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CASE REPORTS

Awake craniotomy for eloquent pial arteriovenous fistula: anesthetic and surgical consideration of a rare case

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Abstract Intracranial pial Arteriovenous Fistulas (AVFs) are rare cerebral vascular lesions composed of one or more arterial connections to a single venous channel. We report a 50-year-old patient with a ruptured pial AVF located in an eloquent area. Microsurgery for pial AVF occlusion was proposed with awake craniotomy for motor function and neurological evaluation. Awake craniotomy is a technique that is especially useful for cerebral vascular lesions in eloquent areas, where an occlusion often compromises or suppresses the blood supply, culminating in ischemia with consequent transient or definitive deficits in neurological function.
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Introduction

Cerebral vascular lesions are associated with high morbidity and mortality rates, especially when they rupture.[1] Subarachnoid hemorrhage after aneurysm rupture is the most catastrophic vascular manifestation. However, there are other lesions that may be associated with severe neurological impairment, such as intracranial Arteriovenous Fistulas (AVFs). Pial AVFs are rare vascular lesions, and approximately 150 cases have been described.[1] They are high-flow vascular malformations that present with a high rupture risk, especially after an initial hemorrhage, and poor neurological outcomes.

Brain hemorrhage at an atypical site in young patients may be a presentation of this disease. Treatment usually involves a neurosurgical approach. Comprehensive knowledge of the brain anatomy before and during the procedure is essential to achieve good results. Neurological monitor-

Abbreviations: AVF, Arteriovenous fistula; CT, Computed tomography; ICG, Indocyanine green.

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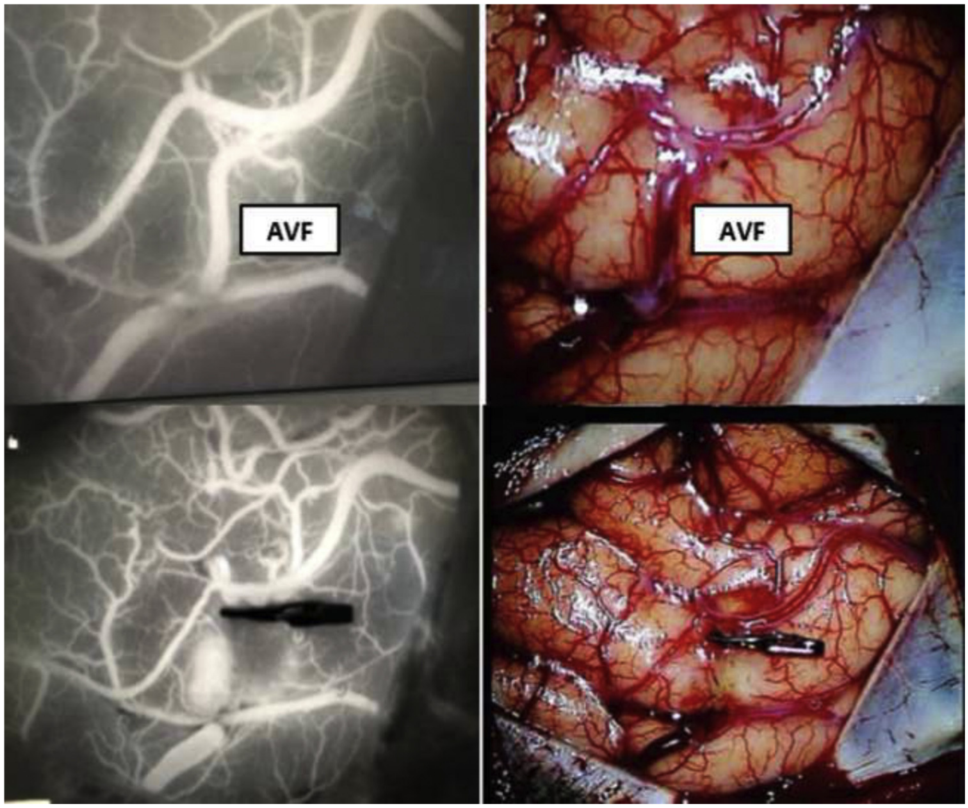

Figure 1 A, Left/superior: intraoperative videoangiography. B, Right/superior: microsurgical view under magnification of the AVF. C, Left/inferior: intraoperative videoangiography with ICG after AVF clipping. D, Right/inferior: microsurgical view after AVF clipping (use of the images authorized by the patient through informed consent). AVF, Arteriovenous Fistula; ICG, Indocyanine Green.

ing may be performed using electrophysiology methods, and awake craniotomy improves the results. Collaborations between neurosurgeons and anesthesiologists are crucial for performing an effective procedure. In the present report, we describe a unique case of a pial AVF that was successfully treated through awake craniotomy.

Case report

A 50-year-old woman presented to the emergency department with the sudden onset of a holocranial headache and mild left hemiparesis (grade 4) associated with a change in superficial sensitivity in the left dimidium with spontaneous regression 5 days after the event. She received analgesic drugs and underwent brain Computed Tomography (CT) and cerebral angiography, which revealed a small-volume intracranial hemorrhage that did not require drainage. During the investigation, complementary examinations were performed (brain resonance imaging, angioresonance, and digital angiography), and the presence of a right frontal pial AVF was confirmed.

Thus, we opted to perform elective surgery while the patient remained awake to continuously assess motricity and sensitivity, since after the occlusion of an AVF, ischemia and acute neurological deficits may occur. There is evidence reported in the literature that during the temporary occlusion of vascular lesions, deterioration in neurological function may occur without concomitant changes in neurophysiological parameters. This information corroborates the indication for awake surgery in cases like the one described.[2]

After occlusion of the AVF using a titanium clip (Fig. 1), we performed motor and sensitive tests on the left dimidium for 15 minutes through active mobilization of the limbs by the patient. In addition, verbal fluency and attention were continuously assessed during the procedure. Vessel patency was evaluated based on the visualization of normal flow after intravenous Indocyanine Green (ICG) administration.

This strategy allowed for the definitive occlusion of the fistula with safety, ensuring the preservation of motor and sensitive functions, and the patient did not develop any neurological sequelae. The duration of the awake procedure was approximately 50 minutes, with a total operative time of approximately 150 minutes. There were no changes in the motor and sensitive evoked potentials. The patient was monitored using electrocardiography, blood oxygen saturation measurements, bispectral index assessments, capnography, and non-invasive blood pressure measurements every 5 minutes during the asleep phases and every 30 minutes during the awake phase. The anesthesiologist placed a venous line (20G catheter) in the forearm of the patient. The patient was comfortably positioned.

The first phase of the procedure (asleep phase 1) was performed with previous oxygenation for 5 minutes, followed by an intravenous dose of 1% lidocaine (0.5 $mg.kg^{-1}$) to reduce or prevent pain at the injection site. Anesthetic induction with propofol (6–8 $\mu g.mL^{-1}$) and remifentanil (4 $ng.mL^{-1}$) was performed, followed by the introduction of a

Figure 2 Awake phase: verbalization (use of the image authorized by the patient through informed consent).

laryngeal mask tailored to the patient. The ventilator was set in a pressure-controlled mode, and the patient received mechanical ventilation. Subsequently, the doses of propofol and remifentanil were immediately reduced to 4 μg.mL^{-1} and 1.5 ng.mL^{-1}, respectively. A new venous site of injection was set using an 18G catheter.

The patient received a urinary catheter, prophylactic antibiotics (cefuroxim), and drugs to control her symptoms (ondansetron, dexamethasone, dipyrone, pantoprazole) according to her body weight.

Subsequently, the anesthesiologist performed a scalp block with 1% lidocaine with no epinephrine and 0.5% bupivacaine with epinephrine of the supraorbital, supratrochlear, major occipital, zygomaticotemporal, and auriculotemporal nerves bilaterally. A total dose of 150 mg of lidocaine plus 75 mg of bupivacaine was used in 50:50 dilutions. After all the surgical preparation, the procedure surgery was initiated.

After craniotomy and duramater exposure, duramater was soaked with a gauze containing 1% lidocaine (100 mg) for 15 minutes, and the venous anesthesia was disrupted.

Once the patient exhibited an increase in the BIS index (up to 70), or to the awake level, the laryngeal mask was removed, and we initiated the second phase of the procedure: awake (Fig. 2).

The neurophysiologist then continuously monitored the neurological function, helping the surgeon to clip the fistula safely. The patient was awake, with good cognition, without pain, and very collaborative.

After complete clipping of the fistula and neurological evaluations, the patient was submitted to the third phase of anesthesia, asleep phase 2. New induction was performed with propofol and remifentanil, the laryngeal mask was easily reintroduced with the patient positioned with cranial pins, and the patient received mechanical ventilation until completion of the procedure. The patient had an estimated bleeding volume of less than 100 mL. Once completed, the patient was awakened again, not reporting pain or other symptoms. She was admitted to the intensive care unit for 12 hours and then to a hospital bed for 24 hours, being discharged after 36 hours with excellent recovery and the absence of neurological deficits.

This case report describes for the very first-time surgical treatment for a pial AVF while the patient was awake.

Discussion

Pial AVF are rare vascular lesions, and approximately 150 cases have been described[1] that required prompt treatment aiming to minimize neurological sequelae. Based on a review of the current literature, the use of awake surgery for the treatment of AVF has not previously been reported.[2] AVFs may be acquired traumatically, iatrogenically, or congenitally. They are typically classified as a subtype of pial arteriovenous malformations; however, they are differentiated by their lack of an intervening nidus between the arterial feeder and venous drainage.[1] Due to their unfavorable natural course, conservative management is associated with a mortality rate of up to 63%.[3] One of the most common presentations is intracranial hemorrhage,[4] such as in this case.

Owing to their high mortality rate, despite being rare lesions, pial AVFs must be considered as a differential diagnosis in young patients with intracranial hemorrhage in atypical topography. Intraoperative resources such as intraoperative videoangiography using ICG and intraoperative monitoring should be used to increase therapeutic success, preventing neurological complications and the need for further surgeries.

Awake craniotomy for lesions in eloquent area is a well-established procedure in the field of neuro-oncology[5]; however, this method is not widely used for vascular lesions.[3] Intraoperative monitoring of motor evoked potentials is a valuable resource that guides neurosurgeons; however, in some cases, it may be insufficient to prevent neurological motor deficits because electrical conduction may be imprecise. When a patient is awake, a neurological examination may be performed in real time to improve the success of the procedure. Neurosurgeons, neuroanesthesiologists, and neurophysiologists must be in perfect synchronism during awake surgical procedures.

In vascular malformations, the anatomy might be distorted, and a neurosurgeon may not know if a manipulated vessel contributes to the supply of eloquent regions, and every method that aim to minimize sequelae is valuable and should be performed by a trained multidisciplinary team, such as awake craniotomy. Good relationships among the team are mandatory for such delicate procedures because collaboration during the awake phase is essential to the success of the surgery.

However, patients who undergo appropriate neuropsychological and motor evaluations and have good relationships with the team may undergo this procedure without encountering complications.

Conclusion

Advanced resources for surgery for tumors in eloquent areas through awake craniotomy are well described in many papers. Appropriate evaluations of patients before these procedures and collaborations among neurosurgeons and anesthesiologists are mandatory. Nevertheless, awake craniotomy remains less used for vascular lesions. Pial fistulas in eloquent areas are rare and may evolve with severe and disabling neurological impairments, especially concerning speech and motricity. This is the first report of successful awake craniotomy for a peri-rolandic fistula wherein advanced neuroimaging methods such as with ICG administration were implemented with excellent results. The use of these resources should be encouraged for brain vascular lesions.

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Statement

All data of the patient in this report case was used just after application of the informed consent form and approval from ethical committee.

Conflicts of interest

The authors declare no conflicts of interest.

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