CASE REPORT

Remote multifocal bleeding points producing a Sylvian subpial hematoma during endovascular coiling of an acutely ruptured cerebral aneurysm

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Accepted 2 November 2016

SUMMARY

Subpial hematoma is a rare type of intracranial hemorrhage with a poor prognosis. A new mechanism to explain subpial Sylvian hematoma formation in acute post-aneurysmal subarachnoid hemorrhage (SAH) was observed during coiling of a posterior communicating artery aneurysm. Multiple small bleeding points from pial branches of the left middle cerebral artery, which were remote from the ruptured aneurysm, were observed on conventional angiography. This bleeding led to the formation of a large expanding subpial Sylvian hematoma. Similar observations have been recently demonstrated on 4D CT angiography (4D CTA). We present a case that adds evidence to the literature in support of a new mechanism of subpial hematoma formation in the setting of acute SAH. This may advocate the early use of 4D CTA and conveys a poor prognosis, which might influence treatment decisions.

BACKGROUND

Subpial hematomas are a rare type of intracerebral hemorrhage with an unclear mechanism and poor prognosis.^{1–4} Previously Sylvian hematoma in subarachnoid hemorrhage (SAH) has been attributed to aneurysm rupture.⁴ Recent case reports using 4D CT angiography (4D CTA)^{5–6} suggest an additional mechanism for the development of Sylvian

hematomas in patients with acute post-aneurysmal SAH that is separate from aneurysmal rebleeding. We present a case of unilateral Sylvian hematoma secondary to multifocal bleeding points detected on conventional angiography during coiling of an acutely ruptured posterior communicating (PComm) artery aneurysm. To our knowledge, this is the first demonstration using conventional angiography of Sylvian subpial hematoma formation secondary to multifocal bleeding points from small pial branches of the left middle cerebral artery (MCA).

CASE PRESENTATION

A 59-year-old woman presented to her local hospital with acute onset of severe headache. Her past medical history included diabetes mellitus, hepatitis C infection and paraproteinemia.

Initial unenhanced CT of the head (figure 1A), performed 2 hours after presentation, demonstrated acute SAH with intraventricular extension. Initial Glasgow Coma Scale score was 15. While awaiting transfer to her local neurosurgical unit, her Glasgow Coma Scale score decreased to 5 and repeat CT of the head indicated a rebleed (figure 1B). On arrival at our institution, the patient underwent insertion of an external ventricular drain and intracranial pressure (ICP) monitoring

Figure 1 (A) Initial presentation plain CT of the head demonstrating diffuse subarachnoid hemorrhage with intraventricular extension but without a large focal hematoma. (B) Repeat of the CT head following a decrease in Glasgow Coma Scale score demonstrating a slight increase in blood load.

To cite: Hilditch CA,
Sonwalkar H,
Wuppalapati S. *BMJ Case
Rep* Published online:
[please include Day Month
Year] doi:10.1136/bcr-2016-
012715

Figure 2 (A) CT cerebral angiogram volume rendered image demonstrating the left posterior communicating artery (PComm) aneurysm and large pseudoaneurysm (labelled). (B–D) Sagittal, axial, and coronal reformats showing a developing left Sylvian hematoma (arrows).

bolt, and was kept sedated and ventilated overnight on the intensive care unit.

CT cerebral angiogram performed during the night showed an acutely ruptured left PComm aneurysm with a large pseudoaneurysm (figure 2A). The CT angiogram also showed a moderate sized left Sylvian hematoma (figure 2B–D). Although the patient's overnight ICP was unstable, it was decided to undertake urgent coil embolization of the aneurysm to prevent rebleed.

TREATMENT

At the start of the coiling procedure, 5000 units of heparin were administered to limit thromboembolic complications. The first angiogram (figure 3) demonstrated the left PComm aneurysm and large pseudoaneurysm but did not show any gross abnormality of the left Sylvian MCA branches. During coiling a check angiogram revealed multiple small bleeds from pial branches of the left MCA (figure 4). On table CT (figure 5) showed the left Sylvian hematoma had hugely expanded with contrast extravasation. Heparin was immediately reversed. Her pupils became fixed and dilated and the procedure was abandoned.

OUTCOME AND FOLLOW-UP

The patient unfortunately died shortly after coiling.

DISCUSSION

It has been suggested that subpial Sylvian hematoma in the setting of acute SAH usually occurs due to rupture of MCA aneurysms.⁷ 4D CTA is a new imaging modality which involves multiple CTA acquisitions or a continuous volume acquisition for a period of time.⁸ Recently, a few case reports have identified multifocal extravasation sites from subpial MCA branches resulting in Sylvian hematoma formation, and to our knowledge, this is the first demonstration of such bleeding using conventional angiography.

Sylvian hematomas may arise secondary to bleeding into the subpial space rather than the subarachnoid space, and an expanding hematoma in this location can cause severe damage to the brain.^{5–9} When an aneurysm adherent to the pia mater bleeds, the blood may spread below the pia and the expanding hematoma may rupture the thin arteries that run in the pial space.⁸ We believe that this is what we visualized during angiography and has been previously demonstrated with dynamic CTA.^{5–6} This bleeding may have been accentuated by the use of

Figure 3 Early arterial phase angiogram. Precoiling angiogram shows the left posterior communicating artery aneurysm and large pseudoaneurysm. The aneurysm was not actively bleeding, and no active bleeding from the Sylvian middle cerebral artery branches was identified at this time.

Figure 5 On-table CT during the procedure demonstrates dramatic expansion of the left Sylvian hematoma with contrast extravasation.

Figure 4 Anteroposterior and lateral check angiogram during coiling. Early arterial, late arterial, and delayed phases (labels) show multiple small bleeding points (arrows) from small pial branches of the left Sylvian middle cerebral artery.

intraoperative heparin and nimodipine flushes attached to guide catheters. Proinflammatory mediators released following SAH¹⁰ may also have played a role. The sequence of events described suggest intermittent hemorrhaging from these small bleeding points from the time of presentation up to the point of coiling.

This case shares some similarities with the few cases previously published.^{5–6} Our patient was of Chinese ethnicity and the previous cases have been published by Japanese authors, hinting at a possible genetic contribution. A past history of diabetes has also been reported.⁶ Three of the previously reported cases had either MCA aneurysms or PComm aneurysms,⁶ and location of the ruptured aneurysm close to the Sylvian fissure may result in blood tracking into the subpial space and triggering a cascade of events leading to hematoma formation.

It has been suggested that judicious reduced doses of anticoagulants should be used when coiling acutely ruptured aneurysms when concomitant Sylvian hematomas are present.⁶ Our standard practice for acute coiling cases is to administer 5000 units of heparin at the start of the procedure and maintain activated clotting time at 2–3 times the base level. Although we do not feel that the use of heparin and nimodipine caused the multifocal bleeding, they may have contributed to hematoma expansion. There is no agreed consensus regarding anticoagulation for coiling of acutely ruptured cerebral aneurysms. Some interventionists advocate delaying heparin administration until the first framing coil is deployed. Our patient did have a large pseudoaneurysm, evident on the pre-coiling CTA and initial angiography, and this indicated an increased risk of intraoperative rupture. Reduced or split boluses of heparin to maintain a lower target activated clotting time level may be appropriate in such instances. Our patient had recently been investigated for paraproteinemia although routine blood parameters were normal.

The primary alternative to endovascular coiling is surgical clipping. Clipping may have been a good option in this case given the patient's unstable ICP and presence of a hematoma. The Sylvian hematoma seen on the pre-coiling CTA could have been evacuated at the same time with the additional option of decompressive craniectomy. Open surgery would have also negated the use of anticoagulation. We do not feel that surgical clipping would have prevented hematoma formation or expansion in this case, given the most likely underlying pathological mechanism described above.

This case adds evidence to the literature regarding a different pathogenesis for expanding subpial Sylvian hematoma in patients with acute SAH, other than aneurysm rebleed. The angiographic images shown here are, to our knowledge, unique and have not been presented previously. Multifocal bleeding in SAH patients is a rare phenomenon and clearly carries a very

poor prognosis. We attempted to coil this patient's aneurysm but the gesture ultimately proved futile, and recognition of an expanding Sylvian hematomas in the setting of acute SAH may advocate the use of early 4D CTA and guide decisions to treat.

Learning points

- ▶ Subpial hematoma has a poor prognosis.
- ▶ Expanding Sylvian hematoma in the setting of acute subarachnoid hemorrhage may be secondary to damage to small pial branches of the Sylvian middle cerebral artery. This bleeding may be separate to and remote from the original aneurysm rupture.
- ▶ These small bleeding points can be demonstrated with conventional angiography as well as 4D CT angiography.

Contributors CAH wrote the case report and reviewed the relevant literature. HS helped to write the case report. SW and CAH performed the coiling procedure.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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