

# Recurrence of a Refractory Chronic Subdural Hematoma after Middle Meningeal Artery Embolization That Required Craniotomy

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Middle meningeal artery (MMA) embolization has been performed to treat refractory chronic subdural hematoma (CSDH) with good reported outcomes. We have treated three cases of CSDH with MMA embolization to date, but there was a postoperative recurrence in one patient, which required a craniotomy for hematoma removal and capsulectomy. MMA embolization blocks the blood supply from the dura to the hematoma outer membrane in order to prevent recurrences of refractory CSDH. Histopathologic examination of the outer membrane of the hematoma excised during craniotomy showed foreign-body giant cells and neovascular proliferation associated with embolization. Because part of the hematoma was organized in this case, the CSDH did not resolve when the MMA was occluded, and the development of new collateral pathways in the hematoma outer membrane probably contributed to the recurrence. Therefore, in CSDH with some organized hematoma, MMA embolization may not be effective. Magnetic resonance imaging (MRI) should be performed in these patients before embolization.

**Keywords:** chronic subdural hematoma, refractory chronic subdural hematoma, middle meningeal artery embolization, craniotomy

## Introduction

Burr hole irrigation and drainage are usually performed as initial treatment for chronic subdural hematoma (CSDH), but recurrences have been reported in approximately 5–33% of these cases.<sup>1–5)</sup> Treatment for recurrences include percutaneous puncture and aspiration, placement of an Ommaya reservoir,<sup>6,7)</sup> a subdural-peritoneal shunt,<sup>6,8)</sup> and craniotomy.<sup>6)</sup> Endovascular embolization of the middle meningeal artery (MMA) blocks blood that is supplied via the dura to the hematoma outer membrane<sup>9)</sup> and is useful as treatment of recurrent CSDH. The case of a patient who had a recurrence after MMA embolization for refractory CSDH who ultimately required a craniotomy for hematoma removal and capsulectomy is reported, along with imaging and histopathologic findings.

## Case Presentation

A 59-year-old male had a history of alcoholic liver cirrhosis and pulmonary emphysema. He presented with right

hemiplegia 2 months previously and was diagnosed with a CSDH by a local physician. Burr hole irrigation was performed after transfusion of fresh-frozen plasma, but because of a recurrence on postoperative day 3, he was referred to our hospital. At the time of arrival, the patient was somnolent and had right hemiparesis. Head computed tomography (CT) showed a thick hematoma in the left central convexity (Fig. 1a). Since T<sub>2</sub>-weighted magnetic resonance imaging (MRI) showed low signal intensity in part of the hematoma cavity, organized hematoma was suspected (Fig. 2a, b).<sup>10)</sup> Because of liver dysfunction and a low platelet count (41,000/μL), fresh-frozen plasma and platelets were transfused during the perioperative period, and burr hole irrigation was again performed. However, on postoperative day 7, head CT showed a second recurrence. MMA embolization was performed under local anesthesia, followed by burr hole irrigation.

After systemic heparinization, a guiding catheter was placed in the external carotid artery, and a microcatheter was guided to the MMA. Superselective MMA angiography revealed an abnormal vascular network (Fig. 3a). Embolization was performed using polyvinyl alcohol (PVA) particles (size: 250–355 μm) and fibered coils. Angiography after embolization no longer showed the MMA (Fig. 3b). The heparinization was reversed, and burr hole irrigation was then performed.

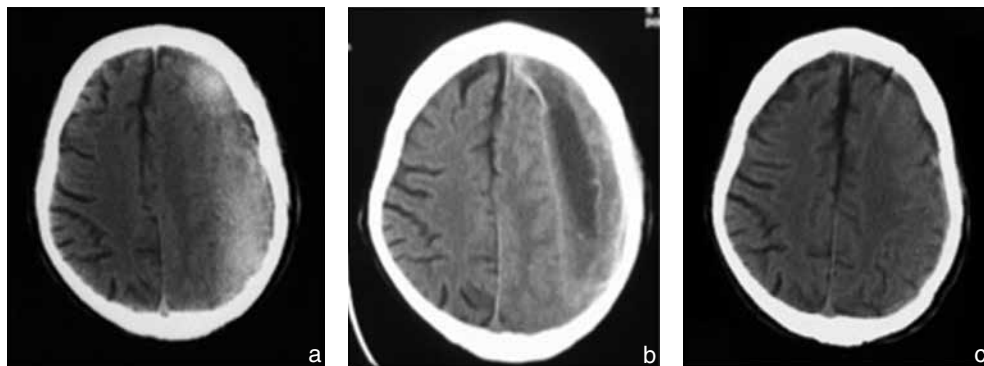
Head CT after burr hole irrigation showed organized hematoma (Fig. 1b). We have a follow up for symptoms had subsided. A recurrence occurred about 3 months later, so a craniotomy for hematoma removal was performed under general anesthesia. Intraoperative findings showed a four-layered structure of the hematoma, including an outer membrane, organized hematoma, liquid hematoma, and an inner membrane; there was also blood flow within the MMA that was occluded (Fig. 4a). The outer and inner membranes were fibrosed, and there were only a few adhesions with surrounding tissue (Fig. 4b). Histopathologic examination showed embolic material, foreign-body giant cells, and neovascular proliferation in the outer membrane (Fig. 5). During 2 years of subsequent follow-up, there has been no hematoma recurrence (Fig. 1c).

## Discussion

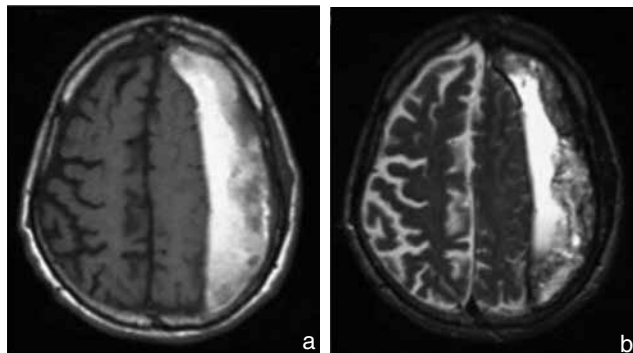
Burr hole irrigation was performed in 381 patients with CSDH at our hospital between January 2000 and March 2011. Of these, 72 patients had a recurrence requiring further surgery, including 15 cases of a second recurrence and four cases with a third recurrence. In three patients,

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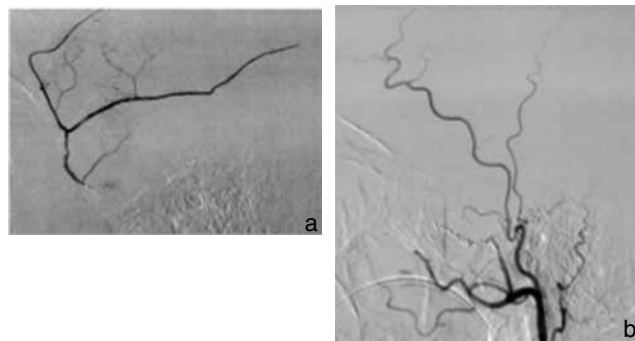
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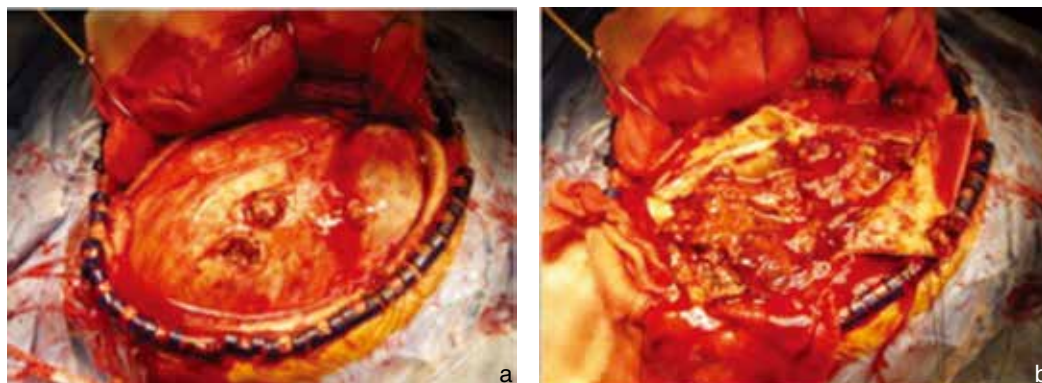
**Fig. 1** a: Plain head computed tomography (CT) before embolization shows a chronic left subdural hematoma with an internal area of isodensity. b: Plain head CT 6 months after craniotomy for hematoma removal and capsulectomy shows no recurrence of the hematoma. c: During 2 years of subsequent follow-up, there has been no hematoma recurrence.



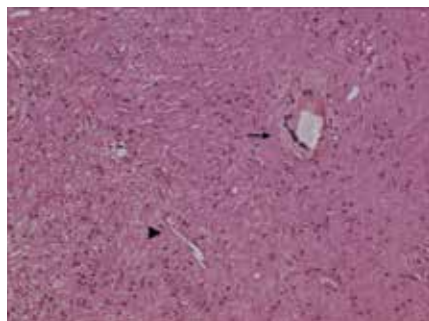
**Fig. 2** a, b: T<sub>1</sub>-weighted (a) and T<sub>2</sub>-weighted (b) head magnetic resonance imaging images before embolization show a mixture of a liquid hematoma component (T<sub>1</sub>- and T<sub>2</sub>-weighted: high signals) and an organized component (T<sub>1</sub>-weighted: isodense signal, T<sub>2</sub>-weighted: low signal).



**Fig. 3** a: Selective middle meningeal artery (MMA) angiography before embolization. There is an abnormal vascular network extending along the dura from the anterior and posterior branches. b: External carotid angiography after embolization. The MMA is completely embolized with polyvinyl alcohol and coils.



**Fig. 4** a: Craniotomy findings. There is no recanalization of the middle meningeal artery where embolization was performed. b: Dural incision findings. The hematoma has a four-layered structure, including an outer membrane, organized hematoma, liquid hematoma, and inner membrane.



**Fig. 5** Outer membrane of the hematoma (×100 magnification). Small branches from the middle meningeal artery that feed the dura are embolized with polyvinyl alcohol, and there are foreign-body giant cells (arrow). In addition, there is proliferation of new blood vessels (neovascularization) with a fragile structure (arrowhead).

including one with a third recurrence and two with a second recurrence in whom there was a high risk of further recurrence, MMA embolization was performed. These three patients were all men, with a mean age of 67 years (range: 59–78 years) (Table 1). Angiography in all these patients showed an abnormal vascular network (Fig. 3a), and among those in whom embolization was performed, two had no further recurrences after follow-up for at least 6 months. However, the patient in this case report had re-enlargement of the hematoma 3 months after embolization, and a craniotomy for hematoma removal with a capsulectomy was performed.

MMA embolization was performed under local anesthesia. After systemic heparinization, a guiding catheter was placed in the external carotid artery, and a microcatheter was guided to the MMA. Superselective angiography showed an abnormal vascular network, but no dangerous anastomoses were seen. The MMA was occluded using 250- to 355- $\mu$ m PVA particles and fibered coils. After embolization, the heparinization was reversed, and then burr hole irrigation was performed.

MMA embolization for refractory CSDH was first reported by Mandai et al. in 2000,<sup>9</sup> and since then, based on our literature review, it has been performed in 42 patients.<sup>9,11–18</sup> Treatment outcomes have generally been

good, and of a total of 45 cases (including our three patients), no recurrences have been reported in 40 patients after treatment (Table 2). In the present patient, a complete cure was not achieved with embolization, so a craniotomy for hematoma removal and capsulectomy was necessary. This is the first case report of histopathologic examination of a hematoma outer membrane after MMA embolization for CSDH.

In reports to date, burr hole irrigation has been performed as initial treatment in all cases, but there has been no uniform consensus regarding when to perform embolization after recurrences. Embolization is generally performed after at least two recurrences or after one recurrence when there is a high risk of further recurrence. This is in general agreement with the policy at our medical center. The reasons for selecting embolization rather than craniotomy have been similar to the reason in the present patient, namely, because of a bleeding diathesis associated with liver failure<sup>9,16,18</sup> or anticoagulant therapy,<sup>12,16</sup> or the presence of risk factors for general anesthesia such as pneumoconiosis<sup>19</sup> or chronic renal failure.<sup>11,13</sup>

Selective MMA angiography is done in all cases when surgery is performed to identify an abnormal vascular network. The embolic materials that are used vary among reports, but they include gelatin sponges, n-butyl-2-cyanoacrylate (NBCA), coils, PVA, gelatin sponges+coils, and PVA+coils.

NBCA and small-sized PVA particles are effective to embolization of abnormal vessels in the dura or hematoma membrane. On the other hand, Ishihara et al. stated that, in MMA embolization, attention must be paid to dangerous anastomoses, such as the recurrent meningeal artery,<sup>13</sup> and that an injection site must be carefully selected for those materials. To avoid embolization through a dangerous anastomosis, we only use PVA particles with a size of at least 250 to 355  $\mu$ m. In addition, because of concern about

**Table 1** List of patients

No.	Age	Sex	Past medical history	Number of recurrences	Further recurrence
1	78	Male	None	3	No
2	64	Male	None	2	No
3	59	Male	Alcoholic liver cirrhosis Pulmonary emphysema	2	Yes

**Table 2** List of reported cases

Report	No. of cases	Past medical history	AVN	Embolic material	Concomitant surgery with embolization	Recurrence	Additional treatment
Mandai et al. (2000) <sup>9</sup>	1	Alcoholic liver cirrhosis	Yes	PVA	No	No	
Takahashi et al. (2002) <sup>17</sup>	2(3)	None	Yes	PVA	No	No	
Hirai et al. (2004) <sup>12</sup>	2	Anticoagulant therapy	Yes	PVA+coil	No	No	
Takeshima et al. (2004) <sup>18</sup>	1	Alcoholic liver cirrhosis	Yes	Gelatin sponge	Drainage	Yes (1/1)	Embolization Drainage
Fushihara et al. (2006) <sup>11</sup>	5	CRF and other diseases	Yes	NBCA	No	No	
Ishihara et al. (2007) <sup>13</sup>	7	CRF and other diseases	Yes	NBCA	No	No	
Fujita et al. (2007) <sup>19</sup>	1	Pneumoconiosis	Yes	PVA+coil	No	No	
Mino et al. (2010) <sup>15</sup>	4	None	Yes	Gelatin sponge+coil	No	No	
Kawaguchi et al. (2010) <sup>14</sup>	7	None	Yes	Coil	No Drainage	No	
Nakajima et al. (2011) <sup>16</sup>	12	Alcoholic liver cirrhosis Anticoagulant therapy	Yes	Not reported	Small craniotomy for hematoma removal	Yes (3/12)	Embolization Drainage
Present series (2013)	3	Alcoholic liver cirrhosis Pulmonary emphysema and other diseases	Yes	PVA+coil	Drainage	Yes (1/3)	Craniotomy for hematoma removal and capsulectomy

AVN: abnormal vascular network, PVA: polyvinyl alcohol, NBCA: n-butyl-2-cyanoacrylate, CRF: chronic renal failure.

possible recanalization of the MMA when using PVA alone, we also use fibered coils for embolization of the proximal MMA.<sup>20</sup> Treatment with embolization alone has been reported.<sup>11,13</sup> The use and type of additional treatment for hematomas after embolization varies among reports. Drainage and a small craniotomy for hematoma removal have occasionally been reported.<sup>14–16,18</sup> To promptly improve symptoms and increase activities of daily living in patients at our hospital, we perform burr hole irrigation immediately after embolization.

Recurrences after embolization have been reported in five cases, including the present patient. Treatment for recurrence has included additional embolization<sup>18</sup> or burr hole irrigation alone with a good outcome.<sup>16</sup> In a patient with recurrence after embolization with gelatin sponges alone, middle cerebral artery recanalization occurred, and the mechanism was thought to have involved resorption of the gelatin sponge.<sup>18</sup> In another case report of recurrence, the embolic material was unknown, and the mechanism of recurrence was unclear.<sup>16</sup> To prevent recanalization, we performed embolization using a combination of PVA and coils. However, we had one patient with a recurrence.

Furthermore, in cases of recurrence after embolization to date, only CT imaging has been performed, and whether organized hematoma was present has not been mentioned.<sup>16,18</sup> Organized hematoma in CSDH appears slightly hypointense on T<sub>1</sub>-weighted and T<sub>2</sub>-weighted MRI. Preoperative CT and MRI in the present patient suggested the presence of organization within the hematoma. With organized CSDH, complete cure with burr hole irrigation alone is achieved in only a small number of patients, and craniotomy for hematoma removal and capsulectomy is usually necessary.<sup>21,22</sup>

Small craniotomy and drainage was applied after the MMA embolization, but CSDH relapsed in this case. We consider that remaining organized hematoma delay the healing, giving a period for the development of collateral flow. The present patient also required a craniotomy for hematoma removal and capsulectomy to achieve healing.

The histopathologic findings of the outer membrane of hematoma in CSDH after MMA embolization have not previously been reported. Histopathologic examination of the hematoma outer membrane in the present patient showed foreign-body giant cells, thought to be a reaction to the embolization material, as well as neovascular proliferation (Fig. 5). These findings suggest the development of collateral pathways during the 3 months before recurrence occurred after embolization. The recurrence was probably due to reperfusion of blood flow from the dura to the outer membrane. The period of 3 months until recurrence after embolization was longer compared to previous recurrences, probably because of the time required for collateral pathways to develop. In an organized hematoma, the absence of healing within a short time suggests the possibility that the effectiveness of MMA embolization is insufficient. To predict the effectiveness of embolization, the presence or absence of organized hematoma should be evaluated preoperatively.

The limitation of this study is that it had no angiographic

evaluation of MMA recanalization or development of collateral flow at recurrence. In addition, embolic material was limited to PVA+coil, it cannot refer to the curing of the organized CSDH in the case of selecting other embolic materials such as NBCA.

## Conclusion

MMA embolization is useful for treating refractory CSDH, with a success rate of more than 90%. However, resolution of a CSDH after embolization may be delayed when there is organized hematoma, and the development of new collateral pathways may decrease the therapeutic effectiveness of MMA embolization. Particularly, MMA embolization with PVA and coil might be disabled to cure it. When considering MMA embolization, preoperative MRI to confirm the absence of organized hematoma should be performed.

## Conflicts of Interest Disclosure

None declared. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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