



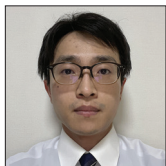
Case Report

Total removal of anaplastic meningioma infiltrating an artery by performing an A3–A3 side-to-side anastomosis

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ABSTRACT

Background: Meningiomas are histologically benign tumors and generally have a good prognosis. However, some are classified as high-grade meningiomas due to their strong invasion of surrounding tissues and high postoperative recurrence rates, resulting in a poor prognosis. Postoperative radiotherapy is often administered for the most malignant anaplastic meningiomas; however, its contribution to improving prognosis and reducing recurrence rates in patients with residual tumors is limited.

Case Description: We present here a 48-year-old man with an anaplastic meningioma that recurred repeatedly and had invaded the right anterior cerebral artery (ACA) despite two postoperative radiotherapy sessions. Dissecting the tumor from the blood vessels was extremely difficult and would only have achieved a partial resection. However, we achieved complete resection by performing a pericallosal artery–pericallosal artery (A3–A3) side-to-side anastomosis and excising the infiltrated blood vessels along with the tumor *en bloc*. No neurological deficits or complications, such as cerebral infarction, were detected postoperatively.

Conclusion: Although reports of performing an A3–A3 side-to-side anastomosis to enable complete resection of tumors invading the ACA are extremely rare worldwide, this procedure should be recognized as a safe and effective treatment option when complete tumor resection is strongly desired, as in the present patient.

Keywords: A3–A3, Adjuvant radiotherapy, Anaplastic meningioma, Side-to-side anastomosis, Vascular infiltration

INTRODUCTION

High-grade meningiomas (World Health Organization [WHO] Grade II or III) are prone to invading surrounding normal tissues and are difficult to treat, often recurring even after a combination of surgical resection and postoperative radiotherapy.^[4,9] When these tumors have invaded surrounding blood vessels and extended beyond the adventitia into the deeper layers, surgically separating them from those blood vessels is challenging.^[6] Preservation of the infiltrated blood vessels may prevent total resection of the tumor at the site of that infiltration. However, such treatment is palliative, with the postoperative recurrence rate and prognosis being inferior to that achieved by complete resection.^[2] When a meningioma has invaded blood vessels, performing bypass surgery before resection can maintain blood flow in the distal parent artery, potentially allowing for complete resection of the tumor along with the involved blood vessels.

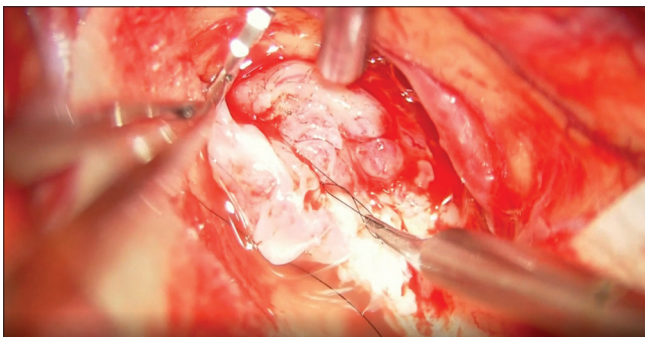
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In the present case, a recurrent falcine meningioma had invaded the right pericallosal artery (A3). We, therefore, performed an A3–A3 side-to-side anastomosis distal to the tumor, enabling its total resection along with the infiltrated A3, resulting in a favorable outcome. Although reports of implementing A3–A3 side-to-side anastomosis for tumor resection are rare, it appears to be an effective treatment option when the tumor has invaded the anterior cerebral artery (ACA), as in the present patient. We, here, report this procedure with an accompanying surgical video [Video 1].

CLINICAL PRESENTATION

While residing in the USA for work, the patient, a 48-year-old man, had been hospitalized urgently 51 months previously with a chief complaint of gradually progressive impairment of consciousness. A 60-mm diameter tumor-like lesion that was strongly compressing the left frontal lobe from the interhemispheric fissure had been found [Figure 1a], and the tumor was resected through a craniotomy. Postoperative magnetic resonance imaging (MRI) confirmed complete resection of the enhancing lesion [Figure 1b]. However, the pathology of the resected specimen was anaplastic meningioma; accordingly, intensity-modulated radiation therapy (54 Gy) was added. After returning to Japan, he visited our Gamma Knife Center for follow-up. No recurrences were detected by MRI up to 18 months after the initial surgery; however, 20 months post-surgery, a recurrent lesion was detected in the corpus callosum near the right A3 within the radiation field [Figure 1c]. Gamma Knife treatment was administered in five fractions with a marginal dose of 30 Gy and a central dose of 40 Gy. Regular imaging follow-up continued, and an MRI 12 months after the Gamma Knife treatment showed that the lesion had enlarged, leading to a second Gamma Knife treatment in 10 fractions with a marginal dose of 40 Gy and a central dose of 50 Gy. Four months after the second Gamma Knife treatment, the



Video 1: This video shows the surgical procedure for completely excising a recurrent anaplastic meningioma infiltrating into the right A3.

lesion had continued to enlarge [Figure 1d], and he was referred to our department for craniotomy and resection. The recurrent lesion showed clear enhancement with gadolinium contrast. It was in contact for approximately 15 mm with the right A3 on the anterior half of the corpus callosum body; no branching vessels were detected in this region. Before surgery, cerebral angiography confirmed that left A3 was patent, and there were no bridging veins that would obstruct tumor resection.

Intraoperative findings

The patient was positioned supine with the head in the midline and secured with a Mayfield three-point fixation device. Somatosensory-evoked potential monitoring was prepared before surgery. The whole procedure was performed through a bicoronal skin incision with a large left frontal craniotomy extending across the midline [Figure 2a]. After reopening and removing the bone flap, the dura was incised with the superior sagittal sinus as the base. Since the left parasagittal frontal region was already encephalomalacic as a result of the previous surgery, we used this route to carefully dissect the interhemispheric fissure until both A3 segments of the pericallosal arteries over the corpus callosum were fully exposed. As shown in Figure 2b, we found that the recurrent tumor was strongly adherent to the right A3. We found no branches from the adhesion sites, and intraoperative indocyanine green (ICG) video-angiography confirmed that the right A3 was patent [Figure 2c].

Initially, we attempted to separate the tumor from the right A3 but had to abandon this plan when we found that the tumor had infiltrated beyond the vascular adventitia and our dissection had inadvertently damaged this artery. The damaged section was trapped and sutured with 9-0 nylon, and hemostasis was achieved. Having determined that separating the tumor from the right A3 was not feasible, we decided to resect the tumor and infiltrated the A3 segment *en bloc*. In addition, since Doppler sonography showed that temporary clamping of the distal right A3 resulted in only a weak backflow, we decided that it was necessary to augment flow to the right ACA territory. The walls of both distal A3 arteries were free of atherosclerosis, making an A3–A3 bypass feasible.

We planned an A3–A3 side-to-side anastomosis distal to the site of tumor infiltration, where both A3 arteries run parallel. We sutured the posterior side of the anastomosis continuously and the anterior side with interrupted knots [Figure 2d]. On releasing the clamp, good blood flow was confirmed (clamp time 24 min). ICG video-angiography confirmed good patency of the anastomosis [Figure 2e]. The right A3 was then clipped and severed on both the proximal and distal sides of the tumor infiltration. The tumor was then

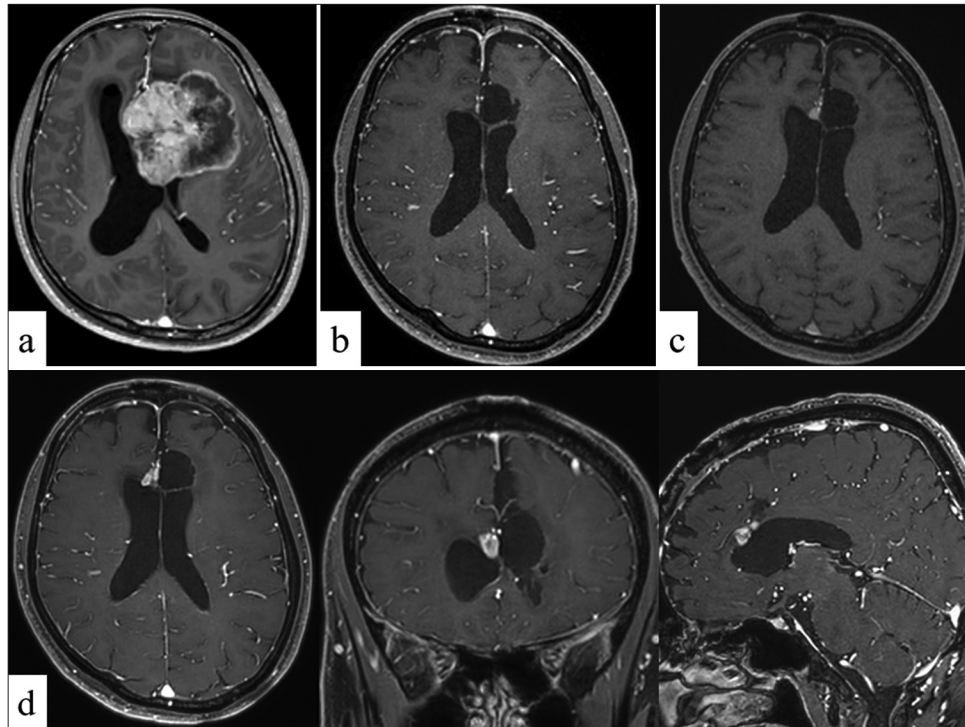


Figure 1: (a) MRI performed before the first operation showing an approximately 60 mm tumor in the interhemispheric fissure of the anterior cerebral region, compressing the left frontal lobe. (b) MRI performed after the first operation showed that the enhanced region had been totally removed. (c) MRI performed 20 months after the first operation showed enlargement of the tumor recurrence in the corpus callosum. (d) MRI performed before the second operation showed a growing tumor infiltrating the corpus callosum and anterior cerebral artery.

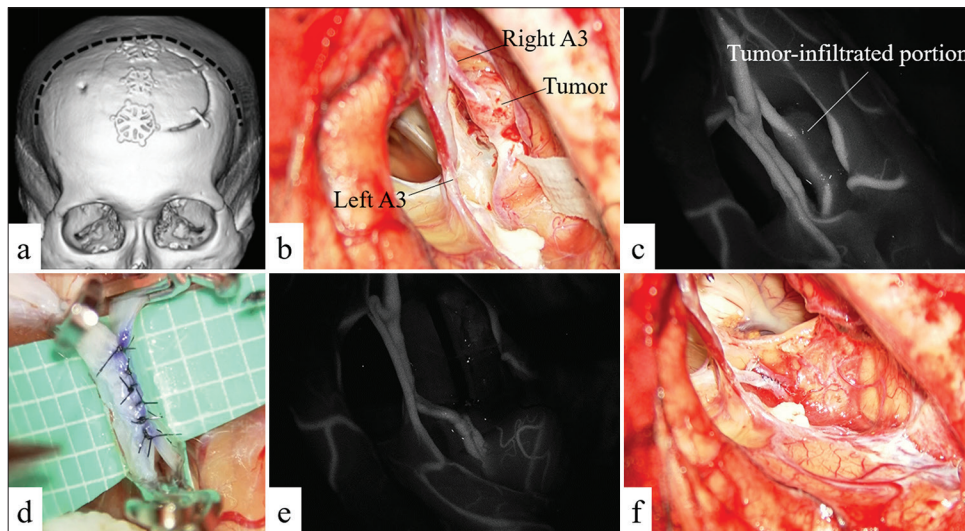


Figure 2: (a) Preoperative CT scan showing frontal craniotomy (previous bicoronal skin incision is shown as a dotted line). (b) Intraoperative photograph showing the right A3(Rt A3) is infiltrated by the tumor. (c) Indocyanine green (ICG) videography showing the tumor infiltrating a portion of A3. (d) Intraoperative photograph showing that an A3–A3 side-to-side anastomosis has been performed. (e) ICG videography showed the patency of the bypass. (f) Intraoperative photograph showing the tumor has been completely resected.

dissected from the surrounding brain and removed *en bloc* [Figure 2f]. Intraoperative somatosensory-evoked potentials

monitoring showed no decline. The surgical procedure is presented in [Video 1].

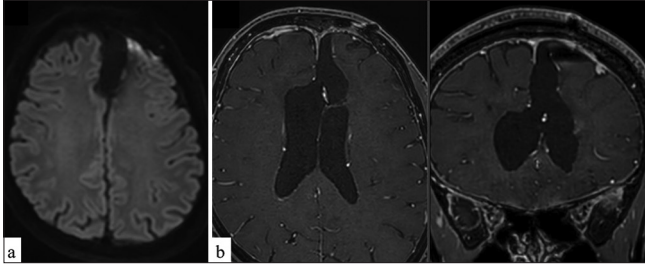


Figure 3: (a) Postoperative diffusion-weighted image (DWI) showing no acute cerebral infarction. (b) Postoperative contrast-enhanced T1 image showing no residual tumor.

Post-operative course

Postoperative MRI showed that there were no complications, such as cerebral infarction [Figure 3a] and that the A3–A3 side-to-side anastomosis was patent. Contrast imaging confirmed complete resection of the tumor. Pathological examination of the resected specimen showed recurrent meningioma involving the cerebrum. Its Ki-67 labeling index was more than 15%. There was no evidence of neurological deficits, and tests of higher cognitive function yielded the following results: 30 points on the mini-mental state examination, 18 points on the frontal assessment battery, 27 s on the trail making test A, and 40 s on the trail making test B, with no significant errors noted. The patient was discharged home on postoperative day 15 with a modified Rankin scale score of 0 and successfully returned to his social activities. The lesion in the corpus callosum had not recurred 7 months postoperatively [Figures 3a and b].

DISCUSSION

Most meningiomas are benign and slow-growing; however, approximately 20–25% are classified as atypical meningiomas (WHO Grade II) and 1–3% as anaplastic meningiomas (WHO Grade III). The latter often grow rapidly, leading to poor outcomes.^[4] High-grade meningiomas (WHO Grade II or III) have high recurrence rates: the 5-year recurrence rates for anaplastic meningiomas are reportedly 72–94% after complete removal and 83–100% after partial removal.^[9] Adjuvant radiotherapy is therefore recommended for all high-grade meningiomas. It has been estimated that adjuvant radiotherapy following total resection of anaplastic meningiomas improves the 5-year progression-free survival rate from 15% to 80%.^[2] The same authors also reported that administering postoperative radiotherapy does not alter the recurrence rate after partial resection (almost all partially resected tumors recur).^[2] In other words, residual anaplastic meningioma correlates strongly with recurrence rate and a poor prognosis.

In the present case, despite undergoing surgery, external radiotherapy, and two rounds of Gamma Knife therapy for

repeated recurrences of anaplastic meningioma, local control was not achieved, prompting a decision to pursue surgical total resection despite the identification of tumor invasion of the right A3. Partial resection without excising the infiltrated portion followed by postoperative radiotherapy was considered; however, it was evident that partial resection would be palliative in this case, given the history of multiple recurrences.

High-grade meningiomas sometimes invade the brain and surrounding tissues. When infiltrating an artery, the tumor first attacks the adventitia, its growth gradually resulting in the disappearance of the adventitia. The tumor then invades the media and elastic lamina and finally changes the morphology of the blood vessel such that there is no clear cleavage plane between the tumor and infiltrated area, making complete separation at the site of invasion difficult.^[6]

In our patient, the tumor was very firmly attached to the arterial wall, and our attempts to remove it were unsuccessful because we were unable to find a clear plane for separation. Thus, it appeared that the only procedure that would achieve complete resection of the tumor at the site of vessel invasion was to remove the entire infiltrated blood vessel after creating an A3–A3 side-to-side anastomosis.

Revascularization to the ACA system can be roughly divided into two types, namely, intracranial–intracranial bypass such as A3–A3 side-to-side anastomosis (also known as *in situ* bypass) and revascularization from extracranial to intracranial arteries (extracranial–intracranial bypass). In the present patient, we also considered one of the latter: superficial temporal artery (STA)–A3 bypass using a free STA graft or an STA–radial artery graft–A3 bonnet bypass, the usefulness of which we previously reported.^[7,8] However, both of these procedures are complicated, involving numerous steps. Furthermore, we anticipated that postoperative skin complications resulting from STA harvest would likely occur, especially given that our patient had previously undergone surgery. Taken together, we decided that A3–A3 side-to-side anastomosis was the best treatment option.

A review of published reports on implementing A3–A3 side-to-side anastomosis revealed very few reports globally,^[3] this procedure primarily having been performed during trapping of ACA aneurysms.^[1,5] Thus, combining A3–A3 side-to-side anastomosis with tumor excision and its usefulness are very rare worldwide and, therefore, worthy of being reported.

CONCLUSION

When a malignant meningioma has invaded a blood vessel, the high recurrence rate of these tumors mandates considering removing the tumor along with the invaded blood vessel. When there are no effective collateral channels to the area of the excised blood vessel, revascularization must be performed.

A3–A3 side-to-side anastomosis should be considered a good option for meningiomas infiltrating the ACA.

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Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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