CASE REPORT

Late onset aneurysm development following radiosurgical obliteration of a cerebellopontine angle meningioma

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Published 14 May 2014; doi:10.1136/bcr-2014-011206

Accepted 15 April 2014

ABSTRACT

The development of de novo intracranial aneurysms following stereotactic radiosurgery for intracranial pathology is a rare complication secondary to vascular injury. Typically, these aneurysms develop within the first few years after radiation surgery. We present the first case of an aneurysm developing 10 years after radiosurgery for a cerebellopontine angle meningioma. This case highlights the importance of careful long-term follow-up of patients who undergo radiosurgery for lesions abutting major vessels and/or who suffer post-radiation complications.

BACKGROUND

De novo development of intracranial aneurysms following stereotactic radiosurgery for vestibular schwannomas, medulloblastoma, and arteriovenous malformations is documented in the literature and has been proposed to result from radiation-induced inflammation of the adjacent vasculature.[1–6] Current reports of incidental findings of aneurysms attributed to radiosurgery typically occur within the first 5 years of follow-up. We present the first reported case of aneurysm development after gamma knife surgery of a cerebellopontine angle meningioma 10 years after treatment, as well as the first commentary regarding the response to radiotherapy as a potential mechanism.

CASE PRESENTATION

A 58-year-old Caucasian woman with a past medical history of hyperlipidemia presented to the Columbia University Medical Center in 2003 with a known cerebellopontine mass for evaluation and treatment. She had been in good health until 1988, when she began to notice intermittent paresthesias in the lower left aspect of her face concomitant with episodes of vertigo. An initial MRI of the brain obtained at an outside hospital was reportedly negative, but a subsequent MRI 2 years later demonstrated a small retroclival mass. Given the slow progression of symptoms and the imaging characteristics of the lesion, it was thought to be either a meningioma or vestibular schwannoma, and the patient opted for observation. Due to incremental worsening of the patient's paresthesias, a subsequent MRI was obtained in 2003. It demonstrated a 1.8×1.2×1.5 cm well-circumscribed enhancing mass in the left pontine cistern (figure 1). She was referred to our institution for neurosurgical consultation.

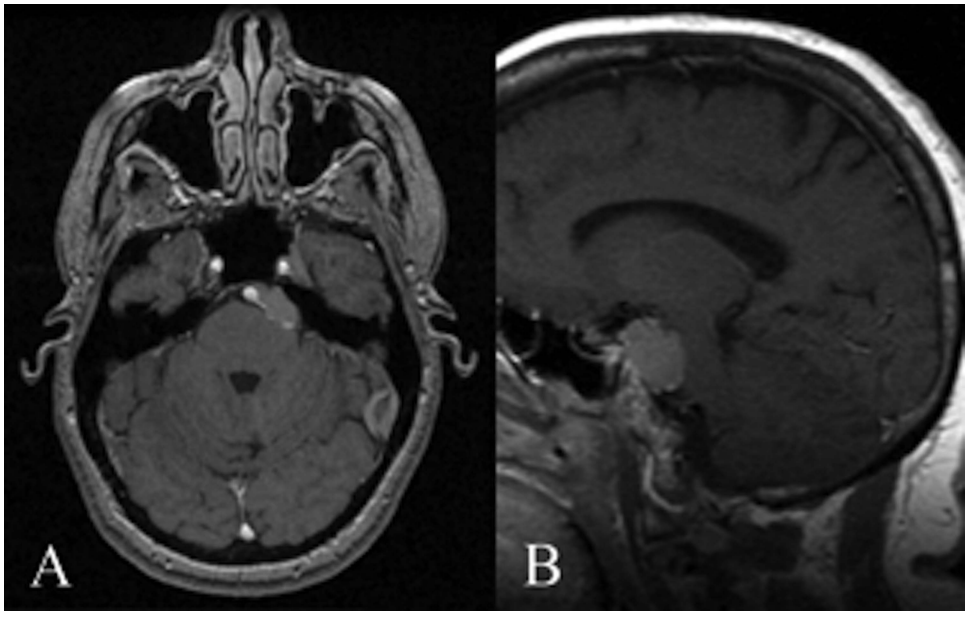

Figure 1 (A) Axial and (B) sagittal T1 MRI with contrast images obtained for gamma knife planning to treat a retroclival meningioma abutting the left superior cerebellar artery.

To cite: Kellner CP, McDowell MM, Connolly ES , *et al*. *J NeuroIntervent Surg* Published Online First: [*please include* Day Month Year] doi:10.1136/neurintsurg-2014-011206.rep

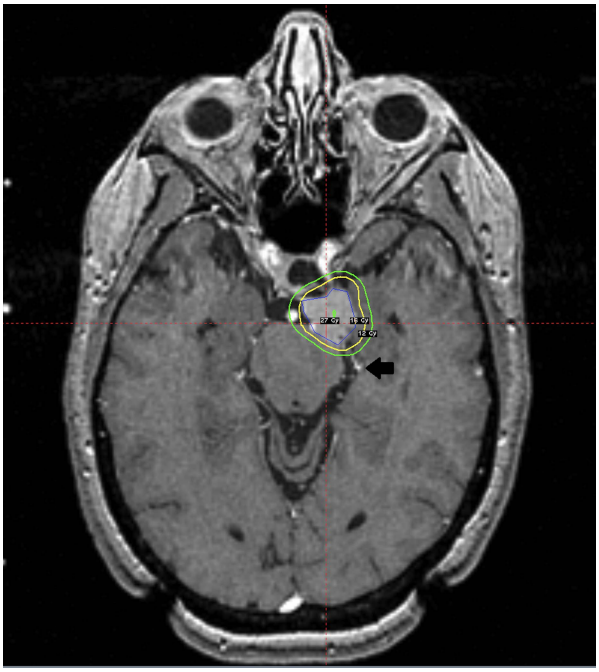

Figure 2 Axial MRI demonstrating the gamma knife surgery plan. At this level the meningioma abuts the left superior cerebellar artery, which can be seen distally (arrow).

The patient had no personal or familial history of malignancy or neurological disease. She denied a history of alcohol or tobacco use. She was found to be hypertensive (160/110), but had an otherwise unremarkable physical examination. Her cranial nerve examination was notable for normal extraocular and pupillary function, no nystagmus, a symmetrical face, strong eye closing, bilaterally intact hearing to gross examination, and bilaterally intact sensation in the orbital, maxillary, and mandibular regions. She had no extremity weakness, pronator drift, dysmetria, or gait instability. Review of the MRI images suggested that the mass was most consistent with a cerebellopontine angle meningioma, given the radiological features such as its adherence to the meninges, homogeneous contrast enhancement, and the presence of a structure consistent with a dural tail. However, other masses such as a vestibular schwannoma could not be ruled out, given the lack of pathological assessment. The patient underwent gamma knife surgery with 16 Gy delivered to the 80% isodense line (figure 2). She tolerated the procedure well and was discharged home the same day. One month after the procedure the patient developed headaches and nausea. A repeat MRI demonstrated edema in the brainstem and cerebellum in the region of the tumor (figure 3). She was prescribed a course of corticosteroids with effective resolution of her symptoms. The patient's baseline symptoms remained stable and serial imaging up to 8 years postoperatively demonstrated gradual obliteration of the mass without recurrence.

INVESTIGATIONS

On her tenth year of follow-up (2013), an MRI demonstrated a small 10×8 × 10 mm mass consistent with an aneurysm, possibly partially thrombosed or a cavernous hemangioma of the brainstem (figure 4). At the time, the patient was completely at her baseline. She was then referred for angiography, which demonstrated an irregularly shaped 6×4×6 mm fusiform aneurysm with saccular components located on the distal left superior cerebellar artery (figure 5).

TREATMENT

Given the fusiform anatomy of the aneurysm and robust collateral flow to the left hemisphere of the cerebellum, the saccular components of the aneurysm were coiled and the parent vessel was sacrificed; the mild fusiform dilation of the posterior cerebral artery in the same region is also appreciated here (figures 6 and 7). The possibility of a bypass procedure was discussed with the open vascular service but it was felt that, given the location and size of the mass, this could not be safely performed. The potential for placement of a flow diversion device was considered, but the size of the superior cerebellar artery was felt to be too small for safe deployment of the device into the posterior circulation. Nonetheless, the rapid progression and abnormal nature of the aneurysm was felt to warrant aggressive attention,

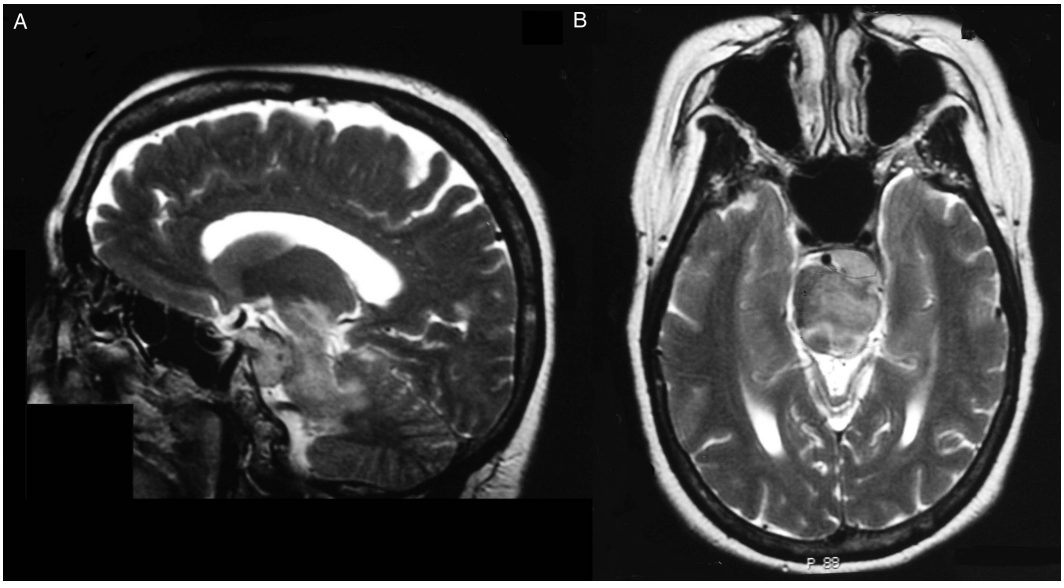

Figure 3 (A) Sagittal and (B) axial fast relaxation fast spin echo MRI demonstrating diffuse brainstem edema proximal to the meningioma extending to the thalamus and cerebellum.

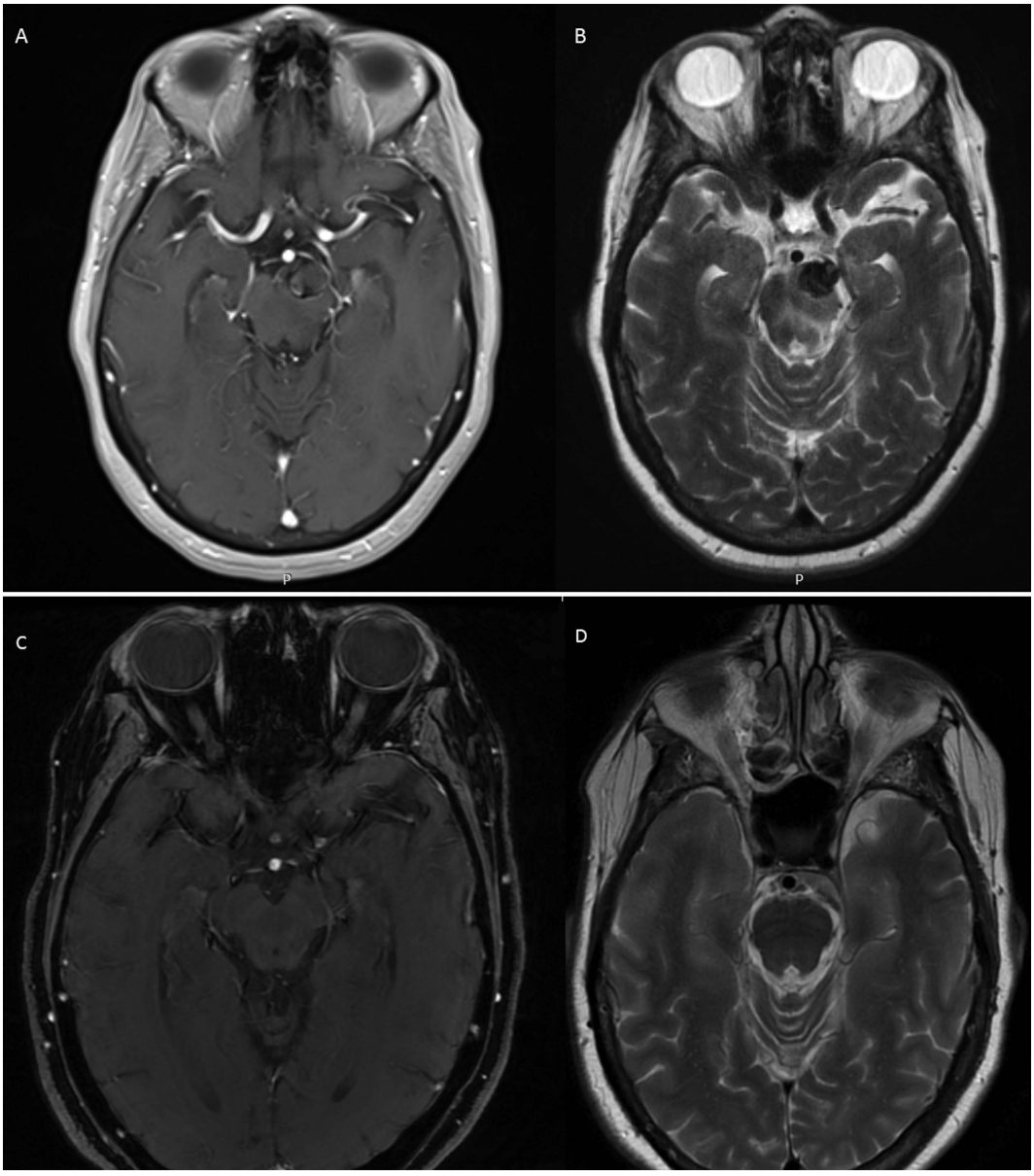

Figure 4 Axial T1 MRI with contrast (A) and T2 image (B) demonstrating a left prepontine mass consistent with a left superior cerebellar artery (SCA) aneurysm or cavernous hemangioma of the brainstem. These are compared with an axial T1 fat suppressed with contrast image (C) from 8 years postoperatively which shows no mass adjacent to the SCA, and a T2 image (D) demonstrating the residual tumor mass but no structure compressing the brainstem.

and the patient was strongly in agreement that she wanted definitive therapy in order to curb the risk of subarachnoid hemorrhage. Balloon occlusion testing was not performed because it would not have changed the management even if ischemia had been produced.

OUTCOME AND FOLLOW-UP

Postoperatively, the patient noted mild dizziness and dysmetria of her left arm and leg but was otherwise at her neurologic baseline. Her dizziness improved and she was discharged to an acute rehabilitation facility primarily for gait issues. At 1 month follow-up she had returned to her baseline (figures 8 and 9). The patient was clinically assessed 6 months later and was found to be in a stable neurological condition. MR angiography (MRA) performed at that time showed stable occlusion of the superior cerebellar artery and no appreciable change in the posterior cerebral arteries, but she has not undergone catheter angiography (figure 10).

DISCUSSION

To our knowledge, this is the first report detailing the development of an aneurysm as a probable complication of stereotactic radiosurgery for treatment of a meningioma. While angiography was not performed prior to radiosurgery, the unusual distal location and long quiescence suggests de novo formation rather than a coincidental aneurysm that was abutting the meningioma prior to treatment.

This aneurysm developed approximately 8–10 years following radiation exposure. The previously documented latest development of an aneurysm following stereotactic radiosurgery was 5 years after treatment, although a patient who was lost to follow-up presented with a ruptured aneurysm that had not

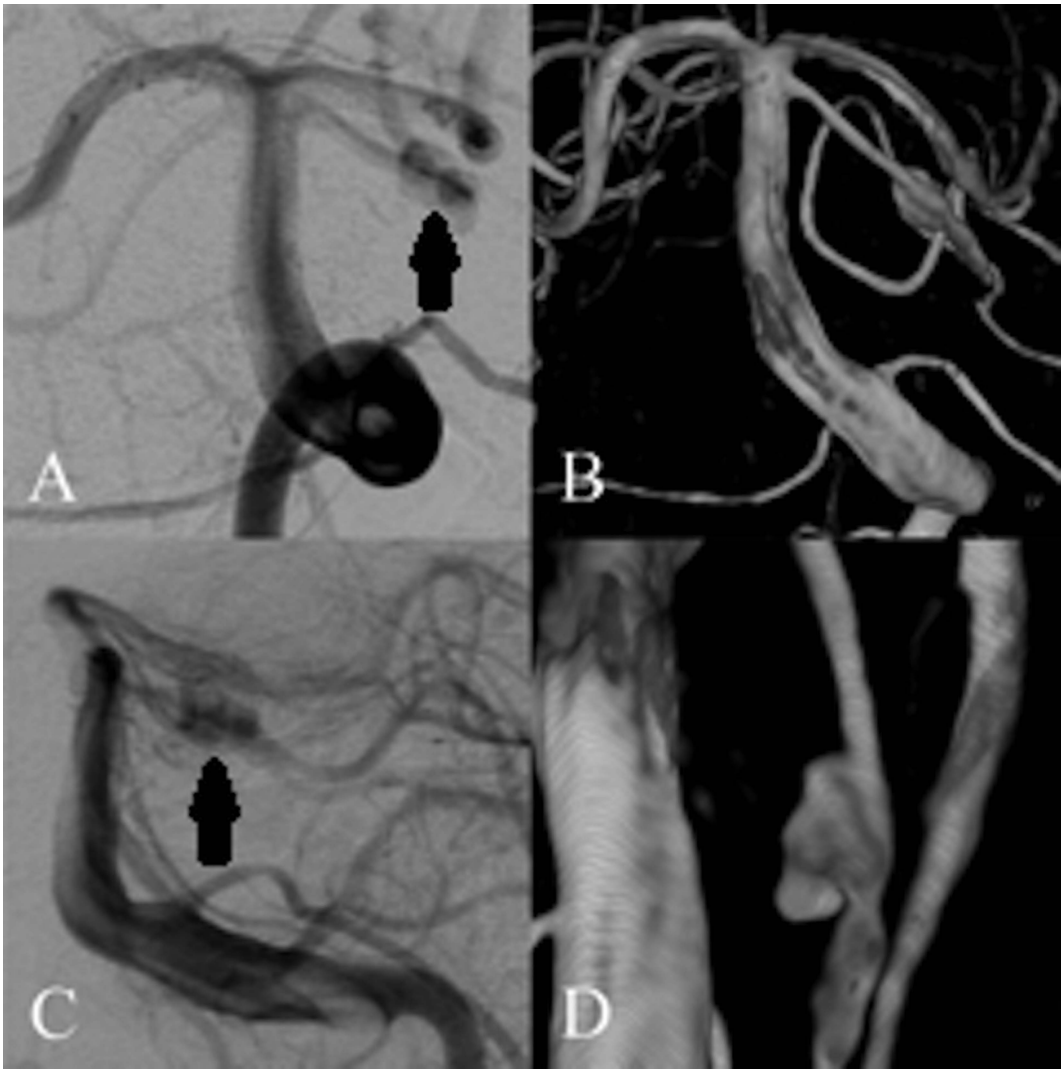

Figure 5 (A) Anteroposterior, (C) lateral and three-dimensional reconstruction (B and D) imaging obtained during diagnostic cerebral angiography showing a fusiform left distal superior cerebellar artery aneurysm with a saccular component and mild fusiform dilation of the posterior cerebral artery in the same region.

been previously diagnosed at 14 years.[1 2 7] The potential development of late onset aneurysms following radiosurgery is a concerning possibility, given that follow-up imaging is often discontinued prior to 10 years after surgery. However, the obvious rarity of this lesion does not justify increased follow-up time for the patient population as a whole. Two factors may have contributed to this patient's aneurysm formation. First, at the time of gamma knife surgery it was clear on the MRI that the meningioma abutted and distorted the left superior cerebellar artery. Second, the patient developed atypically severe radiation-induced edema during the postoperative period. Peritumoral edema is a known adverse effect of radiosurgery and occurs in 10–30% of cases.[8] Given the inflammatory nature of radiation-induced necrosis and edema, individual patient phenotype may play a significant role in the development of post-radiation complications. Patients who are found to have high sensitivity to radiation therapy may require a longer follow-up time to exclude late onset complications. As our understanding of the inflammatory nature of cerebrovascular disease progresses, it may become possible to categorize a patient's risk of vasculopathy and other inflammatory consequences of radiation using genomic data. Studies dedicated to systematic interpretation of genetic data in the context of cerebrovascular disease are already underway at our institution.[9 10] It is important to note, however, that this lesion may also have been secondary to injury from tumor encasement and the subsequent development was secondary to gradual degeneration of the tumor itself rather than radiation exposure.

The pathophysiology of aneurysm formation due to radiation injury is unclear and may be significantly different from typical intracranial aneurysms. Current data suggest that both capillary and large vessel injury is present after radiation exposure, which probably alters hemodynamic and structural factors that may predispose towards the development of vascular pathology.[5] Edematous inflammation may further exacerbate the risk of development or rupture of aneurysms.[11] We feel that, in agreement with past reports, endovascular therapy for these lesions is both effective and indicated. This is the first case of delayed aneurysm formation following gamma knife surgery for a meningioma and demonstrates the benefit of careful long-term follow-up. Perhaps those lesions adjacent to major vascular structures that are treated with stereotactic radiosurgery should have significantly delayed imaging that includes cerebrovascular

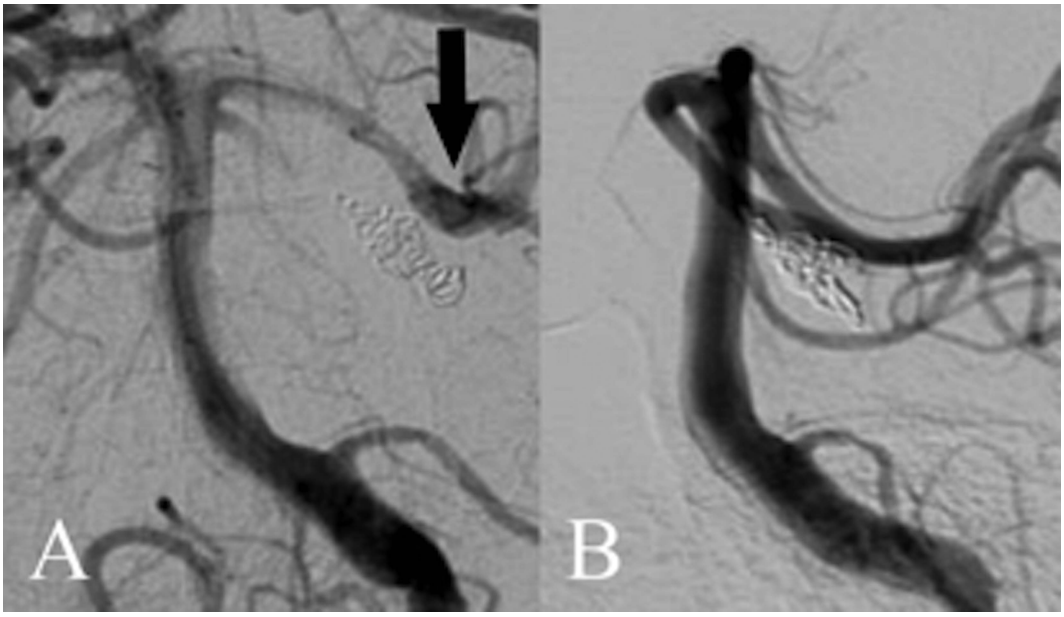

Figure 6 (A) Anteroposterior and (B) lateral angiographic images showing complete occlusion of the aneurysm and superior cerebellar artery sacrifice, and mild fusiform dilation of the posterior cerebral artery in the same region (arrow).

evaluation. The rarity of this incident suggests that the morbidity of the imaging procedure must be taken into account. Adjuvant imaging techniques such as MRA and CT angiography (CTA) during regularly scheduled visits may have merit in this situation. For both new onset and residual aneurysms, the negative predictive value is often greater than the positive using MRA.[12 13] The increasing availability of higher quality MRA images may also help negate the distortion of coil artifact and decrease the false positive rate.[14–16] Small and non-circle of Willis aneurysms are at the greatest risk for being undetectable, however.[13]

CTA appears to have somewhat greater sensitivity and specificity in larger aneurysms, but also comes with the additional concerns of iodine-based contrast and radiation exposure.[17] It is our center's preference to perform MRA routinely in patients with stable long-term follow-up. If a more invasive procedure is deemed necessary based on findings or clinical deterioration, we prefer to use the gold standard catheter angiography.[18]

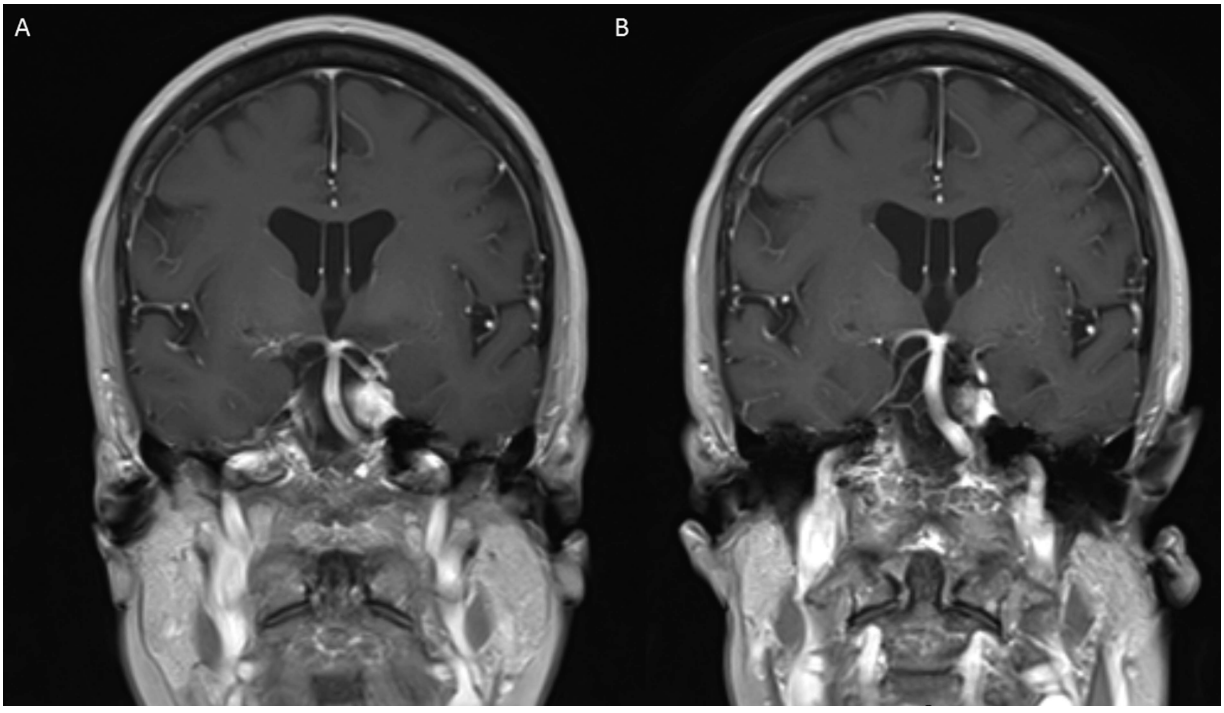

Figure 7 Coronal T1 MR with contrast images demonstrating (A) the presence of a fusiform superior cerebellar artery (SCA) aneurysm preoperatively with (B) subsequent obliteration of the SCA and occlusion of the aneurysm.

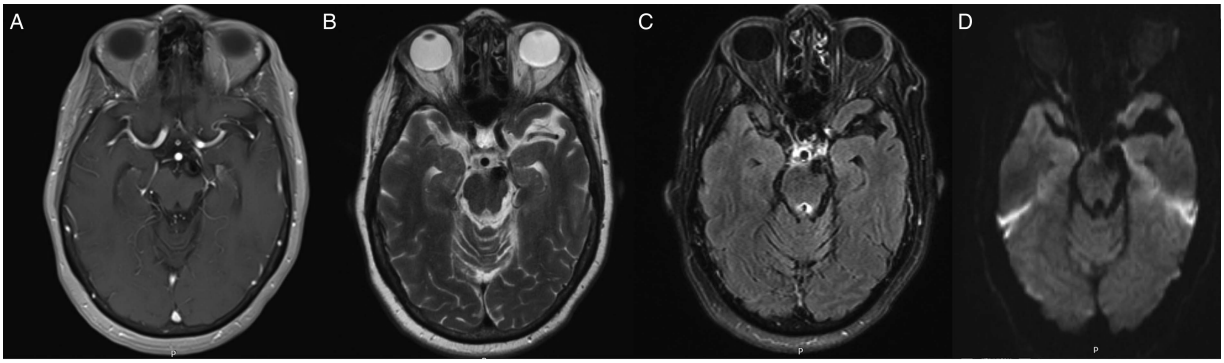

Figure 8 Axial T1 (A), T2 (B), FLAIR (C) and diffusion-weighted (D) MRI demonstrating no apparent infarct or significant edema postoperatively. Distortion from the coil mass is noted anterior to the brainstem.

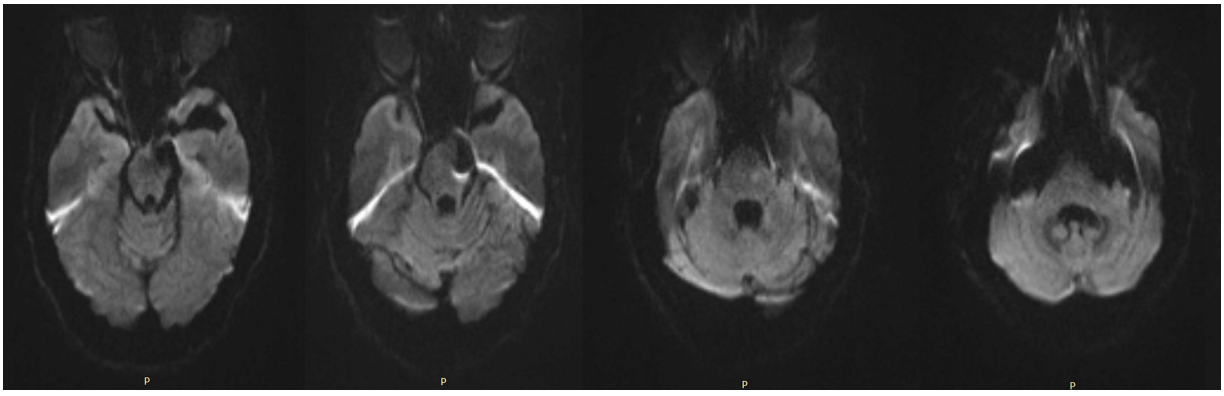

Figure 9 Serial postoperative images demonstrating no apparent infarct or significant edema on diffusion-weighted MRI. Coil mass distortion is again noted anterior to the brainstem.

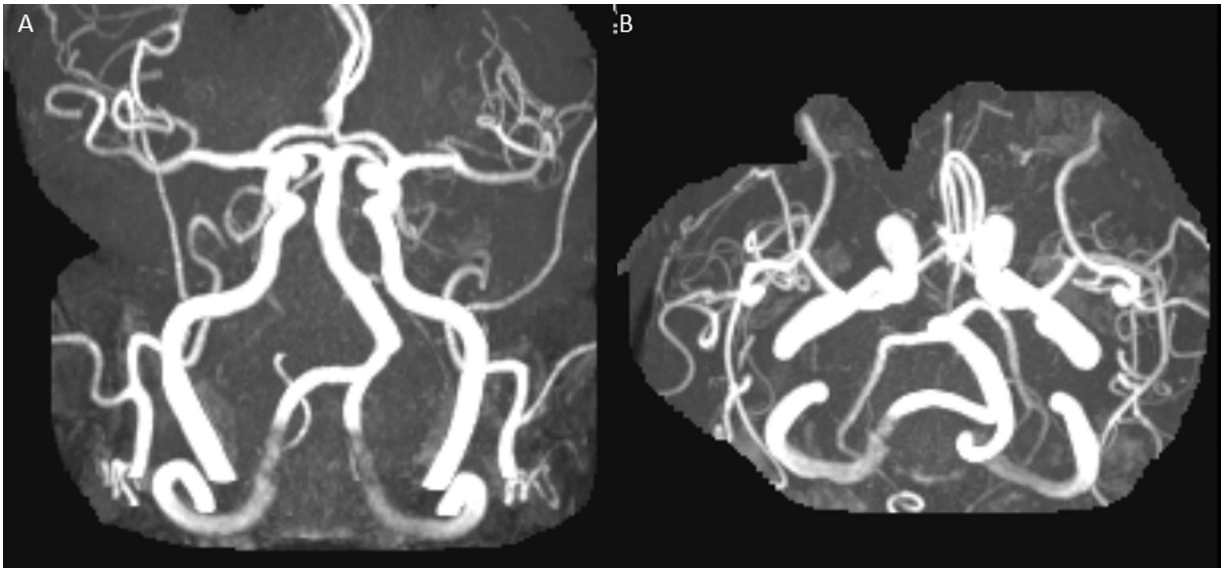

Figure 10 Follow-up MR angiogram demonstrating (A) durable occlusion of the left superior cerebellar artery and (B) no interval change in the posterior cerebral arteries.

Key messages

- Post-radiation complications of neurological disease include de novo aneurysm formation.
- Aneurysms developed after radiation therapy are a risk for hemorrhage and should be managed with microsurgical or endovascular intervention when possible.
- Patients with a greater reaction to radiation therapy may be more likely to develop de novo aneurysms and may warrant a longer duration of follow-up.

Contributors Drafting of manuscript: MMM, CPK. Reviewing/editing manuscript: MMM, CPK, ESC, MBS, SDL. Clinical management/decision making: ESC, MBS, SDL. Analysis of case: MMM, CPK, ESC, MBS, SDL.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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Notes