

Intracranial dermoid cyst rupture-related brain ischemia

Case report and hemodynamic study

Hang Jin, MD, PhD, Zhen-Ni Guo, MD, Yun Luo, MD, Ren Zhao, MD, Ming-Shuo Sun, MD, Yi Yang, MD, PhD*

Abstract

Rationale: Spontaneous rupture of intracranial dermoid cyst is a rare but serious clinical event that can result in cerebral ischemia. Cerebral vasospasm and vasculitis are considered as potential mechanisms of dermoid cyst rupture-related cerebral ischemia. However, the hemodynamic mechanisms between cerebral ischemia and dermoid cyst rupture are not well known.

Patient concerns: A 55-year-old, right-handed man was admitted to our hospital with sudden receptive aphasia and right-sided hypoalgesia. Brain magnetic resonance imaging (MRI) revealed a ruptured dermoid cyst and watershed infarcts in the left hemisphere. Then brain magnetic resonance angiography disclosed mild stenosis in the left middle cerebral artery (MCA), and further high-resolution MRI demonstrated it was caused by an unstable atherosclerosis plaque. Transcranial Doppler of the patient showed a decreasing tendency of peak systolic velocity (PSV) of the left MCA at different time points after the stroke (from 290cm/s at day 6 to 120cm/s at day 30), indicating a transient vasospasm. However, the time course of dynamic cerebral autoregulation (dCA) seemed different from the PSV. The patient's dCA reached its lowest point at day 8 and was restored at day 10. The time course of dCA indicated a "called procedure" of a cerebrovascular regulating function to deal with the stimulation in subarachnoid space.

Diagnoses: A dermoid cyst rupture-related cerebral infarction was diagnosed in this patient.

Interventions: Aspirin (100 mg/d) and atorvastatin (20 mg/d) were given to the patient. A neurosurgical operation was strongly recommended to minimize the risk of further injury of the ruptured dermoid cyst; however, the patient refused the recommended treatment.

Outcomes: The neurological deficit of the patient was significantly improved on 30 days follow-up.

Lessons: We found that the spread of cyst contents through the subarachnoid and/or ventricular system can induce a vasospasm. Then, dCA was "called" to deal with the stimulation in the subarachnoid space. Compromised dCA seems to be one of the compensatory of cerebral vasospasm after a dermoid cyst rupture.

Abbreviations: CA = cerebral autoregulation, dCA = dynamic cerebral autoregulation, MCA = middle cerebral artery, MRA = magnetic resonance angiography, MRI = magnetic resonance imaging, PSV = peak systolic velocity, TCD = transcranial Doppler.

Keywords: brain ischemia, case report, dermoid cyst, hemodynamics

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HJ and Z-NG have contributed equally to the manuscript.

All authors have read and approved the final manuscript. Simultaneously, the patient provided written informed consent for the publication of this case details and the consent procedure was approved by the Human Ethics and Research Ethics committees of the First Hospital of Jilin University.

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Department of Neurology, Stroke Center, the First Hospital of Jilin University, Chang Chun, China.

* Correspondence: Yi Yang, Department of Neurology, The First Hospital of Jilin University, Xinmin Street 71, 130021 Changchun, China (e-mail: doctoryangyi@163.com).

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1. Introduction

Intracranial dermoid cysts are slow-growing benign masses, accounting for <1% of intracranial masses.^[1] Spontaneous rupture of intracranial dermoid cyst is a rare but serious clinical event that results in dissemination of cyst contents from the dermoid cysts into the subarachnoid space and/or ventricles.^[1,2] The cyst contents within the subarachnoid space can induce cerebral vasospasm or vasculitis; thus, cerebral vasospasm and vasculitis are considered as potential mechanisms of dermoid cyst rupture-related cerebral ischemia.^[3–5] However, the hemodynamic mechanisms between cerebral ischemia and dermoid cyst rupture are not well known.

Cerebral autoregulation (CA) is a physiological mechanism that maintains a stable cerebral blood flow within a wide range of blood pressure fluctuations.^[6] Impaired CA has been demonstrated to be a significant contributor to secondary damage of cerebral tissue in several brain injuries.^[7–10] Thus, we sought to determine whether impaired CA plays a role in the pathogenesis of dermoid cyst rupture-related brain ischemia.

2. Case report

A 55-year-old, right-handed man presented with sudden language impairment and right-side numbness. There was no

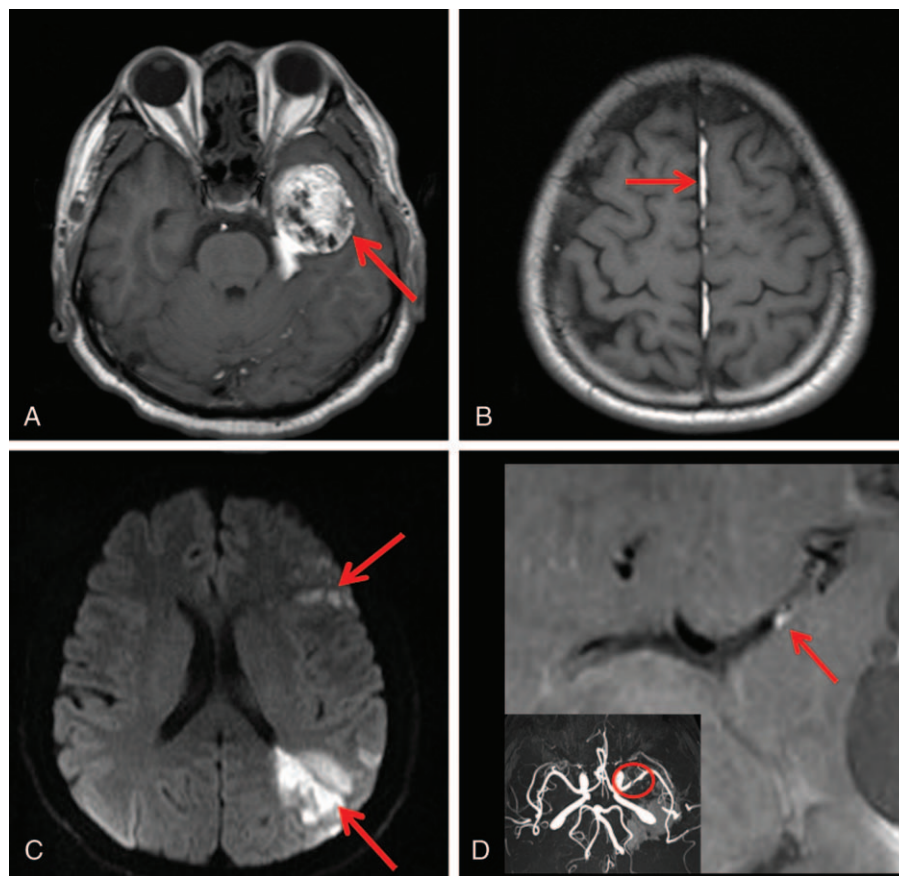


Figure 1. MRI of a dermoid cyst rupture with cerebral infarction. T1-weighted MRI 2 days after onset showed a mixed hyperintense lesion in the left temporal region (A, arrow) with hyperintense droplets scattered into subarachnoid space (B, arrow), simultaneously, diffusion-weighted MRI showed high signal in the border zones between the regions of the MCA and the anterior cerebral artery/posterior cerebral artery, indicating watershed infarcts (C, arrows). Five days later, brain magnetic resonance angiography showed a mild stenosis on the left MCA (D, circle) caused by an eccentricity enhanced plaque showed by high-resolution MRI (D, arrow). MRI = magnetic resonance imaging.

history of trauma and other identified risk factors of a stroke except smoking. Neurological examinations revealed receptive aphasia and right-sided hypoalgesia. Emergent cranial computed tomography showed a hypodense lesion in the left temporal lobe. Two days after an onset, brain magnetic resonance imaging (MRI) revealed a $4.0 \times 3.5 \times 3.5$ cm ruptured dermoid cyst (Fig. 1A and B) and watershed infarcts in the left hemisphere (Fig. 1C). Five days later, brain magnetic resonance angiography (MRA) demonstrated mild stenosis on the M1 segment of left middle cerebral artery (MCA), and further high-resolution MRI examination showed an unstable atherosclerosis plaque in the left MCA (Fig. 1D). Simultaneously, we monitored the blood flow velocity of bilateral MCA (M1 segment, 58 mm) with transcranial Doppler (TCD). The results showed a decreasing tendency of the peak systolic velocity (PSV) of left MCA at a different time points after the stroke (290, 250, 200, 150, and 120 cm/s at 2, 6, 8, 10, and 30 days after the stroke, respectively). By comparison, the PSV of the right MCA was kept stable at all time points (80–91 cm/s) (Fig. 2A). Although the pulsatility index of the left MCA at different time points were almost within normal range (0.65–1.10), it showed a decreasing tendency (0.9, 0.62, 0.65, 0.71, and 0.7 at 2, 6, 8, 10, and 30 days after the stroke, respectively). Dynamic cerebral autoregulation (dCA) examinations of the patient were also performed. The gains in the affected (left side) and unaffected hemispheres were 2.40 versus

0.93 (day 6), 2.52 versus 0.87 (day 8), 1.67 versus 0.973 (day 10), and 1.93 versus 0.94 (day 30), respectively (Fig. 2B). The phase difference in the affected hemisphere versus unaffected side was 34.47° versus 28.60° (day 6), 9.08° versus 21.38° (day 8), returned to 22.35° versus 30.18° (day 10) and then remained normal at 30.46° versus 30.89° at the 30-day follow-up (Fig. 2C). As a result, a dermoid cyst rupture-related cerebral infarction was diagnosed. The patient received aspirin (100 mg/d) and atorvastatin (20 mg/d), and the neurological deficit was significantly improved on 30 days follow-up. Seizure did not occur to this patient during treatment. A neurosurgical operation was strongly recommended to minimize the risk of further injury of the ruptured dermoid cyst; however, the patient had refused the recommended treatment.

3. Discussion

In this study, we reported a case of a cerebral infarction related to dermoid cyst rupture and further explored the inner hemodynamic mechanisms using continuous PSV and dCA detection of bilateral MCA. We have found that the spread of cyst contents through the subarachnoid and/or ventricular system can induce a vasospasm. Then, dCA was “called” to deal with the stimulation in the subarachnoid space. Compromised dCA seems to be one of the compensatory of cerebral vasospasm after a dermoid cyst rupture.

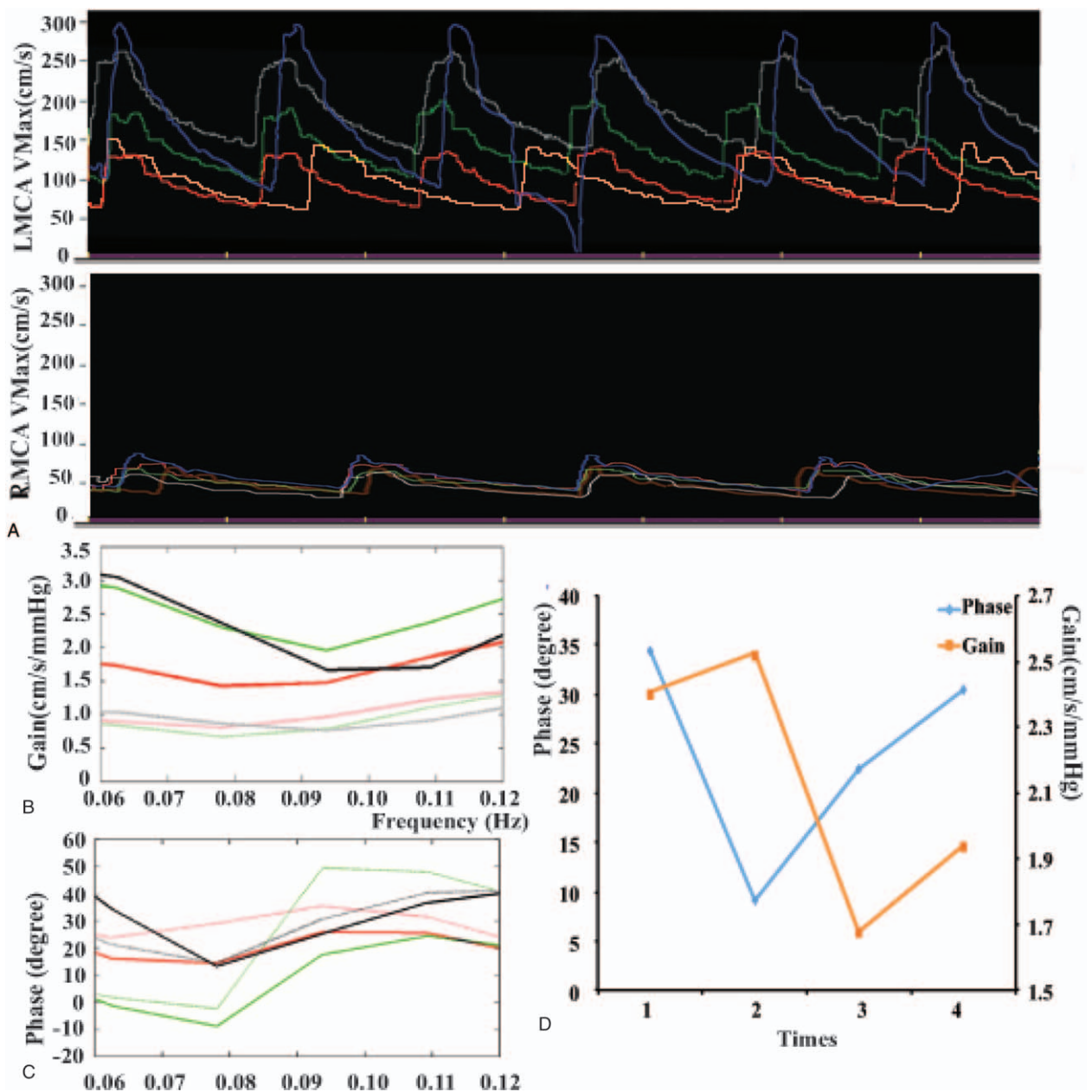


Figure 2. TCD and dCA of the patient. TCD (A) showed the peak systolic velocity of the left MCA (upper) and right MCA (lower) at day 2 (blue lines), 6 (white lines), 8 (green lines), 10 (red lines), and 30 (orange lines) days after the stroke, respectively. Corresponding dCA examinations showed the value of gain (B) and the value of phase at different time points (C) (day 6 black lines, day 8 green lines, and day 10 red lines; full lines for the left MCA and dashed lines for the right MCA). The tendency of phase and gain of the left hemisphere are shown in (D). dCA = dynamic cerebral autoregulation, MCA = middle cerebral artery, TCD = transcranial Doppler.

Intracranial dermoid cysts are congenital ectodermal inclusion cysts that contain varying amounts of ectoderm derivatives.^[11] With the accumulation of glandular secretion and epithelial desquamation, the size of a dermoid cyst grows slowly. Thus, rupture of intracranial dermoid cysts is a rare phenomenon. The rupture of a dermoid cyst leads to the spillage of lipid material into the subarachnoid space and/or ventricles, which could induce cerebral vasospasm or vasculitis. It may cause headache (the most common symptom), seizure, cerebral ischemia, and aseptic meningitis. Rupture of intracranial dermoid cyst eventually caused cerebral ischemia in this patient.

However, the hemodynamic mechanisms between cerebral ischemia and dermoid cyst rupture are not well known. Some

factors were thought to be related to ischemia, for example, vasospasm and vasculitis.^[11] The well-known one is intracranial vasospasm. In our case, a vasospasm course was confirmed by TCD monitoring. The PSV on the second day after the stroke was extremely high, reaching 290 cm/s, then declining over time, and returning to normal at day 30 (120 cm/s). A short-term vasospasm resulted in transient left MCA severe stenosis, which is the cause of cerebral ischemia just like a vasospasm-related delayed cerebral ischemia after subarachnoid hemorrhage.^[12] dCA is defined as the mechanism by which constant cerebral blood flow is maintained despite changes of arterial blood pressure/intracranial pressure, it refers to an intrinsic physiological control of cerebrovascular function. When the vasospasm occurred, the dCA was “called” to

deal with vasospasm. The time course of CA changes in this patient indicated the “compensatory procedure” of dCA to respond to vasospasm. This “called procedure” was gradually increased and reached maximum on the day 8. In this patient, we have found the gain was more sensitive to stimulations (Fig. 2D). “Gain” is another index of dCA, it regards to be an indicator of the change of amplitude in cerebral blood flow velocity corresponding to the changes in arterial blood pressure. In general, increasing the gain indicates poorer autoregulation, while a lower gain is considered to represent improved autoregulation. However, several factors could affect “gain,” such as atherosclerosis, intracranial vascular stenosis, etc., thus the value of gain is variable, which can not accurately represent dCA.

Medical management is indicated for symptom control. This patient received aspirin (100 mg/d) and atorvastatin (20 mg/d) as a standard treatment for preventing further injury in the acute phase of dermoid cyst rupture-related stroke. As to dermoid cysts, surgery is recommended when dermoid cysts lead to mass effect or serious neurological deficits. Recurrence is rare if the mass and the tumor wall are resected completely. Unfortunately, scattered fat droplets following rupture was too difficult to allow for complete removal.^[4]

There were some flaws in this case. For example, we could not get the baseline dCA data because we put a vasospasm as initial pathogenesis. Also, we could not get the pathological data because the patient declined to receive neurosurgical operation. To the best of our knowledge, this is the first case study that is focused on the hemodynamic mechanism of a dermoid cyst rupture-related brain ischemia.

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