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Case Report

Ruptured Intracranial Arteriovenous Malformation Arising from an Isolated Developmental Venous Anomaly

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ABSTRACT

The evolution of *de novo* Arteriovenous Malformations (AVM) is still unclear. A 46-year-old woman with known drug abuse and a Developmental Venous Anomaly (DVA) in the left basal ganglia diagnosed 18 years previously was admitted with loss of consiousness due to a large intracerebral and intraventricular hemorrhage. Imaging demonstrated an AVM spetzler-martin III at the site of the previous DVA fed by branches of the left middle and posterior cerebral arteries. After CSF drainage and stabilization of the patient, neurosurgical resection of the AVM and hematoma evacuation was performed.

DVAs are considered benign lesions that do not require radiological follow-up. This case indicates that long-standing drug abuse or other risk factors may predispose to DVA transformation into more aggressive lesions.

Keywords: Radiological; Malformations; Anomalies; Angiography; Hemodynamic

INTRODUCTION

The evolution of *de novo* Arteriovenous Malformations (AVM) is still unclear [1]. In contrast to potentially dangerous AVMs, Developmental Venous Anomalies (DVAs) are considered benign and usually asymptomatic lesions, which commonly accompany cavernous malformations [2]. Radiological follow-up is not deemed necessary for pure DVAs, although they are thought to be vulnerable to hemodynamic changes or venous hypertension [3-5].

Drug abuse and particularly long-standing cocaine usage has been associated with vascular malformations and intracerebral hemorrhage [6]. We report a case of a ruptured AVM that developed exactly at the site of a previously diagnosed DVA and this evolution was associated with long-term drug abuse including cocaine.

CASE PRESENTATION

A 46-year-old female patient with a medical history of seizures and long-standing consumption of multiple drugs had been diagnosed with an isolated DVA in the basal ganglia in the vicinity of the internal capsule in 2002. No clinical or imaging follow-up was considered necessary and the patient was routinely followed in the drug substitution program. In addition to methadone the patient consumed other opioids, cocaine and cannabis.

In November 2020, the patient was found unconscious at home and admitted to the ER with the suspicion of intoxication. She was instantaneously intubated and stabilized. The pupils were isocor and no reaction to light was observed. The blood results were positive for opioids, cocaine and cannabis. Computed Tomography (CT) demonstrated an intracerebral hemorrhage with massive intraventricular hematoma and moderate midline shift. CT angiography identified an AVM in the left basal ganglia as the bleeding source. An external ventricular drain was inserted for CSF drainage and ICP control and the patient was transferred to the neurocritical care unit. Digital Subtraction Angiography (DSA) confirmed the AVM Spetzler-Martin grade III with a rather diffuse nidus located in the left basal ganglia with drainage into the internal cerebral veins. Small feeding arteries originated from the lenticulostriatal arteries with main feeders from the left posterior cerebral artery and the Percheron artery. Additionally, an asymptomatic second AVM Spetzler-Martin grade I was detected superficially feeding from the inferior truncus of the right middle cerebral artery with a single draining vein in the sigmoid sinus.

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Following medical stabilization and thorough interdisciplinary discussion the patient was prepared for surgical hematoma evacuation and AVM resection. Surgery was performed uneventfully *via* a transcallosal approach several days after hemorrhage. Postoperative CT and DSA demonstrated adequate hematoma evacuation and complete resection. The patient was extubated one week after surgery demonstrating a right-sided hemiparesis (grade 2/5). She is currently undergoing neurological rehabilitation (Figure 1).

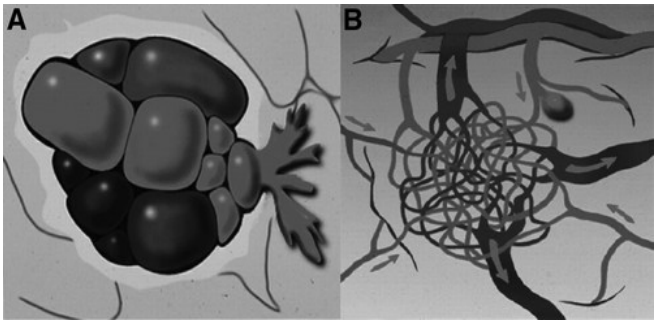

Figure 1: Schematic illustrations of (A) CCM and (B) AVM angioarchitecture.

DISCUSSION

To our knowledge, this is the first description of an AVM development from a DVA associated with polytoxicomania. *De-novo* formations of AVMs are rare and little is known about the pathophysiological mechanisms in these cases.

Hemodynamic factors including high flow rates causing high endothelial shear stress are suggested to promote vascular lesion and AVM formation [7]. Angiogenic factors and especially local overexpression of Vascular Endothelial Growth Factor (VEGF) induce vascular malformation development and increase BBB permeability predisposing for vessel wall rupture [8-11]. Inflammatory cytokines (interleukins, tumor necrosis factor-α), neutrophils and macrophages have been observed as potent stimulators of angiogenesis and BBB disruption leading to AVM development and rupture [12,13].

Drug abuse and alcohol has been associated with neuron inflammation and alterations of the cytoskeletal structures inducing increased Blood Brain Barrier (BBB) permeability. Cocaine is a potent vasoconstrictor and chronic consumption has been observed with ischemic and hemorrhagic strokes, as well as intracerebral hemorrhage [14]. Yin, et al. showed in a rat model the correlation of cocaine and angiogenesis through activation of the HIF-VEGF pathway and microvascular density [15]. Further, cocaine-induced up regulation of proinflammatory cytokines (TNF-α, IL-1 and IL-6) in pericytes and lead to increased expression of proinflammatory markers for autophagy in mice [16]. Therefore, we suggest an association of the cocaine abuse and the AVM formation in this rare case. Presenting with a DVA in 2002 our patient was not followed up radiologically, since DVAs are considered a benign anatomic variant and imaging follow-up is not deemed necessary for pure DVAs. Their vascular structure is composed of dilated draining medullary veins. DVAs can be vulnerable to hemodynamic changes and patho physiologically evolve of the increase inflow through an upstream AVM. DVAs per se should not be treated or obliterated because of the potential risk of the induced venous infarction [17].

CONCLUSION

DVAs are considered benign lesions that do not require radiological follow-up. This case indicates that long-standing drug abuse or other risk factors may predispose to DVA transformation into more aggressive lesions. In these cases repeated imaging may be considered.

DISCLOSURES

The authors declare no conflicts of interest.

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