

CASE REPORT

Spontaneous subarachnoid hemorrhage due to ruptured cavernous internal carotid artery aneurysm after medical prolactinoma treatment

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SUMMARY

Aneurysms of the cavernous segment of the internal carotid artery (ICA) are believed to have a low risk of subarachnoid haemorrhage (SAH), given the confines of the dural rings and the anterior clinoid process. The risk may be greater when the bony and dural protection has been eroded. We report a case of spontaneous SAH from rupture of a cavernous ICA aneurysm in a patient whose large prolactinoma had markedly decreased in size as the result of cabergoline treatment. After passing a balloon test occlusion, the patient underwent successful endovascular vessel deconstruction. This case suggests that an eroding skull base lesion may distort normal anterior cranial base anatomy and allow communication between the cavernous ICA and subarachnoid space. The potential for SAH due to cavernous ICA aneurysm rupture should be recognised in patients with previous pituitary or other skull base lesions adjacent to the cavernous sinus.

BACKGROUND

Incidentally noted aneurysms of the cavernous segment of the internal carotid artery (ICA) are managed conservatively because of a low risk of rupture. The 5-year cumulative rupture rate for carotid cavernous aneurysms is 0% for aneurysms with a diameter of 12 mm or less, 3% for 13–24 mm lesions and 6.4% for lesions 25 mm or larger.¹ In addition, the intradural and extradural portions of the cavernous carotid are separated by a distal dural ring that is adherent to the ICA. This distal dural layer serves as a barrier to extension of blood into the subarachnoid space after rupture of a cavernous ICA aneurysm. Thus, rupture of a cavernous ICA aneurysm typically leads to carotid–cavernous fistula formation rather than subarachnoid hemorrhage (SAH), thereby conferring a lower mortality.^{2–3} There are, however, multiple case reports of SAH from cavernous ICA aneurysms.^{4–8} In one report,⁴ SAH occurred after the dura was opened during a craniotomy to treat a cavernous ICA aneurysm, which coexisted with a prolactinoma under cabergoline treatment; the authors suggested that an aperture to the subarachnoid space was created by shrinkage of the prolactinoma by cabergoline. We report a case of spontaneous SAH due to rupture of a cavernous ICA aneurysm in a patient whose large prolactinoma had shrunk following cabergoline treatment.

CASE PRESENTATION

A 61-year-old man with no significant past medical history presented in 2008 with headache and vision loss in the right eye. Head CT revealed a large sellar mass with a maximum dimension of 6 cm. Brain MRI (figure 1) revealed a large enhancing multilobulated sellar mass and a 1.2 cm aneurysm of the cavernous segment of the right ICA. The prolactin level at initial presentation was >22 000 ng/mL, prompting initiation of cabergoline therapy. Two years later the prolactin level had decreased by 99% and imaging showed a 50% reduction in size of the prolactinoma (figure 2). The cavernous ICA aneurysm had remained stable in size and no intervention was recommended.

In 2014, 6 years after initial presentation, the patient reported a 4-month decline in vision in his right eye. CT revealed further reduction in size of the prolactinoma. CT angiography revealed that the right cavernous ICA aneurysm was partially thrombosed and now measured 3.6 cm with a 2.8 cm intraluminal portion (figure 3).

Over the next year the patient noted further vision loss in his right eye. Neuro-ophthalmological examination confirmed a worsening right optic neuropathy, which progressed from 20/80 to 20/100 best-corrected visual acuity in the right eye. Other aspects of the neuro-ophthalmological examination, including eye movements and alignment, were normal. MR angiography revealed further enlargement of the aneurysm and no change in the appearance of the prolactinoma. Accordingly, vision loss was attributed to the enlarging aneurysm. Endovascular treatment of the aneurysm with

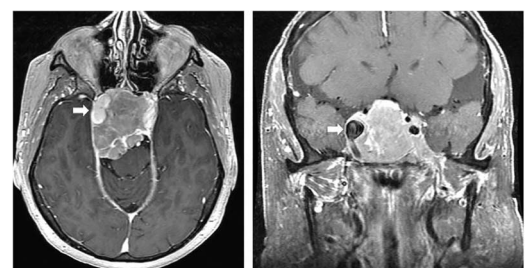


Figure 1 Post-contrast axial (left) and coronal (right) T1-weighted MRIs performed at initial presentation shows a 6 cm enhancing sellar mass considered to be a prolactinoma, based on a very high prolactin level. There is also a 1.2 cm aneurysm of the cavernous segment of the right internal carotid artery (arrows).



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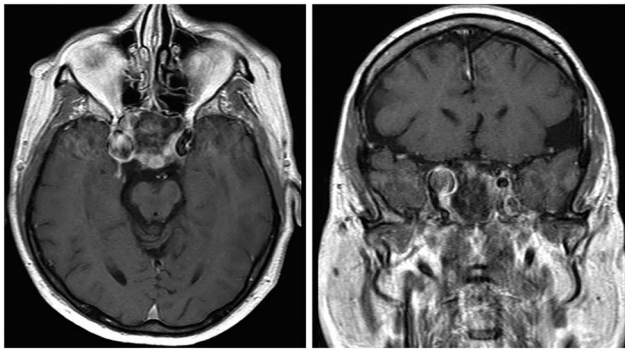


Figure 2 Post-contrast axial (left) and coronal (right) T1-weighted MRIs, obtained after 2 years of oral cabergoline therapy, show reduction in size of the prolactinoma. The cavernous internal carotid artery aneurysm is stable in size.

a flow remodeling device was recommended, but the patient declined pre-procedural angiography because of perceived risks.

Six months later the patient developed a sudden severe headache, right upper lid ptosis, and diplopia. Examination revealed a fixed mid-dilated right pupil with impaired supraduction, infraduction and adduction, consistent with a new right third cranial nerve palsy. Right eye visual acuity was limited to light perception only. CT revealed SAH within the sella that extended into the suprasellar and prepontine cisterns (figure 4). MRI confirmed enlargement of the cavernous ICA aneurysm (figure 5). Angiography (figure 6) of the right ICA demonstrated a fusiform bilobed cavernous aneurysm that measured 4.3 cm in the greatest transverse dimension by 2.3 cm in the craniocaudal dimension. The supraclinoid segment of the ICA above the level of the clinoid process was normal. Bony erosion at the level of the dural ring was evident on CT bone windows (figure 7).

TREATMENT

Balloon test occlusion, including hypotensive challenge, was tolerated by the patient. Given the ectatic and dysplastic nature of this vessel and the aneurysm, he underwent vessel deconstruction with a combination of coils (Axium and Concerto, Medtronic, Minneapolis, Minnesota, USA and Target, Stryker, Kalamazoo, Michigan, USA), MVD microvascular plug (Medtronic), as well as Onyx (Medtronic). There were no complications.

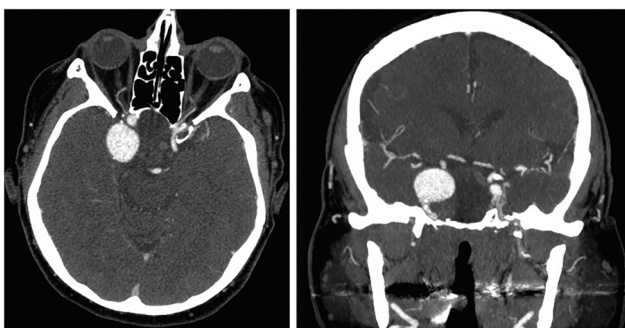


Figure 3 CT angiograms in axial (left) and coronal (right) planes obtained 6 years after initial presentation show interval enlargement of the partially thrombosed aneurysm of the right cavernous internal carotid artery.

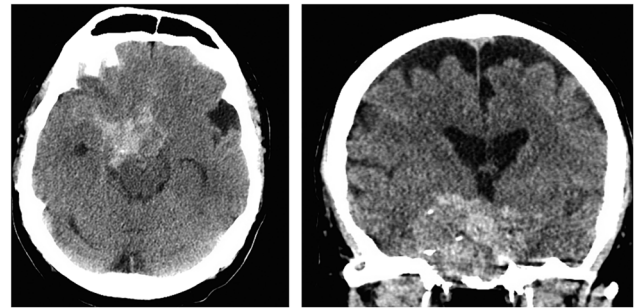


Figure 4 Axial (left) and coronal (right) non-contrast head CTs performed at the time of acute headache and right third cranial nerve palsy show subarachnoid hemorrhage surrounding the aneurysm and extending into the suprasellar and prepontine cisterns, along with the sylvian and interhemispheric fissures.

OUTCOME AND FOLLOW-UP

Neuro-ophthalmologic examination 4 months later revealed that vision had improved to pre-rupture levels, with 20/40 corrected visual acuity in the right eye. The third cranial nerve palsy had fully resolved.

DISCUSSION

Our patient developed a spontaneous SAH from a cavernous ICA aneurysm that appeared radiographically to be entirely extradural. Prior to the rupture the aneurysm had grown and the adjacent pituitary tumour had shrunk after cabergoline therapy.

Case reports have documented SAH from ruptured cavernous ICA aneurysms with a radiographically extradural appearance.⁴⁻⁸ Several mechanisms have been proposed. Dural erosion from giant intracavernous aneurysms has been reported during autopsy⁷ and craniotomy,⁶ although intracavernous aneurysms as small as 1.5 cm have been associated with SAH.⁹ Dural erosion is more likely to occur in cases of giant aneurysms, aneurysms arising from the anterior genu of the carotid siphon and those eroding into the sella turcica.³ In a case of SAH from a cavernous aneurysm without evidence of dural erosion, a loose dural ring was proposed as the mechanism of SAH.⁸ SAH may also occur with carotid cave aneurysms which arise from the ventromedial portion of the proximal intradural ICA and extend into a pouch of subarachnoid space within a more loosely adherent medial aspect of the distal dural ring.¹⁰

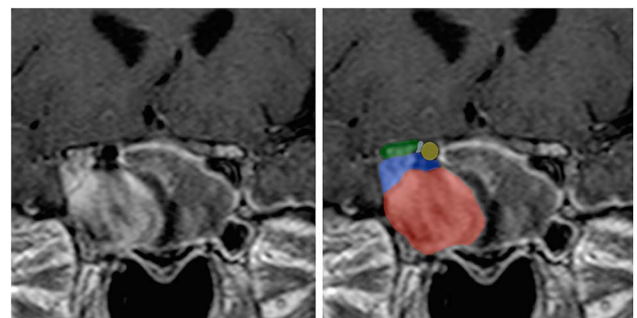


Figure 5 Post-contrast coronal MRI (left) performed at the time of aneurysm rupture confirms cavernous location of the internal carotid artery (ICA) aneurysm. Identical MRI slice (right) with colour labels (green, anterior clinoid; light blue, optic nerve; yellow, ICA distal to aneurysm; blue, displaced cavernous sinus contents; red, partially thrombosed cavernous ICA aneurysm).

Figure 6 Right internal carotid artery (ICA) angiogram (left) performed pre-embolisation reveals a giant cavernous ICA aneurysm. Left common carotid artery angiogram (right) after sacrifice of the right ICA shows excellent filling of the right ICA distribution from the left ICA.

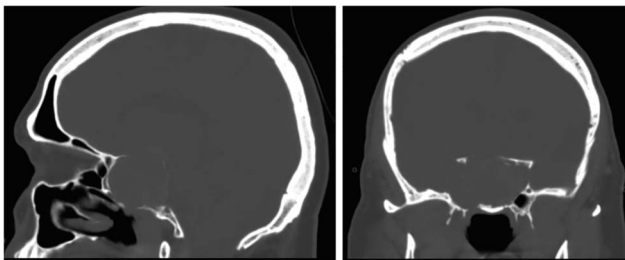
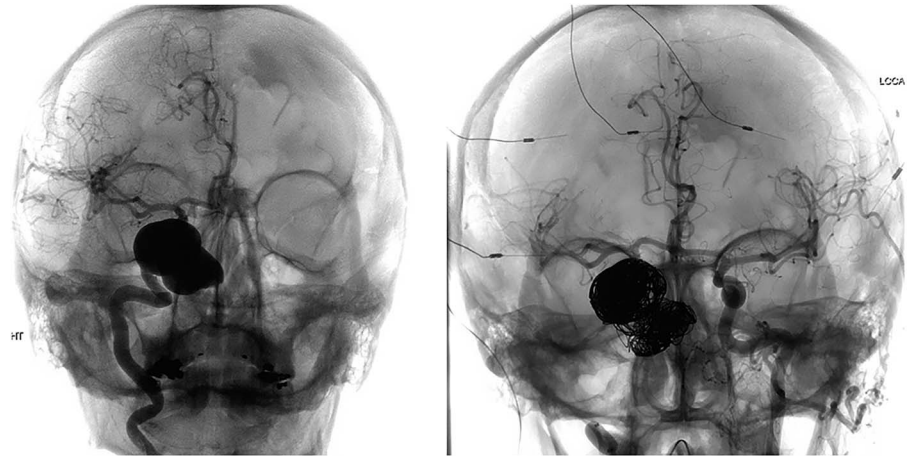


Figure 7 Sagittal (left) and coronal (right) bone windows reveal bony erosion at the level of the dural ring.

Perhaps most pertinent to our case is the report of SAH arising after opening of the dura during a craniotomy to treat a cavernous ICA aneurysm in a patient who also had a prolactinoma treated with cabergoline.⁴ The authors proposed that the initially large prolactinoma may have eroded the dural boundary of the cavernous sinus. Subsequently, shrinkage of this prolactinoma could have left a residual dural defect, allowing communication between the cavernous sinus and the subarachnoid space. In our case, the SAH occurred spontaneously. This is the second

reported example of SAH occurring in a patient with a medically treated prolactinoma. It raises the question of whether treatment of incidental cavernous ICA aneurysms should be considered in cases of concurrent or previously treated large pituitary macroadenomas. In such cases, as suggested previously,⁴ treatment of the aneurysm should be considered prior to initiation of medical therapy for the prolactinoma in order to avoid creating a corridor to the subarachnoid space.

Contributors All authors were involved in acquisition and analysis of the data presented and the patient's case. All authors reviewed the submitted manuscript prior to submission. The manuscript was drafted by SSK and TCH. Critical review was performed by JGG and ASP. JGG and ASP supervised the project.

Competing interests None declared.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

REFERENCES

- 1 Wiebers DO, Whisnant JP, Huston J III, *et al.* Unruptured intracranial aneurysms: natural history, clinical outcome, and risks of surgical and endovascular treatment. *Lancet* 2003;362:103–10.
- 2 Kupersmith MJ, Stiebel-Kalish H, Huna-Baron R, *et al.* Cavernous carotid aneurysms rarely cause subarachnoid hemorrhage or major neurologic morbidity. *J Stroke Cerebrovasc Dis* 2002;11:9–14.
- 3 Linskey ME, Sekhar LN, Hirsch WL Jr, *et al.* Aneurysms of the intracavernous carotid artery: natural history and indications for treatment. *Neurosurgery* 1990;26:933–7.
- 4 Akutsu N, Hosoda K, Ohta K, *et al.* Subarachnoid hemorrhage due to rupture of an intracavernous carotid artery aneurysm coexisting with a prolactinoma under cabergoline treatment. *J Neurol Surg Rep* 2014;75:e73–6.
- 5 Hamada H, Endo S, Fukuda O, *et al.* Giant aneurysm in the cavernous sinus causing subarachnoid hemorrhage 13 years after detection: a case report. *Surg Neurol* 1996;45:143–6.
- 6 Iwaisako K, Toyota S, Ishihara M, *et al.* Intracerebral hemorrhage caused by ruptured intracavernous carotid artery aneurysm. Case report. *Neurol Med Chir* 2009;49:155–8.
- 7 Lee AG, Mawad ME, Baskin DS. Fatal subarachnoid hemorrhage from the rupture of a totally intracavernous carotid artery aneurysm: case report. *Neurosurgery* 1996;38:596–8; discussion 598–9.
- 8 Nishioka T, Kondo A, Aoyama I, *et al.* Subarachnoid hemorrhage possibly caused by a saccular carotid artery aneurysm within the cavernous sinus. Case report. *J Neurosurg* 1990;73:301–4.
- 9 Choulakian A, Drazin D, Alexander MJ. Endosaccular treatment of 113 cavernous carotid artery aneurysms. *J Neurointerv Surg* 2010;2:359–62.
- 10 Kobayashi S, Kyoshima K, Gibo H, *et al.* Carotid cave aneurysms of the internal carotid artery. *J Neurosurg* 1989;70:216–21.

Learning points

- ▶ Cranial base tumours can distort traditional landmarks for vascular anatomy.
- ▶ The potential for subarachnoid haemorrhage due to cavernous internal carotid artery (ICA) aneurysm rupture should be recognised in patients with previous pituitary or other skull base lesions adjacent to the cavernous sinus.
- ▶ Incidental cavernous ICA aneurysms may warrant treatment in cases of concurrent or previously treated large pituitary macroadenomas or other anterior cranial base lesions.
- ▶ Treatment of a cavernous ICA aneurysm should be considered prior to initiating medical therapy for a concurrent large prolactinoma.

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