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Preoperative Endovascular Embolization of Intracranial Hemangioma: A Case Report

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ABSTRACT

Introduction: Hypervascular tumors of the head, neck, and central nervous system are associated with a high risk of bleeding during surgery. Hemangioma is a benign neoplasm that tends to grow rapidly, bleed, and have a high recurrence rate, especially after partial resection. Considering that hemangioma is a type of tumor with high vascularity, preoperative tumor embolization was performed to reduce intraoperative risk and maximize resection success. **Case:** A 33-year-old female came with the chief complaint of chronic progressive headaches and vision loss in the last 5 years. She got a lump on her forehead and had a visual acuity of 1/∞ in both eyes. She had already been diagnosed with hemangioma at a secondary regional hospital, where she bled profusely on the operating table, causing the resection to be discontinued. She was referred to our hospital for preoperative embolization. She underwent an endovascular embolization procedure with PVA particles to seal the feeder artery to the tumor. After embolization, angiography showed a complete devascularized tumor lesion. A total resection was done three days later with minimal bleeding compared to the first surgery. After a three-month follow-up, there was no new neurological deficit, and her headache was resolved completely, although her vision still did not improve. **Conclusion:** Endovascular embolization has developed into an important and effective adjuvant in tumor management before a surgical resection. Advances in catheter-based techniques, in conjunction with the development of the neurointervention field, are expected to raise the number of perioperative embolization procedures performed.

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INTRODUCTION

Surgical management of intracranial hypervascular tumors is a challenging procedure for neurosurgeons due to the risk of massive intraoperative bleeding, which discourages tumor resection with regard to developing postoperative neurological deficits.[1] Endovascular embolization is often used in conjunction with surgical techniques in an attempt to minimize mortality, morbidity and increase the chances of successful tumor resection.[2]

Hekster *et al*. reported the first successful tumor embolization case in 1974, when they used transfemoral catheterization to embolize a meningioma.[3] Since then, many reports and case series have been published describing the advantages of the procedure. However, there is still no standard consensus on how the tumor embolization procedure should be done.[4]

In 1867, Virchow described hemangiomas as a benign vascular tumor that manifests most commonly on the mucosa and skin of the head, face, chest, or back, with onset mainly occurring during childhood or adolescence and rarely growing in adulthood.[5,6]

Histologically, hemangiomas are divided into two main subtypes: capillary and cavernous.[5] Capillary hemangiomas consist of small capillary lobules separated by varying fibrous tissue, in which lobules are formed by clumps of endothelial cells lining the vascular wall and making capillary channels obscure.[5] Cavernous hemangiomas are cystically dilated, thin-walled, larger vessels that are often associated with thrombosis, perivascular hemosiderin deposition, and calcification.[5] Cavernous hemangiomas are more common than capillary types in the central nervous system, notably in the brain lobes. They are usually found in older children and adults.[7]

Hemangioma of the central nervous system is a very rare pathology.[8,9] In March 2020, a PubMed search found only 41 publications containing a total of 52 cases of intracranial hemangioma.[8] The average age was 26 years, with a slightly higher prevalence in women (28 cases, 53%) and adults (28 cases, 53%), with headache being the most prevalent symptom (21 cases, 40%). The actual prevalence of intracranial hemangioma may be higher due to the small number of studies and reported cases, as some cases may be asymptomatic, spontaneously regress, or even go unreported.[6]

From those numbers, only two cases of intracranial capillary hemangioma with preoperative endovascular embolization treatment have been reported: a temporal lobe intracranial hemangioma in a 7-week-old infant and a 28-year-old woman, which were followed by total tumor resection.[8,10] After 6 and 12 months, both cases reported an excellent outcome and no recurrence.[8]

CASE

A 33-year-old woman came in with a chronic progressive headache for the last 8 years and vision loss for the last 5 years. Headache is felt mostly on the right side of the head, with a dull quality, intermittent, and worsening over the last 2 years. Initially, medication relieved the headache, but in the past two years, it had stopped working and a growing lump had appeared on the head (Figure 1). She also complained of progressive vision loss for the last 2 years in both eyes until she could not see anymore. She had no other symptoms or neurologic deficits. A comprehensive neurological physical examination revealed an eye acuity 1/∞ in both eyes, in which the patient could only identify a light source.

A brain CT scan from her previous hospital showed an intracranial mass with hyperostosis and extensive perifocal edema. The patient had a craniectomy, surgical resection, and tissue biopsy, with massive intraoperative bleeding around 1500–2000 cc. With that much blood, surgical resection was discontinued, and only a little part of the tissue was taken for a biopsy. The pathology result showed a small-to-medium-sized blood vessel between the bone trabeculae lined with endothelial cells with non-atypical nuclei and containing erythrocyte cells in the lumen, which resembled a hemangioma. The patient was then transferred to our hospital for further treatment. A brain MRI examination (Figure 2A) showed a heterogeneous soft solid mass in the right frontal lobe around 5x8x5.3 cm with poorly defined and lobulated irregular edges. The post-contrast injection showed a heterogeneous mass with areas of diffusion restriction (Figure 2B). There was an intra mass cystic component with suspected necrosis and prominent vascularization, as well as a bony defect with anteriorly protruding mass growth involving the surrounding meninges. Prominent perifocal edema was seen around the mass, which caused a midline shift to the left by ±1.3 cm, subfalcine herniation to the left side, ventriculomegaly of III and IV, and bilateral ventricular. The patient was then placed on a ventriculoperitoneal shunt as initial treatment and was planned for an embolization procedure prior to tumor resection.

The patient was in stable condition on the day of the procedure, and all of the laboratory test results, especially those for kidney function, the EKG (electrocardiogram), and the chest X-ray, were within normal limits. She underwent an endovascular embolization procedure with a microcatheter parked proximal to the frontal branch of the middle meningeal artery. Extracranial and intracranial collaterals were identified before the embolic agent was injected, and no dangerous collaterals were detected (Figure 3). The feeder arteries to the tumor were sealed using PVA particles that ranged in size from 150 to 250 microns. After embolization, angiography showed a complete devascularized tumor lesion (Figure 4).

Total tumor resection was performed three days after embolization (Figure 5). In comparison to the first operation, there were no significant difficulties with bleeding as much as ±500 cc during this operation. The results of another pathology examination on a part of the tissue were consistent with the presence of a hemangioma. No new post-resection neurologic deficits were found, and she was discharged five days after surgery. Unfortunately, she was immediately returned to her hometown after the operation, and one month later she had a control brain CT scan in her hometown, which revealed extensive perifocal edema in the right cerebral hemisphere and a right frontoparietal bone defect (Figure 6). There were no new complaints or significant complications, but her vision has not recovered.

DISCUSSION

Intracranial hemangiomas can grow in the cerebral lobes, sagittal sinus, cerebellum, sellar area, cavernous sinus, fourth ventricle, or anterior choroid artery.[5] Clinical signs and symptoms vary, ranging from asymptomatic to decreased consciousness, with a common mechanism being the mass effect on adjacent anatomic structures, which results in clinical symptoms based on the location of the lesion and includes headache, seizures, cranial nerve paresis, hemiparesis, and behavioral abnormalities in adults, whereas in children, signs and symptoms are often associated with conditions of increased intracranial pressure.[8,11] Santoro et al. found that headache was the most common symptom (21 cases, 40%), followed by at least one cranial nerve palsy (16 cases, 30%), visual disturbances (10 cases, 19%), nausea or vomiting (9 cases, 17%), seizures (7 cases, 13%), hydrocephalus (7 cases, 13%), limb motor deficits (7 cases, 13%), and decreased consciousness (3 cases, 6%).[8] Our patient demonstrated the most prevalent symptoms found in the literature, such as headaches and visual disturbances. A growing hemangioma in the frontal region that invaded the meninges, compressed the optic nerve, and raised the intracranial pressure was the cause of the symptoms. She also had a lump because of vessel invasion throughout the bone.

Although hemangiomas are benign neoplasms, they tend to grow rapidly, bleed, and are associated with a high recurrence rate, especially after partial resection.[12,13] The first surgery for resection was successfully omitted because of massive bleeding at that time (1500–2000 cc). Preoperative embolization of highly vascularized head and neck tumors was first described in 1974 when Hekster *et al*. reported that meningioma embolization could reduce the amount of bleeding at the time of surgery, increase the likelihood of successful tumor resection, and shorten the operative time and postoperative recovery period.[2] The benefit in regards to reducing intraoperative bleeding was more pronounced, especially for larger, high-grade, and more vascularized tumors such as paraganglioma, angiofibroma, hemangiopericytoma, meningioma, hemangioblastoma, schwannoma, juvenile nasopharyngeal angiofibroma, and hypervascular metastases.[2,4,14] There are several goals of preoperative embolization: (1) to control hardly accessible feeder arteries;[15] (2) to reduce intraoperative bleeding;[15] (3) to shorten procedural time; (4) to increase the chances of successful total resection;[4] (5) to reduce the damage of surrounding normal tissue;[16] (6) palliative therapy to reduce intractable pain;[17] (7) to reduce tumor recurrence; (8) to improve visualization of the surgical field.[18] Considering that hemangioma is a type of tumor with high vascularity, preoperative tumor embolization was performed to reduce intraoperative risk and maximize the success of resection.

Super-selective catheterization of the feeder branch and transarterial embolization with particulate agents are two common techniques that interventionists use for preoperative tumor devascularization. Super-selective angiography of extracranial carotid artery branches can confirm the feeder artery and identify other selective contributions from various branches, especially when multiple feeders are present.[2] Collateral pathways can open due to an increase in intracranial pressure, and the injection of an embolic agent can result in a hazardous neurological deficit.[2] In our case, a feeder from the frontal branch of the middle meningeal artery was identified with no dangerous collaterals.

Tumors with a high vascular supply may be more susceptible to embolization. Embolization with particulate matter, liquid embolic agents, or coils can improve tumor resection with minimal intraoperative bleeding in tumors with high vascularity.[2] Ideal tumor embolization is achieved by occlusion of the distal vasculature within the tumor while maintaining surrounding normal tissue. The goal of embolization is to devascularize tumor arteries and induce tumor necrosis; thus, embolization should be performed as distally as possible.[4] In order to target distal small vessels, it is common practice to begin embolization with smaller particles and then gradually increase the size of the embolic particles.[19]

Polyvinyl alcohol (PVA) is an embolic agent available in various sizes ranging from 45 to over 1000 microns. Smaller particles (45–150 microns) can occlude tumor capillaries and devascularize them by causing inflammatory reactions and angio-necrosis, thereby assisting the devascularization but being prone to unwanted embolization.[18] Larger PVA (150–250 microns) can be used for distal embolization and occlusion at the capillary level since the tumor bed capillaries are typically around 200 microns.[18] If a dangerous anastomotic route is suspected, a larger particle size, such as 250–350 microns or 350–500 microns, can be used to prevent unwanted embolization.[4] In our case, we used PVA (150–250 microns) to achieve tumor devascularization.

Surgery onset after embolization also plays an important role in successful tumor resection. Surgical resection should be performed 1–8 days after embolization to maximize its benefit. Early tumor resection (less than 24 hours) after embolization can reduce the benefits of embolization because there is not enough time for devascularization and tumor necrosis; hence, a greater risk of intraoperative blood loss persists.[4] Resection should ideally be performed 24–72 hours after embolization to allow time for thrombosis of the occluded vessel and prevent recanalization of the vessel.[18] Due to the possibility of vessel recanalization and recruitment of collateral routes, surgery lasting more than 8 days offers no significant benefit.[2] On the other hand, a longer surgical procedure can raise the intracranial pressure and cause compression of the surrounding structures due to edema from tumor necrosis.[2] For large intracranial tumors, steroids may be considered immediately after embolization to minimize edema and the compression effect.[2] To maximize the benefits, we waited 72 hours after embolization to perform the surgical resection without the use of steroids. The surgical result was also satisfying, with successful total tumor resection and relatively controlled intraoperative bleeding (±500 cc) compared to the first surgery.

Successful embolization can be evaluated from a technical and clinical perspective. Technically, a successful embolization is seen by the occlusion of tumor vessels, which results in an 80% reduction of tumor blush is met.[19] Clinically, it can be evaluated by the reduction in expected bleeding during surgery, tumor removal, duration of surgery, and reduction in intra-operative complications.[2] In general, complications can be classified into major and minor categories.[20] Major complications are defined as those requiring additional therapy, a higher level of care, longer hospitalization, death, a permanent neurologic deficit, brain edema, arterial embolism, stroke, intracranial hemorrhage, and contrast-related nephropathy.[20] Minor complications are those that do not require special treatment outside of observation and without further clinical consequences, such as hematoma at the puncture site, pain at the puncture site, and fever.[20] In general, our procedure was considered successful since it resulted in tumor devascularization, no significant intraoperative bleeding, total mass resection, and no major post-embolization complications

CONCLUSION

Preoperative endovascular embolization in intracranial hypervascular tumors is an effective and safe adjuvant prior to surgical resection. Although no specific guideline has been available up to this date, preoperative embolization is one method that can improve surgical success and patient safety. It is hoped that advanced embolization techniques and agents such as intra-arterial chemotherapy may expand endovascular therapeutic options for a wide variety of cancers.

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Author Contribution
Author: examined the patient, contributed to data collection and manuscript description, and wrote the manuscript.
Co-author: examined the patient together with the author, contributed to data collection, and reviewed the manuscript.

Conflict of Interest
The authors have no conflicts of interest to disclose for this report.

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TABLES AND FIGURES

Figure 1. Bony lump on midfrontal region.

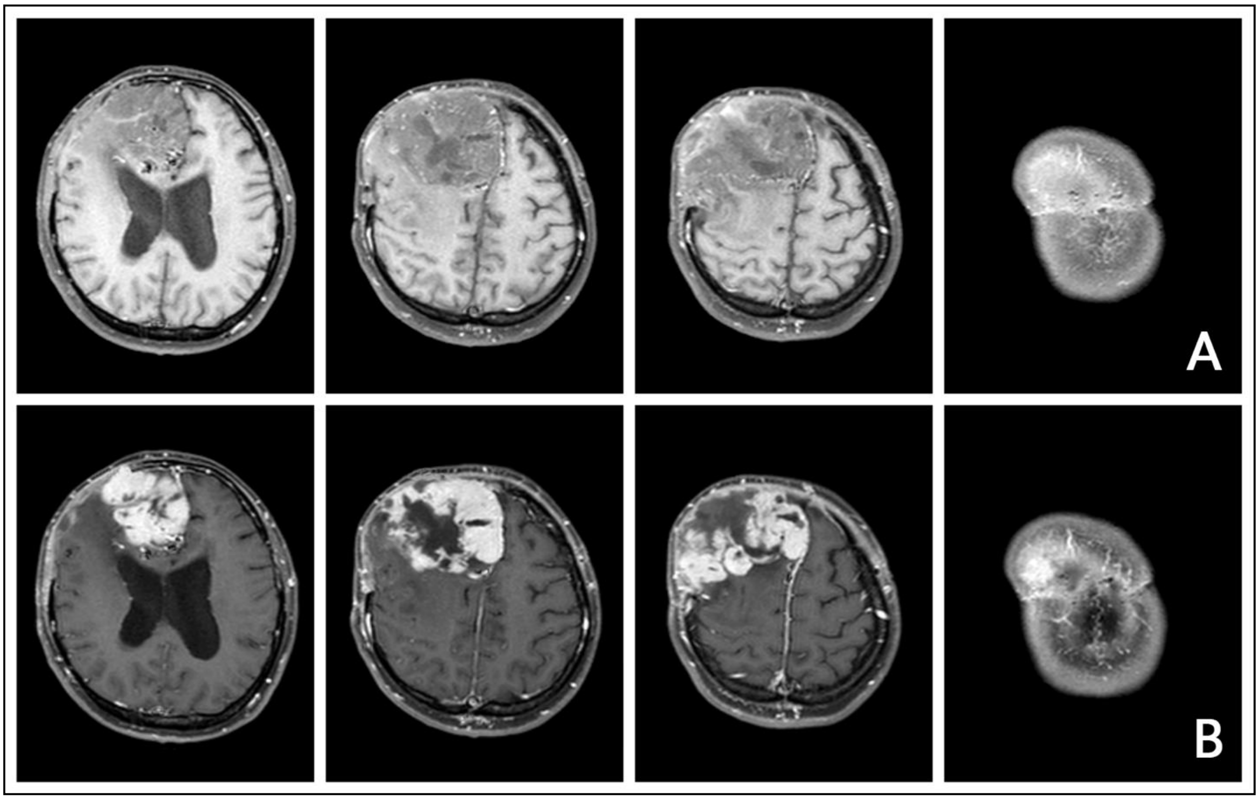

Figure 2. MRI T_1 non-contrast (A) and MRI T_1 post contrast (B)

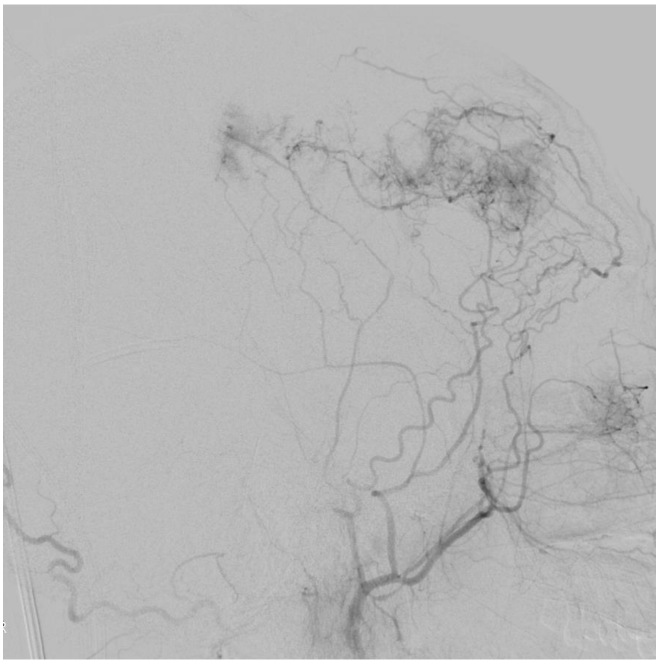

Figure 3. RECA pre-embolization evaluation.

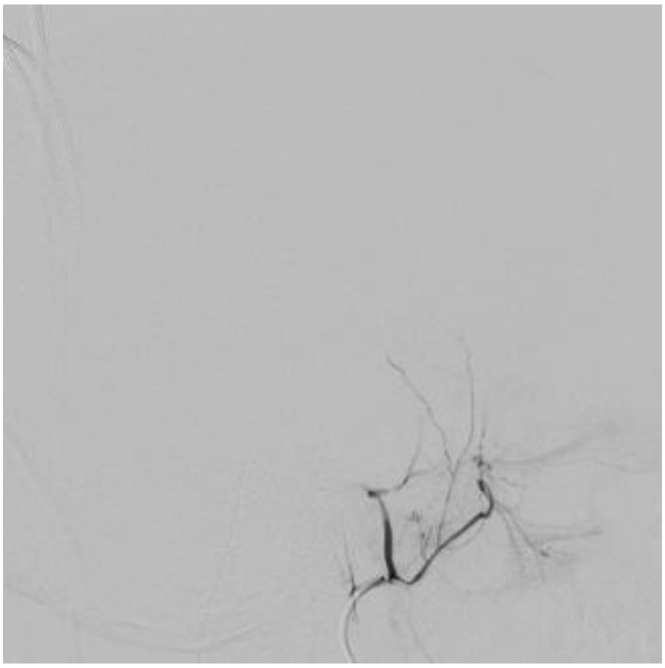

Figure 4. RECA post embolization evaluation.

Figure 5. Total resection hemangioma mass.

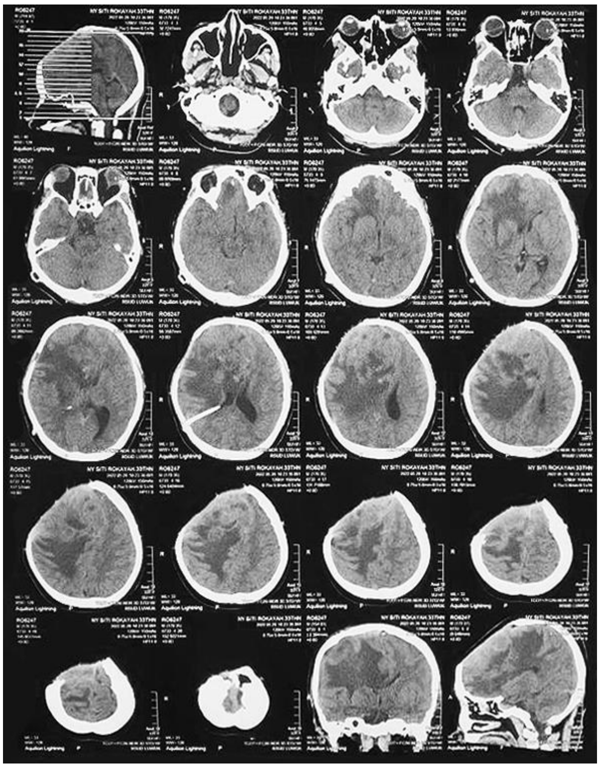

Figure 6. Post resection control CT scan.