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Case Report

Complex arteriovenous malformation of the corpus callosum: Surgical nuances

Mustafa Ismail[1], Saja A. Albanaa[2], Ali M. Neamah[2], Noor F. Hassan[2], Yasir A. Mohialdeen[2], Awfa Aktham Abdulateef[3], Samer S. Hoz[4]

[1]Department of Neurosurgery, Neurosurgery Teaching Hospital, [2]Departments of Surgery, College of Medicine, University of Baghdad, Baghdad, Iraq, [3]Department of Neurosurgery, Tokyo General Hospital, Nakano, Japan, [4]Department of Neurosurgery, University of Cincinnati, Cincinnati, United States.

ABSTRACT

Introduction: Arteriovenous malformations of the corpus callosum (CC-AVMs) are rare and challenging lesions, accounting for 8–9% of all cerebral AVMs. They are associated with a high risk of recurrent hemorrhage and historically were considered inoperable. This case report describes the successful surgical resection of a high-grade CC-AVM following failed stereotactic radiosurgery.

Case Description: A 19-year-old female with an anterior CC-AVM presented with seizures despite being on triple anti-epileptics. The patient had previously undergone conservative management for an intraventricular hemorrhage and stereotactic radiosurgery for the AVM. Pre-operative imaging revealed a 5 cm CC-AVM located at the genu and rostrum of the corpus callosum with intraventricular extension into the third ventricle. The AVM was classified as grade 4 according to the Spetzler-Martin grading system and grade 6 according to the supplementary grading system. The lesion was exposed through the anterior interhemispheric trans-callosal approach, and microsurgical nidal dissection was performed. The large draining vein was identified, coagulated, disconnected, and resected along with the nidus. The total operative time was 12.5 h. Postoperatively, the patient had no neurological deficits and was discharged on day 10. Complete resection was confirmed on postoperative magnetic resonance imaging, and the patient remained seizure-free at the 18-month follow-up visit.

Conclusion: This case report demonstrates the feasibility of complete surgical resection for high-grade CC-AVMs that have failed non-surgical therapy. Careful surgical planning and technique can lead to favorable outcomes in these challenging cases.

Keywords: Cerebrovascular surgery, Arteriovenous malformations, Corpus callosum

INTRODUCTION

Arteriovenous malformations of the corpus callosum (CC-AVMs) are relatively rare vascular anomalies, accounting for 8–9% of all cerebral AVMs.[1] As to clinical presentation, hemorrhage is the most common presenting symptom, with intracerebral and subarachnoid hemorrhage being responsible for 70–100% of the recorded primary presentations, collectively.[1] Other presentations include seizures and headaches. CC-AVMs favor the younger age group compared to the more superficially positioned AVMs, with a mean age at presentation between 25 and 35 years.[1] Besides, due to their deep-sealed location and deep venous drainage, CC-AVMs are more likely to have recurrent hemorrhage compared to more superficially located AVMs.[2]

Historically, CC-AVMs were considered inoperative due to the high incidence of severe neurological deficits and neuropsychological dysfunction following surgical resection.[3] In 1951, Guidetti and Spallone[4] reported the first attempt to resect a CC-AVM surgically which was followed by a considerable development in the treatment options. Current literature informs us of 37 studies reporting on CC-AVM management results, focusing on the use of alternative treatment approaches, including endovascular embolization and stereotactic radiosurgery.[1]

In this paper, we report the successful complete excision of a CC-AVM with extensive intraventricular extension, which showed remarkable growth and new-onset seizures while being treated with stereotactic radiosurgery. Using the success of surgery for such a notoriously tricky lesion as an example, the paper highlights the feasibility of complete surgical resection of high-grade CC-AVMs that have failed non-surgical therapy.

***Corresponding author:** Mustafa Ismail, Department of Neurosurgery, Neurosurgery Teaching Hospital, Baghdad, Iraq. mustafalorance2233@gmail.com

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CASE REPORT

A 19-year-old female with anterior corpus callosum arteriovenous malformation (CC-AVM) presented with a history of three episodes of tonic-clonic seizures over the preceding month while on a full dose of triple anti-epileptics. This is on a background history of a previous admission, in 2018, for an intraventricular hemorrhage that was managed conservatively. Subsequently, the patient received a course of stereotactic radiosurgery (Gamma Knife). The lesion size was noted to have increased at the 9-month follow-up visit, following a period of initial stabilization.

At this admission, the pre-operative imaging showed a CC-AVM with a maximum nidal diameter of 5 cm nidus located at the genu and rostrum of the corpus callosum with intraventricular extension into the third ventricle. The arterial supply came from the pericallosal artery anterior cerebral artery (ACA), and the venous drainage was represented by a very large deep draining vein, which spanned the cavity of the third ventricle to terminate posteriorly at the vein of Galen. According to the Spetzler-Martin grading system, this AVM was a grade 4 (size of 5 cm– 2 points-, deep venous drainage – 1 point-, and eloquent adjacent areas – 1 point). According to the supplementary Spetzler-Martin grading system, the AVM receives an additional two points for the patient's age and nidal diffuseness, giving a combined grade of 6 [Figure 1].

The AVM was exposed through the anterior interhemispheric trans-callosal approach with coagulation and cutting of the anterior third of the falx cerebri. Next, microsurgical nidal dissection was carried out, including identification and cauterization of the feeding arteries. Intraoperatively, a large part of the nidus was within the cavity of the third ventricle. Thus, further dissection and tracking continued until the very large draining vein was identified. At this point, the draining vein, which has now become soft (low flow), was coagulated, disconnected, and resected along with the nidus. Then, an inspection of the AVM cavity was checked for any remnant nidus. Next, hemostasis was achieved, and craniotomy closure took place. The overall operative time was 12.5 h.

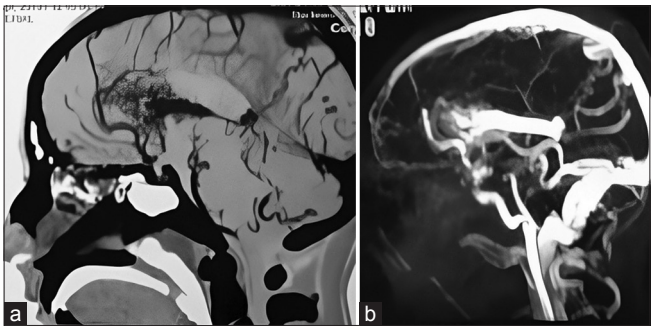

Figure 1: Pre-operative (a) computed tomography angiography with early venous phase and (b) magnetic resonance venography (sagittal views) showing spetzler-Martin Grade 4 arteriovenous malformations of the corpus callosum, located at genus and rostrum of corpus callosum, with extension into the third ventricle. The feeders are visualized coming from the anterior pericallosal artery and a very large deep draining vein terminating at the vein of Gallen is seen.

Postoperatively, the patient was conscious with no neurological deficits. She developed a 1-week history of high-grade fever, which after the exclusion of infectious etiologies, was deemed to be secondary to an anterior hypothalamic injury caused by intraoperative manipulation. The patient progressed as expected and was discharged on day 10. Post-operative computed tomography angiography confirmed complete resection of the AVM with no residual lesion [Figure 2]. The anti-epileptics were tapered to be ceased at 6 months. At the 18-month follow-up visit, the patient remained seizure-free with no treatment and the magnetic resonance imaging showed no new findings.

DISCUSSION

CC-AVMs are relatively rare lesions, accounting for 15% of all surgically resected AVMs.[5,6] Yasargil *et al*. described four groups of CC-AVMs according to their position in relation to the corpus callosum. The first group – to which our case belongs – involves lesions affecting the genu or the anterior part of the corpus callosum. The main arterial feeders of anterior CC-AVMs are branches of the anterior cerebral artery, including the pericallosal arteries. Venous drainage typically involves the inferior sagittal sinus through the callosal and septal veins or Galen veins through the internal cerebral veins. The other three groups include lesions affecting the body, those involving the posterior third of the body or splenium, and holocallosal AVMs. CC-AVMs are most commonly found in the splenium, followed by the body, and genu, with occurrences of 43%, 31%, and 23%, respectively.[5,6]

Picard *et al*.[6] further categorized CC-AVMs into three types based on nidal architecture (a) the "compact" nidus, which is well-defined and restricted to the CC, (b) the "extensive" nidus involving the cingulate gyrus or septum pellucidum along with the corpus callosum, and (c) the "diffuse" nidus, which is poorly demarcated and includes various cortical, subcortical, and intraventricular regions. CC-AVMs with intraventricular extension are notoriously challenging to resect, often requiring drastic procedures, and risking catastrophic neuro-psychological sequelae. In the present case, the nidus showed a remarkable extension into the third ventricle and can thus be defined as "diffuse." According to the supplementary Spetzler-Martin AVM grading scale, the AVM of our patient was a grade six, denoting high procedural risk. Furthermore, the nidus was diffuse and lay adjacent to eloquent areas, further increasing the risk.

Despite the advancements in endovascular and stereotactic radiosurgical treatments, microsurgical excision remains the

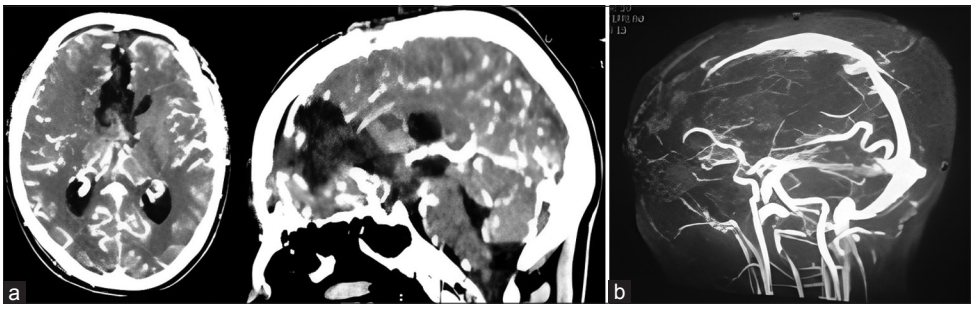

Figure 2: (a) Post-operative computed tomography angiography and (b) a magnetic resonance venography show total resection of the arteriovenous malformations of the corpus callosum with intact surrounding vasculature.

gold standard, offering the highest cure rate and excellent clinical outcomes for both ruptured and unruptured AVMs. In addition, microsurgery provides optimal protection against hemorrhage for low-grade brain AVMs.[7] Typically, a multi-modal treatment approach is advocated. The patient in this case necessitates a more thorough obliteration of the AVM. Opting for an endovascular procedure may result in partial obliteration, thereby heightening the risk of re-bleeding, which is not viable for this patient due to the associated increased mortality risk.

Microsurgical resection of CC-AVMs is difficult due to the deep-seated position of these lesions, their connection to paraventricular structures, and their complex vascular supply, including arterial feeders typically recruited from both anterior and posterior circulation and deep venous drainage. In this case, the draining vein was particularly enlarged and extended deeper, following the course of the internal cerebral vein. Microsurgical circumferential dissection of the nidus proceeded along the route of the vein, requiring a further alteration of the previously planned inter-hemispheric approach to an expanded anterior trans-callosal approach up to the middle of the third ventricle roof at the level of massa intermedia.

Neuroendovascular therapy is frequently considered as a preoperative adjunct to occlude arterial flow through the nidus thus enabling complete surgical resection.[8,9] In our case, the AVM's diffuse nature and the unavailability of neuroendovascular facilities across the country rendered embolization an unfavorable option. Stereotactic radiosurgery, as a stand-alone therapy or as a way to downstage the lesion before surgery, has shown promising results in small nidal size of <3 cm.[10] Given its non-invasive nature, radiosurgery is an enticing technique with a propensity to avoid various potentially crippling complications. However, long treatment courses of 2–3 years are often required to obliterate the AVM completely. In the present case, the patient experienced an increase in the AVM size, followed by a re-hemorrhage during her course of radiosurgery, necessitating definitive surgical intervention.

Microsurgical resection, combined with endovascular embolization or radiosurgery, is the cornerstone of AVM management. However, for CC-AVMs, the precise radio-anatomic and patient-specific parameters that guide treatment decisions remain controversial.[8] Nevertheless, the conclusions of this review were informed by the available literature, consisting mainly of case reports, case series, and retrospective cohort studies, with no prospective cohort or randomized clinical trials.

CONCLUSION

Deep-seated high-grade AVMs represent a surgical challenge, with each lesion processing its unique characteristics. We presented a rare case of complete resection of an anterior CC-AVM with a diffuse nidus without new neurological deficits. The paper highlights the challenging nature of these lesions and the importance of personalized surgical planning and meticulous technique in achieving good surgical outcomes.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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