Mid A1 blister aneurysm presenting with subarachnoid hemorrhage: Case report and review

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Abstract:
Blister aneurysms are uncommon and difficult-to-treat lesions. They are a substantial cause of morbidity and mortality when encountered. Here, we report a blister aneurysm of the mid A1 segment of the anterior cerebral artery presenting with diffuse basal subarachnoid hemorrhage (SAH). The aneurysm was treated by surgical clipping of the parent vessel. Postoperatively, there was no filling of the parent vessel or aneurysm. A treatment algorithm including direct surgical repair and flow diversion for ruptured blister aneurysms is described. A high level of suspicion should be maintained in the setting of angiographic-negative SAH with an asymmetrically diffuse pattern.

Keywords:
Angiography, blister aneurysm, subarachnoid hemorrhage

Introduction
Intracranial blister aneurysms are uncommon and difficult-to-treat cerebrovascular lesions. Blister aneurysms represent 1% of intracranial aneurysms and are usually located at the posterior nonbranching portions of the distal internal carotid artery (ICA). They are small lesions of intracranial blood vessels with only a fibrous layer of tissue in the wall in a hemispheric configuration. Thus, they are not true aneurysms and are typically harder to manage, with one review reporting blister aneurysms having higher hemorrhage, relapse, morbidity, and mortality rates. According to a recent meta-analysis anterior cerebral artery (ACA), blister lesions account for 0.3% of blister aneurysms. In this report, we explain a case of a very rare blister aneurysm of the mid A1 segment of the ACA (A1) presenting with diffuse basal subarachnoid hemorrhage (SAH). To the best of our knowledge, this location has yet to be reported. We also review other reports of blister aneurysms in atypical locations and delineate our multidisciplinary treatment algorithm for treating these unusual lesions, when encountered.

Case Report
Presentation
A 50-year-old woman with past medical history of migraines, myasthenia gravis, and depression presented to the emergency department with a postcoital, worst headache of her life, confusion, and seizures. Her systolic blood pressure was in the 200s and heart rate in the 40s. She presented initially to an outside hospital and was significantly drowsy and was intubated for airway protection. On initial evaluation, she was lethargic but able to follow simple commands in all extremities and had a right sixth cranial nerve palsy consistent with Hunt and Hess Grade 3. She was found to have Fisher Grade 3 SAH and acute hydrocephalus on computed tomography (CT) [Figure 1a]. A right-sided external ventricular drain was placed.
Digital subtraction angiography (DSA) revealed very small left mid A1 1.5 mm × 1.3 mm aneurysm [Figure 1c]. The morphology of the aneurysm was consistent with blood blister-like aneurysm. The neurointerventional and cerebrovascular team recommended microsurgical repair with possible parent vessel sacrifice.

**Treatment**

A standard left pterional craniotomy was performed. The Sylvian fissure was widely split lateral to medial with large amounts of SAH noted. The left optic nerve and ICA [Figure 2a white triangle] were identified as well as the M1, anterior choroidal artery, and proximal left A1 disappearing into clot [Figure 2a white arrow]. The ICA appeared to have areas of atheroma, and the proximal A1 was noted to have areas of hemorrhagic irregularity on its adventitia. While gently dissecting the clot on the backside of the mid A1, an intraoperative rupture of the blister aneurysm occurred. It was controlled with temporary clips at the proximal ICA/A1 junction and distal A1 (yet unexposed). At this point, another area in the vessel distal to the previous bleeding was found to be ruptured. As the whole A1 was likely dysplastic, the A1 was sacrificed with sequential stacking of clips up to the A1/anterior communicating artery (AComm) junction [Figure 2b]. Intraoperative angiography revealed bilateral filling of the ACA territory from the right A1/AComm as well as no angiographic filling of the left A1 or blister aneurysm [Figure 2d]. The patient remained stable during the procedure.

**Postoperative course**

Immediately after surgery, the patient was awake, alert, following commands, and moving all four extremities. Post-SAH day 4, the patient was taken for repeat angiography to assess for vasospasm. Mild spasm was present and treated with verapamil. Post-SAH day 9, the patient had an acute change in mentation and stopped following commands. Emergent angiography revealed severe diffuse vasospasm worse on the right M1 and A1 with perfusion deficits in the left ACA territory. Verapamil was again infused in the left and right carotid as well as vertebral artery, as the posterior fossa also had severe spasm. The diffuse severe spasm improved to moderate spasm with 30%–50% increase in vessel size throughout. The patient was taken the next day as well for intra-arterial verapamil infusion, and balloon angioplasty was completed at a residual focal area of narrowing in the proximal A1. The following day, the patient was able to follow commands once more. A milrinone drip was also started, and systolic blood pressure was allowed to reach the 190s for the duration of her vasospasm period. In addition, over the course of her hospitalization, she underwent multiple sessions of plasmapheresis to treat her myasthenia gravis. Four months after surgery, the patient presented for follow-up. On examination she was alert and oriented to time, place, and person with mild cognitive dysfunction and able to stand with minimal assistance.

**Discussion**

Here, we reported a case of SAH due to rupture of a blister aneurysm in an atypical location. Blister aneurysms are classically located at the posterior nonbranching portions of distal ICA\(^1\). Atypical aneurysms have also been described in other rare anterior circulation areas such as the AComm,\(^4-7\) A1/2 junction dissecting aneurysms,\(^4,8\) proximal A1 artery,\(^9\) dissecting A2 lesions,\(^10\) and A1 fusiform aneurysms.\(^11\) Table 1 exhibits a list of known studies with atypical blister aneurysm location and SAH. Both its location and the nature of blister aneurysms make a case like this a case study in evaluation and treatment options. One study classifying A1 lesions found that these lesions can rupture at small sizes, can be
Table 1: Blister aneurysm in atypical anterior circulation locations presenting with subarachnoid hemorrhage, modified from Peschillo et al., 2015[21]

| Report                        | Location          | Treatment                          | Outcome               | Spasm                  | Rupture (intraoperative) |
|-------------------------------|-------------------|------------------------------------|-----------------------|------------------------|--------------------------|
| Morris and Brophy 2009[4]     | ACOM              | Clipping                           | mRS 2 Hydrocephalus   | No                     | No                       |
| Andalu and Zuccarello 2008[5] | 5 horizontal ACOM blister | Surgical clipping                  | 2 good                | No mention             | 2/5                      |
| Seo et al., 2009[6]           | ACOM              | Surgical clipping with cotton and glue reinforcement | Moderate disability. VP shunt placed | Yes                    | Yes                      |
| Rouchaud et al., 2013[7]      | ACOM              | Bilateral A1-A2 flow diverters     | Good                  | Uneventful hospital course | No                       |
| Le Feuvre and Taylor 2011[8]  | Proximal A1 MCA   | Sundt clip                         | mRS 0                 | No                     | No                       |
| Grant et al., 2014[9]         | MCA × 3           | Stenting (neuroform)               | mRS 0-1               | No                     | No                       |
| Peschillo et al., 2015[10]    | Proximal A1       | Flow diversion (g16 device) flowed by coiling sacrifice | mRS 1                 | No, only transient deficit | No                       |
| Peschillo et al., 2015[11]    | ACOM              | Flow diversion (silk)              | Expired               | No                     | No                       |
| Bulsara et al., 2013[12]      | MCA               | Stenting (neuroform)               | Good                  | No                     | No                       |
| Pistochi et al., 2013[13]     | A2 MCA            | A. Flow diverter (silk × 2)        | mRS 0                 | No                     | No                       |

MRS: Modified Rankin scale, VP: Ventriculoperitoneal, MCA: Middle cerebral artery, ACOM: Anterior communicating artery.

difficult to detect, multiple, and presentation can include intraventricular hemorrhage.[16] Blister aneurysms can be responsible for angiography-negative SAH and may not be detected until the second or third angiograms.[5-6] In addition, CT angiography may be useful in diagnosis if DSA is negative multiple times.[5] In our case, we were able to diagnose the blister aneurysm on the first DSA which allowed timely decision-making and intervention. Had the initial angiogram been negative, we would have treated her medically for SAH and repeated angiogram in 7–10 days to reevaluate. If the second angiogram was negative and there remained high suspicion, then, we would have discussed with the patient the option of exploration through craniotomy.

Treatment options for blister aneurysms in general include clipping[17] which often times narrows the parent vessel, Sundt clip vessel reconstruction,[18] cotton clipping,[19] wrapping,[20] vessel sacrifice,[21] bypass,[22] and primary repair with suture.[23] Endovascular treatment with stents,[24] stent coiling,[25] stent-assisted onyx placement,[26] endovascular electrocoagulation,[27] or flow diversion[27,28,29] have also been described. Although in the setting of a friable vessel, endovascular stenting can lead to perforations.[30] In regard to endovascular treatment, often the need for dual antiplatelet is a deterrent; however, one study reported giving only aspirin during the procedure followed by loading postoperatively. However this study also overlapped/telescoped flow diverters to maximize aneurysm coverage.[26] In addition, some authors have utilized oversized Neuroform stents with only aspirin to circumvent the need for dual antiplatelet medications.[12] Flow diverters have thus far shown good results in the few case series published for ruptured blister aneurysms,[26-28] but long-term follow-up for this treatment is lacking, and delayed enlargement of a previously flow-diverted blister aneurysm has been reported.[29]

Combined surgical and endovascular therapy can also be employed with the use of balloon guides or balloons for flow arrest during surgical clipping.[30] One review from 2014 reported two-third of blister lesions going for open surgery and 21% of patients requiring a second treatment.[31] Careful surgical exploration is the first-line technique in the majority of cases in one series.[31] Surgery for A1 segment lesions should involve minimal retraction. Hematoma evacuation should be kept to a minimum as to not irritate small perforators irrigating the optic nerves, anterior striatum, and basal forebrain. Opening of the interhemispheric fissure or resection of the overlying gyrus rectus can sometimes help give space if needed.[16] Small lesions on the back of the artery such as ours are the most difficult to treat. Temporary clips should be used judiciously to avoid ACA infarction.[32] As in our case, if bilateral A1s are equal in caliber and the AComm patent, then vessel sacrifice may be well tolerated.

In general, our blister aneurysm treatment algorithm is as follows: if the aneurysm is unruptured, we move toward flow diversion. If the aneurysm is ruptured...
and the patient a good surgical candidate, or has a high likelihood of needing additional procedures for which dual antiplatelet medications would be contraindicated, we lean toward surgery. Surgery typically includes direct clipping, cotton clipping, Gore-Tex wrapping, and clipping. If the cerebral hemodynamics are such that a bypass is needed, and the patient not in severe spasm, direct or indirect bypass with trapping may be considered. If the patient is not a good candidate for surgical repair then flow diversion should again be considered. A recent meta-analysis noted higher immediate occlusion rates and follow-up occlusion rates with surgery compared to endovascular treatment. Surgery was however complicated by intraoperative rupture 81% of the time and did carry 20% perioperative morbidity. In addition, there were trends toward overall better outcomes with endovascular therapy. This same trend was also noted by another review article for atypical location blister aneurysms. Thus, there is no real consensus on the best management course.

Future interventions that may hold promise include Pipeline Embolization Devices with Shield Technology, that through mimicry decreases thrombogenicity and perhaps will obviate the need for dual antiplatelet medications.

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.

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Conflicts of interest

There are no conflicts of interest.

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