

Diagnostic and therapeutic challenge in intracerebral hemorrhage due to aneurysm of distal lenticulostriate artery

Reto diagnóstico y terapéutico en la hemorragia intracerebral por aneurisma de la arteria lenticuloestriada distal

José A. Figueroa-Sánchez¹, Enrique Caro-Osorio¹, Héctor R. Martínez-Rodríguez², and José C. Herrera-Castro^{1*}

¹Department of Neurosurgery; ²Department of Neurology. Centro Médico Zambrano Hellion, School of Medicine and Health Sciences, Tecnológico de Monterrey, Monterrey, Nuevo León, Mexico

Abstract

Objective: The objective of this study was to present an atypical case of deep intracerebral hemorrhage caused by the rupture of an aneurysm of the distal lenticulostriate artery. A 42-year-old woman presenting with the right-sided mild hemiparesis was diagnosed with an acute left-sided hemorrhage within the basal nuclei. Cerebral angiography revealed a microaneurysm of the distal portion of the left lenticulostriate artery. Conservative treatment under close neurological surveillance was recommended. After a few days of hospital stay, the patient's neurological symptoms improved, and she was discharged. The patient has remained asymptomatic for more than 6 months. Aneurysms of the distal lenticulostriate artery are extremely rare.

Keywords: Aneurysm. Cerebral hemorrhage. Lenticulostriate artery.

Resumen

Objetivo: Presentar un caso atípico de hemorragia intracerebral profunda causada por la ruptura de un aneurisma de la arteria lenticuloestriada distal; se revisa la bibliografía y se analizan la fisiopatología y el tratamiento. Se presentó a la sala de emergencias una mujer de 42 años con hemiparesia leve del hemicuerpo derecho. Se la diagnosticó con una hemorragia aguda en los núcleos de la base izquierdos. La panangiografía cerebral reveló un microaneurisma de la porción distal de la arteria lenticuloestriada izquierda. Se recomendó tratamiento conservador bajo estrecha vigilancia neurológica. Después de unos cuantos días de hospitalización, los síntomas neurológicos de la paciente mejoraron y la paciente recibió el alta. La paciente ha permanecido asintomática durante más de seis meses. Los aneurismas de la arteria lenticuloestriada distal son extremadamente raros.

Palabras clave: Aneurisma. Hemorragia cerebral. Arteria lenticuloestriada.

Correspondence:

*José C. Herrera-Castro

Avda. Batallón de San Patricio, 112

Col. Real San Agustín

C.P. 66278, San Pedro Garza García, N.L. México

E-mail: jcherrera.c4@gmail.com

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Background

Hemorrhagic strokes account for 10-20% of all cerebral vascular events (CVE)¹⁻³. Deep intracerebral hemorrhage (ICH) in the basal ganglia is common and can develop due to multiple causes, most often related to systemic arterial hypertension (SAH). Rupture of a lenticulostriate artery (LSA) aneurysm is an extremely rare event, and even more when it occurs in the distal portion of the vessel⁴. Cerebral angiography is the most sensitive study to detect LSA aneurysms; however, it is almost never performed in patients with deep ICH². At present, there is no consensus on the treatment of this condition due to the low incidence of LSA aneurysms rupture as a cause of deep ICH unrelated to SAH. This document reports a case of distal LSA aneurysm rupture that was conservatively treated.

Clinical case

A previously healthy 42-year-old woman presented to the emergency room with the right-sided hemiparesis (4/5 on Lovett scale), subtle right central facial paralysis, without aphasia, with a National Institute of Health Stroke Scale (NIHSS) CVE score of 4 and modified classification score of 2. On her arrival, the CVE care team was activated. Computed axial tomography (CT) of the skull revealed hemorrhage in the left basal ganglia (Fig. 1A, B). The patient was alert and hemodynamically stable. She was admitted to the intensive care unit for medical treatment, and a contrasted brain magnetic resonance imaging (MRI) was performed (Fig. 1C, D).

Laboratory tests were normal or negative, including platelet count, coagulation profile, autoimmunity profile (antinuclear antibodies, ANCA, C-reactive protein, and C3-C4-C50 supplement), C-protein, S-protein, anti-thrombin III, and Leiden factor V. The hematoma volumetric analysis was 8 cm³. After ruling out the main causes of ICH, a cerebral angiogram was requested 3 days after admission, in which a small aneurysm was found in the distal portion of the left lenticulostriate artery. The possibility of an aneurysm of infectious cause was ruled out due to the absence of risk factors, non-specific clinical findings and negative laboratory tests for infection, and antibiotic therapy was, therefore, not administered. Supraselective endovascular embolization was considered for the aneurysm, although vascular anatomy of the LSA proximal portion was found to be narrow and tortuous,

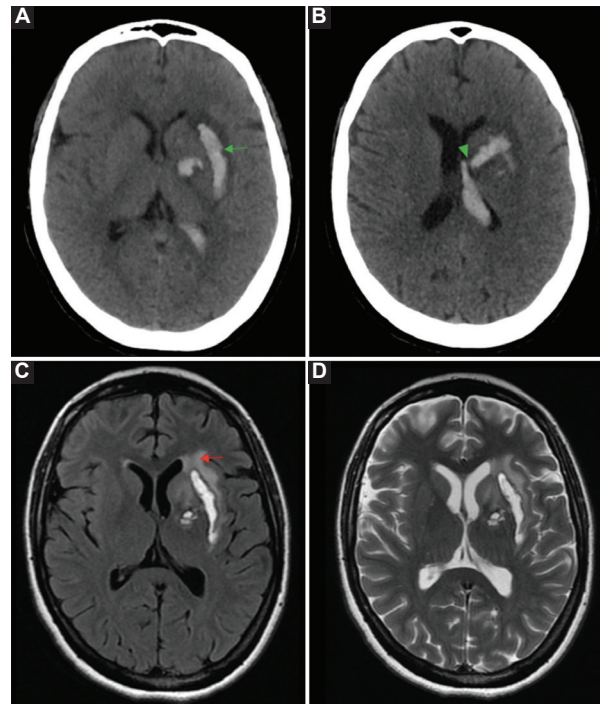


Figure 1. Cranial CT axial section showing an 8-mL deep ICH that mainly compromises the left putamen (green arrow). **A:** intraventricular extension (arrowhead). **B:** brain MRI: axial section T1-sequence. **C:** left basal ganglia hemorrhage with perilesional edema (red arrow). **D:** the T2-sequence axial MRI image did not identify aneurysm or vascular malformation.

and thus, this therapeutic approach was disregarded given that it was considered to be of high risk (Figs. 2 and 3).

Wada functional test showed dominance of the left brain hemisphere. A multidisciplinary team analyzed the case and, after considering the patient's wishes, a conservative treatment approach was decided.

Seven days after admission, the patient was discharged with slight right-sided hemiparesis and no facial paralysis. On clinical follow-up at 6 and 12 months, the patient was asymptomatic (NIHSS 0, mRS 0).

Conservative treatment in this patient was based on the type and size of the aneurysm, deep location and eloquence and, although she remains asymptomatic 1 year after the episode, follow-up neuroimaging studies were suggested, in which the patient refused.

Discussion

In patients with spontaneous ICH, surgical treatment and other invasive treatments remain controversial. However, recommendations in current guidelines

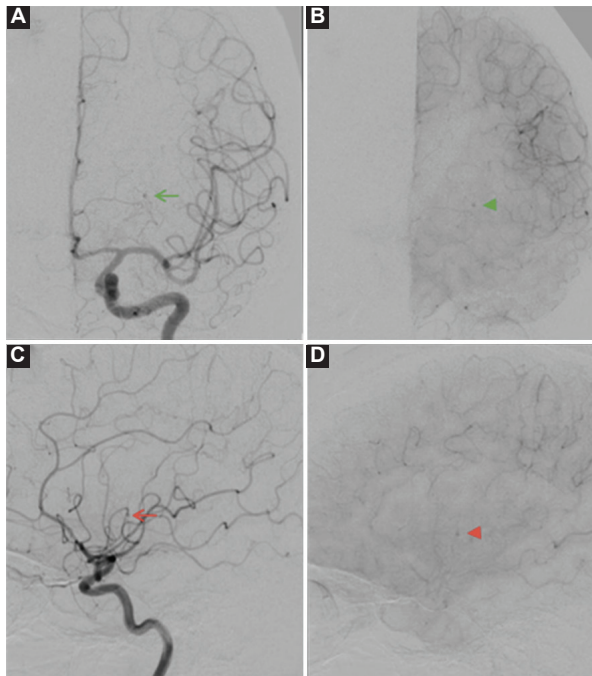


Figure 2. Cerebral angiography. Left internal carotid artery early arterial phase. **A:** AP projection showing distal LSA subtle abnormal dilatation (green arrow), which corresponds to a $1.4 \times 1.5 \times 1.5$ mm saccular microaneurysm, with an approximate volume of 0.15 cm^3 . **B:** late arterial phase AP projection; the contrast material remains within the aneurysm (green arrowhead). Early arterial phase. **C:** lateral view showing distal LSA subtle abnormal dilatation (red arrow). Late arterial phase. **D:** lateral view; the contrast remains within the aneurysm after the early arterial phase (red arrowhead).

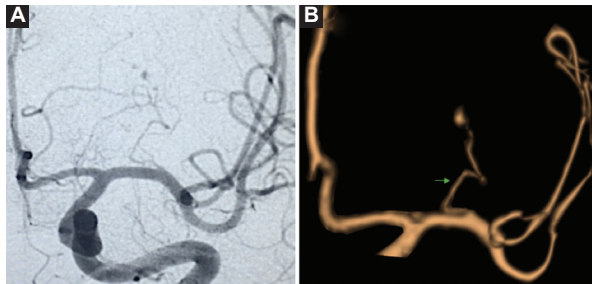


Figure 3. Cerebral angiography. **A:** left carotid angiogram early arterial phase with first plane on LSA. **B:** three-dimensional reconstruction showing a narrow and tortuous aspect of LSA proximal portion angioconfiguration.

do not include patients with structural lesions that cause ICH such as brain aneurysms².

In deep ICH physiopathology, microaneurysms have been described in patients with SAH, which usually affect the perforating arteries (Charcot–Bouchard aneurysms), out of which 80% have been observed in the LSA; however, there is little evidence of microaneurysms in LSAs of young patients without SAH^{5–7}.

Vargas et al.⁸ made the most exhaustive description of LSA aneurysms. They presented a series of 48 cases of LSA aneurysms and classified them into three types. Type 1 includes aneurysms arising from the middle cerebral artery next to the place of LSA origin; in type 2, the aneurysm originates in LSA proximal portion; and type 3 comprises distal LSA aneurysms. Based on this classification, this case was established to be type 3⁸.

LEA aneurysms have been associated with several diseases, such as SAH, Moya–Moya disease, and systemic lupus erythematosus^{8,9}. In usual diagnostic approach to deep ICH, in patients, in whom cerebral angiography is usually not performed, CT angiography has been shown to be sensitive in the detection of small aneurysms; however, some lesions may not be observed. Tan et al.¹⁰ demonstrated the “spot sign” on contrasted CT (contrast enhancing spot within the hematoma) in a patient with basal ganglia ICH caused by a LSA aneurysm.

In young patients with deep ICH, in whom other causes of bleeding are ruled out, it is important to confirm this type of LSA distal aneurysms. Notwithstanding, a small distal LSA aneurysm may go unnoticed, even on cerebral angiography¹¹.

Due to the reduced number of cases reported in medical publications, there is no consensus on the best treatment approach for LSA aneurysms. Some authors have tried to use stents with rather dissimilar results. Other options such as radiosurgery^{12–14} or endovascular therapy⁸ have also been described, but without sufficient evidence to generalize this type of treatments. Prospective and multicenter studies are required to analyze a larger number of patients, generalize the results, and issue diagnostic and therapeutic recommendations.

Conflicts of interest

The authors declare that they have no conflicts of interest.

Ethical disclosures

Protection of human and animal subjects. The authors declare that no experiments have been performed on humans or animals for this research.

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained informed consent of the patients or subjects referred to in the article. This document is in the possession of the corresponding author.

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