Modified Transanterior Perforated Substance Approach Using the Medial Territory to Thalamomesencephalic Cavernous Malformations: A Case Report

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BACKGROUND AND IMPORTANCE: Thalamomesencephalic lesions remain a challenge to neurosurgeons for their eloquent anatomy and postoperative neurological deficits. With limited cases in the literature, these lesions are still managed in a case-by-case fashion.

CLINICAL PRESENTATION: Here, we present a case of an otherwise healthy man who had a 46-day history of impaired vision and right-sided weakness and numbness caused by ventrally located thalamomesencephalic cavernous malformations (CMs). A modified transanterior perforated substance approach using the medial territory was performed for gross total resection of the CMs. No new-onset neurological deficits were observed postoperatively. Apart from visual improvements, the patient's muscle strength constantly improved and recovered full strength on 14-month follow-up.

CONCLUSION: The authors believe that the transanterior perforated substance approach through the medial territory can be considered as an option for ventrally located thalamomesencephalic CMs.

KEY WORDS: Anterior cerebral artery, Anterior perforated substance, Brainstem, Cavernous malformation, Thalamus, Thalamomesencephalic

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For lesions limited to the midbrain, the safe entry zone theory is well established.[1-4] While in thalamus, current theories are adequate for lesions limited to 1 of the 6 thalamic regions.[5] However, there is no consensus on management of thalamomesencephalic lesions.[6-11] In 2008, Feiz-Erfan et al[9] introduced the transanterior perforated substance (TAPS) approach using the lateral territory of anterior perforated substance (APS) to resect a thalamomesencephalic cavernous malformation (CM). The surgery was successful despite residual neurological deficits on follow-up. In this work, we applied a modified TAPS approach using the medial territory to completely remove extensive thalamomesencephalic CMs. To the best of our knowledge, this case was the first of its kind to be managed with this modified approach.

ABBREVIATIONS: APS, anterior perforated substance; **CM,** cavernous malformation; **CST,** corticospinal tract; **DWI,** diffusion weighted imaging; **HDFT,** high-definition fiber tractography; **ML,** medial lemniscus; **OD,** left eye; **OR,** optic radiation; **OS,** right eye; **RLL,** right lower limb; **RUL,** right upper limb; **TAPS,** transanterior perforated substance; **TMJ,** thalamomesencephalic junction.

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CLINICAL PRESENTATION

The patient was a 31-year-old otherwise healthy Asian male white-collar worker who presented in another hospital with a 46-day history of progressively impaired vision and right-sided weakness and numbness. Despite treatment with mannitol, valproic acid, and physical therapies including acupuncture and occupational therapies, his symptoms deteriorated and was referred to this hospital, a tertiary referral center. Past and family history was unrevealing. On admission, he was escorted in office with a wheelchair. Timeline of preadmission events can be referred to Figure 1. On physical

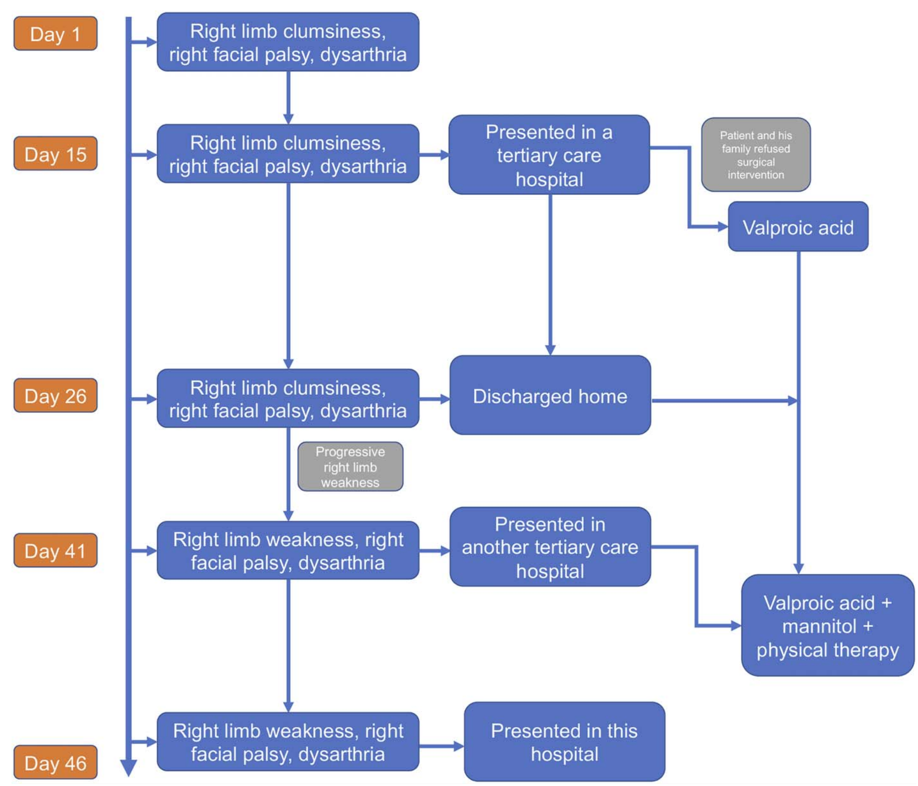

FIGURE 1. *Flowchart of significant events leading up to this patient's visit at our office.*

examinations, his vital signs were stable. He was alert and oriented. Detailed physical examinations can be referred to Table. Visual field test showed right-sided hemianopia. Right-sided homonymous hemianopia was confirmed (Figure 2). MRI revealed left-sided thalamomesencephalic CMs (Figure 3). High-definition fiber tractography (HDFT, **Supplemental Digital Content 1**, http://links.lww.com/NEUOPEN/A56) was reconstructed with DSI Studio (http://dsi-studio.labsolver.org), revealing disrupted and laterally displaced corticospinal tract and medial lemniscus (Figure 4). The left-sided optic radiation was displaced posteriorly (Figure 4).

Family meetings were held to explain risks and benefits of surgery. Informed consent was signed by the patient and his family. This report was further approved by the institutional review board and followed Surgical Case Report (SCARE) guidelines.[12] The surgery (Figure 5, Video) was performed by the senior author (M.L.) on day 6 of the patient's hospitalization. Intraoperative cranial nerve VII and motor-evoked and somatosensory-evoked potentials were monitored. Detailed descriptions of the pterional craniotomy were not exhaustive here because the techniques have been well established.[13,14]

Entry point was confirmed using neuronavigation (StealthStation S7, Medtronic). An incision was made in the medial territory of APS through A1 perforators. Microinstruments were maneuvered parallel to the course of perforators. Care was taken to keep perforators in situ whenever possible to prevent forced traction or occlusion of perforators. Blood clots were first evacuated on opening of APS. A retractor and microdissectors were used to improve visualization. The CM was resected in a piecemeal fashion after the dissection plane established between the CM and the perilesional hemosiderin deposits. The other CM was confirmed at the midbrain using neuronavigation and was removed in the same resection cavity. The associated developmental venous anomaly was preserved. After resection of the CM, the hemosiderin was excised to prevent postoperative epilepsy.[15,16] Several measures were taken throughout the procedure to prevent vasospasm (detailed in Video).

The patient was closely monitored at the dedicated neurosurgery intensive care unit postoperatively, and no new-onset neurological deficits were found. Complete removal of the CMs was confirmed (Figure 6). Postoperative course was uneventful, and the patient was discharged on postoperative day 9 for rehabilitation at a local health care facility, where the patient received rehabilitation therapy and instructions for 1 month and was then discharged for home rehabilitation which he performed consistently until the latest follow-up. His neurological functions constantly improved (Table). On 14-month follow-up, visual field test was near normal (Figure 7). High-definition fiber tractography indicated anatomic restoration of fiber tracts (Figure 8). The patient is now employed full-time, and the modified Rankin Scale score is 0.

TABLE. Comparisons of the Work by Feiz-Erfan et al 2008[9] and This Study

Study	Feiz-Erfan et al 2008[9]	This study
Presentations	Progressive right upper limb tremors, mild hemiparesis; CM at TMJ followed for 5 y	6-wk history of progressive impaired vision and right-sided weakness and numbness
Physical examinations	Right-sided limb tremors and paresis (not graded)	•Right-sided CN VII HB grade II •RUL strength grade 2 •RLL strength grade 3 •Right-sided proprioception ↓ •Right-sided Babinski (+) •20/63 (OD), 20/50 (OS) •Right-sided hemianopia •mRS = 4
Imaging findings	CM at TMJ, size not reported	Two CMs: one 45 × 38 mm thalamomesencephalic CM including hemorrhage, the other 8 × 6 mm tegmental CM
Craniotomy	Orbitozygomatic	Pterional
Approach	TAPS using the lateral territory, between M1 perforators	TAPS using the medial territory, between A1 perforators
Complete removal?	Yes	Yes
New-onset neurological deficits	Transient hemiplegia a few days postoperatively, which spontaneously resolved	None
Surgical outcomes	•10-mo follow-up, there was still residual weakness of the right limbs (not graded) •At last (timeline unmentioned) follow-up, the patient resumed daily activities.	•3-mo follow-up ○RUL and RLL strength 4 ○Right-sided proprioception slightly ↓ •14-mo follow-up ○Right-sided CN VII HB grade II ○Bilateral limbs strength normal ○Right-sided Babinski (−) ○20/25 (OS) and 20/32 (OD) ○Visual field test was near normal ○mRS = 0

↓, decreased; (+), positive; (−), negative; CM, cavernous malformation; CN, cranial nerve; HB, House-Brackmann; mRS, modified Rankin Scale; OD, left eye; OS, right eye; RLL, right lower limb; RUL, right upper limb; TAPS, transanterior perforated substance; TMJ, thalamomesencephalic junction.

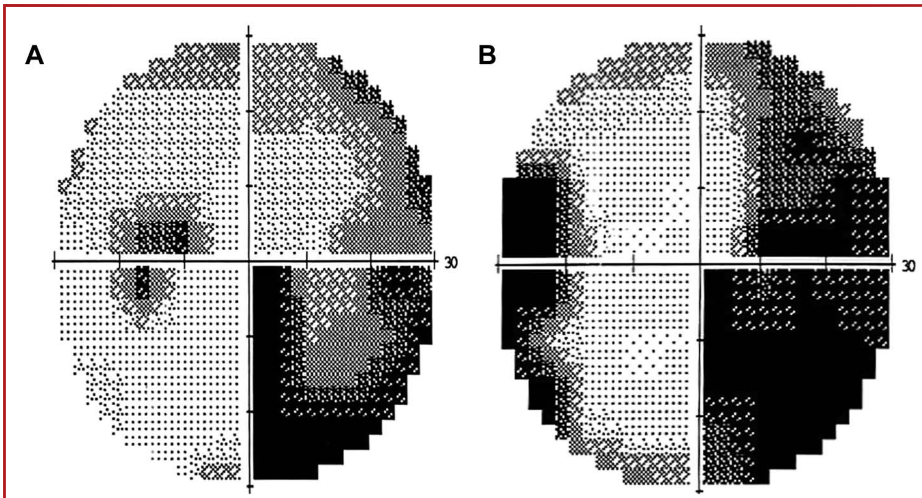

FIGURE 2. *Preoperative visual field test.* **A**, *Left eye visual field test revealed right-sided hemianopia;* **B**, *Right eye visual field test also revealed right-sided hemianopia.*

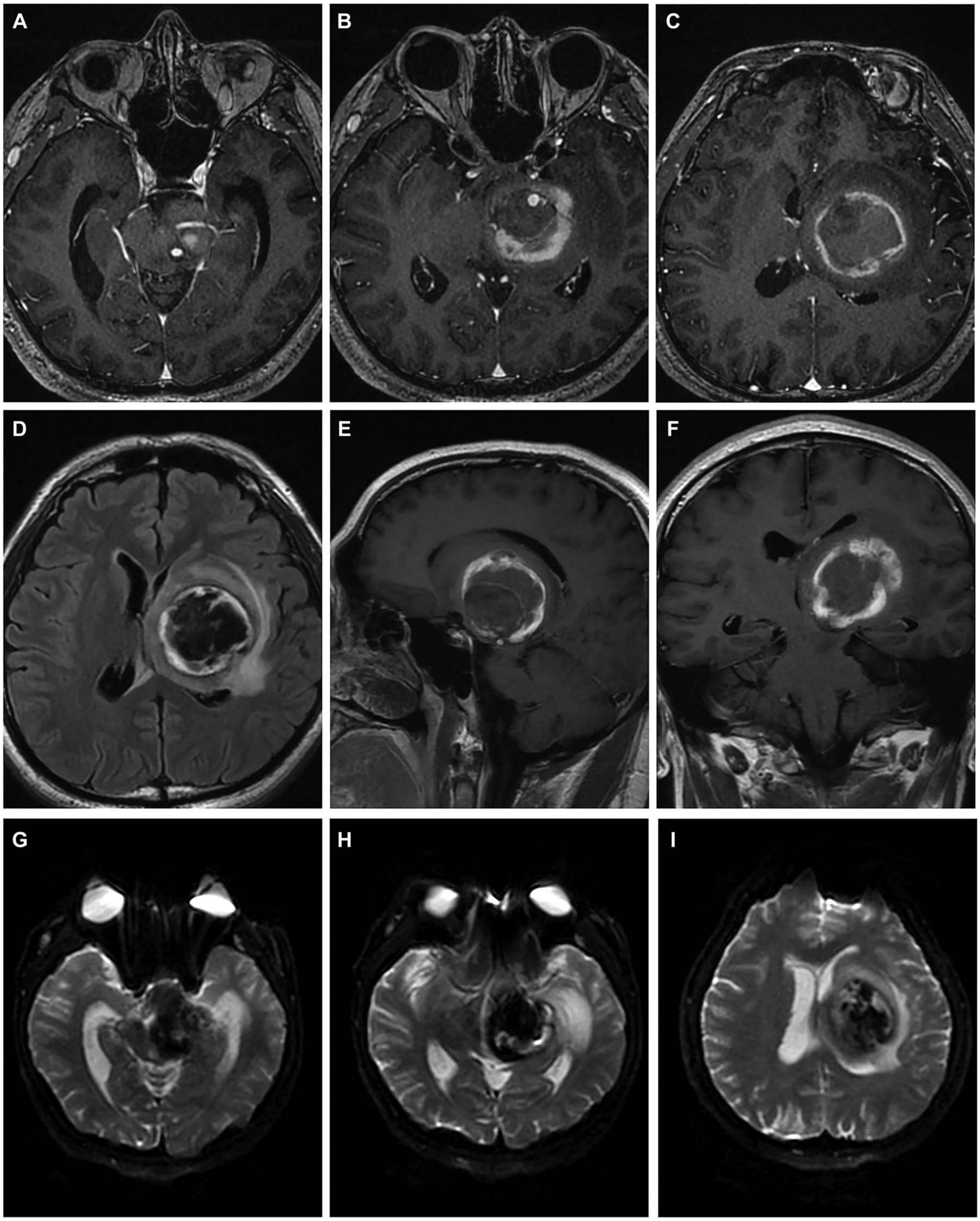

FIGURE 3. *Preoperative MRI studies revealed ventrally located thalamomesencephalic CMs (Zabramski Type II). The predominant CM including hemorrhage was 45 × 38 mm while the other 8 × 6 mm CM was at tegmentum.* **A**, *At the midbrain level, the CM, a developmental venous anomaly, and hematoma were obvious on T1WI with contrast.* **B**, *At the level of optic chiasm, the CM and hematoma were revealed.* **C**, *The hematoma cavity was extensive at the level of thalamus.* **D**, *On T2WI, the hematoma and surrounding edema compressed the left lateral ventricle.* **E** *and* **F**, *The lesions and the hematoma cavity were extensive from the thalamus to midbrain.* **G-I**, *Preoperative DWI study. CM, cavernous malformation; DWI, diffusion-weighted imaging; T1WI, T1-weighted imaging; T2WI, T2-weighted imaging.*

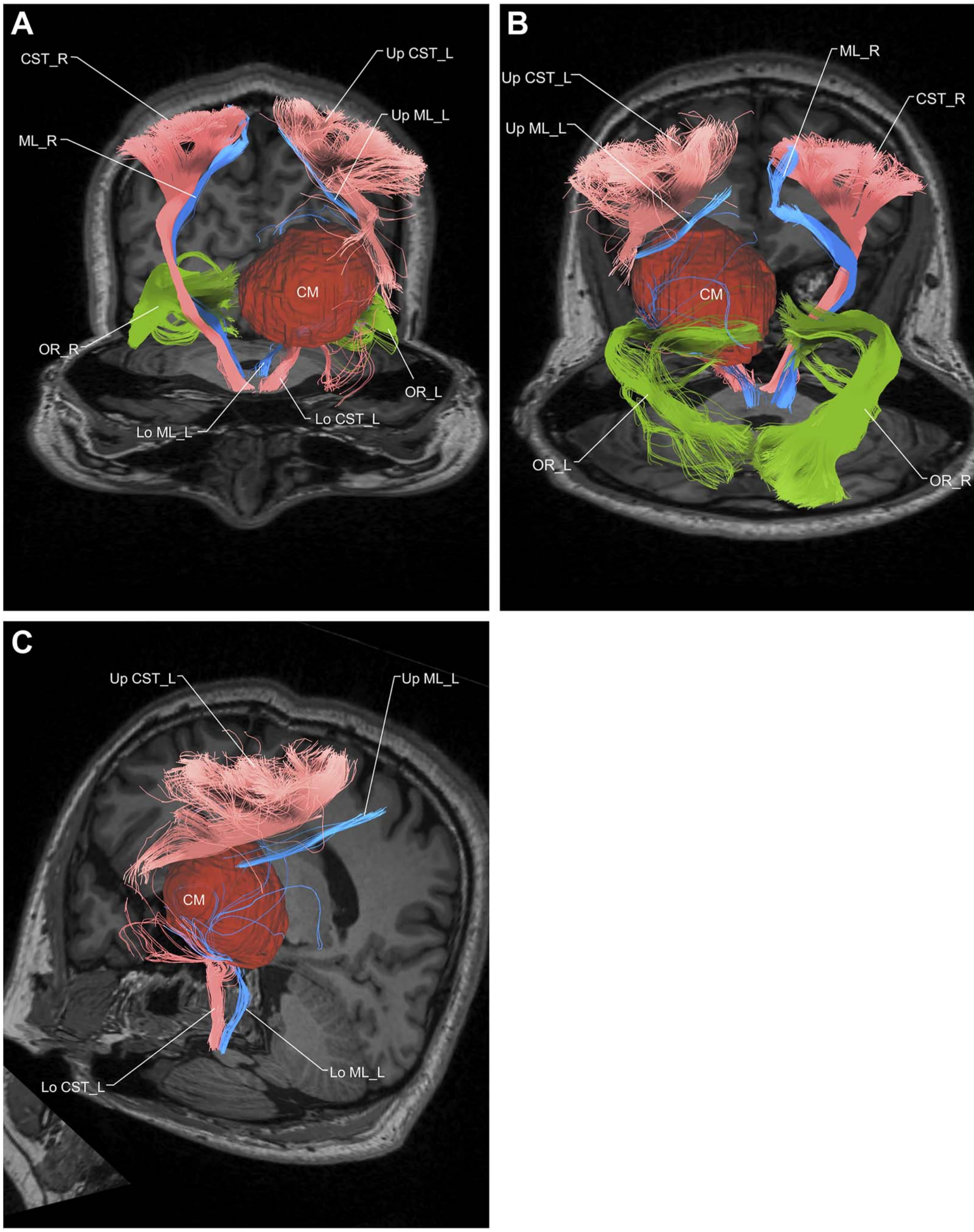

FIGURE 4. *Preoperative HDFT. The CMs and their related hemorrhage were manually segmented to build a 3-dimensional model. The optic tract of the diseased side was unable to be reconstructed because of mass effects. Color coding of the fiber tracts is assigned by the authors.* **A**, *The anterior view showed laterally displaced left CST and ML by the CMs and hematoma.* **B**, *The posterior view showed posteriorly displaced left OR.* **C**, *Left lateral perspective indicated disrupted and dispersed left CST and ML because of mass effects. CMs, cavernous malformations; CST, corticospinal tract; HDFT, high-definition fiber tractography; L, left-sided; Lo, lower portion; ML, medial lemniscus; OR, optic radiation; R, right-sided; Up, upper portion.*

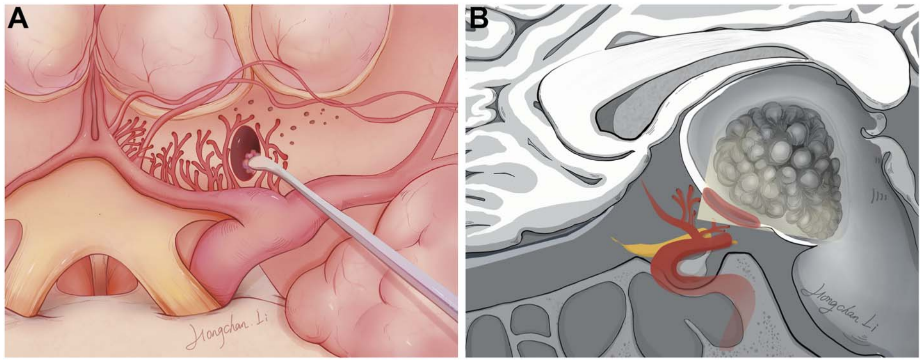

FIGURE 5. *Artist's (Hongchan Li) illustration of the transanterior perforated substance approach using the medial territory. The inferior limit of incision was A1. Perforators of the recurrent artery and M1 were omitted for better view.* **A**, *Anterior view shows the incision into the medial territory of anterior perforated substance, between the A1 perforators.* **B**, *Lateral view demonstrates that after frontal lobe retraction, the angle of attack (dim yellow light) through the incision covers both the lower thalamus and ventral midbrain. Courtesy of Hongchan Li.*

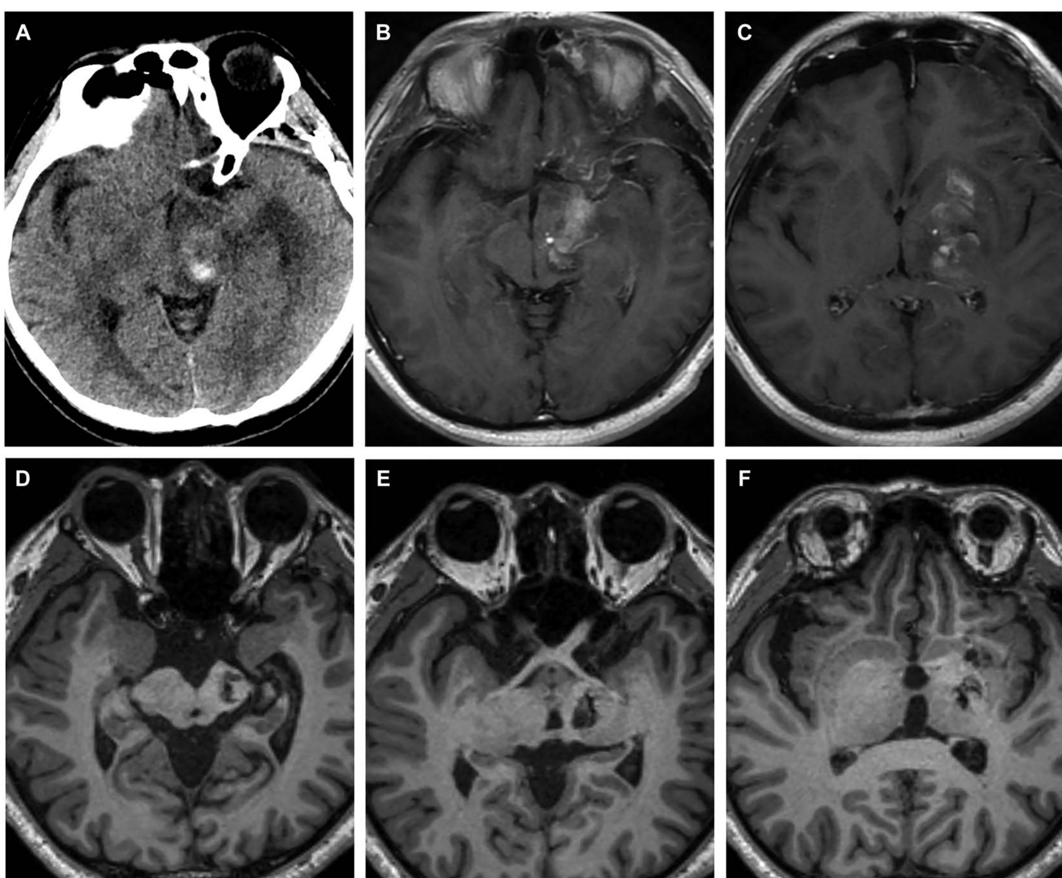

FIGURE 6. *Postoperative imaging studies revealed complete removal of the lesions.* **A**, *Head computed tomography obtained on 1 day postoperatively revealed a region of high density at the level of thalamomesencephalic portion. This was due to implants of absorbable hemostats.* **B** *and* **C**, *T1WI scanned on 1 week postoperatively indicated surgical implants in the resection cavity.* **D-F**, *On 14-month follow-up, the resection cavity was obvious on T1WI. No sign of rehemorrhage was noted. T1WI, T1-weighted imaging.*

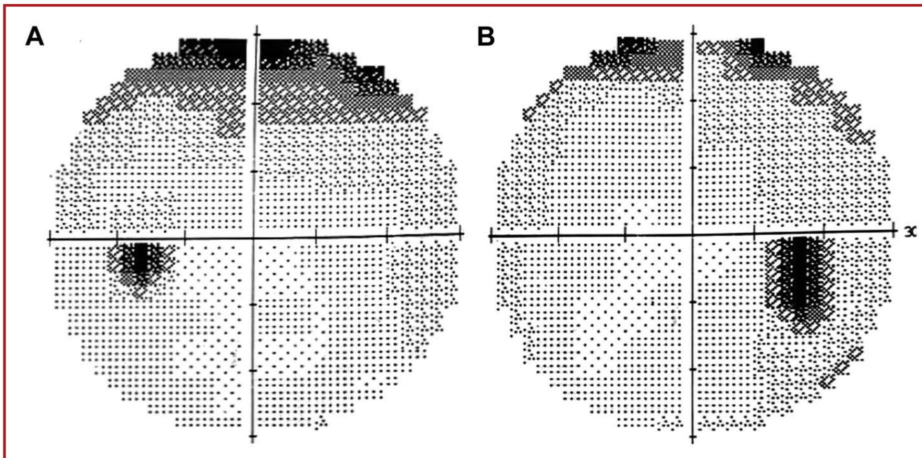

FIGURE 7. *Visual field test on 14-month follow-up.* **A**, *Left eye visual field test revealed recovered hemianopia.* **B**, *Right eye visual field test also revealed recovered hemianopia.*

DISCUSSION

Surgery remains a mainstay treatment for thalamus and brainstem CMs, but it should be noted that not all CMs need surgery, and observation for stable lesions without neurological deficit is acceptable and most appropriate course.[17-21] This case was not suitable for posterior approaches, including the suboccipital supracerebellar infratentorial approach, given the posteriorly displaced optic radiation. Subtemporal or other transcortical approaches such as transinsular approach may risk damaging already displaced fiber tracts including the corticospinal tract and optic tract.[22-25] The trajectory of the anterior ipsilateral interhemispheric transcallosal or anterior contralateral interhemispheric transcallosal approach, on the other hand, would be relatively long, which may not be ideal for this patient with 2 CMs in a deep-seated location. A supracarotid-infrafrontal approach[5,26] to attack the anteroinferior thalamus may suffice but visualization of the midbrain portion would be limited.

According to a cadaveric study by Rosner et al,[27] APS can be divided into 2 territories (medial and lateral) or 3 zones (anterior, middle, and posterior). The medial territory is perfused by perforators of A1, C4, and recurrent artery while the lateral territory is perfused by perforators of M1.[27] Perforators of A1 and recurrent artery mostly supply the genu of the internal capsule, parts of the putamen, caudate nucleus, and globus pallidus. This was further confirmed in a case series of unruptured A1 aneurysms by Lee et al,[28] where postoperative computed tomography study after A1 perforator occlusion showed infarction in these regions.

This case was different from the work by Feiz-Erfan et al[9] in several ways (Table). First, we used a pterional instead of orbitozygomatic craniotomy, also achieving adequate exposure and brain relaxation. Second, our incision was made in the anterior and middle zone of the medial territory instead of lateral territory. Thus, we could avoid ischemic events of the posterior limb which was most susceptible to injuries of M1 perforators in the lateral territory.[27] Third, despite worse presentations and more extensive lesions, no new-onset neurological deficits were observed postoperatively, and the patient's muscle strength recovered to normal after 14 months. Finally, in case of more cephalically located thalamic lesions, the incision in TAPS can be extended frontally to bridge the incision of the supracarotid-infrafrontal approach.

Limitations

To our best knowledge, this case was the first of its kind managed with a TAPS approach using the medial territory. The patient complied with treatment and rehabilitations and was satisfied with the outcomes. However, this report was still limited. First, intraoperative mapping was not used. In addition, we failed to export tractography from DSI Studio to the navigation system because of incompatibilities. Second, we removed the perilesional hemosiderin deposits in this patient, but it should be noted that the role of hemosiderin excision in eloquent areas remains elusive.[15] Third, inadvertent injuries of A1 perforators could insult the corticobulbar tract, which may cause dysarthria, facial palsy, or dysphagia.[27,29-31] However, it is noteworthy that the genu of the internal capsule is perfused by perforators of both A1 and C4 branches, the later entering the APS below the level of A1 and M1—the inferior limit of our approach. Finally, given the limited literature, the indications of this approach for lesions other than CMs are unclear. Above all, we believe that this approach should be reserved for situations when safer options are unavailable. Further anatomic and human studies are warranted.

CONCLUSION

In experienced hands, a TAPS approach using the medial territory can be considered for ventrally located thalamomesencephalic CMs.

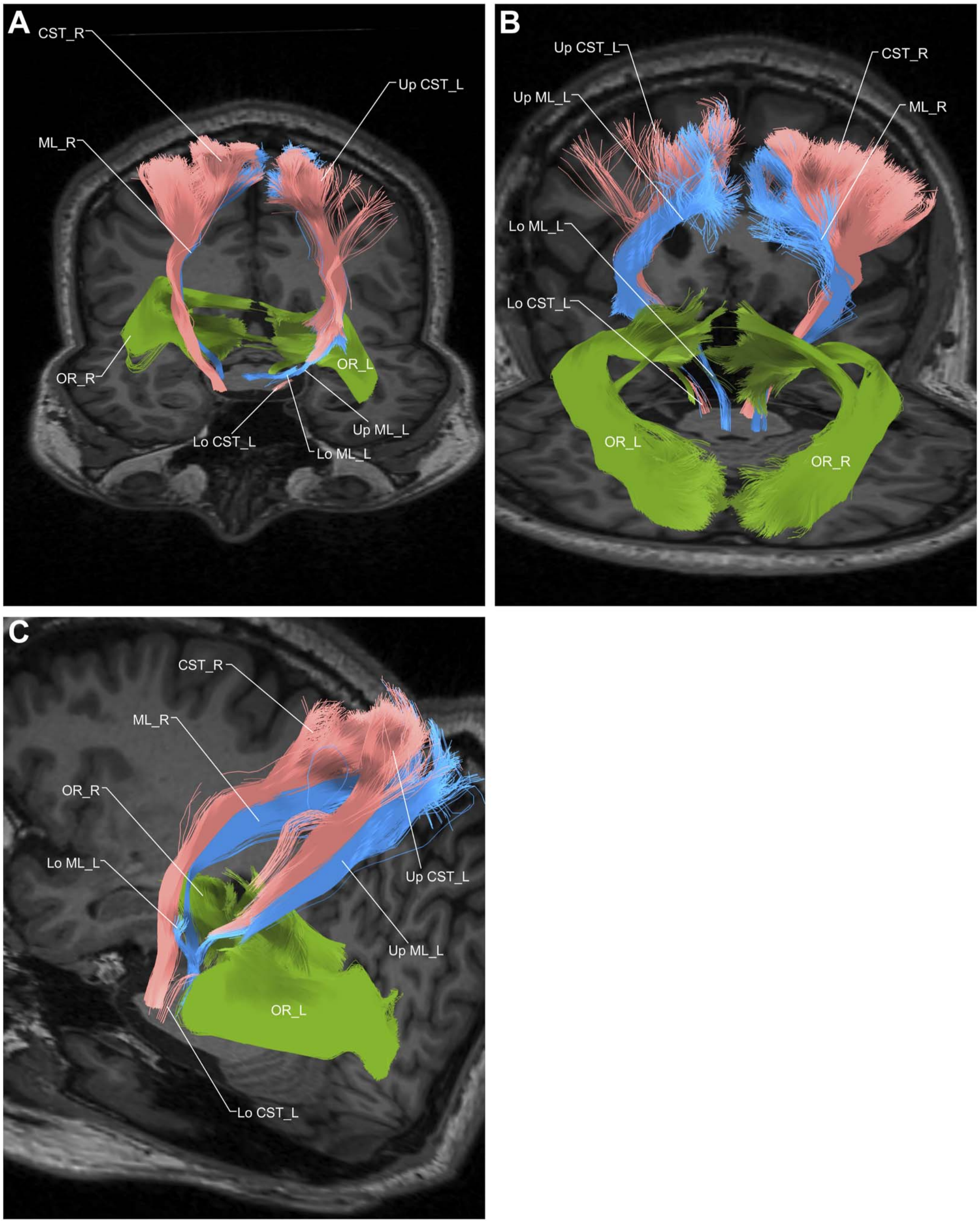

FIGURE 8. *HDFT on 14-month follow-up.* **A**, *From the anterior view, the left-sided CST and ML were less displaced as compared with preoperative HDFT.* **B**, *From the posterior view, increase of left OR volume was obvious.* **C**, *Lateral view revealed still displaced and disrupted CST and ML. CST, corticospinal tract; HDFT, high-definition fiber tractography; ML, medial lemniscus.*

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Disclosures

The authors have no personal, financial, or institutional interest in any of the drugs, materials, or devices described in this article.

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Supplemental digital content *is available for this article at neurosurgerypractice-online.com.*

Supplemental Digital Content 1. Institutional protocol of high-definition fiber tractography acquisition and reconstruction.

VIDEO. Modified transanterior perforated substance approach using the medial territory to thalamomesencephalic cavernous malformations: operative video.

COMMENTS

The authors present the resection of a challenging cavernous malformation via a trans-anterior perforated substance (APS) corridor and achieve significant improvement in preoperative neurological deficits. A discussion of some surgical strategies, including use of tractography and description of microsurgical dissection, is presented. Certainly, the lesion's most superficial aspect was anterior and offered the shortest corridor. In addition, various other surgical approaches including the suboccipital supracerebellar infratentorial, interhemispheric transcallosal, and transcortical approaches were considered by the authors but had drawbacks. While I imagine no neurosurgeon would be excited to recreate an APS approach for their own patient, the critical questions are how the patient tolerated this approach and whether these insights can be helpful for future patients? A healthy respect for the anterior perforators generated from years of study and reporting of clinical outcomes[1a-4a] as well as development of microsurgical technique has surely helped inform the treatment of this very difficult case.

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