found	Not successful	DSA images missing	Giant lobulated thrombotic MCA aneurysm and M2 originating from within aneurysmal lobule	Emergency surgery with thrombectomy and clipping
Istanbul, Turkey	2005 - 2006 (2 separate times)	Did not use the drug regularly	Rize, Turkey	**Died from diffuse vasospasm**

that the bleeding vessel was M1 itself and the parent artery. After the parent artery was closed with clips, the bleeding ceased. The thrombectomy was completed in 6 minutes.

2.2. Prognosis

The aneurysm had reached a giant size. It was formed of the MCA bifurcation and had remained within the M2 and its branches. No atheroma plaque to prevent the clip insertion was seen. At the end of the process, there was no pulsation in the right cerebral hemisphere as had been seen at the start of the surgery. The necessary precautions were taken to increase the patient's blood pressure. In case edema developed in the brain, duraplasty was performed via a galeal graft, and the cranium was not closed. Following the operation, the patient was taken to the inten-

sive care unit. His blood pressure did not increase despite the discontinuation of opiates. Dopamine was started. No radiological follow-up was performed since the patient's hemodynamics were failing. His anisocoria recovered in the postoperative early period, during which he began to localize the pain with the help of his right arm. Shortly after that, at the end of the postoperative 12th hour, his response to pain suddenly deteriorated. Eventually, the patient lost spontaneous respiration. He had rapid cardiac arrhythmia, followed by cardiac arrest. Deep bradycardia and hypotension were assumed to be due to ischemia affecting the brain stem.

Figure 3. Surgical Images
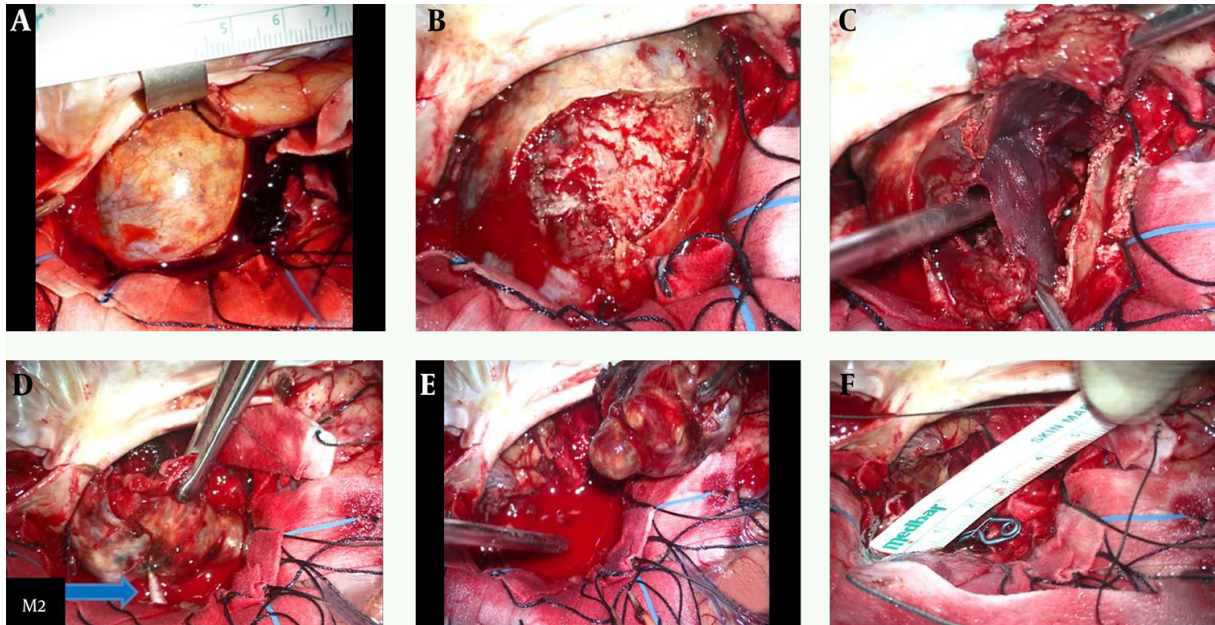
A, appearance of the aneurysmal wall surface on the proximal Sylvian sulcus; B, Aneurysmal wall opening; C, old laminar thrombus; 3d, aneurysmal wall is moved after thrombectomy and M2 is found M2 originating from second lobule of aneurysm. E; base of aneurysm is reached; F, measurement within the cavity with ruler after clipping.

3. Discussion

The main objective during treatment of all intracranial aneurysms is to leave the aneurysm completely out of the circulation by protecting the blood flow of the parent artery, thus eliminating the possibility of hemorrhage (4-8). Different techniques have been used so far for the surgical treatment of giant aneurysms. Yet, the optimal strategy for the treatment of thrombosed giant intracranial aneurysms is still unclear. It may be impossible to clip the neck directly or to fill it with a coil via endovascular routes (9). Surgical methods include direct clipping, clipping via thrombectomy, and occlusion or trapping with and without bypass (7, 8). Separately, a simple observation without any intervention is also possible along with endovascular methods. However, in a patient in whom the cerebral herniation process has just begun, the chosen treatment must be started immediately. Direct surgery is generally difficult and is associated with high surgical morbidity and mortality rates, but is urgently required in a patient with a herniation. For this purpose, classifications are required to allow the determination of which current treatment option is the most appropriate. In order to be able to determine the best appropriate treatment method, Lawton et al. divided giant thrombosed aneurysms into six subtypes according to lumen morphology and the position of the thrombus (10). These subtypes are: concentric, eccentric, lobulated, complete, canalized, and coiled. According to this classification, it was decided that the aneurysm in our case was Type 3. We also noted that it was much larger than the other giant aneurysms in the literature, and it would even be more accurate to call it a huge aneurysm.

Type 1 concentric thrombotic aneurysms have a spherical shape. Within the aneurysmal body, thrombi are present on all sides; these are classic thrombotic aneurysms. The thrombi are generally layered, resembling an onion formed by layers of blood from different points in time. In this type of aneurysm, the thrombus within grows and expands toward the aneurysmal neck. This characteristic distinguishes Type 1 from Type 2 (10). Direct clipping usually fails in these aneurysms (7).

Type 2 aneurysms have an eccentric thrombus. The trunk and the lumen of the aneurysm are elliptical, and the thrombus is typically located distally, at the dome. However, there is no thrombus covering the interior part of the lumen as in Type 1, and the thrombus does not extend toward the aneurysmal neck. The thrombus in these cases also prevents closure via direct clipping (10).

Type 3 aneurysms have a saccular morphology and multiple lobes. Within them are lobulated thrombi. A giant lobulated thrombotic aneurysm may have the appearance on angiography, CT, or MRI of a Type 2 eccentric

aneurysm formed of only one lobe (10). However, these lobulated thrombotic aneurysms are different from the others in that the thrombus within extends down the aneurysmal neck. It is sometimes difficult to notice such distinctions on radiological images prior to surgery. In the present case, the actual size and lobular structure of the aneurysm could be determined during the surgery, but there were two lobules connected to each other, rather than different lobules originating from the MCA.

All Type 4 aneurysms are thrombosed, and their lumens cannot be seen on angiography. Type 5 aneurysms, with their fusiform and dolichoectatic morphologies, have canalized thrombi extending within the lumen. Most canalized thrombotic aneurysms are treated with bypass and occlusion. Type 6 aneurysms, on the other hand, are those that have been treated via endovascular methods, and that have an intraluminal thrombus and iatrogenically deployed foreign material of the coils (10, 11). There was no foreign material within the aneurysm in our patient.

Almost-complete thrombus formation in giant aneurysms is very rare (1, 2). The thrombus formation is based on the proportion between the aneurysmal neck and the size of the dome (2). In aneurysms with a narrow neck and a large dome, the blood-flow rate within may decrease and a thrombus may occur (2). It was noted that the development of the aneurysm did not cease even in the presence of a thrombus that completely filled it (5, 7). An enormous intra-aneurysmal thrombus poses an obstacle against closing the aneurysm from the outside. Thrombectomy prevents the large mass effect of the thrombus and increases the visibility of the aneurysmal neck (3). With this attempt, it is also possible to avoid occlusion of the parent artery and the distal MCA, and it is ensured that the vascular anatomy is kept under control (3, 10). Yet, in the case presented, the aneurysmal morphology impeded the protection of the parent artery and its branches. During thrombectomy, tapping the blood flow of the parent artery for a short time may have an ischemia-causing effect. In order to follow-up on this, it is recommended that motor-evoked potential (MEP) monitoring be performed (3, 7). However, it is impossible to perform this in emergency cases. In recent years, the cavitron ultrasonic surgical aspirator (CUSA) has been widely used in microsurgery for particularly persistent organized and laminated thrombi. It has been reported that such an application is possible because the CUSA probe does not cause any damage to the aneurysmal wall (3).

When the amount of thrombus that fills an aneurysm is too much, both microvascular and endovascular surgical treatments can cause difficulties. A small number of cases that cannot be embolized have been reported in the literature (10-13). Also, because of the aneurysm's shape in the present case, originating from the aneurysmal wall of the M2 artery branch and being almost completely thrombosed, coil embolization was contraindicated. This complex situation could have a different solution, such as STA-MCA (to the M2 segment) bypass; after this operation, the aneurysm could possibly be completely occluded (13, 14). In our case, we performed emergency surgery intended to save the patient's life.

During the surgical procedure, it was observed that the thrombus layers in the form of lamina could be quite easily removed via tumour forceps without causing any damage to the aneurysmal wall during thrombectomy (Figure 3C). Endovascular treatment methods are also performed in the treatment of giant aneurysms (14). Due to problems such as failure to completely obliterate the aneurysm, the possibility of a late subarachnoid hemorrhage, the high incidence rate of distal thromboembolism, and failure to eliminate aneurysmal mass effect, endovascular treatment is only recommended in cases with surgical contraindications (5, 15). In fact, endovascular treatment had previously been attempted in the present case. It is assumed that a common practice model was not established in the former treatment attempt. Therefore, there was no chance to perform a surgical intervention under elective circumstances. The belief that the patient's aneurysm would never bleed was obvious. Moreover, it is reported that successful results have been achieved via detector devices that have come into use through endovascular methods in giant aneurysms with thrombus (14). Separately, there is always the possibility of microembolus formation toward the lumen of the parent artery in the natural developmental process of a giant aneurysm. This sort of problem was not noted in the medical records of our patient. For the treatment of a giant lobulated thrombotic MCA aneurysm, a combined approach of surgical and endovascular management may yield a better outcome than surgery or endovascular management alone (15, 16).

3.1. Article's Strengths and Weaknesses

3.1.1. Strengths

There are different and interesting radiological and surgical images. There are only nine articles in the literature about giant lobulated thrombosed aneurysms. The lobulation shape is different from others in the literature. The article can provide solutions to emergency problems; it is one of the few articles reporting a failure of endovascular treatment, and it may help to solve this problem. Giant lobulated thrombotic aneurysms are important because they are life-threatening. This patient is an important example demonstrating the necessity of combination

treatment for giant aneurysms. The contents of the thrombosed aneurysm were described in detail.

3.1.2. Weaknesses

Old radiological images of the patient are missing (especially DSA). Changes in the hemodynamics and thrombus-size of the giant aneurysm could not be determined. After emergency surgery, radiological control evaluations could not be performed. The surgery did not achieve its clinical objective.

3.2. Conclusion

Thrombectomy and clipping of giant intracranial aneurysms with proper morphologies can prevent hemorrhage in the proximal vessel. This method is quite secure and applicable in emergency cases as long as the necessary precautions are taken. However, the most important goal is to find a safer way to treat these patients with combined treatment before a bleed occurs.

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Footnote

Conflicts of Interest: The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

References

1. dos Santos ML, Spotti AR, dos Santos RM, Borges MA, Ferrari AF, Colli BO, et al. Giant intracranial aneurysms: morphology and clinical presentation. *Neurosurg Rev.* 2013;**36**(1):117–22. doi: 10.1007/s10143-012-0407-0. [PubMed: 22791075] discussion 122.
2. Krings T, Alvarez H, Reinacher P, Ozanne A, Baccin CE, Gandolfo C, et al. Growth and rupture mechanism of partially thrombosed aneurysms. *Interv Neuroradiol.* 2007;**13**(2):117–26. [PubMed: 20566139].
3. Sugita M, Kinouchi H, Nishiyama Y, Kanemaru K, Yoshioka H, Horikoshi T. Direct clipping of a thrombosed giant cerebral aneurysm after thrombectomy without bleeding to minimize the temporary occlusion time-technical case report. *Neurol Med Chir (Tokyo).* 2009;**49**(12):600–3. [PubMed: 20035137].
4. Sughrue ME, Saloner D, Rayz VL, Lawton MT. Giant intracranial aneurysms: evolution of management in a contemporary surgical series. *Neurosurgery.* 2011;**69**(6):1261–70. doi: 10.1227/NEU.0b013e31822bb8a6. [PubMed: 21734614] discussion 1270-1.
5. Bohnstedt BN, Ziemba-Davis M, Edwards G, Brom J, Payner TD, Leipzig TJ, et al. Treatment and outcomes among 102 posterior inferior cerebellar artery aneurysms: a comparison of endovascular and microsurgical clip ligation. *World Neurosurg.* 2015;**83**(5):784–93. doi: 10.1016/j.wneu.2014.12.035. [PubMed: 25541085].
6. Shin SH, Choi IS, Thomas K, David CA. Combined surgical and endovascular management of a giant fusiform PCA aneurysm in a pediatric patient. A case report. *Interv Neuroradiol.* 2013;**19**(2):222–7. [PubMed: 23693047].
7. Nanda A, Sonig A, Banerjee AD, Javalkar VK. Microsurgical management of giant intracranial aneurysms: a single surgeon experience from Louisiana State University, Shreveport. *World Neurosurg.* 2014;**81**(5-6):752–64. doi: 10.1016/j.wneu.2012.12.010. [PubMed: 23246634].
8. Sharma BS, Gupta A, Ahmad FU, Suri A, Mehta VS. Surgical management of giant intracranial aneurysms. *Clin Neurol Neurosurg.* 2008;**110**(7):674–81. doi: 10.1016/j.clineuro.2008.04.001. [PubMed: 18490100].
9. Yasargil MG. General operative techniques, Special operative problems. In: Yasargil MG, editor. Microneurosurgery. 1. New York: Georg Thieme Verlag; 1984. .
10. Lawton MT, Quinones-Hinojosa A, Chang EF, Yu T. Thrombotic intracranial aneurysms: classification scheme and management strategies in 68 patients. *Neurosurgery.* 2005;**56**(3):441–54. [PubMed: 15730569] discussion 441-54.
11. Nakajima H, Kamiyama H, Nakamura T, Takizawa K, Ohata K. Direct surgical treatment of giant intracranial aneurysms on the anterior communicating artery or anterior cerebral artery. *Neurol Med Chir (Tokyo).* 2013;**53**(3):153–6. [PubMed: 23524498].
12. O'Grady A, Denton J, Laing A. Microsnare-assisted microcatheter navigation for flow diversion of a challenging giant intracerebral aneurysm: a novel technique. *J Med Imaging Radiat Oncol.* 2015;**59**(1):70–3. doi: 10.1111/1754-9485.12267. [PubMed: 25565420].
13. Shi X, Qian H, Fang T, Zhang Y, Sun Y, Liu F. Management of complex intracranial aneurysms with bypass surgery: a technique application and experience in 93 patients. *Neurosurg Rev.* 2015;**38**(1):109–19. doi: 10.1007/s10143-014-0571-5. [PubMed: 25154436] discussion 119-20.
14. Kivipelto L, Niemela M, Meling T, Lehecka M, Lehto H, Hernesniemi J. Bypass surgery for complex middle cerebral artery aneurysms: impact of the exact location in the MCA tree. *J Neurosurg.* 2014;**120**(2):398–408. doi: 10.3171/2013.10.JNS13738. [PubMed: 24286147].
15. Alderazi YJ, Shastri D, Kass-Hout T, Prestigiacomo CJ, Gandhi CD. Flow diverters for intracranial aneurysms. *Stroke Res Treat.* 2014;**2014**:415653. doi: 10.1155/2014/415653. [PubMed: 24967131].
16. Lanzino G, Fang S. Challenges and changes in the endovascular treatment of very small intracranial aneurysms. *World Neurosurg.* 2014;**81**(5-6):678–9. doi: 10.1016/j.wneu.2013.07.126. [PubMed: 24067741].