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A case of recurrent hemangioblastoma receiving blood supply from the mastoid and transosseous branches of the occipital artery

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Case Report

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ABSTRACT

Background: Hemangioblastomas (HBs) typically receive their blood supply from the branches of the intracranial arteries; however, meningeal branches or arteries originating from the external carotid artery are rare because of the intramedullary subpial location of the HBs. Although HB embolization is effective, it carries complication risks.

Case Description: A 71-year-old man with a history of incomplete HB resection consulted our hospital with headache, vertigo, and nausea. Neuroimaging revealed a recurrent HB. Angiography demonstrated that the HB was fed by mastoid branch (MB) and transosseous branches (TOBs) from the occipital artery (OA), in addition to the superior cerebellar artery (SCA) and the posterior inferior cerebellar artery (PICA). The patient underwent preoperative embolization with n-butyl-2-cyanoacrylate through the SCA and PICA branches. After embolization for TOBs, which led to feeder occlusion of the OA, the meningeal branch of the MB from the OA was revealed. We hesitated to perform embolization targeting this MB, considering the risk of potential anastomosis to the vertebral artery. Total resection through an enlarged craniectomy was conducted with minimal bleeding. Postoperative magnetic resonance imaging revealed no remnant tumor; however, infarction was observed in the area perfused by the SCA due to embolization of the SCA branches. The symptoms improved after surgery, and the patient was discharged following rehabilitation, with slight ataxia as a sequela.

Conclusion: This is a rare case of recurrent HB receiving a blood supply from the MB and TOBs from the OA. Thus, embolization for TOBs is safe and effective for recurrent HB resection.

Keywords: Embolization, Hemangioblastoma, Infarction, Mastoid branch, Transosseous branch

INTRODUCTION

Hemangioblastomas (HBs) are benign vascular tumors of the central nervous system, representing 1–2.5% of all brain tumors, and commonly arise in the posterior cranial fossa. Cerebral angiography is a standard diagnostic method for HBs that detect feeding arteries and venous drainage. HBs typically receive blood supply from the branches of the intracranial arteries, such as the superior cerebellar artery (SCA), anterior inferior cerebellar artery (AICA), and posterior inferior cerebellar artery (PICA). Blood supply to HBs from the meningeal branches or

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arteries from the external carotid artery (ECA) is rare owing to the intramedullary subpial location of the HBs.^[11]

The aim of HB treatment is complete resection, which could be curative. However, complete resection of HBs is challenging because internal decompression or piecemeal resection could lead to devastating intraoperative bleeding.^[3,6] Preoperative endovascular embolization before HB resection is often indicated to minimize surgical blood loss and increase the success rate of complete resection.^[3,7,9-11] While embolization for HBs has been reported to be effective and safe,^[3,7,9-11] some studies do not recommend this owing to the high risk of complications without benefit.^[1]

Herein, we present the rare case of a patient with recurrent HB who received blood supply from the mastoid branch (MB) and transosseous branches (TOBs) of the occipital artery (OA) and underwent surgical resection after embolization.

CASE DESCRIPTION

A 71-year-old man was referred to our department with headache, vertigo, and nausea that had persisted for several days. The patient had undergone craniotomy for cerebellar HB at another hospital 9 years ago. The tumor had not been completely resected, and the patient underwent follow-up at the hospital. However, the patient discontinued follow-up after the 3-year visit. Figures 1a and b illustrate the magnetic resonance imaging (MRI) performed before the first surgery and at the final follow-up at the previous hospital. Computed tomography showed a tumor mass, and gadolinium-enhanced MRI showed a cystic-solid tumor in the right cerebellar hemisphere [Figure 1c and d]. The patient was diagnosed with recurrent HB and underwent tumor resection after endovascular embolization.

Tumor embolization was conducted under general anesthesia. Right vertebral angiography revealed a tumor receiving blood supply from the branches of the right SCA and PICA [Figure 2a]. The microcatheters were individually inserted into the branches of the SCA and PICA near the tumor, and n-butyl-2-cyanoacrylate (NBCA) was injected, leading to a considerable reduction in the tumor blush [Figure 2b]. Right external carotid angiography showed TOBs from the OA as the feeding arteries through the skull gap created by the previous craniotomy [Figures 2c and d]. The microcatheter was guided into the OA near the TOB origin, and NBCA was injected, leading to feeder occlusion [Figure 2e]. Repeat external carotid angiography following occlusion demonstrated the MB from the OA as a tumor feeder [Figure 2f]. An angiogram obtained from a previous hospital before the first resection revealed the MB as a tumor feeder, similar to that in our angiogram [Figure 2g]. Embolization targeting this MB was not performed because of the risk of migration of the embolic material to the vertebral artery through potential anastomosis, and the procedure was terminated at this point. Surgical resection was conducted on the same day as the embolization through suboccipital craniectomy enlargement. The bleeding from the skull and dura was minimal due to embolization for TOBs. The tumor was strongly adhered to the dura, which was then detached carefully. The tumor was dissected from inside the cyst and then dissected circumferentially, resulting in the en bloc complete resection. The embolic material in the feeding arteries was visible during resection, making it easy to orient the surgical field. Once the tumor was sufficiently embolized, little blood loss was noted during the surgery.

Postsurgical MRI revealed no remnant tumor [Figure 3a]; however, an infarction was detected in the area perfused by the SCA, which was attributed to the embolization of the SCA branches [Figure 3b]. The infarction did not worsen the patient's symptoms, and the condition improved. The patient was discharged after rehabilitation 2 months after surgery, with slight ataxia as the only sequela (Karnofsky Performance Status: 80%).

DISCUSSION

HBs are benign intramedullary vascular tumors that are often situated near the pia mater. Therefore, they generally



Figure 1: (a) Gadolinium-enhanced magnetic resonance imaging (MRI) performed before the first surgery revealed a solid tumor in the right cerebellar hemisphere. (b) MRI taken 3 years after the first surgery showed a small enhanced nodule at the cerebellar surface. (c) Computed tomography at our hospital revealed a tumor recurrence. (d) MRI showing a cystic-solid tumor.



Figure 2: (a) Lateral view of the right vertebral angiography showing the tumor blush from the branches of the superior cerebellar artery (SCA) (black arrow) and posterior inferior cerebellar artery (PICA) (white arrows). (b) The tumor blush was reduced after embolization through the SCA and PICA. (c) Lateral view of the right external carotid angiogram showing the transosseous branches (TOBs) (black arrowheads) from the occipital artery (OA) as the feeding arteries. (d) Three-dimensional external carotid angiography revealing TOBs through the skull gap created by the previous craniotomy. (e) N-butyl-2-cyanoacrylate was injected into the OA near the origin of the TOBs, leading to feeder occlusion. (f) External carotid angiography after feeder occlusion revealed the mastoid branch from the OA as a tumor feeder (white arrowheads). (g) An angiogram obtained at a previous hospital revealed the mastoid branch, which was similar to the angiogram performed in our hospital (white arrowheads).



Figure 3: (a) Postsurgical magnetic resonance image revealing no evidence of remnant tumor. (b) Diffusion-weighted image showing infarction in the area perfused by the superior cerebellar artery.

receive blood supply from the intracranial arteries, such as the branches from the PICA, AICA, and SCA; however, blood supply from the meningeal branches or arteries originating from ECA is relatively rare.^[11] Previous studies have reported that HBs with dural attachment or invasion due to the effects of previous surgery, recurrence, or bleeding could receive blood supply from the dural arteries.^[5,11] Yamada *et al.* reported two cerebellar HBs with minor blood supply from the dural branch of the external and internal carotid arteries.^[11] Recently, Varga-Urbina *et al.* also reported that primary and recurrent HBs can

receive blood supply from the middle meningeal artery and OA, which successfully underwent embolization targeting the vessels.^[10] Contrastingly, the neuromeningeal branches of the ascending pharyngeal arteries from the ECA could feed the HBs situated at the cerebellopontine angle because this branch supplies the meninges near the internal auditory meatus.^[2,5]

The present case of recurrent cerebellar HB was rare because the BM and TOBs fed it from the OA. The BM from the OA was noted as a feeding artery before the first surgery performed 9 years before the present one. The tumor could not be completely resected during the first surgery, and the MB may have remained as one of the feeding arteries, facilitating tumor recurrence. TOBs have been speculated to develop and flow into the tumor through the skull gap during tumor regrowth. The TOBs in the present case were not "true" transosseous because the branches did not penetrate the skull; however, they passed through the skull gap created by craniotomy. Thus, these TOBs may be considered part of the meningeal branches in the present case.

With the development of endovascular treatment, preoperative embolization for HBs is considered an effective treatment to minimize intraoperative blood loss and increase complete resection rate and is now widely performed.^[3,7,9-11] However, a systematic review concluded that embolization

for HBs only increased complications and did not provide any beneficial contributions to surgical resection.^[1] In addition, previous studies regarding embolization targeting any brain tumor type have revealed that embolization for tumors other than meningiomas, targeting vessels other than the ECA, and the use of liquid material were significant factors for developing complications.^[4,8] In our patient, infarction occurred in the SCA area due to embolization without worsening the symptoms. The complications also occurred due to liquid material injection into the SCA for tumors other than the meningiomas and were consistent with the findings of the aforementioned studies.

The favorable outcome in our patient was attributed to complete resection. In general, reoperations are more complicated than initial operations due to adhesions and anatomical transformations that occur due to initial interventions. In this case requiring reoperation, complete resection was achieved with minimal bleeding despite strong adhesion. This was because bleeding from the tumor, as well as bleeding during craniotomy, was minimal. Considering that the TOBs developed with the tumor recurrence through the skull and dura, bleeding would have increased during craniotomy if TOBs had not been embolized. Thus, although TOBs are not the main feeders of HBs, embolization for TOBs can be safe and effective for craniotomy, dural incision, and tumor dissection, especially in recurrent HBs.

CONCLUSION

This is a rare case of recurrent HB with blood supply from the MB and TOBs from the OA. While preoperative embolization of branches from the intracranial arteries carries the risk of ischemic complications, embolization for TOBs is safe and effective for craniotomy and resection for recurrent HB.

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