Successful removal of a huge hypervascular tentorial cavernous angioma after preoperative endovascular embolization

Case report

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The authors report a rare case of a huge hypervascular tentorial cavernous angioma treated with preoperative endovascular embolization, followed by successful gross-total removal. A 15-year-old girl presented with scintillation, diplopia, and papilledema. Computed tomography and MRI studies revealed a huge irregularly shaped tumor located in the right occipital and suboccipital regions. The tumor, which had both intra- and extracranial components, showed marked enhancement and invasion of the overlying occipital bone. Angiography revealed marked tumor stain, with blood supply mainly from a large branch of the left posterior meningeal artery. Therefore, this lesion was diagnosed as a tentorium-based extracranial tumor. For differential diagnosis, meningioma, hemangiopericytoma, and malignant skull tumor were considered. Tumor feeders were endovascularly embolized with particles of polyvinyl alcohol. On the following day, the tumor was safely gross totally removed with minimum blood loss. Histopathological examination confirmed the diagnosis of cavernous angioma. To date, there have been no reports of tentorium-based cavernous angiomas endovascularly embolized preoperatively. A tentorial cavernous angioma is most likely to show massive intraoperative bleeding. Therefore, preoperative embolization appears to be quite useful for safe maximum resection. Hence, the authors assert that the differential diagnosis of tentorium-based tumors should include tentorial cavernous angioma, for which preoperative endovascular embolization should be considered.

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Key Words: • cavernous angioma • tentorial tumor • embolization • hypervascular • profuse bleeding • vascular disorders

Abbreviations used in this paper: CA = cavernous angioma; PVA = polyvinyl alcohol.
ed in the right occipital and suboccipital regions, that is, in both the supra- and infratentorial spaces, in association with destruction of the inner table of the overlying occipital bone (Fig. 1). Magnetic resonance imaging indicated the precise extension of the tumor, that is, both intradural and extradural components as well as parts located in both the supra- and infratentorial regions. The tumor showed hypointensity on T1-weighted images, hyperintensity on T2-weighted images with signal flow void because of the large intratumoral vessels, and isointensity on diffusion-weighted images. The tumor demonstrated marked and irregular enhancement with Gd but no obvious dural tail sign (Fig. 2). Angiography demonstrated marked tumor stains, with blood supply from a well-developed branch of the left posterior meningeal artery, the right occipital artery, and parietal branches of the right middle meningeal artery. There was no early-phase arteriovenous shunting indicative of a dural arteriovenous fistula. The flecked tumor stains persisted in the late venous phase. Moreover, the right transverse sinus was occluded by the tumor (Fig. 3 upper).

**Operation.** Preoperative endovascular embolization of the left posterior meningeal artery, right occipital artery, and right middle meningeal artery was safely performed with polyvinyl alcohol (PVA) particles (Fig. 3 lower). On the following day, with the patient in the left park-bench position, we performed a right occipital and suboccipital craniotomy with a large T-shaped skin incision. The tumor rose from the cerebellar tentorium near the right transverse sinus, extended into both the right occipital lobe and the infratentorial space, and invaded the occipital bone on the right side of the inion, eroding the outer layer. The tumor was dark reddish and multilobulated (Fig. 4) and had smooth, well-demarcated margins.

![Fig. 1. Noncontrast CT scans (upper) showing a hyperdense tentorial mass with coarse calcification extending into the right occipital lobe. Contrast CT scans (lower) revealing marked enhancement.](image-url)
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We could perform internal decompression with minimum blood loss (725 ml) because of the preoperative embolization of feeders, requiring a minimal blood transfusion (560 ml of red blood cell concentrates). Although the tumor attachment on the venous confluence was coagulated to avoid injury to the venous sinus, the tumor bulk was gross totally removed (Fig. 5).

Pathological Examination. Histopathological examinations revealed that the tumor was composed of dilated and fibrous-walled vascular channels lined with a single-cell layer of endothelium without intervening brain tissue (Fig. 6 left). Immunohistochemical stain with epithelial membrane antigen was negative for meningothelial cells in the stroma. Elastica-Goldner stain was also negative for internal elastic lamina in the vascular wall (Fig. 6 right). These findings confirmed the diagnosis of CA.

Fig. 2. Axial T1-weighted MR image (A) demonstrating a hypointense mass in the right occipital lobe. Tumor extended into the skull. Axial T2-weighted MR image (B) showing hyperintensity with signal flow void in the tumor. Axial enhanced T1-weighted MR image (C) revealing marked and irregular enhancement. Enhanced sagittal (D) and coronal (E) T1-weighted MR images demonstrating a huge tentorial tumor with both supratentorial and infratentorial extensions.

Fig. 3. Left vertebral artery angiogram (upper) showing the large feeder of the posterior meningeal artery and the marked stain retained until the late venous phase. After embolization (lower), the thick feeder was well embolized, and the tumor stain disappeared.

Fig. 4. Intraoperative photograph showing a dark reddish, well-demarcated, multilobulated tumor extending into the right occipital lobe.
Postoperative Course. The patient had a good postoperative course and recovered from her symptoms except for the left homonymous scotoma, and delayed healing of the wound was observed as well. She resumed junior high school. The tumor did not recur for more than 1 year.

Discussion

As mentioned above, only 9 tentorial CAs have been reported in the literature,6,14,16,17,19,20,26 6 cases in males and 3 in females (age range 0–60 years; Table 1). Common symptoms were headache due to increased intracranial pressure and visual field disturbances. The patient in our case also presented with these symptoms, which had a relatively long-standing history.

Neuroimaging of Tentorial CA

The MRI appearance of intraaxial CA is distinct enough to allow confident diagnosis on the basis of imaging studies alone. Usually, CAs show a reticulated core with mixed signal intensity surrounded by an obvious hemosiderin ring with faint or no enhancement. However, dural CAs, including tentorial CAs, are frequently misdiagnosed as meningiomas because of their similar radiographic findings. Plain CT findings of tentorial CAs included an iso- or hyperdense mass and homogeneous enhancement in only half of the cases (Table 1). Their MRI findings consisted of hyperintensity on T2-weighted images and iso- or hypointensity on T1-weighted images with homogeneous enhancement and an occasional dural tail sign. Generally, it is difficult to distinguish a tentorial CA from a meningioma or hemangiopericytoma. The present case showed almost the same findings as those in the tentorial CAs described previously, except for the coarse calcification on plain CT, irregular enhancement, and no dural tail sign on MRI. Moreover, in the differential diagnosis for the present tumor, skull tumors such as chondrosarcoma had to be considered because of the skull invasion. Hence, tentorial CA, albeit a rare tumor, should be considered in the differential diagnosis of tentorium-based tumors, including meningiomas and/or hemangiopericytomas and skull tumors.

Preoperative Endovascular Embolization

Angiographic information was available in 5 of the 9 previously reported cases (Table 1).7,14,16,19,23 Among these were 3 cases showing hypervascularity;7,14,19 patients in 2 of these cases were evaluated for intraoperative bleeding from tumors, and both experienced massive bleeding during surgery.7,19 Thus, most tentorial CAs were hypervascular and had profuse intraoperative bleeding. To our knowledge, no study has reported preoperative feeder embolization for tentorial CA. In the present case, the tumor was hypervascular, and blood was supplied by feeders such as a large meningeal branch of the left vertebral artery. The tumor was so large that we considered performing internal decompression or piecemeal removal at surgery. We planned to deliver embolic material deep inside the tumor and inserted a microcatheter to a distal
portion of the posterior meningeal artery. Thus, we could use PVA without reflux into the vertebral artery instead of proximal ligation of the posterior meningeal artery via a Guglielmi detachable coil. The feeders were embolized preoperatively, resulting in successful gross-total removal of the tumor with minimal blood loss. Hence, preoperative feeder embolization is quite useful for maximum safe resection of a tentorial CA.

Conclusions

Herein, we describe the first case of a huge hypervascular tentorial CA treated with preoperative endovascular embolization, followed by successful gross-total removal. A tentorial CA is most likely to be preoperatively misdiagnosed as a meningioma or hemangiopericytoma and carries a high risk of massive intraoperative bleeding. Therefore, it is important to remember that the differential diagnosis of tentorium-based tumors should include tentorial CA, although it is a rare tumor. Further, preoperative endovascular embolization should be considered for tentorial CA.

Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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