Abstract
Subarachnoid hemorrhage (SAH) with multiple intracranial aneurysms is common, but the difficulties often arise in determining treatment strategy in the acute phase. We experienced a case of SAH with distal anterior cerebral artery aneurysm coexisting with middle cerebral artery and anterior communicating artery aneurysms, in which it was difficult to identify the precise rupture site preoperatively, and both pterional approach and interhemispheric approach were required in the acute phase of SAH. However, we could treat whole aneurysms in one stage and obtained an excellent outcome using our surgical procedure with ipsilateral frontotemporal and frontal parasagittal craniotomies through a single skin incision.

Keywords: Aneurysm, clipping, intracranial, multiple, subarachnoid hemorrhage

Introduction
Regarding patients with subarachnoid hemorrhage (SAH) with multiple intracranial aneurysms (MIAs), it is the most important to diagnose the precise rupture site preoperatively and treat the ruptured aneurysm promptly. If the rupture site is misidentified, more invasive multiple treatment approaches may be required in the acute phase of SAH possibly worsening the prognosis. Therefore, treatment design and strategies to reduce brain invasion are essential. We present an excellent outcome case of SAH with MIAs treated with our surgical design and strategies, in which we could not determine the precise rupture site preoperatively, and both pterional approach (PA) and interhemispheric approach (IHA) were required in the acute phase of SAH.

Case Report
The patient was a 71-year-old female presenting with severe headache and disturbance in consciousness. She was admitted to our hospital with a systolic blood pressure of 106 mmHg, Glasgow Coma Scale 12 (E3V3M6), and no focal symptoms. Computed tomography (CT) scan revealed diffuse SAH, particularly thickened at the right Sylvian fissure and basal cistern [Figure 1a]. We diagnosed as SAH, the World Federation of Neurological Surgeons Grading System for SAH Grade 4. The patient was admitted and underwent digital subtraction angiography (DSA). DSA revealed three aneurysms: right middle cerebral artery (MCA) aneurysm, anterior communicating artery (AcomA) aneurysm, and right distal anterior cerebral artery (ACA) aneurysm. Dome size of the MCA aneurysm, AcomA aneurysm, and right distal ACA aneurysm was 3.5, 2, and 4.1 mm. All the aneurysms were irregularly shaped having no bleb. Dome/neck ratio of the right MCA aneurysm, AcomA aneurysm, and right distal ACA aneurysm was 1.75, 1, and 2.7 [Figure 1b]. We diagnosed the right MCA aneurysm as the rupture site according to the uneven distribution of SAH and performed right PA first.

The patient was positioned 15° semi-Fowler position; the head was rotated at approximately 30° contralaterally, skull vertex in the neutral position and fixed in Mayfield skull clamp. We opened right frontotemporal skin incision and reflected anteriorly, placed a burr hole at right Kocher’s point, and inserted an external ventricular drain into the right anterior horn in standard fashion.[1] We performed a right frontotemporal craniotomy and dissected to open the Sylvian fissure and reflected anteriorly, placed a burr hole at right Kocher’s point, and inserted an external ventricular drain into the right anterior horn in standard fashion.[1] We performed a right frontotemporal craniotomy and dissected to open the Sylvian fissure and reflected anteriorly, placed a burr hole at right Kocher’s point, and inserted an external ventricular drain into the right anterior horn in standard fashion.[1] We performed a right frontotemporal craniotomy and dissected to open the Sylvian fissure and reflected anteriorly, placed a burr hole at right Kocher’s point, and inserted an external ventricular drain into the right anterior horn in standard fashion.[1] We performed a right frontotemporal craniotomy and dissected to open the Sylvian fissure and reflected anteriorly, placed a burr hole at right Kocher’s point, and inserted an external ventricular drain into the right anterior horn in standard fashion.[1] We performed a right frontotemporal craniotomy and dissected to open the Sylvian fissure and reflected anteriorly, placed a burr hole at right Kocher’s point, and inserted an external ventricular drain into the right anterior horn in standard fashion.[1] We performed a right frontal lobotomy in consideration of latter

For reprints contact: reprints@medknow.com
possible IHA. We exposed and clipped the MCA aneurysm which was apparently an unruptured aneurysm with a round shape and no bleb [Figure 2a].

Subsequently, we continued to approach the AcomA aneurysm through trans-Sylvian approach, opened the Sylvian fissure completely, and released the anchorage between the frontal lobe and frontal base. We exposed and clipped the AcomA aneurysm which was located on fenestrated AcomA and was also apparently an unruptured aneurysm with a round shape and no bleb [Figure 2b]. As a result, the distal ACA aneurysm was identified as the rupture site and continuously treated through subsequent IHA. Performing IHA, the operating table was rotated 30° to affected side laterally and 15° vertically up. We extended the frontotemporal skin incision to the contralateral side and performed a right frontal parasagittal craniotomy using the puncture point of the external ventricular drain. We dissected to open the interhemispheric fissure widely and exposed and clipped the right distal ACA aneurysm with obvious rupture point and fibrin plug [Figure 2c]. Consequently, we could treat the whole aneurysm in the acute phase with minimally invasive.

Figure 3a shows DSA on day 7, in which all aneurysms were obliterated and moderate vasospasm presented. However, the patient was not complicated with apparent
neurological deficit, and the vasospasm improved after the acute phase. Figure 3b shows the head CT at the time of discharge revealing no visible brain contusion on the approach route. The patient was discharged home after rehabilitation in a state of modified rankin scale (mRS) 1 and is returning for a follow-up visit now.

Discussion

MIAs have an occurrence frequency of 14%–34% and have been known that the risk of poor outcome increases in the setting of SAH.\[2-5\] However, there are no rigid guidelines for the methods to diagnose the rupture site, treating coexisting unruptured aneurysms and approach to treatment.

Preoperative diagnosis of the rupture site

Regarding preoperative diagnosis of the patients with SAH associated with MIAs, the aneurysmal morphological factors (the largest aneurysm, dome/neck ratio >1.6, presence of bleb, and irregular shape) are known as the predictors of the rupture site. In addition, the previous studies reported that the rupture sites could be estimated in 45%–59% of cases by uneven distribution of SAH or the location of intracerebral hemorrhage on CT.\[7,11\] However, there are several cases, in which these predictors are not reliable, and the precise rupture site cannot be estimated preoperatively.\[11,12\]

In our case, the distribution of SAH on CT suggested the right MCA aneurysm as the rupture site; however, the size and dome/neck ratio were largest in the right distal ACA aneurysm. In addition, it was expected that the rupture rate of a small aneurysm in MIAs was not negligible and that locations with the highest probability of rupture in MIAs were the AcomA.\[6,7,13\] Therefore, in our case, it was difficult to estimate the precise rupture site preoperatively, and the treatment design and strategies taking into consideration, the possibility of miscegenation the rupture site was essential.

Treatment strategies

Ruptured aneurysms used to be treated with surgical clipping, but recently, surgical clipping procedures remained stable while the number of aneurysms treated by means of endovascular procedure doubled.\[14\] Although the safety of endovascular one-stage coiling for ruptured aneurysm has been reported,\[15,16\] endovascular treatment for AcomA aneurysm, distal ACA, and MCA aneurysms still remains challenge because of the distal location, incorporated branching, and wide neck shape. Furthermore, it was reported that intraprocedural complications during endovascular coiling of ruptured aneurysm more often occur in patients with these aneurysm locations.\[17-20\]

In our case, there were three aneurysms considered unsuitable for endovascular coiling. Furthermore, in endovascular coiling, we had to treat all the aneurysms to reliably treat the precise rupture site because it was impossible to identify the ruptured aneurysms with the fibrin plug at the rupture point under direct vision. In surgical clipping, on the other hand, we were able to judge whether to treat other unruptured aneurysms simultaneously in consideration of the damage to the brain at the time of treating the rupture site. For these reasons, we performed surgical clipping for this case.

Surgical design

Several studies have reported that the unruptured aneurysms in MIAs have a higher risk of rupture than that of ordinary unruptured aneurysms.\[21-23\] Other studies also reported that one-stage clipping of MIAs within 72 h after SAH can be performed without increasing the risk of cerebral vasospasm and symptomatic vasospasms.\[24\] In addition, when treating the other unruptured aneurysms in the chronic phase, the likelihood of infection or skin disorder associated with reoperation and the operational difficulties due to arachnoid thickening after SAH may occur.\[25\] Therefore, if other unruptured aneurysms in MIAs are able to be observed in one stage, they are desirable to be treated simultaneously with the rupture site. Indeed, multiple unilateral supratentorial aneurysms are commonly treated in one stage through frontotemporal craniotomy,\[26\] and on the other hand, multiple bilateral intracranial aneurysms may be treated in one stage if anatomically feasible but usually treated in two stages.\[27,28\] However, the presence of distal ACA aneurysm coexisting with unilateral MCA or internal carotid artery (ICA) aneurysm complicates the treatment strategies because both PA and IHA require separate procedures that the skin incision and the fields of craniotomies interfere with each other.

The previous literature showed that 47% of multiple aneurysms are on opposite sides and 29% have one in the midline and one on the side.\[29\] Other reports showed that distal ACA aneurysm coexisted with other aneurysms 35% of the time compared with other aneurysm locations.\[30\] In our institution, 48 patients of SAH with MIAs were identified between January 2005 and March 2016, and only 3 (6%) patients had distal ACA aneurysms coexisting with MCA or ICA aneurysms. Thus, the distal ACA aneurysm coexisting with unilateral MCA or ICA aneurysm is uncommon, and few studies have focused on the concrete surgical method. Although the methods with large bone flap or two separated bone flaps through a single skin incision have been reported,\[26,31\] we present below our surgical design modifying the previously reported methods.

First, the patient is positioned 30° semi-Fowler position, the head is rotated at approximately 30° contralaterally, skull vertex in neutral position, and fixed in Mayfield skull clamp. When performing IHA, the operating table is rotated 30° to affected side laterally and 15° vertically up to accommodate to approach the distal ACA aneurysm.
Regarding the skin incision and craniotomies, we make a coronal skin incision spreading PA side widely to be possible to accommodate the frontotemporal craniotomy and ipsilateral frontal parasagittal craniotomy, and separate the field of the two craniotomies [Figure 4a]. We can accommodate to contralateral PA or basal IHA by symmetrically extending the skin incision to the contralateral side. Considering surgical stress on the frontal lobe by two approaches, we design the craniotomy of the temporal region to not expose the frontal lobe to the extent possible. We can maintain the strength and blood flow of the cranial bone by setting two bone flaps and prevent the postoperative infection. In addition, the remaining bone in a bridge shape is responsible for anchoring dura preventing acute epidural hematoma after the surgery and is used as a hand placement for improving the stability of the surgical operation [Figure 4b].

Distal ACA aneurysms are able to be observed from either the right or left side of falx but are usually treated from the ipsilateral side of the aneurysm existing. However, in cases of MIAs, where distal ACA aneurysm coexists with MCA or ICA aneurysm, we should treat the distal ACA aneurysm from the ipsilateral side of coexisting MCA or ICA aneurysm to protect the contralateral frontal lobe by the falx preventing the surgical stress on the bilateral frontal lobe. Furthermore, AcomA aneurysm is able to be observed by either PA or IHA, but in cases of MIAs, where AcomA aneurysm coexists with unilateral MCA or ICA aneurysm, the AcomA aneurysm is desirable to be treated simultaneously with the other aneurysms by PA to avoid the surgical stress on the bilateral frontal lobe. However, if it is difficult to treat the AcomA aneurysm by PA due to the large size or upward direction, AcomA aneurysm may be treated by IHA or additional endovascular coiling of necessity.

**Conclusion**

We experienced a case of SAH with MIAs. We could treat all the aneurysms in one-stage operation and obtain a good outcome using the described surgical design and strategies. Surgical clipping with ipsilateral frontotemporal and frontal parasagittal craniotomies through a single skin incision is a useful procedure for the cases of MIAs, in which distal ACA aneurysm coexists with other aneurysms on the side.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

**Acknowledgments**

The authors wish to acknowledge Dr. Kazuo Tsutumi, Assistant Chief of Showa General Hospital, for his helpful advice on various surgical techniques discussed in this paper.

**Financial support and sponsorship**

Nil.

**Conflicts of interest**

There are no conflicts of interest.

**References**

Tsunoda, et al.: Surgical design for SAH with MIAs


