Less Invasive Management of Endovascular Embolization and Neuroendoscopic Surgery for a Dural Arteriovenous Fistula Presenting with Acute Subdural Hematoma

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Key Messages
- A relatively rare case of a DAVF resulting in ASDH and intracerebral hemorrhage in the left occipital lobe is presented.
- This is a successful case of DAVF managed less invasively with endovascular embolization and hematoma removal under neuroendoscopy with small craniotomy.
- Our surgical strategy can be a less invasive option for DAVF manifesting ASDH.

Abstract
Acute subdural hematoma (ASDH), which causes midline shift of the brain, rarely arises from a dural arteriovenous fistula (DAVF). Herein, we report the first case of a DAVF manifesting ASDH, which was treated less invasively with endovascular embolization of a drainer of the DAVF and hematoma removal under neuroendoscopy. A 59-year-old man with a sudden onset of headache was transported to our hospital. Left ASDH and intracerebral hematoma in the left occipital lobe were detected. A cerebral angiogram revealed a DAVF fed by the petrosquamous branch of the left middle meningeal artery and jugular branch of the right ascending pharyngeal artery. The shunting point in the lateral tentorial DAVF drains through the internal occipital vein to the superior sagittal sinus. A varix was recognized in the draining vein (Borden type 3, Cognard type 4). The DAVF was embolized with Onyx (Medtronic, Minnesota, USA), and the left ASDH was removed with a small craniotomy under neuroendoscopy. No origin of the left ASDH was apparent in the surgical field. The patient was discharged from the hospital on postoperative day 18. The patient’s status was modified Rankin scale 1 on discharge. Our management of combined endovascular treatment and neuroendoscopic hematoma removal may be useful and less invasive for hemorrhagic DAVF.

Keywords
► dural arteriovenous fistula
► endovascular
► Onyx
► neuroendoscope
► acute subdural hematoma
Introduction

Dural arteriovenous fistula (DAVF) accounts for approximately 10 to 15% of intracranial arteriovenous malformations. A high risk of hemorrhagic events is estimated in DAVFs with retrograde cortical venous drainage. Following intracranial bleeding such as intracerebral hemorrhage (ICH), occurrence of subarachnoid hemorrhage has been described, but acute subdural hematoma (ASDH) with midline shift of the brain is a relatively rare hemorrhagic manifestation of DAVF. To our knowledge, reports on the minimally invasive management of DAVF presenting as ASDH seem to be scant to date. Herein, we present the first case of DAVF manifesting ASDH and ICH, which was less invasively managed with endovascular embolization and direct surgery under neuroendoscopy.

Case Description

A 59-year-old man was transferred to our hospital after a sudden onset of headache without any head trauma. On arrival, his Glasgow Coma Scale score was 15. Neurological examination revealed right hemianopia. Left ASDH and ICH in the left occipital lobe were detected using computed tomography (CT). Apparent bony fractures were not detected. CT angiography revealed an arteriovenous shunt. The bleeding origin of the left ASDH was not found in the subdural space of the anterior or middle cranial fossa. To rule out a traumatic event, the patient was examined using magnetic resonance imaging (MRI). No apparent contusions were observed in the brain. However, the left ASDH, ICH in the left occipital lobe, and middle ASDH were clearly observed on the MRI (Fig. 1C, D). We considered that the middle ASDH could have resulted from the increased ICH due to an arteriovenous shunt. Further, since there was no apparent contusion or bony fracture, we considered that the left ASDH could have been related to the arteriovenous shunt. As the patient’s neurological status did not aggravate, we examined the arteriovenous shunt using cerebral angiography under general anesthesia. The arteriovenous shunt was supplied by the left petrosquamous branch of the middle meningeal artery and jugular branch of the right ascending pharyngeal artery. The arteriovenous shunt had a shunting point at the lateral tentorial sinus over the cerebellar tentorium. The shunt was retrogradely drained into the internal occipital vein through the shunting point. The drainer had a varix and bleb, which we suspected were caused by hemorrhagic events. The flow of the transverse sinus and sigmoid sinus was anterograde (Fig. 2A–F). Thus, we diagnosed the lesion as tentorial DAVF (Cognard type 4, Borden type 3). Following diagnostic cerebral angiography, endovascular embolization was performed. Activated clotting time was maintained between 150 and 200 seconds with systematic heparinization. The drainage of the arteriovenous fistula was embolized using Onyx (Medtronic, Minnesota, USA). The varix was partially embolized (Fig. 2G, H). Embolization with Onyx was completed in 10 minutes. After endovascular surgery, the arteriovenous shunt had disappeared. The total operation time, including diagnostic angiogram, was 1 hour and 25 minutes. Heparinization was reversed with protamine after endovascular surgery. Postoperative CT revealed that the left ASDH had not increased (Fig. 3A). Following endovascular surgery, we planned hematoma removal to release the compression of the brain due to the left ASDH. The patient was then moved to an operating room. We planned hematoma removal with a small craniotomy using a neuroendoscope. A skin incision was designed to be extended for external decompression (Fig. 3B). Intraoperatively, the brain did not show a remarkable swelling and remained slack after removal of the left ASDH. No bleeding origin was identified in the surgical field. Therefore, we concluded that external decompression was not necessary. The midline shift of the brain improved (Fig. 3C). Total intraproductive bleeding was 15 mL. The patient was extubated the day after confirming the absence of postoperative hemorrhage. Postoperatively, the patient’s status improved with rehabilitation therapy. Right homonymous hemianopia was observed, but it did not impede the patient’s occupation. The patient was discharged 18 days after the surgery. The modified Rankin Scale score was 1 at discharge from our hospital. Recurrence of the arteriovenous fistula was not observed on cerebral angiography performed 6 months after discharge. Upper right hemianopia persisted, but the patient could drive a car to renew his driver’s license.

Fig. 1 (A) Intracerebral hematoma in the left occipital lobe and left acute subdural hematoma are observed. (B) A vascular lesion was suspected near the inner surface of the left occipital lobe. (C, D) An increase in left and middle acute subdural hematoma is observed. There is no evidence of a contusion.
Subdural hematoma is a rare hemorrhagic manifestation of DAVF. Similar cases of concurrent ASDH and ICH in the occipital lobe, as in our case, have been described as an even more rare manifestation\(^4\)–\(^7\) (\(\textbf{Table 1}\)). Due to the rarity of the concurrence of ASDH and ICH in the occipital lobe as a bleeding pattern from DAVF, standardized management has not yet been established. Matsuzaki et al embolized a DAVF and did not perform surgery.\(^6\) This is possibly because the brain was not strongly compressed by the bilateral ASDH. Kitazono et al performed direct surgery of a DAVF using intraoperative subtraction angiography.\(^7\) Both Suyama et al and Saito et al reported similar cases of hemorrhagic DAVF treated with endovascular embolization and direct surgery (hematoma removal and external decompression).\(^4,5\) Suyama et al performed direct surgery prior to endovascular embolization, while Saito et al performed endovascular embolization prior to direct surgery.\(^4,5\)

In our case, obliteration of DAVF and removal of ASDH were both necessary. The bleeding point of the DAVF in our case was located medially, while the ASDH was located laterally. If we had planned to manage both the shunting point of the DAVF and ASDH surgically in one session, the craniotomy would have been large, and the surgery in one session could have been invasive. In our case, the arteriovenous fistula and varix were first embolized. As the shunting point of the DAVF was obliterated, we thought that a small conventional craniotomy would be sufficient to remove the left ASDH. The small conventional craniotomy in our case was successful as less invasive management.

Owing to the rarity of ASDH resulting from a DAVF, a standardized surgical management for a DAVF accompanied by ASDH is yet to be established. In our case, endovascular embolization of the drainer of the DAVF preceded the removal of the left ASDH. Our decision was made based on the patient’s neurological status (Glasgow Coma Scale score of 15).
Table 1 Reported cases of dural arteriovenous fistula manifesting intracerebral hemorrhage in the occipital lobe and acute subdural hematoma

<table>
<thead>
<tr>
<th>Article (author, y)</th>
<th>Case (age, sex)</th>
<th>Symptoms</th>
<th>Hematoma location</th>
<th>Feeder</th>
<th>Drainage route</th>
<th>Surgical strategy</th>
<th>Outcome</th>
</tr>
</thead>
<tbody>
<tr>
<td>Matsuzaki et al 2009</td>
<td>66 y, male</td>
<td>Mild alexia and agraphia</td>
<td>Bilateral ASDH ICH in the left temporo-occipital lobe</td>
<td>Multiple feeders of the left occipital artery</td>
<td>Temporo-occipital veins</td>
<td>1 observation (1 week) 2 endovascular embolization</td>
<td>mRS 1 (alexia and agraphia remained)</td>
</tr>
<tr>
<td>Kitazono et al 2010</td>
<td>68 y, male</td>
<td>Headache</td>
<td>Left ASDH ICH in the left occipital lobe</td>
<td>A parietal branch of the left middle meningeal artery</td>
<td>The superior sagittal sinus through cortical veins on the occipital lobe</td>
<td>1 observation 2 obliteration of the drainer and removal of ICH</td>
<td>Right hemianopia, discharged 25 days after the onset</td>
</tr>
<tr>
<td>Saito et al 2014</td>
<td>56 y, male</td>
<td>Clouded consciousness difficulty in opening eyes</td>
<td>Right ASDH ICH in the right occipital lobe</td>
<td>The right middle meningeal artery, a parietal branch of the superficial temporal artery, and a meningeal branch of the right occipital artery</td>
<td>The superior sagittal sinus and vein of Galen through cortical veins on the occipital lobe</td>
<td>1 endovascular embolization 2 external decompression and removal of ASDH 3 cranioplasty</td>
<td>Communicating with conversation, walking with aid</td>
</tr>
<tr>
<td>Suyama et al 2018</td>
<td>61 y, female</td>
<td>Headache and unconsciousness (Glasgow Coma Scale 6)</td>
<td>Left ASDH ICH in the left occipital lobe</td>
<td>The left posterior meningeal artery</td>
<td>The internal occipital vein</td>
<td>1 external decompression and removal of ASDH 2 endovascular embolization (4 days after the surgery) 3 cranioplasty (1 month after the surgery)</td>
<td>Discharged 2 weeks after cranioplasty Neurologically intact at a 3-year follow-up</td>
</tr>
<tr>
<td>Present case</td>
<td>59 y, male</td>
<td>Headache</td>
<td>Left ASDH ICH in the left occipital lobe</td>
<td>A petrosquamous branch of the left middle meningeal artery, and a jugular branch of the right ascending pharyngeal artery</td>
<td>The lateral tentorial sinus and internal occipital vein</td>
<td>1 endovascular embolization 2 removal of ASDH under neuroendoscope</td>
<td>mRS 1 (upper right hemianopia remained)</td>
</tr>
</tbody>
</table>

Abbreviations: ASDH, acute subdural hematoma; ICH, intracerebral hemorrhage; mRS, modified Rankin Scale.
Although the left ASDH and middle ASDH increased after MRI, the patient’s neurological status did not aggravate. Thus, we decided to first evaluate the bleeding origin of the intracranial hematoma. Our surgical strategy has the advantage that the bleeding origin of ASDH is already obliterated prior to direct surgery. Because systematic heparinization is necessary in endovascular embolization, ASDH existing preoperatively can increase after endovascular embolization. If the ASDH continues to compress the brain, it must be removed. Removal of ASDH can be unnecessary in cases where ASDH either does not increase or only increases slightly. If ASDH is not removed, the patient should be monitored with repeated CT examinations.

However, if the patient is in a comatose state due to cerebral herniation resulting from ASDH, the ASDH should be removed. In cases where the ASDH is removed first, surgeons should consider the possibility that ASDH can increase because the bleeding origin is not obliterated. Thus, external decompression with a large craniotomy should be performed. Suyama et al. chose this surgical strategy.4

Our institute has previously reported the usefulness and less invasiveness of removing ASDH under neuroendoscopy in elderly patients.8 The present case was not an elderly patient, and his brain was not atrophic. Thus, intraoperatively, we should always consider the necessity of external decompression. To prepare for the intraoperative need of external decompression, the skin incision should always be designed as a partial line of a large skin incision. However, as shown in our case, the ASDH arising from a DAVF can be removed only with a neuroendoscope after obliteration of the shunting point of DAVF.

**Conclusion**

A combination of endovascular embolization and hematoma removal under neuroendoscopy may be a less invasive therapy for cases of DAVF manifesting ASDH and ICH.

**Informed Consent**

We obtained written informed consent from the patient.

**Conflict of Interest**

None declared.

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**References**


