Case Report

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Rare clinical presentation of a ruptured spinal bulbomedullary arteriovenous malformation: a case report

Daniel A. Vega-Moreno, MD[a], Rodolfo P. Molina-Martínez, MD[a], Yolanda Aburto-Murrieta, MD[b], Beatriz Méndez-González, MD[b], Gervith Reyes-Soto, MD[c], Mónica Serrano-Murillo, MD[c], Ulises García-González, MD[a], Bipin Chaurasia, MS[d,*]

Introduction and importance: Spinal arteriovenous malformations (AVMs) are a rare condition that has a high risk of bleeding and complications. The authors present the case of a spinal arteriovenous malformation in an unusual location and presentation.
Case presentation: A 67-year-old man with subarachnoid hemorrhage due to a ruptured spinal arteriovenous malformation type IVa, with associated bulbomedullary aneurysm, which was managed conservatively due to the high risk of complications and mortality.
Clinical discussion: Spinal AVMs have had different management and treatments over the years, so conservative management remains an option when arterial cannulation is complex and surgery carries a high risk of complications.
Conclusion: Due to the high risk of complications of surgery in this location, conservative treatment is an option for the management of such cases with good outcomes.

Keywords: craniovertebral junction AVM, medullary AVM, spinal AVM

Introduction

Spinal arteriovenous malformations (AVMs) are a heterogeneous group of abnormalities developed from spinal, radicular, or medullary blood vessels, and they are associated with a high risk of hemorrhage and mortality. These can be classified depending on the vascular characteristics and angiographic appearance in grades I, II, III, and IV, with grade IV subdivided into a, b, and c depending on the vessels involved and the characteristics of the venous drainage system[1]. The clinical presentation of these malformations is very variable and will depend on the location. However, it has been described that they can present with some motor or sensory syndrome or even, in rarer cases, a subarachnoid hemorrhage[2]. These malformations are treated using endovascular therapy and sometimes surgical treatment.

HIGHLIGHTS

- Spinal arteriovenous malformations are a rare condition, that have a high risk of bleeding and complications.
- We present the case of a spinal arteriovenous malformation of unusual location and presentation.
- A 67-year-old man with subarachnoid hemorrhage due to a ruptured spinal arteriovenous malformation type IVa, with associated bulbomedullary aneurysm, which was managed conservatively due to the high risk of complications and mortality.

Although embolization can be considered the first treatment option due to its location, it entails several risks and technical difficulties[3]. We present the atypical case of a 67-year-old man with spontaneous subarachnoid hemorrhage secondary to rupture of a bulbomedullary IVa spinal arteriovenous malformation that was managed conservatively.

Case report

A 67-year-old man presented to the emergency department with neck pain and headache of sudden onset. The Visual Analog Scale of pain was 10/10, accompanied by vomiting on two occasions; minutes later, dysarthria and sudden loss of awake state were added. He was admitted with a 3-point Glasgow, with preserved brainstem reflexes except for gag or cough reflexes, without motor or sensory response in the four limbs, so emergency endotracheal intubation was decided. Given the clinical suspicion and diagnosis of a probable subarachnoid hemorrhage, it was

[a]Department of Neurosurgery, Hospital Central Sur de Alta Especialidad, PEMEX, [b]Neurological Endovascular Therapy Department, Instituto Nacional de Neurología y Neurocirugía 'Dr. Manuel Velazco Suarez', [c]Neuro-oncology Department, National Institute of Oncology, Tlalpan, Ciudad de México, CDMX, Mexico and [d]Department of Neurosurgery, Neurosurgery Clinic, Birgunj, Nepal

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*Corresponding author. Address: Department of Neurosurgery, Neurosurgery Clinic, Birgunj, Nepal. Tel.: +97 798 454 54636. E-mail: trozexa@gmail.com (B. Chaurasia).

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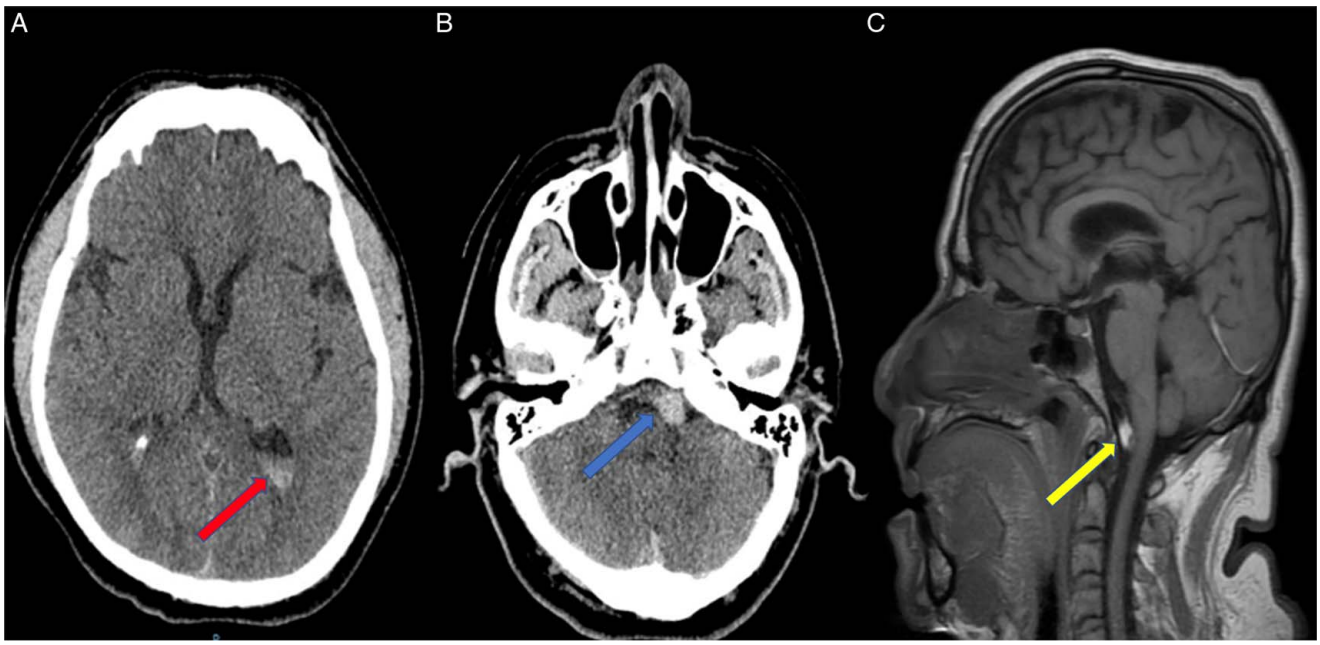

Figure 1. (A) Red arrow. Presence of blood density in the left and right occipital horn of ventricular system. (B) Blue arrow. Dense clot on the premedullary cistern and medullary-cerebellum cistern. (C) Yellow arrow. Sagittal MRI in T1 sequence where hyperintensity is observed in the bulbomedullary junction.

decided to perform a simple tomography at the beginning and, subsequently, MRI (Fig. 1).

Given the diagnosis of Fisher IV subarachnoid hemorrhage, with an image of a dense clot in the prepontine and medullary cisterns, it was decided to perform a diagnostic cerebral angiography of six vessels (Fig. 2).

During the angiography procedure, multiple attempts were made to cannulate the feeding artery of the arteriovenous malformation. However, due to the minimal caliber of the vessel and the imminent risk of vasospasm and thrombosis of the anterior spinal artery, it was decided not to embolize the AVM and stop the angiographic study. In addition, due to the location in the bulbomedullary junction and the severe clinical conditions of the patient, it was decided not to perform neurosurgical management either due to the high risk of complications. The patient was managed with early rehabilitation during his hospitalization. He was discharged 43 days after his admission, with tracheotomy and gastrostomy, with a global strength of 4/5 on Daniel's scale

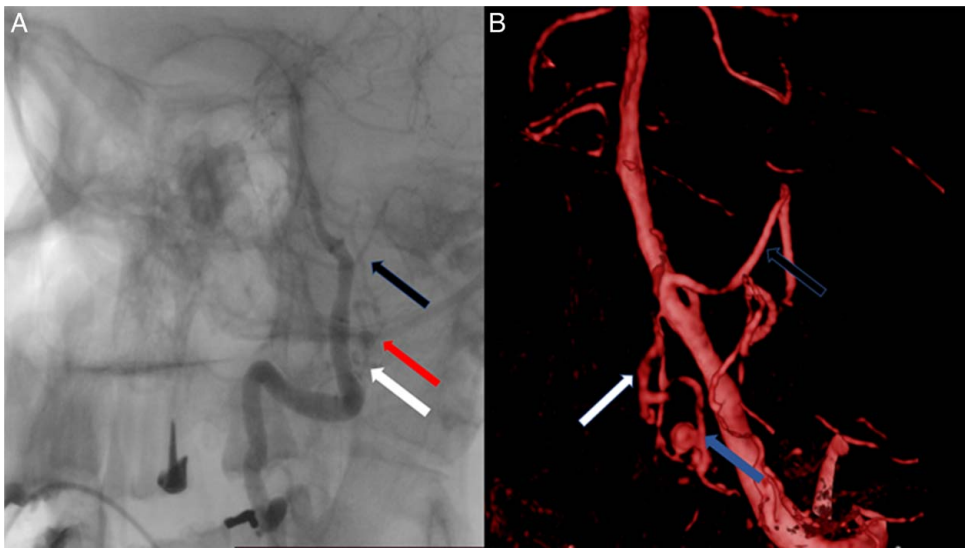

Figure 2. (A) Cerebral angiography with selective shot of the left vertebral artery. White arrow. Feeding arterial branch dependent on anterior spinal artery. Red Arrow. Aneurysm associated with the AVM. Black arrow. Deep venous drainage by single drainage vein. (B) 3D reconstruction. Black arrow. Posterior inferior cerebellar artery. White arrow. Anterior spinal artery. Blue arrow. AVM-associated aneurysm.

and sensory deficit recovery, with a Glasgow Outcome Score of 3. The patient has adequate neurological functional recovery, with preserved strength for walking, and is being followed up with serial imaging studies.

This case report was written under the Surgical CAse Report criteria. All the work regarding this case report has been reported per the Surgical CAse Report 2023 criteria[4].

Discussion

Spinal AVMs represent 3–4% of all spinal cord lesions[5]. These have presented different classifications over the years; all of these classifications are based on vascular characteristics, their location within the extraintradural space, whether it is intramedullary or peri medullary, and the origin of the feeding arteries. In addition, the transition from an artery to a vein can have its angioarchitecture origin as a network (vascular nidus) or can be direct (fistula)[6]. The clinical presentation can be presented in two types: those that present acutely (complete or incomplete spinal cord syndrome, subarachnoid hemorrhage) or those that present progressively (myelopathy, pain, or radicular syndrome), also being diagnostic as an imaging finding, although less frequently[7]. Typically, spinal AVMs are located in the lower thoracic, lumbar, or conus medullaris. The cervical location is quite infrequent; also, due to the high pressures that the cervical vascular system handles, the presentation as hemorrhage is more frequent than in other areas, being practical, in most cases, right arterial branches coming from the vertebral artery, the participants in the malformation and rupture[8].

Regarding the treatment of malformations, spontaneous closure of AVMs is extremely rare, although it has been reported. Therefore, treatment is based on endovascular management (minimally invasive) or surgical treatment. Regarding surgical treatment, laminectomy plus resection of the AVM via a posterior approach is the one that presents the best results. Those in the anterior medullary region are especially difficult[9,10]. Regarding endovascular treatment, embolization of the vascular nidus and the proximal portion of the drainage vein is necessary. Because of the pure embolization of the main feeding artery carries the risk of recurrence and rupture due to changes in vascular flow[11]. Although endovascular treatment has advanced in many areas, the spinal vasculature has a different architecture than the cerebral vessels, with spinal vessels being smaller and more likely to be accidentally obliterated[12]. Therefore, considering that the lesion was located right at the junction of the medulla oblongata and the spinal cord, the risk of vasospasm of the anterior spinal artery or migration of the embolization material was very high, so we decided not to give invasive this time. In this case, the decision not to offer more treatment was the one considered because it was not possible to advance the guide beyond the vascular nidus due to the minimal caliber of the vessels, in addition to being a single nutrient artery and of great importance such as the anterior spinal artery, it was decided not to continue with the procedure. In the study by Young-Jun Lee *et al*., published in 2014, 44 patients with spinal AVMs of various locations underwent different treatment modalities. There were five patients for whom conservative management was decided, none having deteriorated in a 7-year follow-up. However, the rate of deterioration or complications for those who underwent endovascular treatment was 26%; for surgical treatment, it was 0%; and for conservative management, it was 0%. Although the characteristics of the patients guided to conservative management are not specified, it is clear that the decision not to intervene in a patient is based on the patient's conditions, quality of life, comorbidities, and risk of complications[13].

On the other hand, although radiosurgery for treating these lesions has shown controversial results in some series, some with favorable clinical results, no angiographic cure has been found in such cases. Therefore, considering our case's complex and delicate location, we do not consider this treatment modality acutely due to the high risk of complications and permanent deficit[14]. The anterior spinal artery irrigates two-thirds of the spinal cord, so an infarction in the bulbomedullary junction, specifically of the anterior spinal artery, is considered a severe lesion with high rates of disability and mortality[15,16]. Therefore, surveillance is the best option in this case.

Conclusions

We show an unusual location of a type IVa spinal AVM. The clinical presentation, in this case, was infrequent since subarachnoid hemorrhage in the occipital horn was a rare location due to a rupture of a spinal AVM, in this particular case and due to a single feeding vessel derived from the anterior spinal artery, which, due to the risk of bulbomedullary infarction and patient comorbidities, was decided to manage conservatively.

Ethical approval

This article was written for academic purposes according with the privacy agreement with the patient, who was informed and signed the consent form in agreement.

Consent

Written informed consent was obtained from the patients for publication and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

All the authors contributed to the investigation, development and writing of this article.

Conflicts of interest disclosure

The authors declare no conflicts of interest.

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