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A 15-year follow-up of permanent intraoperative internal carotid artery occlusion for hemostasis in a giant cavernous sinus hemangioma: a case report

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Abstract

Background Cavernous sinus hemangiomas are rare vascular tumors characterized by significant intraoperative bleeding. Their preoperative diagnosis is challenging and often leads to initial misdiagnosis as other middle fossa lesions, such as tumors or aneurysms.

Case presentation We report the case of a 16-year-old Persian male presenting with progressive left-sided visual loss, cranial nerve palsy, with an intracranial lesion originating from the left cavernous sinus, extending into the sella, and occupying half of the middle fossa. He had previously undergone surgery with a preliminary diagnosis of a cerebral aneurysm, but the procedure was aborted owing to excessive intraoperative bleeding. The patient was subsequently transferred to our institution for further intervention. During the second surgical attempt, persistent bleeding and hemodynamic instability necessitated the permanent ligation of the left internal carotid artery to achieve hemostasis. Postoperative pathological examination confirmed the diagnosis of a giant cavernous sinus hemangiomas. Remarkably, the patient exhibited no postoperative neurological deficits. Over a 15-year follow-up period without adjuvant radiotherapy, he remained asymptomatic, with no evidence of tumor recurrence or neurological deterioration.

Conclusion The emergent nature of the situation in this case necessitated carotid artery ligation, which was performed without long-term neurological compromise. However, assessing the patient's tolerance to carotid artery ligation preoperatively is crucial in ensuring a safe outcome. A proactive, well-prepared surgical approach, including thorough preoperative vascular assessment, is essential for optimizing intraoperative safety and plays a pivotal role in managing the risks associated with severe intraoperative bleeding in cavernous sinus hemangiomas cases.

Keywords Cavernous sinus hemangioma, Middle fossa, Massive hemorrhage, Carotid artery ligation, Case report

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Introduction

Cavernous sinus hemangiomas (CSHs) are rare and benign extra-axial lesions that arise from the dural sinuses. This subset of vascular tumors is situated within the anatomically intricate cavernous sinus at the skull base. Incidence studies reveal a predilection for CSHs in adults, particularly middle-aged females, though

comprehensive epidemiological data are limited due to their rarity [1].

These tumors, accounting for less than 2% of cavernous sinus neoplasms, pose unique challenges in both diagnosis and treatment. CSHs often present with a spectrum of neurologic symptoms, including but not limited to headache, visual disturbances, or cranial nerve palsies [1]. The slow-growing nature of these tumors contributes to a gradual onset of symptoms, making early diagnosis challenging. CSHs are preliminarily diagnosed by compatible clinical presentation and imaging results. However, imaging study results may mimic meningioma, schwannoma, chordoma, metastasis, lymphomas, abscess, sarcoidosis, Tolosa–Hunt syndrome, and cerebral aneurysms. Prognostically, CSHs are generally considered benign, and complete surgical resection often leads to favorable outcomes. The primary treatment options for these cases are surgical resection and radiosurgery. However, Gamma Knife radiosurgery may not be suitable for large and giant cavernous sinus hemangiomas (CSH) owing to limitations in its effectiveness. In addition, surgical resection can be challenging for these lesions owing to the risk of neurovascular injury and severe intraoperative bleeding, which occurs in up to 40% of cases [2]. Moreover, intraoperative mortality is reported to be as high as 12.5% due to high risk of uncontrolled bleeding [3]. This case report details a young man with a massive cavernous sinus hemangioma, initially misdiagnosed as an aneurysm and the ensuing challenges encountered during surgical resection.

Case presentation

A 16-year-old Persian male sought medical attention owing to a 2-year history of progressive left-sided visual loss and proptosis. Brain computed tomography (CT) imaging revealed a mass, prompting his referral to a secondary care hospital. The initial workup, including a relevant family history of cranial surgery in distant relatives and additional imaging studies, led the primary surgeon to provisionally diagnose a middle fossa malignancy.

The patient was scheduled for surgical removal of the identified lesion within a few weeks. Following a preliminary tumor diagnosis, surgery proceeded without further investigation beyond routine preoperative laboratory tests and imaging. However, the procedure was abruptly halted due to unexpected and substantial bleeding during the attempted lesion removal. Given a high suspicion of an aneurysm, the lesion was packed, and the surgical field was temporarily sealed. Owing to limited resources and surgical expertise for managing this critical situation, the patient was urgently transferred to our institution for further intervention and comprehensive care.

On examination, the patient exhibited profound blindness in the left eye along with palsy of the oculomotor (CN III), trochlear (CN IV), abducens (CN VI), and ophthalmic (V1) and maxillary (V2) branches of the trigeminal nerve, strongly suggesting cavernous sinus involvement. No cognitive deficits were observed. A head CT scan performed at the referring hospital revealed a large hyperdense mass occupying the middle fossa within the left cavernous sinus, extending into the lateral and third ventricles. In addition, the imaging showed significant mass effect and midline shift, with a hypodense core in the left temporal lobe (Fig. 1A).

Both magnetic resonance imaging (MRI) and CT angiography were performed. MRI revealed a large lesion originating from the left cavernous sinus, extending into the sella, and occupying half of the middle fossa (Fig. 1B–D). The lesion appeared isointense on T1-weighted images and hyperintense on T2-weighted images, with a central dark, hypointense signal. In addition, peripheral enhancement with a non-enhancing core was observed. CT angiography ruled out an aneurysm, and no major feeding vessels to the lesion were identified (Fig. 1; top right corner).

A reoperation was performed within the same year under the diagnostic impression of a middle fossa tumor. The previous temporoparietal craniotomy was extended to include a frontobasal approach. Removal of the deformed sphenoid ridge and orbital bones exposed an extra-axial, interdural, broad-based lesion originating from the cavernous sinus. The lesion appeared firm with a purple-to-bluish hue (Fig. 2A). An attempt at surgical resection followed; however, despite employing various hemostatic techniques—including the use of surgical and absorbable gelatin sponges—severe pulsatile bleeding ensued after partial lesion resection. Efforts to control the bleeding through packing were unsuccessful.

Despite the use of various hemostatic measures, persistent surgical field bleeding and hemodynamic instability necessitated ligation of the left internal carotid artery (ICA) as a last resort to achieve hemostasis. A Yasargil clip was applied to permanently ligate the proximal segment, followed by careful packing of the tumor bed to control bleeding. Given the patient's overall condition, no further attempts were made to achieve complete resection beyond a subtotal excision (Fig. 2B). The patient's condition was carefully monitored throughout the remainder of the procedure until wound closure. The recovery phase was smooth and uneventful, with no observed new neurological deficits postoperatively. Histological examination identified the lesion as a large CSH (Fig. 2C, D). Subsequent MRI

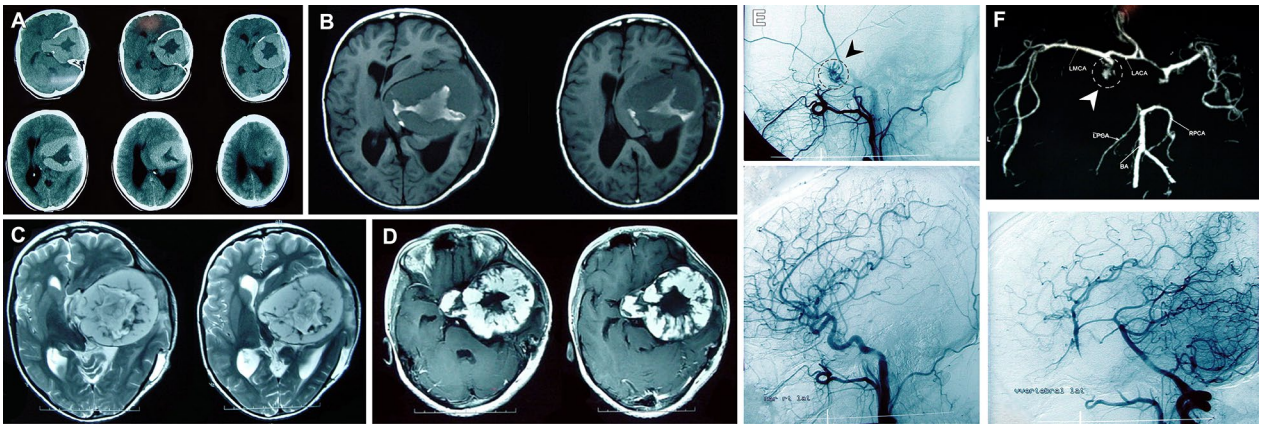

Fig. 1 Preoperative computed tomography imaging with previous craniotomy evident laterally (**A**). The lesion exhibited isointense signal on T1W (**B**) and hyperintensity on T2W (**C**), with a central necrotic core with a dark, hypointense signal. A sponge-like lobulated well-demarcated lesion with heterogeneous enhancement was seen in T2-weighted (**C**) and gadolinium-enhanced T1-weighted (**D**) images. Computed tomography angiography prior to surgical intervention (**F**) revealed diminished-to-no arterial feeding (top right corner—dashed line and arrow head). Postoperative digital subtraction angiography (**E**) revealed a faint blushing of the left middle meningeal artery (dashed lines and arrow heads) and closure of the ipsilateral internal carotid following clipping. Contralateral and vertebral views are given below the image as reference

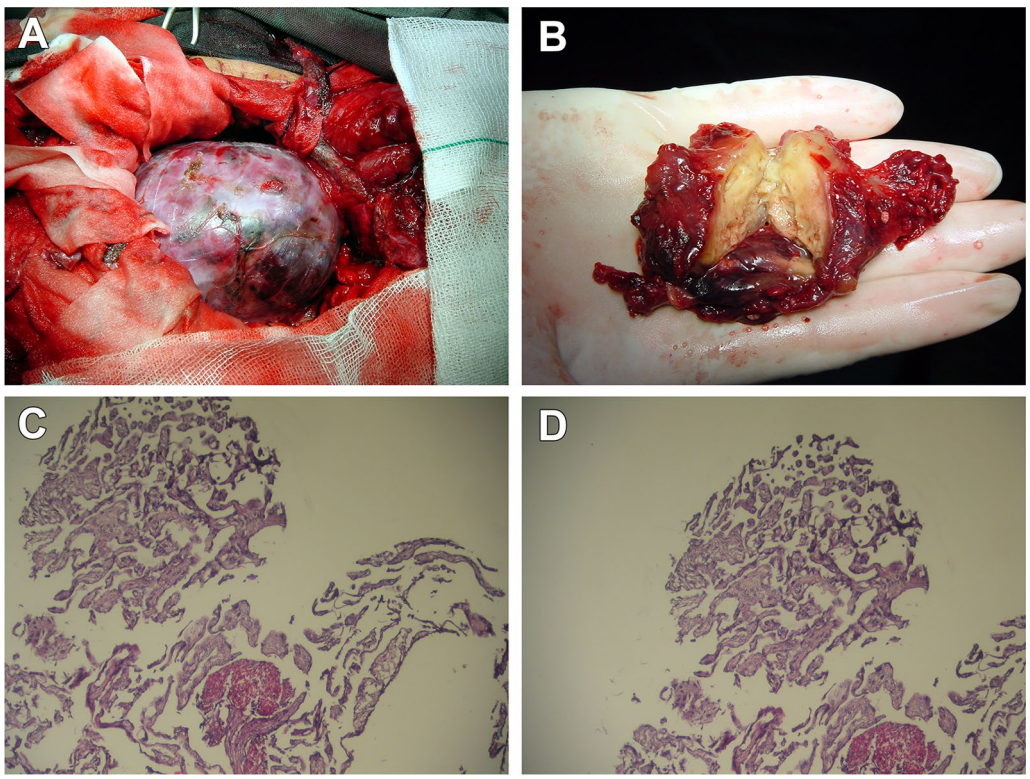

Fig. 2 **A** Surgical view of the cavernous sinus hemangioma in an inter-dural position. **B** Gross histology of the mulberry-like subtotal excised tumor. **C**, **D** Histological features of the excised lesion were sinusoidal vessel cavities and the surrounding connective tissue, compatible with cavernous sinus hemangioma (hematoxylin–eosin stain)

confirmed the partial removal of the tumor (Fig. 3). A follow-up cerebral angiography indicated a faint blush originating from the middle meningeal artery at the tumor base (refer to Fig. 1; right side of the figure). Upon discharge and resolution of neurological symptoms, the patient was counseled on the potential benefits

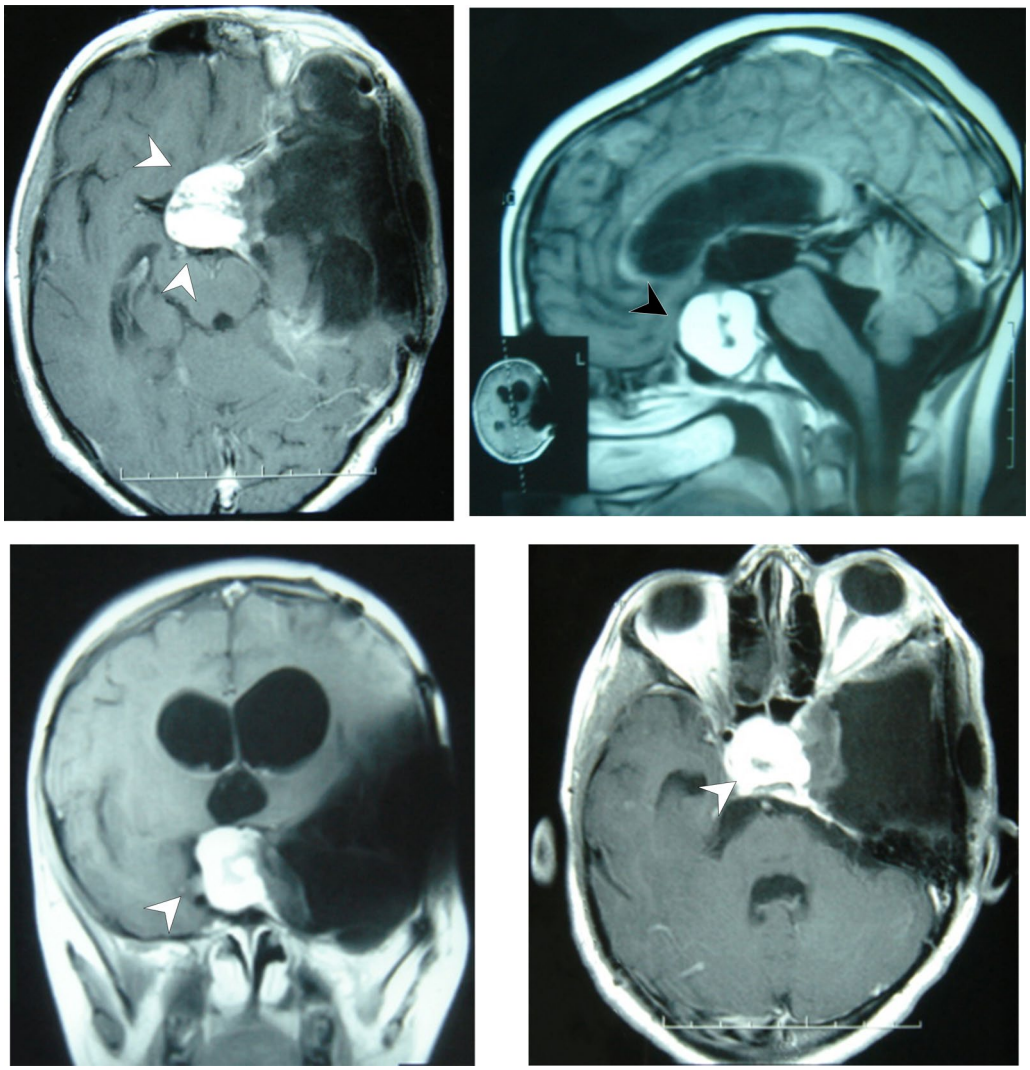

Fig. 3 Postoperative magnetic resonance imaging scans of the patient years after the surgical intervention. The residual mass (arrowheads) did not expand beyond what is shown here

of radiotherapy for a comprehensive treatment plan. However, after being informed about potential side effects, he opted against radiotherapy. As a result, he was strongly advised to adhere to regular follow-up visits, including monthly assessments and annual evaluations, to monitor tumor remission.

Over the subsequent 15 years of clinicoradiological follow-up, the patient—aged 31 years at the last evaluation—remained neurologically intact, with no signs or symptoms of recurrence. Sequential brain MRIs showed no detectable changes in residual tissue (Fig. 3). Written informed consent was obtained from the patient for the publication of anonymized clinical and imaging data.

Discussion

CSH comprise 3% of all benign cavernous sinus tumors [4]. With these lesions representing one of the cerebral vascular malformations, excluding other presumptive diagnoses in the adjacent region in imaging studies is rather difficult, as meningiomas, schwannomas, sarcoid granulomas, and venous thrombi may also form in or in close proximity to the cavernous sinus. Nevertheless, the sensitivity of MRI imaging alone in diagnosing CSH is 89.5% [5]. Extensive background knowledge is paramount to establish a presurgical diagnosis. CSHs are isodense or hyperdense in CT imaging. In MRI, these lesions are recognized by well-circumscribed low or iso-signal intensity in T1 and sponge-like heterogeneous

hyperintensity with contrast-enhanced, T2-weighted, and fluid-attenuated inversion recovery images (Table 1).

A filling-in effect or anteromedial shift of C3–C4 segment of the internal carotid artery in angiography may also be suggestive of CSHs [6]. Digital subtraction angiography (DSA) may reveal diverse flow patterns in the cavernous sinus hemangioma (CSH). The most common finding is arterial staining during the arterial phase. Other observed patterns include delayed arterial enhancement, no arterial uptake, and late venous filling. In addition, the humoral blush receives blood supply from branches of the external carotid artery or the cavernous portion of the internal carotid artery.

The primary objective of surgical intervention for cavernous hemangiomas is to alleviate neurological symptoms rather than achieving complete resection of tumor foci. The symptoms observed in each patient primarily stem from the tumor's compressive effect on adjacent neurovascular components. However, achieving total resection through traditional surgical approaches can be challenging due to the lesion's location and the potential for life-threatening hemorrhage. Conservative measures such as induction of hypotension may be insufficient to prevent such complications [3]. In situations involving substantial blood loss following lesion manipulation, therapeutic occlusion or embolization of the main feeding artery may emerge as a critical, life-saving intervention [7]. While this practice is often applied to giant aneurysms in the vertebral region, basilar artery at the upper pontine border, carotid-cavernous fistulas, or certain other skull-based neoplasms, intraoperative ICA closure could be employed in emergent conditions to achieve hemostasis in CSHs. Nonetheless, abrupt non-selective ICA occlusion carries a considerably high risk of subsequent cerebral infarction and significant morbidity and mortality [8]. The established method for preoperative risk assessment of ICA ischemic tolerance involves angiographic balloon test occlusion, coupled with either imaging modalities or clinical evaluation to assess patient tolerance [9]. In light of the complications observed in our case, we advocate for the judicious application and widespread adoption of preoperative ICA ligation tolerance screening in all cases of cavernous sinus hemangiomas undergoing surgical resection, especially in those with no apparent vascular feeding in imaging.

Although the urgent nature of the patient's presentation at our center precluded a thorough examination before surgical intervention, proper testing could have enhanced the surgeon's confidence, influenced the surgical approach, and mitigated the potential for intraoperative exsanguination in this patient. Although the current literature advocates for adjuvant therapies for further reduction in tumor size and recurrence rate, we did not observe any further growth beyond that of the residual tumor despite adequate follow-up. In light of high rate of complications, the question arises whether surgical or radiological intervention or adjuvant therapies may be necessary and are superior to vigilant follow-ups in highly-vascular and non-symptomatic cases.

Conclusion

Even with clear visualization of supplying or draining vessels, attempting surgical intervention on CSH carries a significant risk catastrophic intraoperative bleeding. This potential for life-threatening hemorrhage necessitates meticulous preoperative planning and a multi-pronged approach to mitigate this risk, to ensure the safety of intraoperative ligation of the carotid artery as the last resort. Preoperative angiography, angioembolization of the related vasculature, and occlusion tolerance tests of the internal carotid artery may be utilized as indispensable preoperative tools in this regard. In similar emergent cases where a thorough

Table 1 Imaging findings associated with CSH

CT-scan	Well-demarcated hyperdense lesion. May be occasionally dumbbell-shaped with sellar extension and/or dural tail
T1-weighted MRI	Low or isointense pre-contrast signal
T1 contrast enhanced MRI	Lobulated structure and homogeneous or progressive filling-in enhancement of the lesion from the periphery to center. Homogeneous filling-in may more likely be seen with subtype A
T2-weighted, fluid-attenuated inversion recovery (FLAIR) and proton-density MRI	Extra-axial hyperintense lesion. T2-weighted signal intensity similar to CSF signal
Apparent diffusion coefficient (ADC) map and diffusion-weighted MRI (DWI)	No restriction of diffusion normally seen in ADC map. Isointense to adjacent brain tissue in DWI
Magnetic resonance spectroscopy (MRS)	Small choline and N-acetyl-aspartate peaks without frank alanine peak. A large lipid peak could be seen at 0.9–1.3 ppm
Digital subtraction angiography (DSA)	Delayed tumoral blush irrigated by dural branches of the carotid siphon and middle meningeal artery. One-third of CSHs may be angiographically silent

workup or comprehensive cerebrovascular history is lacking, intraoperative ICA ligation should be considered as a last resort, balancing the preservation of life against the potentially catastrophic consequences of unsafe ICA occlusion.

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Author contributions
Mehdi Chavoshi-Nejad: Conceptualization, methodology, data curation, writing—review and editing, and visualization. Reza Molahoseini: validation, resources, data curation, and project administration. Ali Baradaran Bagheri: validation, writing—review and editing. Sepehr Aghajanian: writing—original draft.

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Data availability
The anonymized clinical data and imaging files are available from the corresponding author on reasonable request.

Declarations

Ethics approval and consent to participate
Informed consent was obtained from the patient. Ethical approval was waived due to direct consent and the absence of preoperative planning of interventions beyond the standard of care.

Consent for publication
Written informed consent was obtained from the patient's legal guardian for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Competing interests
The authors have no relevant affiliations or financial involvement with any organization or entity with a financial interest in or financial conflict with the subject matter or materials discussed in the manuscript.

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