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Vessel Wall Imaging in Angiogram-Negative Diffuse Subarachnoid Hemorrhage Reveals a Ruptured Lenticulostriate Aneurysm

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A patient presented with acute onset headache and subsequent unconsciousness. The neurologic exam showed left-sided myoclonic jerking and right flaccid hemiparalysis. Noncontrast computed tomography revealed diffuse subarachnoid hemorrhage (SAH) with acute hydrocephalus. Initial digital subtraction angiography (DSA) showed no culprit source for SAH. Repeat DSA on day 7 after initial presentation raised suspicion for left internal carotid artery ophthalmic segment and left lateral lenticulostriate artery (LSA) aneurysms. A magnetic resonance vessel wall imaging (VWI) exam was performed given the presence of multiple potential culprit aneurysms. Vessel wall enhancement around the dome of the left LSA aneurysm suggested rupture, which then facilitated treatment with surgical clipping. LSA aneurysms are exceedingly rare and challenging to treat. Given the associated high degree of morbidity, expedient diagnosis is critical to direct management. VWI could be a valuable tool for detecting ruptured aneurysms in the setting of angiogram-negative SAH.

Key Words: Magnetic resonance imaging; Circle of Willis; Aneurysm; Basal ganglia cerebrovascular disease; Subarachnoid hemorrhage; Cerebrovascular imaging

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INTRODUCTION

Lenticulostriate artery (LSA) aneurysms are rare vascular aberrations with only 112 cases reported in the literature and are associated with tumors or vascular diseases such as hypertension, systemic lupus erythematosus vasculopathy, and moyamoya disease.[1,2] While unruptured aneurysms can be found incidentally, LSA aneurysm rupture causing intracranial hemorrhage or subarachnoid hemorrhage (SAH) is the most common presentation.[3] Ruptured LSA aneurysms are difficult to treat and highly morbid, highlighting the need for expedient diagnosis.[4] Intracranial magnetic resonance vessel wall imaging (VWI) has shown promising utility in detecting angiographically occult ruptured intracranial aneurysms, subtle dissecting aneurysms, or small vascular malformations in the setting of SAH.[5-8] We report a case of computed tomography (CT) and catheter cerebral angiogram-negative SAH from a ruptured LSA aneurysm detected by VWI.

CASE REPORT

A patient called emergency services with an acute onset headache. The patient, a nonsmoker with no past medical history except hypertension, was found unconscious and transported to the emergency department. The neurologic exam showed flaccidity on the right and left-sided myoclonic jerking with an upgoing Babinski sign. Pupils were dilated 5 mm with left gaze.

Head CT revealed diffuse pattern SAH (Hunt-Hess grade 4/modified Fischer scale grade 3 [HH4/mF3]) with acute hydrocephalus (Fig. 1A). The initial computed tomography an-

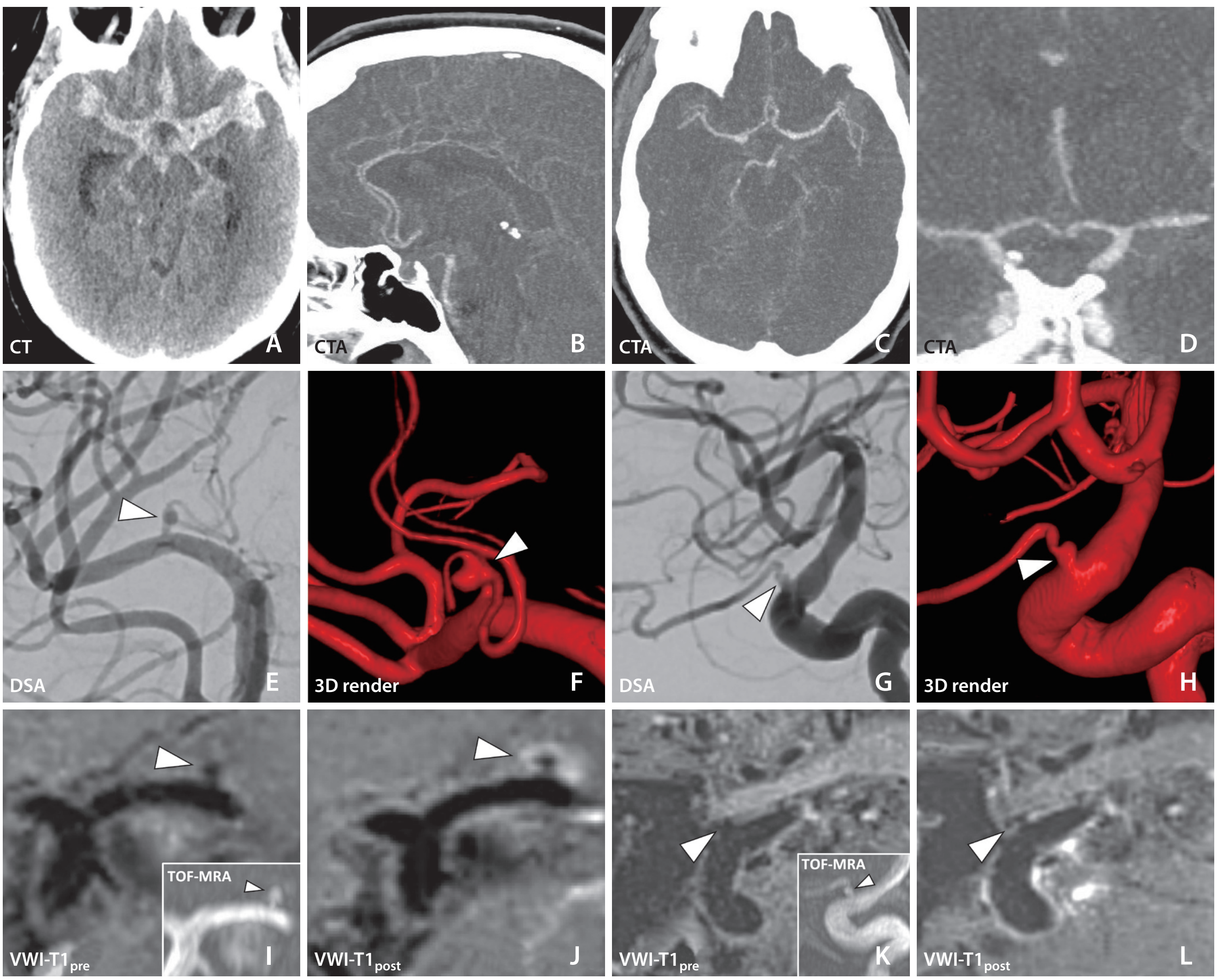

Fig. 1. (**A**) Unenhanced head computed tomography (CT) shows diffuse subarachnoid hemorrhage (SAH) in the basal cisterns, Sylvian fissures and sulci, and acute hydrocephalus with enlargement of the ventricles. (**B**) Sagittal, (**C**) axial, and (**D**) coronal views of the initial computed tomography angiography (CTA) show no saccular aneurysm or sources for SAH. (**E**) Lateral views of repeat digital subtraction angiography (DSA) on day 7 showing an aneurysm (arrowhead) of the left lenticulostriate artery (LSA). (**F**) Three-dimensional (3D) volume rendering of the left LSA aneurysm (arrowhead). (**G**) Lateral view of repeat DSA on day 7 visualizing aneurysm of the ophthalmic segment of the left internal carotid artery (ICA) (arrowhead). (**H**) 3D volume rendering of the left ICA ophthalmic segment aneurysm (arrowhead). (**I**) Pre-contrast T1-weighted vessel wall imaging (VWI) showing focal irregularity at the origin of the left LSA (arrowhead). Inset shows the time-of-flight magnetic resonance angiography (TOF MRA) of the left LSA (arrowhead). (**J**) Post-contrast T1-weighted VWI shows a rim of enhancement around the LSA aneurysm dome (arrowhead), suggesting rupture. (**K**) Pre-contrast T1-weighted VWI showing aneurysm at the left ICA ophthalmic segment (arrowhead). Inset shows the TOF MRA of the left ICA ophthalmic segment (arrowhead). (**L**) Post-contrast T1-weighted VWI of the left ICA ophthalmic segment aneurysm reveals no wall enhancement (arrowhead).

giography (CTA) was read as showing no saccular aneurysm (Fig. 1B–D). The patient was transferred to a tertiary care center, where an emergent ventriculoperitoneal shunt was placed and an initial digital subtraction angiography (DSA) showed no culprit sources of SAH. The patient was transferred to the neuro-intensive care unit during which an intracranial VWI was attempted 6 days after the DSA but aborted due to motion. A second DSA was performed on day 7, which showed left lateral lenticulostriate and left internal carotid artery (ICA) ophthalmic segment aneurysms (Fig. 1E–H). A high-resolution cone-beam CTA demonstrated a dysplastic left lateral lenticulostriate aneurysm associated with a lenticulostriate perforator. Given the possibility of multiple culprit aneurysms, VWI was re-attempted on the same day while keeping the patient intubated. VWI demonstrated focal outpouching at the left LSA origin with a subtle rim of enhancement around the aneurysm dome (Fig. 1I, J). There was no vessel wall enhancement associated with the left ICA ophthalmic segment aneurysm (Fig. 1K, L).

Based on VWI, the patient underwent emergent left pterional craniotomy for aneurysm clipping. The LSA aneurysm was found to be ruptured with fresh thrombus at the ruptured dome. Intra-operative DSA and cone-beam CTA were performed to assess clip placement. These demonstrated occlusion of the aneurysm of the parent circulation with patency of the LSA and middle cerebral artery.

DISCUSSION

We present a case of diffuse SAH due to a ruptured LSA aneurysm diagnosed by VWI. LSA aneurysms are exceedingly rare and present diagnostic challenges due to their small size and location. Of the 112 LSA aneurysm cases reported in the literature in a recent systematic review, 78% were ruptured and 30% presented as SAH.[1] Distal ruptured LSA aneurysms were associated with intracranial and intraventricular hemorrhage (47% and 40% of cases respectively), while proximal LSA aneurysms tended to present with SAH, with several patients having more than 1 of these manifestations. Angiography was used to confirm the diagnosis of LSA aneurysm in all cases reported.[1]

Identifying the cause of bleeding in diffuse SAH can be challenging. While CTAs are effective in diagnosing ruptured aneurysm sources for SAH, DSA is the gold standard, as small aneurysms or vascular lesions may not be detectable on CTA due to limited spatial resolution, venous contamination, or thrombus occluding the aneurysm sac at the time of initial imaging.[6,9] However, even DSA has limited sensitivity. In a single-center case series of 95 patients with diffuse pattern SAH, DSA was unable to identify a hemorrhagic source in 85% of cases, while the remaining 15% of cases were diagnosed and treated for ruptured aneurysms/pseudoaneurysms.[10] Additionally, previous studies have reported a 15–20% incidence of initial negative DSA in spontaneous SAH, with repeat DSA identifying bleeding source in only 7% of cases.[11,12] Because timely treatment of aneurysmal SAH is also key to preventing further rupture, morbidity, and mortality, comprehensive diagnostic assessment for a culprit source is critical for patient care. Improved outcomes, including earlier discharge to home and increased 12-month survival, can be achieved with earlier treatment.[13] These previous findings highlight the need for rapid and accurate diagnostics for aneurysm rupture, especially in cases of diffuse SAH.

VWI has shown the ability to identify ruptured intracranial aneurysms. A recent systematic review highlights several studies where VWI found vessel wall enhancement in ruptured aneurysms.[14] By contrast, there are few studies and case reports regarding the use of VWI in angiogram-negative diffuse SAH. Among 13 patients with initial angiogram-negative diffuse SAH, Yoon and colleagues[7] found VWI was able to show arterial wall focal enhancement in 10 patients (76.9%), with repeat DSA identifying culprit lesions in 5 of those 10 patients (2 aneurysm dissections, 2 blood-blister like aneurysms and 1 small perforating aneurysm). The utility of VWI in identifying the site of rupture among multiple aneurysms in the setting of aneurysmal SAH has also been reported, where unruptured intracranial aneurysms showed no vessel wall enhancement compared to those that ruptured.[15] However, care in the diagnostic VWI interpretation is needed given the possibility of false positives. Yoshikawa et al.[16] evaluated VWI in the setting of multiple aneurysms and found false positive cases of wall enhancement in unruptured aneurysms due to atherosclerotic buildup in the aneurysm wall. Thus, further investigations into the utility of VWI in the use case of multiple aneurysms is needed. In this case, VWI was able to identify vessel wall enhancement of the left LSA aneurysm favoring rupture from a rare site. With no wall enhancement observed at the ophthalmic segment of the left ICA, VWI helped confirm the diagnosis of rupture at the LSA and directed treatment.

Due to the low incidence of LSA aneurysms, there is no consensus on optimal treatment. Of the reported ruptured LSA aneurysm cases in the literature, 51% were treated surgically, 34% conservatively (non-operatively), 15% with endovascular repair, and 1 case treated with gamma-knife radiation.[1] While endovascular interventions are becoming increasingly advanced, there is still no evidence of benefit over the surgical approach, and treatment should be decided based on contraindications to surgery, anatomic complexity, as well as patient preference. Open surgery for aneurysm clipping was chosen in our case because of concerns the aneurysm was too small to coil and uncertainty around the effectiveness of a flow-diverting stent, given proximity to the LSA and branching vessels directly off the aneurysm. Another contraindication to endovascular treatment is the risk associated with post-procedure dual antiplatelet therapy in a patient with already high-grade SAH.

In summary, we report VWI findings in a patient with HH4/mF3 SAH due to a rare ruptured LSA aneurysm. VWI helped in clinical decision-making, facilitating the steps towards treatment for a highly morbid condition. With the increasing accessibility of advanced imaging technology, VWI has the potential to be an effective tool in the setting of angiogram-negative SAH.

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None.

Ethics Statement

This report was written in concordance with the Penn Medicine Institutional Review Board. The requirements for institutional Review Board approval and consent were waived for a case report. We anonymized patient information, including sex and age, to ensure that individual patients cannot be identified.

Conflicts of Interest

The authors have no conflicts to disclose.

Author Contributions

Concept and design: HQP and JWS. Analysis and interpretation: HQP, SGA, SBR, OAC, and JWS. Data collection: HQP, SGA, and JWS. Writing the article: HQP and JWS. Critical revision of the article: HQP, SGA, SBR, OAC, and JWS. Final approval of the article: HQP, SGA, SBR, OAC, and JWS. Overall responsibility: HQP and JWS.

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