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Case Report

Acute spontaneous subdural hematoma secondary to ruptured arteriovenous malformation: A rare entity

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ABSTRACT

Introduction: Acute spontaneous subdural hematoma (ASSDH) due to ruptured arteriovenous malformation (AVM) is exceptional. There were only four reported cases. In this paper, we present a successful multimodality treatment of the ASSDH secondary to ruptured AVM.

Case presentation: A 21-year-old healthy man with no history of trauma presented to our hospital with complaints of severe headache for 12 hours before admission. On examination, he was alert and oriented. He had no intracranial hypertension, meningismus, and neurological deficits. Computed tomography illustrated a right acute subdural hematoma 8mm in thickness with a 5mm midline shift and a right frontal intraparenchymal hemorrhage 40 × 25mm in size. Digital subtraction angiography showed a 2 × 3 cm right frontal AVM, Spetzler-Martin grade I. The feeding arteries were cortical branches of the right anterior cerebral artery, and drain veins were cortical veins. He received emergency preoperative embolization followed by hematoma evacuation and total excision of the malformation. His headache was relieved and disappeared after a week. No postoperative neurological deficits were reported.

Clinical discussion: Elective surgical resection of AVM after 4–6 weeks was preferred in patients with no risk factors of rebleeding. Emergent surgery was only indicated for significant mass effect or acute hydrocephalus. Preoperative embolization is helpful for the presence of intra-nidal or peri-nidal aneurysm, AVM with high grades, reducing intraoperative blood loss and occlusion of deep vessels.

Conclusion: ASSDH due to ruptured AVM is rare and easy to omit in clinical settings. Preoperative embolization and surgical excision are effective treatments.

1. Introduction

Arteriovenous malformations of the brain (AVM) are intraparenchymal clusters of abnormal connecting blood vessels containing both artery and vein elements but no capillaries. Ruptured arteriovenous malformation (AVM), one of the most common causes of stroke in younger patients, is often seen in hemorrhagic stroke. Clinical presentation of AVM with hemorrhage occurs in nearly half of cases [1,2]. The annual hemorrhagic risk is estimated at 1–3% for unruptured AVM and 4–6% for ruptured AVM [3,4]. Risk factors for spontaneous hemorrhage include prior rupture/hemorrhage, deep venous drainage, deep and infratentorial location of AVM nidus, concurrent aneurysm, and patients' age [5].

While the typical manifestations of ruptured AVM are intraparenchymal, subarachnoid, and intraventricular hemorrhages, acute spontaneous subdural hematoma (ASSDH) is exceptional. There were only four reported cases [6] [–] [8]. This paper presents a successful multimodality treatment of the acute spontaneous subdural hematoma and intraparenchymal hemorrhagic stroke secondary to ruptured arteriovenous malformation.

The work has been reported in line with the SCARE criteria [9].

2. Presentation of case

A 21-year-old man with a healthy history presented to our hospital with complaints of severe headache for 12 hours prior to admission. He

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had neither history of head trauma nor other medical, surgical, family, psychosocial, and pharmacologic histories. His headache was sharp, persistent, progressive, and bilateral. This pain had no alleviating factor. He denied nausea and vomiting. On examination, he was alert and oriented (Glasgow coma scale 15 points). He had no intracranial hypertension and meningismus. Neurological examination was intact. Fundoscopy showed no papillary edema and hemorrhage. Other physical examinations were normal.

Computed tomography (CT) illustrated a right acute subdural hematoma 8mm in thickness with a 5mm midline shift and a right frontal intraparenchymal hemorrhage 40 × 25mm in size (Fig. 1). The multislide computed tomography (MSCT) with contrast demonstrated a right frontal ruptured arteriovenous malformation (AVM) (Fig. 1). Digital subtraction angiography (DSA) showed a right frontal AVM with 2 × 3 cm in size, Spetzler-Martin grade I and Lawton-Young grade IV. The feeding arteries were cortical branches of the right anterior cerebral artery, and draining veins were cortical veins.

He received emergency preoperative endovascular embolization with precipitating hydrophobic injectable liquid (PHIL) and then underwent a surgical operation (Fig. 2). Before the operation, a dose of prophylaxis antibiotic (cefotaxime 1g, intravenous injection) was given. We used a right frontal approach to evacuate the hematoma and excise the malformation. The surgical operator was the head of the department of neurosurgery and spine surgery at a tertiary teaching hospital. In addition, he was an associate professor of neurosurgery with an advanced level of surgical experience. Furthermore, he trained as a qualified neurosurgeon at the University of Tours and Louis Pasteur University in France. Histopathological examination illustrated arteriovenous malformation (Fig. 3).

After the operation, the patient received analgesics (acetaminophen 1g, intravenous administration three times per day) and saline solution (sodium chloride 1000ml per day). The postoperative course was insignificant. His headache was relieved and disappeared after a week. He also experienced no postoperative neurological deficits. He was happy to return to work and daily activities.

3. Discussion

While acute subdural hematoma (ASDH) is usually associated with traumatic brain injury, ASDH without trauma is rare. Acute spontaneous subdural hematoma (ASSDH) was first described by Munro et al. in 1936. Regarding etiology, ASSDH is usually caused by disruption of bridging veins in the subdural space; on the other hand, acute subdural hematoma of arterial origin is more infrequent. As reported in previous studies, the causes of ASSDH included rupture of the cortical artery or pial arteriovenous fistulae (AVF), arteriovenous malformation (AVM), or an intracranial aneurysm [6] [–] [8,10] [–] [12], cocaine abuse [13], falx meningioma [14], coagulopathy and moyamoya disease [15]. To the best of our knowledge, there were only four case reports of ASSDH due to ruptured AVM [6] [–] [8] (Table 1). Table 1 illustrates the clinical summary of reported ASSDH secondary to ruptured AVM.

As shown in Table 1, all five cases were males, with the youngest man at the age of one [6]. Severe headaches, seizures, and loss of consciousness were the most common presenting symptoms. Until now, digital subtraction angiography (DSA) and computed tomography angiography (CTA) remain the gold standard for the diagnosis of AVM. Despite that, CTA and DSA were obtained in a merely reported case [7]. This can be explained that most of the cases had no clue of AVM on plain CT. Another reason was the small size of AVM and the compression of the adjacent hematoma, which made the ruptured AVM obscure on plain CT. Therefore, we recommended that when a healthy patient presented with an ASSDH, prompt CTA or magnetic resonance angiography (MRA) should be considered initial screening tools if it is available to rule out a ruptured AVM. Besides, if CTA or MRA were suspicious, DSA should be indicated within 24 hours for definitive diagnosis because of its high sensitivity and specificity for AVM diagnosis. Moreover, by doing this, feeding artery or nidal aneurysms, which increased the risk for rebleeding, might be embolized during catheter-based angiography [16].

Regarding the treatment of ruptured AVM, four cases, including our patient, had AVM resection except for the one-year-old patient. Two of four patients received preoperative embolization followed by surgical

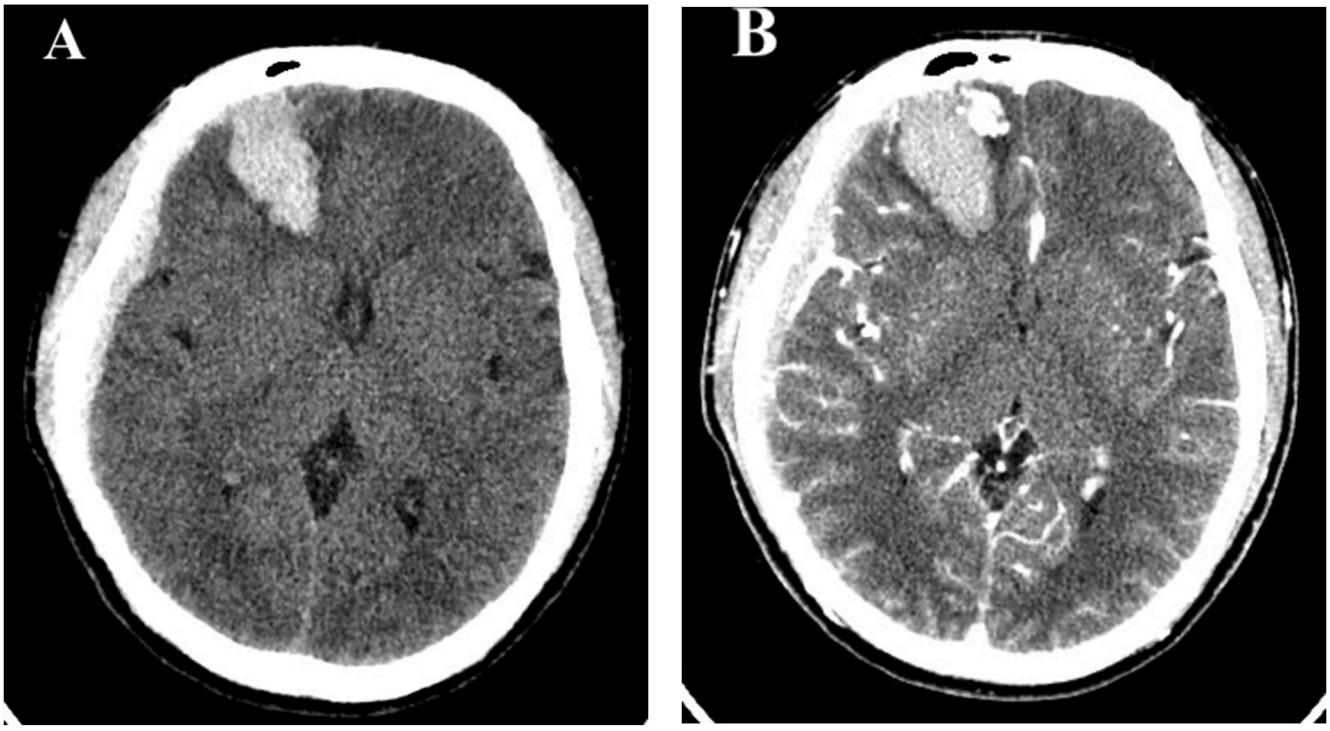

Fig. 1. (A) Computed tomography scan of the head showed an acute subdural hematoma on the right side and an intraparenchymal hemorrhage in the right frontal lobe. (B) The multiple-slice computed tomography with contrast demonstrated a right frontal ruptured AVM.

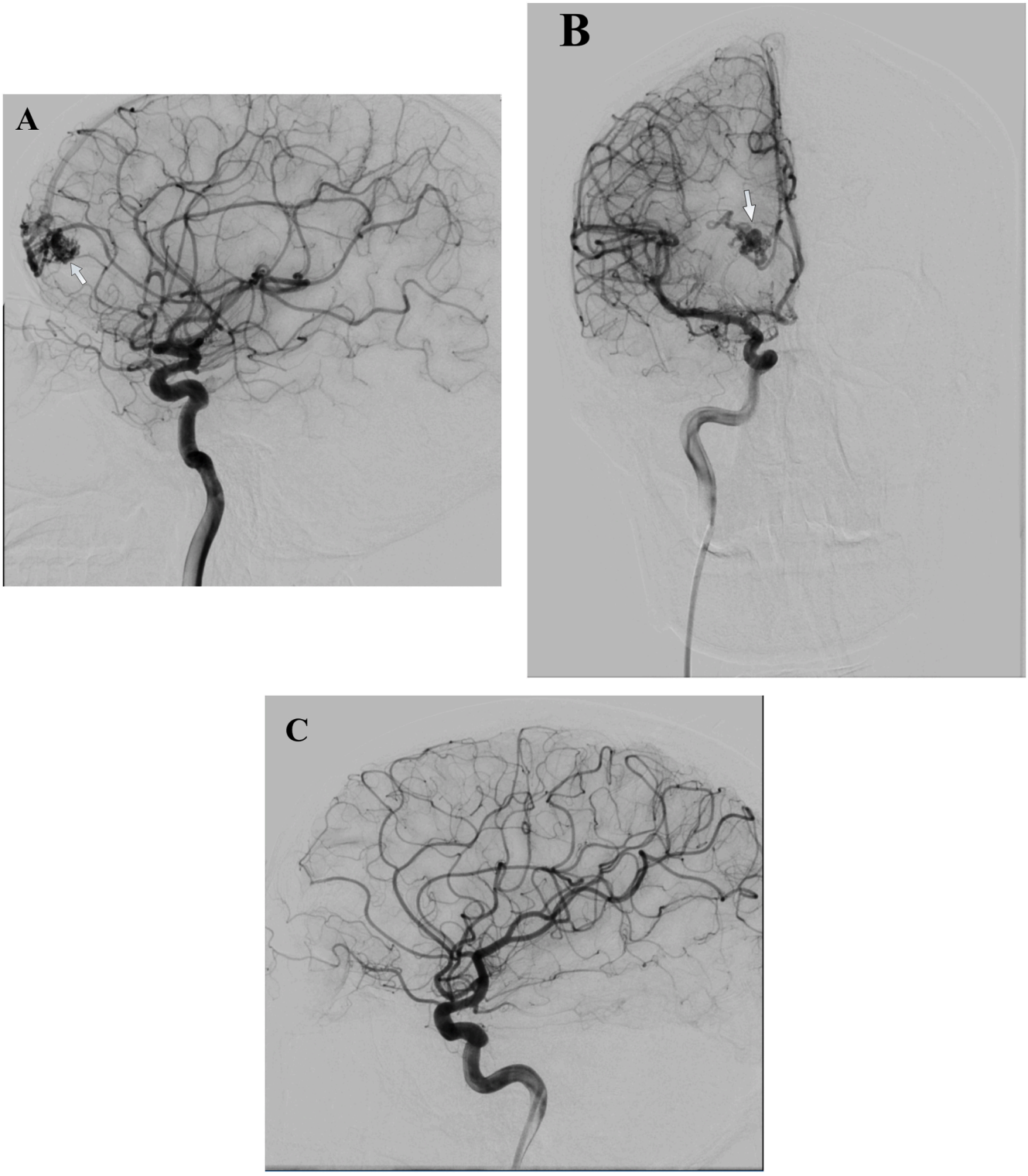

Fig. 2. Digital subtraction angiography (DSA) showed a right frontal AVM with 2 × 3 cm in size, Spetzler-Martin grade I and Lawton-Young grade IV. The AVM was embolized totally by precipitating hydrophobic injectable liquid (PHIL). (A, B) Before embolization. (C) After embolization.

excision of AVM [7]. The ruptured AVMs causing acute spontaneous subdural hematoma were usually small and classified as Spetzler-Martin grade I (Spetzler-Ponce Grade A), which were favorable to surgery. In general, initial stabilization should be obtained first for most ruptured AVM in the intensive care unit, ideally with advanced neurological capabilities. The initial stabilization should pay attention to reversal of coagulopathy, management of blood pressure, glucose, temperature, and seizures [16]. After that, definitive treatment is made based on the weighing risks and benefits of various treatment options, including open microsurgery, endovascular embolization, radiosurgery, and observation. Elective surgical resection of AVM after 4–6 weeks of conservative treatment was preferred if the ruptured AVM had no risk factors of rebleeding [16]. Emergent surgery was only indicated if patients had a significant mass effect and progressive neurological deficits due to a

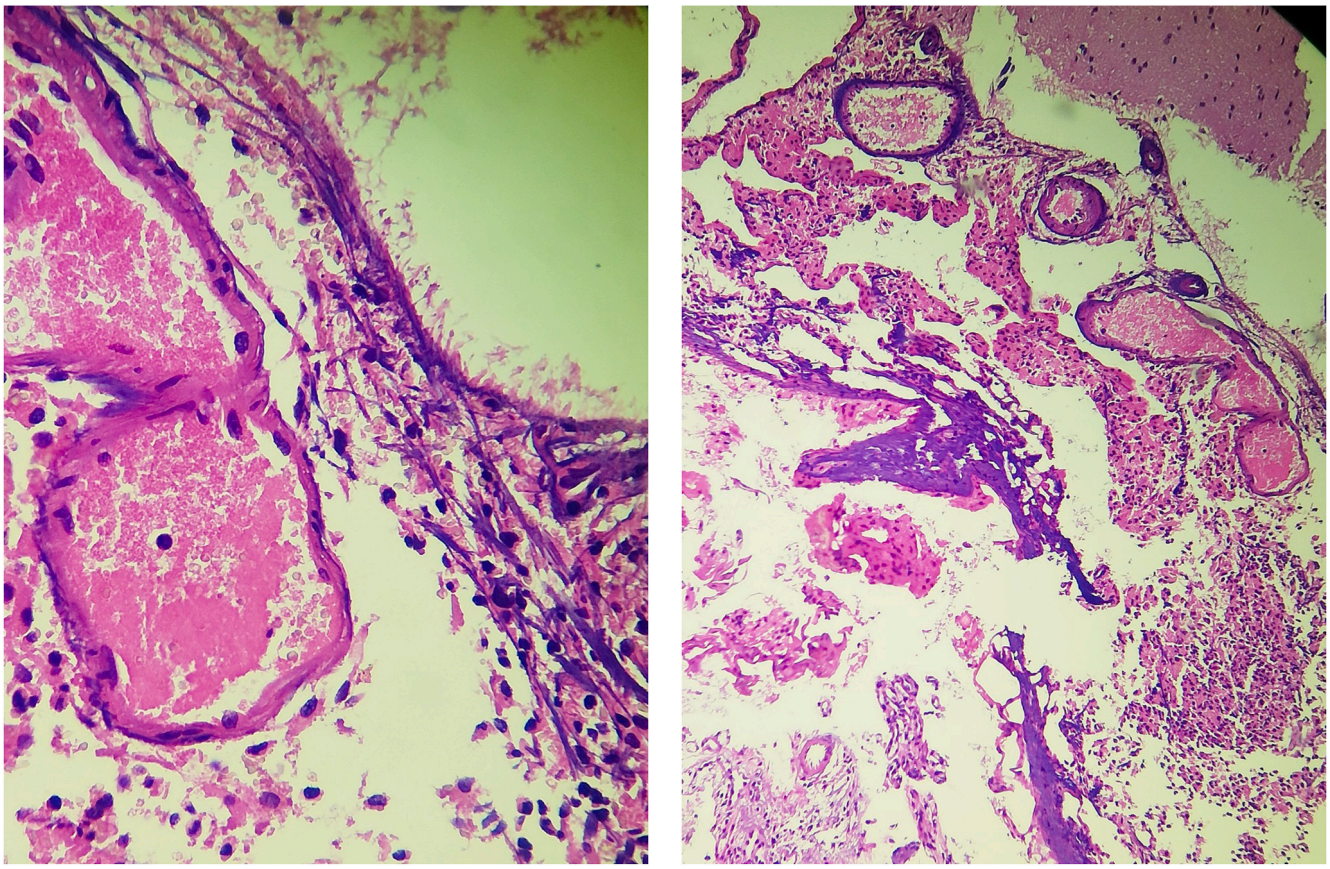

Fig. 3. Histopathological examination illustrated arterio-venous malformation.

large hematoma or acute hydrocephalus. In addition, preoperative embolization is extremely helpful for the presence of nidal or peri-nidal aneurysm, AVM with high grades, reducing intraoperative blood loss and occlusion of deep vessels, which are difficult to approach. Despite these treatments, the patients with ASSDH due to ruptured AVM still might suffer from severe postoperative complications (sepsis, septic shock, pneumonia), permanent neurological deficits (paralysis, diplopia), and even death [6]. Last but not least, stereotactic radiosurgery is reserved for the patients with deep, small AVMs (typically <3 cm) or for elderly patients with severe co-morbidities unable to tolerate general anesthesia. This is because radiosurgery requires a latency period between treatment and complete obliteration, which may last up to three years. Moreover, the proportion of hemorrhage during this latency period is similar to that of the natural history without treatment. In addition, there is no clear obliteration in the first six months after radiosurgery and no more obliteration beyond five years [17,18].

4. Conclusion

Acute spontaneous SDH due to ruptured AVM is quite rare and easy to omit in clinical settings. When a healthy patient was presented with an acute spontaneous SDH, prompt CTA or MRA should be considered initial screening tools when available to rule out a ruptured AVM. Preoperative embolization and surgical excision are the mainstays of treatment of ruptured AVMs.

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Ethical approval

The study was approved by the Research Ethics Committee of Hanoi Medical University. The procedures used in this study adhere to the tenets of the Declarations of Helsinki.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Credit author statement

Hung Dinh Kieu: Conceptualization, Methodology, Investigation, Writing – review & editing, Supervision; Tam Duc Le: Conceptualization, Methodology, Investigation, Writing – original draft, Writing – review & editing, Visualization, Data collection; Tan Minh Hoang: Investigation, Data collection, Writing – review & editing; Hung Dinh Kieu:have contributed equally to this work and Tam Duc Le have contributed equally to this work.

Research registration

Not applicable – this is a single case report, not a systematic review or meta-analysis. Moreover, we attest that it is not a 'first in man' study, either.

Guarantor

Tam Duc Le.

Table 1
Reported cases of acute spontaneous SDH secondary to ruptured AVM.

Author/ Year	Age (years)/ sex	Medical History	Clinical symptoms and signs	Radiological features	Treatment	Complications and Outcome
Hyuk Jin Choi et al., 2015 [8]	51, Male	Healthy	Headache, loss of conscious (GCS 5pts), dilatation of both pupils	CT: acute SDH with severe midline shift CTA and DSA: No	Emergent decompressive craniectomy: SDH evacuation and AVM excision	Glasgow Outcome Scale 3pts
Nor Fadhilah Madon et al., 2018 [6]	1, Male	Glucose-6-phosphate dehydrogenase (G6PD) deficiency	Seizures and loss of consciousness, GCS 6pts, the sluggish light reflex of both pupils. Hypertonia of all four limbs. Extensive bilateral retinal and pre-retinal hemorrhages.	CT: right SDH with severe cerebral edema Serial CT scans showed worsening of SDH and cerebral edema CTA and DSA: No	Resuscitation, no surgical intervention	Death 2 days later
Narendra Datta et al., 2000 [19]	48, Male	Healthy	A severe headache followed within minutes by a deep coma. GCS 3pts. The pupil size was small (2 mm), and the pupils were unresponsive. Then his pupils had dilated bilaterally. His limbs were flaccid when he was transferred to the operating room	CT: acute posterior fossa SDH associated with hydrocephalus CTA and DSA: No	External ventricular drain then suboccipital craniectomy. The external ventricular drain was maintained for 14 days until a ventriculoperitoneal shunt was placed	He was discharged after three months admission. Diplopia but no ataxia.
Matthew Parr et al., 2020 [7]	66, Male	Hypertension, hyperlipidemia, hepatitis C virus infection, and atrial fibrillation chronically anticoagulated with apixaban	After a fall, he had dizziness, GCS 15pts, no neurological deficit. Then his headache got worse, and focal left arm seizure occurred.	CT: a 6 mm right frontal SDH with no midline shift, as well as a right medial orbital wall fracture. Serial CT showed expansion of hematoma 15mm with an 8 mm midline shift. CT and CTA head: right frontal intraparenchymal hemorrhage measuring 5.2 cm × 3.3 cm but no vascular lesion or anomaly. Cerebral catheter angiography: Frontal AVM, Spetzler-Martin Grade 1 (Spetzler-Ponce Grade A)	First operation: Burr holes for evacuation of the SDH and placement of the subdural drain Second operation: Preoperative embolization and AVM resection.	Ventilator-dependent respiratory failure septic shock secondary to *Pseudomonas* bacteremia. Aspiration pneumonia. Paralysis of left upper or bilateral lower extremities.
Our case	21, Male	Healthy	Severe headache, GCS 15pts. No neurological deficits.	CT: A right acute subdural hematoma 8mm in thickness with 5mm midline shift and a right frontal intraparenchymal hemorrhage 40 × 25mm in size DSA: A right frontal AVM with 2 × 3 cm in size, Spetzler-Martin grade I.	Emergency preoperative embolization followed by AVM excision.	His headache was relieved and disappeared after a week. No postoperative neurological deficits.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Declaration of competing interest

None.

Acknowledgments

Nothing to declare.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.102613.

References

[1] B.A. Gross, R. Du, Natural history of cerebral arteriovenous malformations: a meta-analysis ; Clinical article, J. Neurosurg. 118 (2013) 437–443, https://doi.org/10.3171/2012.10.JNS121280.
[2] M. Korja, D. Bervini, N. Assaad, M.K. Morgan, Role of surgery in the management of brain arteriovenous malformations: prospective cohort study, Stroke 45 (2014) 3549–3555, https://doi.org/10.1161/STROKEAHA.114.007206.
[3] H. Kim, A.A. Abla, J. Nelson, C.E. McCulloch, D. Bervini, M.K. Morgan, C. Stapleton, B.P. Walcott, C.S. Ogilvy, R.F. Spetzler, M.T. Lawton, Validation of the supplemented Spetzler-martin grading system for brain Arteriovenous malformations in a Multicenter cohort of 1009 surgical patients, Neurosurgery 76 (2015) 25–31, https://doi.org/10.1227/NEU.0000000000000556.
[4] J.P. Mohr, M.K. Parides, C. Stapf, E. Moquete, C.S. Moy, J.R. Overbey, R.A. S. Salman, E. Vicaut, W.L. Young, E. Houdart, C. Cordonnier, M.A. Stefani, A. Hartmann, R. Von Kummer, A. Biondi, J. Berkefeld, C.J.M. Klijn, K. Harkness, R. Libman, X. Barreau, A.J. Moskowitz, Medical management with or without interventional therapy for unruptured brain arteriovenous malformations (Aruba): a multicentre, non-blinded, randomised trial, Lancet 383 (2014) 614–621, https://doi.org/10.1016/S0140-6736(13)62302-8.
[5] B.A. Gross, R. Du, Natural history of cerebral arteriovenous malformations: a meta-analysis ; Clinical article, J. Neurosurg. 118 (2013) 437–443, https://doi.org/10.3171/2012.10.JNS121280.

[6] N. Madon, A. Hasmi, K. Zainun, H. Nawawi, Spontaneous subdural hemorrhage due to ruptured arteriovenous malformation in a child, J. Forensic Sci. Med. 4 (2018) 174–178, https://doi.org/10.4103/jfsm.jfsm_93_17.
[7] M. Parr, N. Patel, J. Kauffmann, F. Al-Mufti, S. Roychowdhury, V. Narayan, M. Nosko, A. Nanda, G. Gupta, Arteriovenous malformation presenting as traumatic subdural hematoma: a case report, Surg. Neurol. Int. 11 (2020), https://doi.org/10.25259/SNI_160_2019.
[8] H.J. Choi, J. Il Lee, K.H. Nam, J.K. Ko, Acute spontaneous subdural hematoma due to rupture of a tiny cortical arteriovenous malformation, J. Korean Neurosurg. Soc. 58 (2015) 547–549, https://doi.org/10.3340/jkns.2015.58.6.547.
[9] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, for the SCARE Group, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230.
[10] K. Yamauchi, S. Takenaka, T. Iida, H. Sakai, A case of spontaneous acute subdural hemorrhage caused by a dural arteriovenous fistula on the convexity without cortical venous reflux, Case Rep. Neurol. 11 (2019) 312–318, https://doi.org/10.1159/000504290.
[11] G. Li, Y. Zhang, J. Zhao, X. Zhu, J. Yu, K. Hou, Isolated subdural hematoma secondary to Dural arteriovenous fistula: a case report and literature review, BMC Neurol. 19 (2019) 1–6, https://doi.org/10.1186/s12883-019-1272-z.
[12] D. Kondziolka, M. Bernstein, K. ter Brugge, H. Schutz, Acute subdural hematoma from ruptured posterior communicating artery aneurysm, Neurosurgery 22 (1988) 151–154, https://doi.org/10.1227/00006123-198801010-00029.
[13] T.M. Keller, E.T. Chappell, Spontaneous acute subdural hematoma precipitated by cocaine abuse: case report, Surg. Neurol. 47 (1997) 12–14, https://doi.org/10.1016/S0090-3019(96)00380-1.
[14] S. Okuno, H. Touho, H. Ohnishi, J. Karasawa, Falx meningioma presenting as acute subdural hematoma: case report, Surg. Neurol. 52 (1999) 180–184, https://doi.org/10.1016/S0090-3019(97)00028-1.
[15] B. Depreitere, F. Van Calenbergh, J. Van Loon, A.D. Mendelow, A clinical comparison of non-traumatic acute subdural haematomas either related to coagulopathy or of arterial origin without coagulopathy, Acta Neurochir. 145 (2003) 541–546, https://doi.org/10.1007/s00701-003-0020-7.
[16] B.E. Zacharia, K.A. Vaughan, A. Jacoby, Z.L. Hickman, D. Bodmer, E.S. Connolly, Management of ruptured brain arteriovenous malformations, Curr. Atherosclerosis Rep. 14 (2012) 335–342, https://doi.org/10.1007/s11883-012-0257-9.
[17] H. Kano, J.C. Flickinger, H.C. Yang, T.J. Flannery, D. Tonetti, A. Niranjan, L. D. Lunsford, Stereotactic radiosurgery for Spetzler-Martin Grade III arteriovenous malformations, J. Neurosurg. 120 (2014) 973–981, https://doi.org/10.3171/2013.12.JNS131600.
[18] H. Kano, L.D. Lunsford, J.C. Flickinger, H.C. Yang, T.J. Flannery, N.R. Awan, A. Niranjan, J. Novotny, D. Kondziolka, Stereotactic radiosurgery for arteriovenous malformations, Part 1: management of Spetzler-Martin Grade I and II arteriovenous malformations: clinical article, J. Neurosurg. 116 (2012) 11–20, https://doi.org/10.3171/2011.9.JNS101740.
[19] N.N. Datta, K.Y. Chan, J.C. Kwok, C.Y. Poon, Posterior fossa subdural hematoma due to ruptured arteriovenous malformation, Case report., Neurosurg. Focus. 8 (2000), https://doi.org/10.3171/foc.2000.8.6.10.

Abbreviations

ASDH: Acute subdural hematoma
ASSDH: Acute spontaneous subdural hematoma
AVM: Arteriovenous malformation
CT: Computed tomography
CTA: Computed tomography angiography
DSA: Digital subtraction angiography
GCS: Glasgow coma scale
MRI: Magnetic resonance imaging