CASE REPORT

Spontaneous Subdural Haematoma as a Rare Presentation of Ruptured Vascular Lesions: A Case Report of Three Patients

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ABSTRACT

Background: Spontaneous acute subdural hematoma (sASDH) is a rare presentation of aneurysmal and other vascular malformation rupture. There are only few cases reported in the literature, especially in the absence of SAH. The annual haemorrhagic risk is estimated at 1–3% for unruptured AVMs and 4–6% for those ruptured. We share three rare cases to highlight the significance of initial early vascular studies in unique clinical presentations and demographics of resource limited settings such to prompt new management protocols.

Case presentation: The first patient was a 60-year-old female with an unremarkable medical history who presented with a ruptured internal carotid-posterior communicating artery (ICA-PcomA) junction aneurysm and a temporal lobar intracerebral hematoma (ICH). Craniotomy with hematoma evacuation and aneurysm clipping was performed in a single surgery. She recovered with minimal neurological deficit.

The second patient was a 66-year-old female, known hypertensive with poor compliance who presented with ruptured anterior communicating artery (AcomA) aneurysm. She had a frontal lobe hematoma, Fischer 3 SAH, and multiple haemorrhagic lesions involving the brainstem. Unfortunately, she was not a good surgical candidate and therefore managed conservatively in the acute period.

The third patient was a 40-year-old male who presented with an extensive spontaneous subacute subdural hematoma. He underwent craniotomy and hematoma evacuation when a small cortical AVM was incidentally found intraoperatively and scheduled for delayed resection after CT angiography.

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Conclusions: Spontaneous acute subdural hematoma is a rare presentation of a ruptured vascular malformation and often under diagnosed in resource limited settings where non-contrasted CT brain scans are first imaging of choice in the acute emergency setting. The reported cases serve as a reminder to the neurosurgeon, neurologist, radiologist and other relevant clinical specialists the need for adequately initial radiological investigation of patients with suspected spontaneous SDH. Additionally, CT angiography should be considered in future protocols for all patients presenting with signs of acute nontraumatic SDH to exclude vascular malformations. However, in cases where angiography is not readily available, emergency haematoma evacuation should not be delayed if indicated.

INTRODUCTION

Vascular malformations are relatively rare disorders associated with a rupture mortality of about 50% and a 30%–50% neurologic morbidity rate among the survivors.[1, 2] The typical presentation of a ruptured aneurysm is subarachnoid haemorrhage (SAH) with or without Intracerebral haematoma (ICH), depending on the location of the aneurysm. Acute subdural hematoma (ASDH) is a rare presentation. ASDHs are commonly seen in traumatic brain injury, with a mortality rate of up to 65%. Spontaneous ASDH is rarer, occurring in 0.7% to 4.9% of cases.[3, 4] In these cases, ASDH is a complication of aneurysmal SAH and occurs between 0.9% and 5.8%.[4] Pure ASDH without radiological features of SAH is seen in only 0.1%–2.9% of patients with a ruptured intracranial aneurysm. Missori *et al.* reported that the most significant factors for a good outcome were early surgical treatment, a high Glasgow Coma Scale (GCS) score on admission, good papillary reactivity, and a younger age in a review of 82 patients, showed that urgent surgical decompression and immediate occlusion of the aneurysm seem to be acceptable treatment strategies in order to achieve a better outcome.[5] However, two-stage management with immediate decompression and delayed coiling has also been reported with good recovery. The pathophysiology of spontaneous SDH includes rupture of the cortical artery or of the pial arteriovenous fistulae, an arteriovenous malformation (AVM), or intracranial aneurysm, as noted by Hyuk Jin Choi *et al*.

Furthermore, Torne *et al*. and Schuss *et al*., concluded that despite the poor initial condition and the high mortality rate of comatose ASDH patients in the acute phase, the prognosis of survivors would not be worse than patients with SAH alone who also present with coma. We therefore present three cases of ASDH associated with vascular malformations.

CASE PRESENTATIONS

Case 1

A 60-year-old woman was taken to the hospital after losing consciousness while on public transportation. She had no history of trauma, and her past medical history was unremarkable. On arrival, her GCS was 7. She was admitted to the ICU and intubated. Neurological examination showed anisocoria with left mydriasis. A brain CT scan was done on arrival, which showed a left-sided subdural hematoma and a small temporal lobe intraparenchymal hematoma. There was no obvious connection between the subdural and intraparenchymal hematomas (**Fig. 1**).

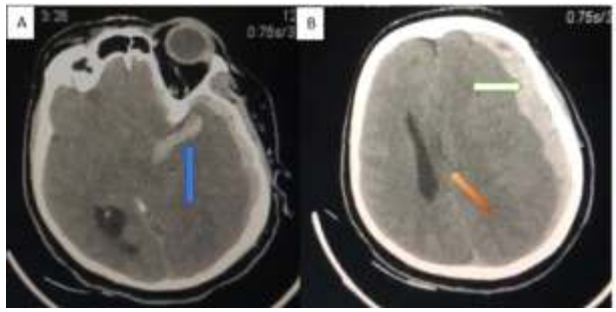

Figure 1 Initial CT showing intraparenchymal haemorrhage (blue arrow), subdural hematoma with no features of subarachnoid haemorrhage (white arrow). There was almost complete left lateral ventricle compression with >10mm midline shift.

There were no CT features of subarachnoid haemorrhage (SAH). Based on the non-traumatic history, a CT angiogram was performed, and an

aneurysm at the ICA-PcomA junction was identified (**Fig. 2**).

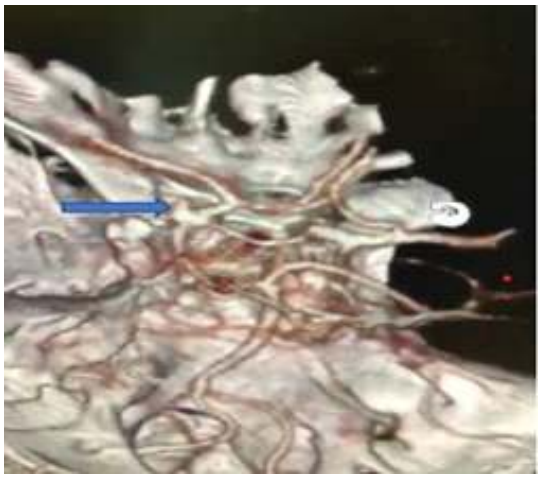

Figure 2 CT angiography shows an aneurysm at the ICA-Pcom junction (blue arrow).

A craniotomy was performed after patient stabilization. About 130mls of ASDH was evacuated, and the aneurysm was clipped in the same sitting. The aneurysm ruptured intraoperatively with minimal manipulation after evacuation of the hematoma, which was easily controlled. There were no features of SAH. The temporal intraparenchymal haematoma was in close proximity with the haematoma, and it was evacuated as well (**Fig. 3**). A control CT showed acceptable postoperative changes. The postoperative period was unremarkable, and the patient recovered with minimal neurological deficit.

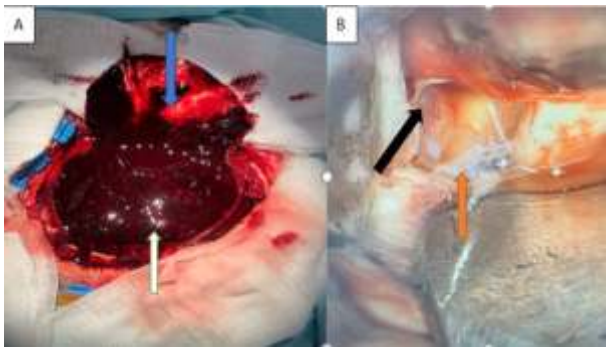

Figure 3 Intraoperative picture of the acute subdural hematoma during evacuation (white arrow) with the dura reflected inferiorly (blue arrow). The aneurysm was clearly visualized (black arrow) with adhesions showing evidence of previous microhaemorrhage (orange arrow).

Case 2

A 66-year-old woman was brought to the hospital by ambulance after being found unconscious at home. She was a known hypertensive with poor compliance. En route to the hospital, she regained consciousness and was able to answer questions in monosyllables. She had repeated episodes of projectile vomiting followed by loss of consciousness. She had no stigmata of trauma. At the time of the neurological evaluation, she had anisocoria, a GCS of 7, and an NIHSS of 30pts. An emergency CT scan revealed a 9-10mm thick right-sided subdural hematoma with 10mm midline shift, a right frontal intraparenchymal haematoma (IPH) with Fischer 3 SAH, a 9x20mm haemorrhagic lesion in the brainstem, and a 5mm petechial haemorrhage in the corpus callosum area. CT angiography revealed an aneurysm of the anterior communicating artery (AcomA) measuring 3.7 x 3.4 mm (**Fig. 4**).

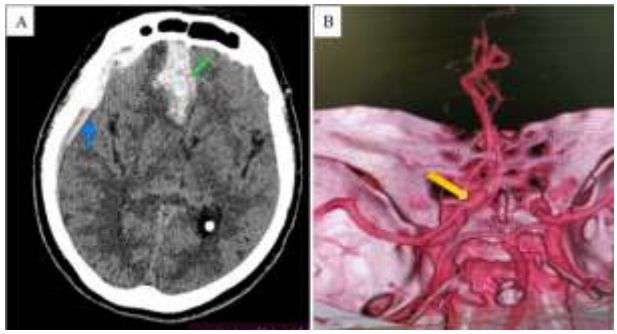

Figure 4: A) Initial CT showing an aSDH (blue arrow) and the typical frontal lobe hematoma (green arrow). B) CT angiography shows an anterior communicating artery (AcomA) aneurysm.

The patient was intubated and admitted to the ICU, where she developed aspiration pneumonia, confirmed by chest CT. Considering the CT findings and other comorbidities, surgery was contraindicated, and the patient was managed expectantly.

Case 3

Male 40 years old presented with one week history of a fall and no prodromal signs or prior seizure. He subsequently developed confusion, amnesia, with aphasia and vomiting that persisted three days later. His past medical history was unremarkable and denied smoking and alcohol abuse. On physical examination, GCS was 14, aphasia but no lateralizing signs or focal neurological deficits.

Initial urgent plain CT brain imaging revealed a left fronto-temporo-parieto-occipital crescent shaped heterogeneously mixed density: low density anteriorly and high density posteriorly, distinct margins, with early ensuing loculations consistent with an evolving chronic subdural from separated subtype to multiloculated laminar subtype. Its thickness 11mm and midline shift 5mm (F**ig 5**).

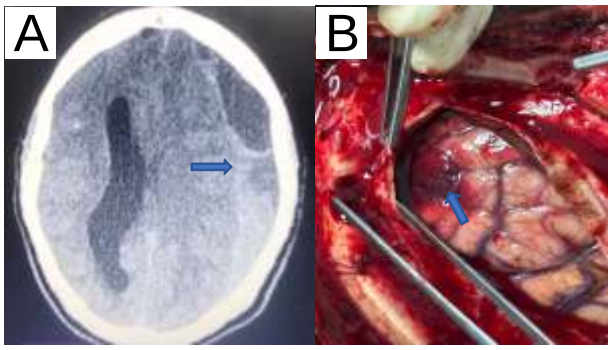

Figure 5: A) Non contrasted CT brain with left heterogeneous laminar multiloculated subdural collection. B) Intraoperative image with arrow showing small cortical arteriovenous malformation as likely cause of the evacuated spontaneous subdural hematoma.

A standard craniotomy and hematoma evacuation was done. Intraoperative findings included mildly tense dura, layered membranes, engine oil fluid mixed with thick dissolving blood. About 60mls was evacuated and membranectomy done with poor compliance to expansion of the brain.

A small < 3cm size cortical arteriovenous malformation was incidentally identified on the surface of the brain with clearly visible feeding arteries and draining veins as well as en-passant vessels as above (F**ig 5**)

Decision was made to consider resection after adequate preoperative preparation with CT angiographic vascular studies.

Informed consent

Informed consent was obtained from the caregiver.

DISCUSSION

The above three cases presented with aneurysms, i.e., the ICA-PcomA junction, AcomA and cortical aneurysms. The acute SDH hematomas complicating aneurysms have been seen frequently in the ICA system, primarily affecting the PcomA and rarely in the vertebrobasilar system. This is because the Lilliquist membrane covers the basilar system and does not rupture easily.[6] However, Biesbroek *et al*., suggested that the presentation of an aneurysmal rupture is not only dependent on location but also its anatomy and perianeurysmal environment.[7] Aneurysmal rupture may result in disruption of the arachnoid membrane and subsequent ASDH and/or SAH when an aneurysm protrudes into the basal cisterns with associated adhesions from previous sentinel haemorrhages. This might explain the presentation of the first case where adhesions between the aneurysm and the arachnoid membrane were seen intraoperatively. Conversely, aneurysm rupture into the subarachnoid space with an intact arachnoid membrane result in purely acute SDH without SAH.

Other mechanisms include: 1) adherence of the aneurysm to the falx or dura with resultant subdural haemorrhage, 2) distal vascular lesions in the cortical areas with accompanying rupture of arachnoid membrane, 3) a high systolic pressure causing subpial and subarachnoid vascular rupture into respective spaces eroding the sinus wall.[6,7]

Associated ICH is seen in 24-27% of cases of aneurysmal rupture.[8] However, in some cases, there is no link between ICH and ASDH or absent SAH, as seen in Case 1. Isolated ASDH is extremely rare, with only a few cases reported in the literature. It is linked to a better long-term outcome because the absence of SAH prevents the development of

complications such as vasospasms and hydrocephalus.

Initial CT scans were performed in the cases presented above and clearly showed the ASDH. Subsequent evaluation with CT angiography was performed and proved invaluable in visualizing the aneurysm. This was emphasized by Marbacher *et al*., who concluded that a contrast CT scan and/or 3-dimensional CT angiography done upon presentation are useful to exclude cerebral aneurysm and vascular malformation.[9] This is especially important as some cases of ruptured aneurysms may not present with SAH or ICH typical of aneurysm rupture, leading to a missed diagnosis. In these cases, decompression without managing the vascular disorder may be detrimental. However, some authors have argued that there is no significant change in transmural pressure to cause intraoperative bleeding and absence of angiography should not delay emergency haematoma evacuation if indicated.

In a systematic review of 85 cases of spontaneous ASDH Akioka *et al*., proposed that early surgical intervention was important even if Glasgow coma scale had been above 12.[10] This is because the acute bleeding of arterial origin results in higher risk of rapid haematoma enlargement.[11] Our first patient recovered well with mild neurological deficits. Unfortunately, due to the brainstem haemorrhage and severe aspiration pneumonia, our second patient was not a good candidate for surgery. The third patient showed marked recovery with resolution of focal deficits.

Additionally, most patients with CSDH present in geriatric age group having either history of trivial trauma, chronic illness or alcohol abuse not consistent with case 3 above. Awareness of recurrent spontaneous CSDH in young adults caused by cortical AVM warranting an urgent preoperative CTA and high index of suspicion.

CONCLUSIONS

Spontaneous acute subdural haematoma is a rare presentation of aneurysmal rupture. As a result, it is usually a missed diagnosis. These cases serve as a reminder to the neurosurgeon, neurologists, radiologists and other surgical specialists to adequately evaluate patients with spontaneous ASDH. In addition, CT angiography should be performed in all patients presenting with non-traumatic ASDH to exclude bleeds from vascular lesions. However, in cases where angiography is not readily available, emergency haematoma evacuation, if indicated, should not be delayed. A multidisciplinary approach would prompt formation of protocols that will ultimately enhance patient care.

Key points:

1. Spontaneous acute subdural hematoma should be investigated fully with angiography prior to surgery if the patient is stable.
2. The absence of angiography should not delay hematoma evacuation and AVM resection if indicated.
3. The operating surgeon must be aware of the risk of encountering an aneurysm or AVM and prepare accordingly.

Competing interests

The authors declare that they have no competing interests.

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Authors' contributions

All authors read and approved of the final manuscript.

List of Abbreviations

ASDH- Acute subdural haematoma
CSDH- Chronic subdural hematoma
AVM- Arteriovenous malformation
AcomA- Anterior communicating artery
PcomA- Posterior communicating artery
ICA- Internal carotid artery
ICH- Intracerebral hematoma
SAH- Subarachnoid haemorrhage

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