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Summary: I encountered 4 cases of blood blister-like aneurysm (3 ruptured and 1 unruptured) during the past 7 years. The unruptured one was accompanied by a ruptured internal carotid-ophthalmic aneurysm. These aneurysms constitute 3.4% of the total of 117 aneurysms managed by open or endovascular surgery during the same period. In these 4 cases, the lesion arose at the nonbranching site on internal carotid artery. The 4 aneurysms were operated upon via a transsylvian approach. A combination of both wrapping and clipping was done in 3 cases, and for the remaining 1 (ruptured) clipping only. The aneurysm managed by clipping only suffered postoperative massive bleeding because of an inappropriate obliteration of the proximal aneurysmal neck. The other 2 ruptured aneurysms that underwent both wrapping and clipping did not suffer any bleeding postoperatively, and 1 patient with an unruptured aneurysm has been in good condition. I detail the course of these 4 cases and problems concerning management for blood blister-like aneurysm.

Key words: blood blister-like aneurysm, subarachnoid hemorrhage

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Introduction

The incidence of the blood blister-like aneurysm (blister aneurysm) is very small, and the diagnosis and the management have been reported to be difficult because the blister aneurysm arises on an unusual site and has very thin wall with an obscure neck. There is high surgical risk of massive intraoperative bleeding because of fragile component of the aneurysm. Surgery for the lesion requires specific surgical technique because blister aneurysm has an obscure neck involving arterial wall of the parent artery. In cases of intraoperative rupture, special techniques such as repair of a tear at the base of the aneurysm with suturing and/or encircling clip is required. Consequently, surgical result and the prognosis have been reported to be associated with generally poor outcomes. During the past 7 years, the author experienced 4 cases of blister aneurysm, in which 3 cases manifested subarachnoid hemorrhage (SAH) and 1 case was unruptured (accompanied with ruptured internal carotid-ophthalmic aneurysm). These 4 blister aneurysms arose at non-branching sites of the internal carotid artery (ICA). This report summarized 4 blister aneurysms which the author encountered during the past 7 years.

Patients and case reports

In the past 7 years between January 1, 1998 and March 31, 2006, the author treated 103 aneurysm patients (including cases with multiple aneurysms), 31 men and 72 women, aged 24 to 80 (mean 64 years), with 66 ruptured aneurysms and 51 unruptured aneurysms (exclusive of cases of dissection of the cerebral artery), in which 100 aneurysm patients were treated by open surgery, and the remaining 3 patients by endovascular surgery. Fifty unruptured aneurysms were saccular in nature, and located at branching site of the cerebral artery, and the remaining 1 was an unruptured blister aneurysm located at ICA (accompanied by ruptured right IC-ophthalmic aneurysm). In ruptured aneurysms,
Fig. 1  A 75-year-old female manifesting subarachnoid hemorrhage (A), and cerebral angiograms revealing an aneurysmal bulge (arrowheads) on ventral wall of the right internal carotid artery and an aneurysm on the right middle cerebral artery (B, C). Intraoperative photos showing a blood blister-like aneurysm arising at ventral wall of the right internal carotid artery (D), direct clipping at the ventral side (E, F), and wrapping secured by a clip around the lesion (G). Postoperative angiograms revealing complete obliteration of the aneurysms (H, I). Arrow (D, F) indicates an intraoperative mirror, in which aneurysmal bulging of blister aneurysm (D), and clip branches applied on ventral carotid wall (F) are seen. Arrow (C) indicates an unruptured middle cerebral aneurysm. Arrowhead (D) indicates right internal carotid artery manipulated using a spatula. Arrow (E) indicates right posterior communicating artery. Arrow (G) indicates surgical cotton secured by a clip for an additional wrapping.
there were 3 cases of blister aneurysm arose at non-branching site of ICA.

**Case illustrations**

Case 1: A 73-year-old female with a history of liver cirrhosis and hypertension manifested SAH (Fig. 1A) with grade 3 (according to Hunt & Kosnik scale\(^3\)). Cerebral angiography revealed an aneurysmal bulge on the ventral wall of the right ICA and a saccular aneurysm on middle cerebral artery (MCA) (Fig. 1B, C). From the distribution of SAH, the aneurysmal bulge on the ICA was considered to be ruptured. On day 1 (the day of onset of SAH is day 0), right transsylvian approach was performed to identify unruptured right MCA aneurysm, and a blister aneurysm on the ventral wall of the right ICA (Fig. 1D, Fig. 2A). Arteriosclerotic change was seen on the MCA. With an inspection using a mirror, the ICA lesion was identified to locate between posterior communicating and anterior choroidal arteries. It appeared to have very thin wall with broad neck involving the wall of ICA. Rebleeding during craniotomy was considered to be likely because unexpectedly thick SAH clots were found around the lesion. Although a fenestrated clip was applied to put the clip branches on the normal wall of the ICA (Fig. 1E), a part of the thin wall of the aneurysm was seen to be residual around PcoA (Fig. 1F). Therefore, wrapping using surgical cotton (which was secured by a clip) was added including PcoA (Fig. 1G). The MCA aneurysm was also obliterated using a clip. Postoperatively, she recovered to be conscious without any neurological deficit. On day 8, while she became lethargy, angiography revealed complete obliteration of the aneurysms, spare of perforating arteries, and not any vasospasm (Fig. 1H, I). The cause of disturbed consciousness was considered to be metabolic due to hepatic dysfunction, because blood parameters such as prothrombin and partial thromboplastin times exhibited much longer (worse) values. Subsequently, she suffered no ischemic episode. Two months after the admission, she underwent a lum-
boperitoneal shunting for a development of normal pressure hydrocephalus (NPH). She, however, suffered heavy intellectual impairment.

Case 2: A 61-year-old female with a history of hypertension manifested SAH with grade 3. Cerebral angiography revealed an aneurysmal change on lateral wall of the right ICA. Early surgery for the lesion was done via a right pterional transsylvian approach on day 1. The aneurysm was identified on lateral wall of the right ICA (Fig. 2B). It appeared red, and had very thin wall with an obscure neck. Branching artery was not found around the lesion. Arteriosclerotic change was seen on ICA and MCA. The aneurysm was wrapped with surgical cotton, and then the surgical cotton was secured using a curved clip. Postoperative hospital course was uneventful, and 2 months after the admission, she was discharged without any neurological deficit after a shunting for a development of NPH.

Case 3: A 49-year-old female manifested SAH with grade 5 because of rebleeding just before hospitalization. Emergent barbiturate therapy (using pentobarbital of 200 mg per hour) was begun after orotracheal intubation. On day 1, cerebral angiography revealed an aneurysmal bulge on dorsal wall of the right ICA. Early surgery via a right transsylvian approach was done to find an aneurysmal bulge on dorsal ICA (Fig. 2C). The lesion appeared to have very thin wall with an obscure neck (broad) involving the wall of the ICA. During manipulation around the lesion, intraoperative bleeding from the aneurysm occurred. Temporary clips were applied to proximal and distal sides of the lesion to form trapping. A right-angled clip was used to apply the clip branches below the base of aneurysm. However, the clip branches within the margin of normal arterial wall appeared to be causative of severe stenosis or obstruction of the ICA. Therefore, the clip branches were shifted to avoid carotid stenosis or obstruction. After relief of the trapping, no further bleeding was observed during the surgery. The patient, however, suffered postoperative bleeding with a resultant cerebral death on day 13. An insufficient obliteration of the proximal neck after the shift of the clip was considered to be causative for the postoperative bleeding.

Case 4: A 28-year-old male suffered SAH followed by head injury (cerebral contusion) with consciousness disturbance. On day 15 after the ictus, open surgery for right IC-ophthalmic ruptured aneurysm (partially thrombosed) was done to obliterate the aneurysm. An unruptured blister aneurysm on dorsal wall of the ICA (Fig. 2D) close to the ruptured aneurysm was found (the blister aneurysm was not identified in preoperative angiography on day 13). The lesion was managed with wrapping secured by a clip. The ruptured aneurysm was managed with neck clipping after anterior clinoidectomy. Postoperative course was uneventful, and he was discharged 1 month after the surgery.

In summary, 4 (3.4%) out of 117 aneurysms obliterated were blister aneurysm. One suffered intraoperative and postoperative bleeding with a resultant cerebral death, 1 suffered intraoperative bleeding during the procedure of craniotomy and resulted in heavily intellectual impairment, and other 2 exhibited good recovery. Until the day of March 31, 2006 (follow-up period is from 4 months to 5 years after surgery), the 2 survived patients after SAH showed no further bleeding, and 1 unruptured case had been in a stable condition.

Discussion

Incidence of blister aneurysm has been reported very rare.\textsuperscript{10} Reportedly,\textsuperscript{10} among 557 cerebral aneurysms surgically treated, 29 (5.2%) were unrelated to arterial divisions and 5 (0.85%) were blister aneurysms. Four out of the 5 cases were reported to suffer intraoperative bleeding with poor clinical outcomes.\textsuperscript{10} In cases of aneurysms unrelated to arterial division, only 10% were located on the ventral wall of the artery.\textsuperscript{10} Therefore, blister aneurysm of ventral wall of the ICA as shown in case 1 is considerably rare (presumably less than 0.1% in incidence). Blister aneurysm has very thin wall, the lesion is very fragile, and pathological findings in a blister aneurysm is reported to show a laceration of the arterial wall based on degeneration of the internal elastic lamina.\textsuperscript{2}

The surgical treatment of ruptured blister aneurysm is still controversial. Several problems as for blister aneurysm have been reported: 1. open surgery for the lesion carries a high risk of intraoperative massive bleeding because of danger of whole arterial wall rupture. 2. specific surgical approach and techniques such as subpial dissection of the dome or positioning the clip branches below the base of aneurysm within the margin of normal arterial wall are necessary. 3. there are cases with difficulty to diagnose the lesion in angiography because of an unusual location with obscure neck.\textsuperscript{15}

As the direct surgical approach for blister aneurysm carries a high risk, endovascular management or observation for the lesion has been discussed in recent several reports. A case who underwent endovascular oblit-
eration of a ruptured blister aneurysm followed by proximal occlusion is reported with a favorable result. A case is reported, in which an observation with serial angiograms for a ruptured blister aneurysm revealed a formation of a saccular aneurysm with subsequent successful obliteration using a clip. Combination of endovascular with neurosurgical approaches, in which preliminary balloon occlusion of the ICA followed by surgical trapping of the ICA beyond the lesion is reported to be a safer alternative to primary surgery of blister aneurysm.  Abrupt growth of a blister aneurysm after wrapping for the lesion in acute stage is reported, in which subsequent endovascular embolization was successful.

In cases of intraoperative bleeding to produce large arterial defect, a repair technique such as suturing of the arterial wall may be necessary. Concerning surgical methods, reported statistical analysis indicated that clipping on wrapping materials had better result compared with other methods such as clipping, wrapping or ICA trapping.

In the present study, wrapping using surgical cotton followed by securing the cotton with a clip was successful in case 2. In case 3, clipping without wrapping was ineffective because of an insufficient positioning of clip branches. In this case, shift of the clip to avoid carotid stenosis was inappropriate, and it is thinkable that additional wrapping should have been done to support the clipping, and to prevent postoperative bleeding. In case 1, although direct clipping for the blister aneurysm was performed, a part of the thin wall of the aneurysm was identified to remain around PcoA. Therefore, additional wrapping (secured by a clip) involving PcoA was necessary, and this maneuver seemed to prevent further bleeding. Although case 3 suffered postoperative bleeding and case 1 suffered intraoperative bleeding during the procedure of craniotomy, careful transsylvian approach to prevent inappropriate retraction of the frontal lobe exposed the lesion successfully, so that it is thinkable that the problem in the direct surgery for blister aneurysm might not the method of surgical approach but the modality of obliteration of the lesion.

If possible, clipping within a normal wall of parent artery involving blister aneurysm may have high surety, but this method carries a risk of stenosis (or obstruction) of the parent artery, and shift of clip branches to avoid the complication may result in an insufficient clipping to ensure intra- or postoperative bleeding. According to pertinent literatures, and considering the present experiences, combination of both clipping and wrapping, especially clipping on wrapping material is considered to have much safety, and exhibit much better result.

The author reported 4 cases of blister aneurysm located on ICA in 117 cerebral aneurysms (103 aneurysm patients). In these 4 cases, 3 were ruptured. Careful transsylvian approach should successfully expose the lesion. The problem in managements for blister aneurysm seems to depend on the modality of the obliteration. From the outcomes of the present cases and a review of the pertinent literatures, combination of both clipping and wrapping was considered to be one of the most favorite managements for blister aneurysm.

References
要 旨

脳血管非分岐部に生じた脳動脈瘤：血豆状脳動脈瘤 4例の報告

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著者は、過去7年間に103例の脳動脈瘤（117脳動脈瘤）を経験し（100例は直達手術、3例は血管内手術を施行した）、うち4例（3例が破裂、1例が未破裂）が血豆状脳動脈瘤であった。これらはすべて内頸動脈の非血管分岐部に生じた。すべて経シルピウス到達法にて手術を行い、wrappingとclippingの組み合わせによる脳動脈瘤処理を3例に行い、1例ではclippingのみを行った。結果としてはclippingのみを行った破裂症例にて術後に再出血を認めた。wrappingとclippingを行った破裂症例（2例）では、術後出血は認めていない。1例の未破裂例では著変なく経過している。これらの4症例について詳しく報告し、血豆状脳動脈瘤手術に関しての問題点に関し文献的考察を加えて報告する。