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Case Report

Ruptured distal anterior inferior cerebellar artery aneurysm years after stereotactic radiosurgery for vestibular schwannoma: A case report and literature review

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ABSTRACT

Background: Aneurysmal formation after stereotactic radiosurgery (SRS) for vestibular schwannoma (VS) is a rare complication. Its characteristics and the best treatment strategies remain controversial, and the clinical course is especially unknown because reported aneurysms are first incidentally detected, or aneurysmal rupture occurs suddenly, and they are treated immediately.

Case Description: A 68-year-old man who underwent SRS for VS 20 years ago presented with subarachnoid hemorrhage (SAH) due to rupture of a radiation-induced fusiform anterior inferior cerebellar artery aneurysm. He was treated with parent artery occlusion, resulting in a modified Rankin scale grade 2. This report illustrates the first case of detected aneurysm formation before rupture with retrospective magnetic resonance imaging evaluation.

Conclusion: We describe the possible risk of rapid progression and rupture of aneurysms, focusing on the interval from SRS to aneurysmal formation. The period of formation of SRS-induced aneurysms is suspected to vary from years to decades regardless of radiation doses; however, aneurysms estimated as pseudoaneurysms have an extremely high risk of rupture within a few years, even when small in size. If aneurysms are discovered unruptured, there are some advantages in not only the prevention of poor prognosis due to SAH but also in the availability of optional therapeutic strategies using revascularization. Long-term annual follow-up, including vessel examination, is warranted not only to assess tumor status but also for early detection of any vascular lesions.

Keywords: Aneurysm, Endovascular surgery, Stereotactic radiosurgery, Subarachnoid hemorrhage, Vestibular schwannoma

INTRODUCTION

Stereotactic radiosurgery (SRS) is a well-established therapeutic modality for treating vestibular schwannoma (VS). Although radiation-induced vasculopathy is known as a delayed complication after SRS, delayed aneurysm formation is rare, and information on its incidence, clinical course, and the best treatment strategy is lacking. We present here a case of a ruptured

distal anterior inferior cerebellar artery (AICA) aneurysm induced by stereotactic irradiation to treat VS and discuss its clinical course and treatment indications with a review of the literature.

CASE PRESENTATION

The patient provided informed consent for the publication of his anonymized data. A 68-year-old man underwent linear accelerator radiotherapy-based SRS of 20 Gy in three fractions for right VS [Figure 1], which was radiographically diagnosed without tissue biopsy. No vascular lesion of the AICA was detected in pre-radiotherapeutic magnetic resonance imaging (MRI). The patient responded well to radiosurgery, except for the development of progressive severe hearing loss in the right ear, and annual MRI follow-up at another hospital demonstrated partial regression of the tumor. Twenty years after SRS, the patient presented with a sudden onset of severe headache associated with nausea and vomiting. His cranial nerve and sensory-motor function were intact, but the right hearing loss was present. Computed tomography revealed a diffuse subarachnoid hemorrhage (SAH) with a predominance of bleeding in the right posterior fossa cisterns [Figure 2]. Digital subtraction angiography showed a fusiform aneurysm arising from the meatal loop of the right AICA. The radiation field of the previous SRS included the aneurysm site. The aneurysm was approximately 2.9 mm in size and was located at a nonbranching point of the right main AICA [Figure 2]. The right superior cerebellar artery and the posterior inferior cerebellar artery were large and seemed to supply a large area of the ipsilateral cerebellar hemisphere. After a thorough discussion in our department of neurosurgery, we performed endovascular aneurysmal coil embolization and parent artery occlusion (PAO) with n-butyl cyanoacrylate. A Marathon microcatheter (Medtronic; Minneapolis, MN, USA) was placed into the aneurysm. The aneurysmal dome and part of the AICA, as the parent artery, were embolized using seven ED coils (Kaneka, Osaka, Japan) followed by PAO of the nonbranching section of the AICA with n-butyl cyanoacrylate [Figure 2]. Postoperative MRI revealed an infarction on the right side of the pons and brachium pontis [Figure 2], causing mild cerebellar ataxia, severe right facial nerve palsy (House-Brackmann Grade 4), and abducens palsy. Except for moderate right hemifacial palsy, the additional neurological disorders recovered fully in 3 weeks. The patient, with a modified Rankin Scale Grade 2, was transferred to a second hospital on the 24th postoperative day to continue rehabilitation.

DISCUSSION

We report here a case of distal AICA aneurysm occurring with SAH 20 years after SRS for VS. Although it is challenging to assess the incidence of such aneurysms, Umekawa *et al.* estimated the incidence at 0.3% based on data from 360 cases of SRS for VS.[13] Only 12 cases with radiation-induced aneurysms after SRS for VS have been previously described [Table 1].[1,3,5-7,9-14] Very importantly, the current case and two other cases showed that no aneurysm was present before SRS, based on prior vascular assessments.[9,10] In all reported cases, the aneurysms showed radiographical

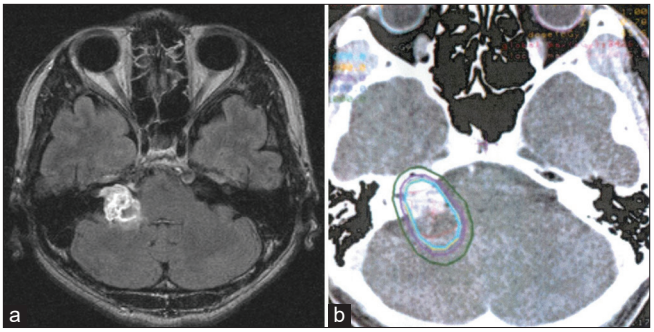

Figure 1: (a) Magnetic resonance imaging performed before stereotactic radiosurgery showing a vestibular schwannoma and (b) axial dose distribution on a vestibular schwannoma target volume superimposed onto the computed tomography scans. The isodose curves corresponding to 20 Gy (cyan) and 10 Gy (green) are shown.

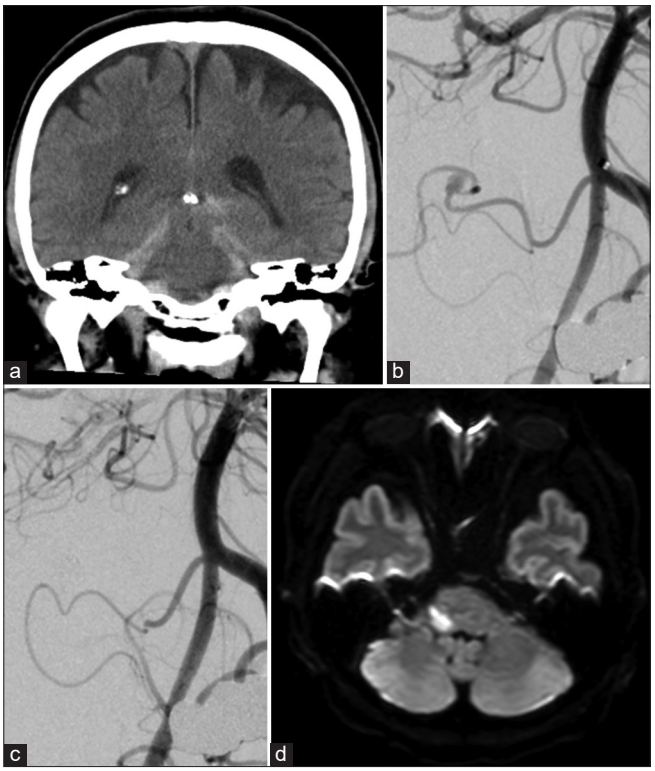

Figure 2: (a) Computed tomography on admission, (b) left vertebral angiogram showing a fusiform aneurysm of the distal anterior inferior cerebellar artery, (c) angiogram after endovascular aneurysmal coil embolization and parent artery occlusion, and (d) postoperative diffusion-weighted magnetic resonance imaging.

Table 1: Summary of published cases with aneurysmal formation of the anterior inferior cerebellar artery after stereotactic radiosurgery for vestibular schwannoma.

Reference	Age, Sex	Radiation therapy, Dose parameters	Years*	Onset	Treatment	Complications	Outcome
Takao *et al.*, 2006[12]	63, F	GKS, 12 Gy	6	SAH	PAO	Facial palsy, hearing loss	mRS 2
Akamatsu *et al.*, 2009[1]	75, F	GKS, 12 Gy	8	SAH	Trapping	N/A	N/A
Park *et al.*, 2009[9]	69, F	GKS, 12 Gy	5	SAH	Conservative care (Failed endovascular)	None	mRS 0
Yamaguchi *et al.*, 2009[14]	67, F	3D-CRT, 50 Gy/25 fractions	6	SAH	Trapping	Facial palsy	mRS 2
Sunderland *et al.*, 2014[10]	50, F	GKS, 1st 13 Gy, 2nd 12 Gy	10	SAH	PAO	Dysarthria, bulbar palsy	mRS 4
Matsumura *et al.*, 2015[6]	49, F, 29, F	N/A, N/A	15, 16	SAH, SAH	PAO, PAO	Hydrocephalus, N/A	mRS 4, mRS 0
Hughes *et al.*, 2015[3]	47, F	GKS, 13 Gy	10	Incidental	Clipping	None	N/A
Mascitelli *et al.*, 2016[5]	59, M	N/A	6	SAH	PAO	Cerebellar/pontine infarction	mRS 2
Murakami *et al.*, 2016[7]	61, M	GKS, 18 Gy	12	SAH	PAO	Cerebellar infarction, facial and abducens palsy	mRS 1
Umekawa *et al.*, 2018[13]	59, M	GKS, 12 Gy	19	Facial palsy	OA-PICA bypass and AICA trapping	None	mRS 1
Takahashi *et al.*, 2023[11]	66, M	GKS, 18 Gy	28	SAH	Trapping	Facial palsy	mRS 1
present study	68, M	LINAC, 20 Gy/3 fractions	20	SAH	PAO	Pontine infarction, facial palsy	mRS 2

AICA: Anterior inferior cerebellar artery, GKS: Gamma knife surgery, LINAC: Linear accelerator radiotherapy, mRS: Modified Rankin Scale, OA: Occipital artery, PAO: Parent artery occlusion, PICA: Posterior inferior cerebellar artery, SAH: Subarachnoid hemorrhage, 3D-CRT: 3-dimensional dynamic conformal radiotherapy. *Years between radiation therapy and detection of the aneurysm. N/A: Not applicable.

characteristics of pseudoaneurysms.[1,3,5-7,9-14] All aneurysms had developed at nonbranching sites within the irradiated field over 5 years after SRS. In total, 84.6% (11 of 13 cases) were ruptured aneurysms; however, their sizes were generally small (2.9–3.3 mm; median, 3.0 mm). The relatively high risk of rupture in small aneurysms also suggests the possibility of pseudoaneurysms.

The pseudoaneurysms were diagnosed using intraoperative findings in five cases[1,3,11,13,14] and pathological features in two.[1,14] Although the relationship between radiation and aneurysm formation remains controversial, it is hypothesized that radiation degrades the parent artery vessel wall, making it vulnerable to hemodynamic stress and leading to aneurysmal formation.[8] This hypothesis is supported by pathological examination findings demonstrating a loss of elastic lamina in the aneurysm wall.[1,14]

No definite treatment has been established for distal AICA aneurysms. Open surgical and neuroendovascular interventions are the two main modalities selected for treatment. As the previously reported aneurysms were located at a nonbranching portion of the AICA, endovascular PAO or surgical trapping without revascularization was the most performed treatment in ten of the 13 cases, including the current case [Table 1]. The use of neuroendovascular therapy is increasing, likely because cannulation of small-diameter, tortuous, and distant vessels has become safer with modern improvements in device technology. When performing endovascular PAO or internal trapping without revascularization, the main concern is the risk of subsequent severe neurological deficits caused by ischemia in the brain stem and/or cranial nerves. The risk of brain infarction is dependent on the anatomical variability of the AICA or collateral flow in the cerebellum. Simple AICA occlusion is not always warranted because there is no established method to evaluate the extent of collateral flow. According to previous reports, severe disability of Grade 3 or more on the modified Rankin scale was seen in only two cases, but the total rate of complications after PAO at this location was high [Table 1]. Particularly, disorders of cranial nerves whose blood supplies mainly depend on the AICA occurred in 50% of cases (5 of 10). Thus, preservation of the parent artery

or concomitant revascularization using neck clipping or extracranial-intracranial bypass with microsurgical trapping should be recommended as essential.

Open surgical interventions at an irradiated and/or postoperative site are extremely challenging due to adhesions and residual tumor necrosis and carry the risk of aneurysmal rupture or trauma to the nerves. In this review of the literature, all of the aneurysms found during surgical exploration were within the tumor.[1,3,11,13,14] The aneurysms were so strongly adherent to the tumor and the adjacent seventh and eighth cranial nerves that dissection and direct clipping or surgical trapping were difficult. This was more challenging in cases of ruptured aneurysms. Thus, no surgical clipping or extracranial-intracranial bypass with microsurgical trapping was able to be used in these cases of SAH.

Meanwhile, the necessity of PAO for such ruptured aneurysms was demonstrated in the case reported by Choi *et al.*[2] They treated a distal AICA aneurysm at the meatal loop with parent artery preservation. The dome of the aneurysm was occluded; however, recurrent hemorrhage from the recanalized aneurysm occurred 1 month after embolization.

It would be reasonable to attempt detecting such aneurysms in unruptured states, considering the negligible risk of causing SAH and the availability of surgical options such as extracranial-intracranial bypass. However, the long-term vascular complications after SRS have seldom been studied. Nanney *et al.* reported that a high radiation dose could be associated with a shorter lag time until aneurysm formation,[8] although there seems to be no association between radiation dose and the latency until aneurysm diagnosis in reported cases of aneurysms after SRS for VS [Table 1]. Many of the aneurysms in these cases were detected after the onset of SAH, and information on the occurrence of the aneurysms remains scarce. Lorenzoni *et al.* evaluated MRI data in patients with trigeminal neuralgia to study changes in the superior cerebellar artery after SRS.[4] Although no definite vascular changes were found, the mean follow-up period was relatively short at 25.2 months, and the manifestation of vascular injury may be expected to take longer, as in reviewed cases [Table 1]. Interestingly, the present case was the only one in which a radiation-induced aneurysm was detected before the onset of SAH using a retrospective review of MRI data; annual follow-up MRI showed no aneurysm for 18 years after SRS but revealed vasodilation in the 19th year [Figure 3]. The period between the onset of vasodilation and SAH onset was only 2 years. Takahashi *et al.* reported a radiation-induced *de novo* aneurysm occurring and rupturing within 5 years of radiotherapy.[11] This may suggest that the period until formation of SRS-induced aneurysms varies, but once it has occurred, the aneurysms have a relatively high risk of rupture within short periods. Vascular imaging is not common in follow-up after SRS; however, long-term follow-up is warranted for all such cases, with special attention paid not only to tumor status but also to the existence of vascular lesions.

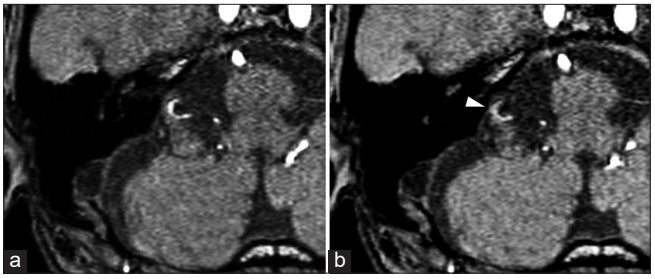

Figure 3: (a) Annual magnetic resonance angiography 18 years after stereotactic radiosurgery showed no aneurysm, but (b) retrospective review revealed the formation of an aneurysm (arrowhead) in the following year.

CONCLUSION

Distal AICA pseudoaneurysm formation after SRS for VS is extremely rare, but it carries a high risk of rupture within a few years after occurrence. Endovascular PAO or surgical trapping without revascularization for such aneurysms has the risk of brain stem infarction, but the risk of severe disability is relatively low, and no other therapeutic options may be available in cases of rupture. Because the latency until aneurysm formation after SRS is not fully understood, surgeons must be aware of this complication, and long-term screening is recommended for adjacent vascular structures along with follow-up of tumor status.

Ethical approval

The Institutional Review Board approval is not required.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

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