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SNI: Unique Case Observations

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Case Report

Awake resection of a right motor cortex arteriovenous malformation in a pediatric patient: A case report and review of the literature

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Received: 16 March 2024
Accepted: 13 November 2024
Published: 06 December 2024

DOI
10.25259/SNI_192_2024

Quick Response Code:

ABSTRACT

Background: Intracranial arteriovenous malformations (AVMs) are extremely rare in the pediatric population, with an estimated prevalence of 0.014–0.028%. About 75–80% of pediatric AVMs present with intracranial hemorrhage, a source of significant morbidity and mortality. Awake craniotomy (AC) has become the standard approach for resecting eloquent area intracranial lesions in the adult population. Its use, however remains limited in the pediatric population and has very rarely been reported for an AVM of the motor cortex in this age group.

Case Description: We report the case of a 17-year-old, right-handed boy who presented to our setup with a 2-month history of left-sided hemiparesis and left facial hypoesthesia following an episode of acute loss of consciousness (ALOC) while playing football. A computed tomography scan done after ALOC revealed an AVM in the right frontoparietal cortex with associated acute hemorrhage. Digital subtraction angiography (DSA) was done which revealed a right-sided grade II AVM with arterial supply from the right middle cerebral artery and venous drainage into the superior sagittal and cavernous sinuses. The patient underwent elective neuronavigation-guided right frontoparietal AC and resection of AVM. Postoperative DSA revealed no residual disease. The patient's neurologic deficits showed improvement in the first few days following surgery. He was discharged with advice to follow up in a neurosurgery clinic to monitor his postoperative recovery and ensure compliance with physiotherapy.

Conclusion: This case represents only the second pediatric patient in the available medical literature to have ever undergone AC for intracranial AVM resection. Pediatric AVMs are a rare entity and pose the risk of significant morbidity and mortality. Awake surgery has the potential to reduce iatrogenic neurological deficits in the pediatric population significantly. More work must be done to increase pediatric patient compliance with awake surgery.

Keywords: Arteriovenous malformation, Awake craniotomy, Pediatric arteriovenous malformation (AVM)

INTRODUCTION

Intracranial arteriovenous malformations (AVMs) are extremely rare in the pediatric population, with an estimated prevalence of 0.014–0.028%.[1] They do, however, carry the potential to cause excessive morbidity and mortality.[2] Approximately 75–80% of pediatric AVMs present with intracranial hemorrhage, a figure much higher than the 50–65% frequency of adult AVMs presenting with hemorrhage.[5]

Awake craniotomy (AC) is a surgical technique wherein the patient being operated on remains awake during the procedure to allow his/her neurologic function to be monitored.[9] It is utilized in cases where lesions lie in or near eloquent brain regions.[1] In addition to reducing the risk of iatrogenic neurologic injury, AC helps avoid the physiologic disturbances and postoperative nausea and vomiting often associated with general anesthesia (GA).[9] Despite being accepted as a standard of care in adult patients, AC is still seldom utilized in the pediatric population.[1] In this article, we review the case of a pediatric patient with a right motor cortex AVM who underwent awake resection. According to the available medical literature, this is only the second pediatric patient in history to have undergone AC for intracranial AVM resection.[1]

CASE DESCRIPTION

A 17-year-old right-handed boy with no prior known comorbidities presented to the emergency room with complaints of left-sided hemiparesis and left facial hypoesthesia following an episode of acute loss of consciousness (ALOC) while playing football. A computed tomography (CT) scan head without contrast and a CT angiogram were done, which revealed an AVM in the right frontoparietal cortex with associated acute hemorrhage. The patient was conservatively managed with blood pressure control and neuromonitoring. He was subsequently discharged with advice to initiate physiotherapy and follow-up in the neurosurgery clinic for digital subtraction angiography (DSA) and possible planned elective AVM resection after the acute hemorrhage resolved. A decision was made to delay the DSA until the time of surgery to allow preoperative embolization to be done alongside it.

A month after the ALOC, elective DSA was performed, which revealed a right-sided grade II AVM with arterial supply from the right middle cerebral artery (MCA) and venous drainage into the superior sagittal and cavernous sinuses [Figure 1]. The patient was thereby planned for interval AVM resection, and it was decided not to perform preoperative AVM embolization as this could result in an MCA branch infarct. After 6 weeks of his intracranial hemorrhage, the patient underwent neuronavigation guided right frontoparietal AC and resection of AVM. Intraoperatively, the AVM was anatomically found at the right inferior frontal gyrus and was surrounded by gliotic tissue. Postoperatively, after ensuring hemodynamic stability, the patient was shifted to the special care unit for neuro-observation. Repeat DSA was performed postoperatively, which revealed no residual disease [Figure 2]. The patient experienced no significant complications in the post-operative period other than nausea, for which anti-emetic medications were optimized, and mild wound dehiscence, for which the defect had to be secured with a stitch.

Neurorehabilitative measures were instituted early on for the patient. His left-sided hemiparesis showed significant improvement in the first few days following surgery, from a Medical Research Council (MRC) muscle strength score of 3/5 in the left upper and lower limbs preoperatively to a score of 4/5 postoperatively. He was thus discharged with advice to follow up in a neurosurgery clinic to monitor his recovery and ensure compliance with physiotherapy. The patient's motor powers continued to improve on regular clinic follow-ups. He exhibited powers of MRC muscle strength score 4+/5 in his left upper and lower limbs in his latest clinic follow-up, allowing him to walk with minimal assistance.

DISCUSSION

Although they are, in essence, congenital lesions,[3] intracranial AVMs are mostly diagnosed in the adult population.[4] The potential for hemorrhage, however, is significantly greater in the pediatric population, which, when combined with the longer life expectancy of children, adds to the morbidity and mortality posed by AVMs in this age group.[5]

The accepted goal of treatment of intracranial AVMs is the complete removal of both the nidus and all of its arteriovenous shunts to eliminate all pathologic angiogenic capacity at the lesion site.[6] There are currently three treatment modalities that can be employed to achieve this purpose: microsurgical resection, stereotactic radiosurgery, and endovascular embolization.[6] Microsurgical resection has been the longest used of the three modalities in the treatment of intracranial AVMs. It remains the first-line therapy due to it possessing the greatest potential to provide a complete cure. However, it does come with the associated risk of causing iatrogenic neurological injury to surrounding brain tissue.[7] Stereotactic radiosurgery offers a non-invasive method of treating brain AVMs; however, its response is rather delayed as the aberrant vasculature takes time to sclerose and involute after radiation to the affected area is applied, and so the risk of hemorrhage remains significant till the AVM persists.[8] Endovascular embolization is mostly employed as an adjunct to surgery and radiation therapy to reduce nidus size before definitive treatment.[7] The chances of achieving complete lesion obliteration are significantly lower with angio-embolization and so it remains mostly an ancillary modality of treatment than the one of choice.[10]

AC has become the standard of care for adult patients with eloquent brain lesions in need of resection;[1] however, it has thus far very rarely been practiced in the pediatric population – in one recent extensive systematic review, a total of 142 pediatric patients could be identified to have undergone

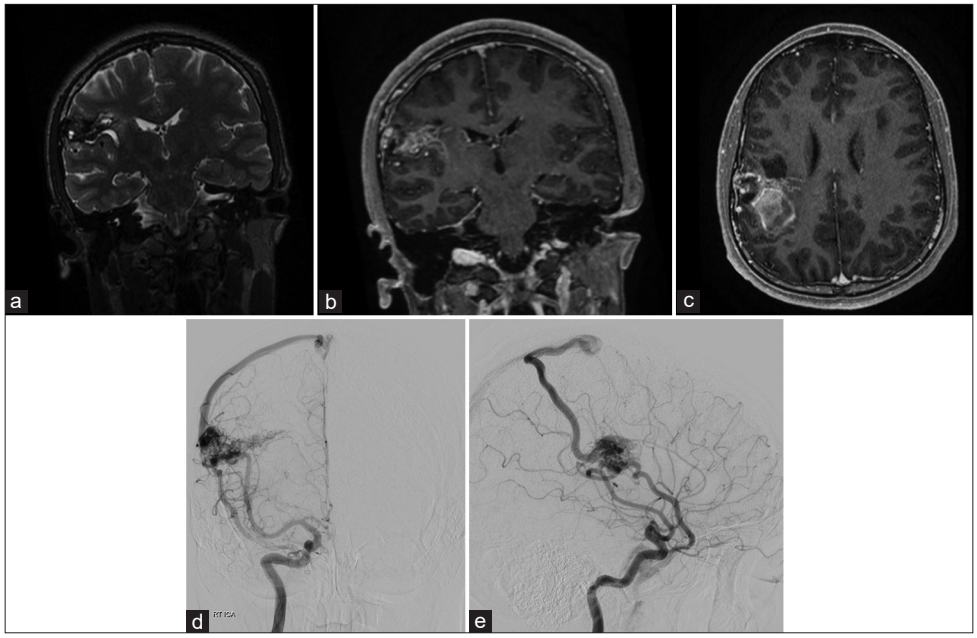

Figure 1: Preoperative imaging of the right frontal arteriovenous malformation. (a) Magnetic resonance imaging (MRI) T2-weighted Coronal view, (b) MRI T1-weighted with contrast, Coronal view, (c) MRI T1-weighted with contrast, axial view, and (d and e) digital subtraction angiography shows the nidus supplied by the right middle cerebral artery and with early draining into the superior sagittal sinus in the arterial phase.

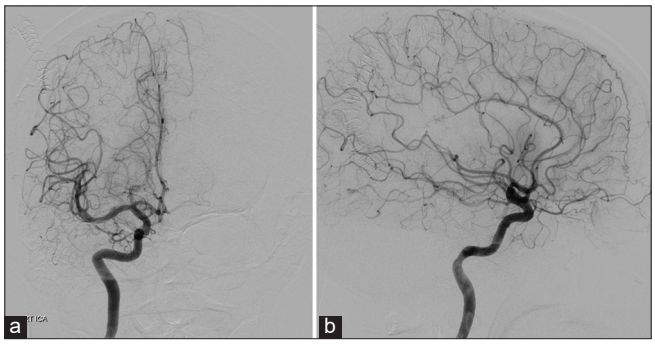

Figure 2: (a and b) Postoperative digital subtraction angiography showing absence of nidus or early draining vein in early capillary/late arterial phase.

AC to date.[1] The indications for AC were as follows: tumor resection (n = 110, 77.46%), seizure/epilepsy (n = 23, 16.20%), insertion of deep brain stimulation electrodes (n = 8, 5.63%), and AVM resection (n = 1, 0.70%).[1] In this review, the youngest age at the time of surgery was identified as 7, while the mean age of the study population was 12.23 years.[1]

The main limiting factor in the application of AC in the pediatric population is the cognitive immaturity of children and the resulting exaggerated stress and anxiety they experience in the operating room setting.[12] The need for conversion from local to GA in the pediatric population, however, does not seem too immense, as reported by a systematic review of pediatric ACs by Bhanja *et al*., who found only four of the 98 cases (4.08%) of pediatric ACs they included in their review ended in conversion to GA with subjective pain, agitation, and discomfort being the main reasons for doing so.[2] Of these four patients, one had to undergo conversion to GA due to a residual tumor in a non-eloquent area that needed GA for optimal resection.[2] Of note, however, is that in the same review, it was found that 19 of 92 pediatric patients (20.65%) found it difficult to perform all monitoring tasks.[2] Nineteen of 98 patients (19.39%) were reported to have developed postoperative complications, including aphasia (n = 4, 4.08%), hemiparesis (n = 2, 2.04%), sensory deficit (n = 3, 3.06%), motor deficit (n = 4, 4.08%), or others (n = 6, 6.12%).[2] Only three of these 98 patients (3.06%), however, continued to experience long-lasting post-operative complications: [2] For one of them, the complication was not strictly neurologic but rather psychologic, as they suffered from major anxiety disorder after surgery.[2] The rest of the 16 patients saw their complications resolve during the immediate postoperative period and subsequent follow-up.[2]

Psychological issues have been studied and reported as major complications following ACs in both adult and pediatric patients.[13] To reduce peri- and intraoperative anxiety and improve patient compliance, Labuschagne *et al*., report using simulated theater experiences to introduce pediatric

patients to the surgical experience, explore their limitations to complying with the monitoring protocols, and tailor the experience to suit their individual needs.[11]

To combat intraoperative anxiety and resulting non-compliance in our case, we ensured the team member responsible for intraoperative neuromonitoring established a good rapport with the patient before the procedure and continuously kept speaking to the patient on topics of his interest during the procedure while monitoring for any deficits. Doing so ensured the patient remained at ease and was willing to follow the neuromonitoring protocol throughout his surgery. Our patient was, however, at the older end of the pediatric age spectrum, so the strategy we employed to make the AC experience more comfortable for him may not be equally applicable to younger patients who, despite having a member of the surgical team by their side to keep them calm during the procedure, might still find the operating room environment too stressful and discomforting to be able to comply to all the requirements and demands of intraoperative neuromonitoring. It is thus warranted to invest in efforts to preoperatively condition pediatric patients scheduled to undergo ACs to the operating room environment and, in the process, perhaps even modify the environment to ensure better intraoperative comfort for and conformity from the patient. More nuanced and subjective strategies, tailored to individual patients' requirements, might be of benefit in this regard.

CONCLUSION

Intracranial AVM is a rare pediatric pathology that bears massive potential for morbidity and mortality. Surgical resection remains, to date, the gold standard to achieve complete resection of the lesions. AC is gradually becoming the standard for eloquent brain lesion resection. In contrast to its common use in adult patients, AC remains seldom utilized in the pediatric population, with intraoperative stress and anxiety being significant limiting factors to compliance with AC in children. More work needs to be done to explore ways to make the AC experience less stressful and more bearable for pediatric patients.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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How to cite this article: Nadeem S, Gujrati A, Mubarak F, Khan A, Enam S. Awake resection of a right motor cortex arteriovenous malformation in a pediatric patient: A case report and review of the literature. Surg Neurol Int. 2024;15:453. doi: 10.25259/SNI_192_2024