CASE REPORT

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A young male with spontaneous ruptured cerebral arteriovenous malformation

Vitorino M. Santos,¹,²* Leandro P. Flores,³ Isabella A. Barros,¹ Priscilla S. Faria,¹ Carolina P. Lamounier,¹ Renata A. Casasanta¹

ABSTRACT

Background: Cerebral arteriovenous malformation (AVM) is considered a congenital condition characterized by arterial-venous connections in the absence of intermediate capillaries with circulatory and functional changes, inclusive on the cerebrospinal fluid (CSF) dynamics.¹,²

Case: A previously healthy young man with abrupt onset of intense headache followed by incoercible vomiting and generalized tonic-clonic seizures. On admission, he was in Glasgow scale IV with respiratory gasping, signs of decerebration and anisocoric mydriasis, and right paresis. Computed tomography showed left intraparenchymal hematoma and intraventricular blood. The abnormal vessels and blood clots were removed by neurosurgery. In early postoperative phase a large cerebrospinal fluid leak developed, and was aspirated and further controlled. The patient had respiratory complications, including pulmonary infection by Pseudomonas.

Conclusions: After longstanding hospitalization, he was referred to other medical institution and home care.

Keywords: Brain arteriovenous malformation, cerebral hemorrhage, cerebrospinal fluid


INTRODUCTION

Cerebral arteriovenous malformation (AVM) is considered a congenital condition characterized by arterial-venous connections in the absence of intermediate capillaries,¹,² with circulatory and functional changes, inclusive on the cerebrospinal fluid (CSF) dynamics.³ Hence, brain edema, mass effect, and hydrocephalus may occur even in non-ruptured AVM.⁴ Hemorrhage is the most common initial manifestation, found in up to 77% of affected people; other frequent clinical features are headache, seizures, and progressive neurological deficits.¹,³

Worthy of note, the annual rate of cerebral hemorrhagic episodes may be up to 4%; moreover, AVM represent one major cause of intracranial bleeding in younger groups of individuals.¹,³ The diagnosis characterization depends on high suspicion index and of brain imaging studies; computed tomography (CT) and digital subtraction angiography have been the best tools.¹,³

The current management of cerebral AVM includes isolated or combined microsurgical resection, stereotactic radiosurgery, and embolization, in addition to watchful expectancy.¹,³ Each individual option should be based on the size, location and morphology of the lesions; and lifestyle, general health, and life expectancy of the patient, and inherent treatment risk.¹,³ As a whole, early intervention is the key of success in patients with ruptured cerebral AVM.⁴

The purpose of the present case report is to stimulate further studies to increase the knowledge about natural history, involved risks and outcome of current AVM management.¹,³

CASE REPORT

A 21-year-old Brazilian male previously healthy was admitted on the Emergency Ward because of intense holocranial headache since three hours before, evolving with incoercible vomiting and generalized tonic-clonic seizures barely responsive to intravenous diazepam. On admission, he was unconscious with respiratory gasping, left signs of decerebration, left anisocoric mydriasis, and right paresis. He underwent orotracheal intubation with mechanical ventilation, and received close semi intensive support.

Immediate computerized tomography of the brain revealed left voluminous intraparenchymal hematoma (Figure 1A) that was associated with bilateral intraventricular hemorrhage.

Further neurological procedure disclosed a huge cerebral AVM localized on the left frontal lobe; abnormal vessels were excised; and the clots of the hematoma were drained (Figure 1B). Because the patient was hemodynamically unstable, and was evolving with aspiration pneumonia, his early postoperative period was followed at the Intensive...
CASE REPORT

Care Unit with vasoactive drugs and tracheostomy, in addition to the course of ceftriaxone and clindamycin.

Worthy of notice, a large leak of CSF (Figure 1C) developed on the immediate postoperative phase and was partially controlled by needle aspiration (Figure 1D). After the first week of admission, high fever and return of leukocytosis were observed, and the antimicrobials were changed by piperacillin plus tazobactam, without improvement.

Imaging studies showed sphenoid sinusitis and mastoiditis, and the culture of tracheal aspirate revealed *Pseudomonas aeruginosa* sensible to polymyxin B and amikacin. Hence, the antimicrobial schedule was accordingly modified, and there was slowly progressive improvement in his clinical condition.

The fever was controlled after a week, and antibiotic course was maintained for a total of 21 days. The laboratory controls are showed in Table 1. On Day 30 of admission, in regular clinical conditions, he was transferred to Clinical ward. Brain images of control showed progressive improvement of CSF leak (Figures 1E and 1F). During hospitalization, the patient had vocal rehabilitation and reintroduction of oral liquid diet, in addition to achieve good recuperation of the right sided paresis by physiotherapy.

Currently, he is under specialized follow-up in other medical institution and with home care.

DISCUSSIONS

The young man herein reported presented acute spontaneous bleeding in congenital AVM manifested by headache, vomiting and seizures, which are the most usual features.\(^1,3\) The extension of supratentorial hematoma might explain the severe neurological changes initially observed on hospital admission, commonly related to intracranial hypertension. Morbidity and mortality rates may be up to 40% and 29% in the hemorrhagic group of AVM.\(^3\) However, the immediate neurosurgery procedure in association with the young age and previous normal health of the patient can have played an important role in the favorable outcome, in spite of the remarkable complications.\(^5\) Conspicuous CSF leak was an early-onset event causing extracranial changes, as well as intracranial images mimicking hydrocephalus.\(^2\)

Rossitti (2013) extensively reviewed diverse physiopathology features of increased CSF pressure, which are associated with AVMs, and emphasized the hydraulic hypothesis.\(^2\) It means that chronic increase of CSF pressure near to breakpoint of the pressure-volume curve may worsen the adverse effects on pathophysiological mechanisms related to cerebral AVM.\(^2\) Intracranial hypertension is equal to CSF hypertension, and is due to increased cerebral blood volume secondary to venous hypertension, or increased CSF volume by reduced absorption.\(^2\) Hydrocephalus development might be related to hypertension in deep brain venous

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**Figure 1** (Computed tomography studies). A: image (in D1) of huge intraparenchymal hematoma in the left frontoparietal region. This change was associated with intraventricular hemorrhage and subfalcine herniation; B: images (in D3) of neurosurgical procedure on the left frontal area, and evidence of residual intraventricular clots; C: images (in D12) showing reduction of the intracranial extradural collection in the left frontal area, and presence of bilateral extraaxial collection suggestive of hygroma, in addition to extensive subgaleal collection with suggestive features of seroma; D: control images (in D22) of the intracranial collections, showing that the changes remain stable.

**Figure 2** Computed tomography image of control (in D 62) revealing significant improvement of the postoperative intracranial changes.
system and/or reduced CSF absorption, either in arachnoid villi or cerebral venules and capillaries. Bir et al. evaluated overall outcomes of early interventions done for ruptured AVM with cerebral hematomas in 78 patients submitted to microsurgical resection, and concluded that early intervention is the corner stone to achieve successful results in this patient group. Surgical procedures after 48 hours of the hemorrhagic event were related to poor outcomes, while ages between 11 and 40 years and supratentorial location of AVM had positive effects. Vilalta et al. evaluated 100 adult patients submitted to surgical treatment for cerebral AVM; the mean age was 34 (15-71) years, and 79% of cases were located in supratentorial area. Hemorrhage occurred in 60% of cases, elective surgery was done in 78% of the patients and seven patients had emergency operations; the outcome was considered good in 75% of cases.

Repeated respiratory infections also occurred, including pneumonia by multiresistant Pseudomonas, which required prolonged courses of antimicrobials guided by sensitivity tests. These infectious complications are often detected in patients treated in Intensive Care Units; therefore, his long hospitalization time represented relevant medical and economic burden.

**CONCLUSION**

Cerebral AVM is a developmental disorder of the brain vascular bed, which more often evolve unsuspected and may be associated with variable clinical picture and outcome. Characterization of diagnosis depends on the index of suspicion and brain imaging studies. Since each therapeutic option is accurately selected, surgery is the best way to treat AVM, and timing of the intervention play a significant role in outcomes of secondary hematomas. Case studies may enhance the suspicion index of primary care workers about this condition.

**REFERENCES**