

Trigeminal Neuralgia Caused by Nerve Compression by Dilated Superior Cerebellar Artery Associated with Cerebellar Arteriovenous Malformation: Case Report

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Abstract

Intracranial arteriovenous malformation (AVM) is a rare cause of trigeminal neuralgia (TGN). In this presented case, successful resolution of AVM-related TGN following embolization and gamma knife radiosurgery (GKRS) was obtained. A patient suffered from TGN on the left side, which was thought to be caused by root entry zone compression by dilated superior cerebellar artery (SCA) associated with cerebellar AVM. The cerebellar vermis AVM was embolized in endovascular surgery. The AVM was reduced in size and TGN was partially relieved. The patient subsequently underwent GKRS for the residual nidus. TGN was completely resolved within one year and a half. GKRS following embolization of the nidus improved the flow-related dilation of the SCA and completely relieved TGN.

Key words: arteriovenous malformation, embolization, neuralgia, radiosurgery, trigeminal nerve

Introduction

Trigeminal neuralgia (TGN) is an infrequent but important cause of facial pain.¹⁾ One of the causes of TGN is nerve compression by vessels. Furthermore, an underlying arteriovenous malformation (AVM) is rarely identified as the cause of TGN. Nerve root entry zone may be compressed either by the nidus itself or by a proximal artery such as basilar or superior cerebellar artery (SCA) with flow-related dilation. Aside from carbamazepine therapy and microvascular decompression (MVD), in cases of AVM-related TGN, treatment options include invasive surgical AVM extirpation, endovascular embolization, and stereotactic radiosurgery (SRS). Endovascular embolization usually offers only an adjuvant treatment to reduce the AVM size¹⁾ prior to craniotomy or radiosurgery, but it may relieve TGN quickly and relatively safely. In this presented case, TGN was incompletely relieved by partial embolization of the nidus and then was completely relieved after subsequent gamma knife radiosurgery (GKRS).

Case

A 69-year-old male physician presented with a severe left-sided facial pain. Magnetic resonance imaging (MRI) showed a vermis AVM, which was subsequently confirmed by catheter angiography (Figs. 1a, b, 2a, b). Compression around the root entry zone of the left trigeminal nerve by dilated left SCA was also detected (Fig. 3a, b). Medication of carbamazepine relieved facial pain for 5 years but then TGN deteriorated. The patient elected to have endovascular embolization for AVM. The AVM was partially embolized and TGN was partially relieved though the patient suffered from temporary truncal ataxia after embolization (Figs. 1c, d, 2c, d). One month later subsequent GKRS was performed at the residual nidus (Fig. 4). His left-sided facial pain was reduced to about half after the embolization. It was completely resolved within 1½ years after GKRS. He did not develop any new complaints and continued to enjoy freedom from pain without medication for 6 years after GKRS. He denied evaluation by transarterial angiography. MRI showed remarkable reduction of the abnormal vessels around the nidus and improvement of the dilation of the left SCA (Figs. 3c, 5, 6).

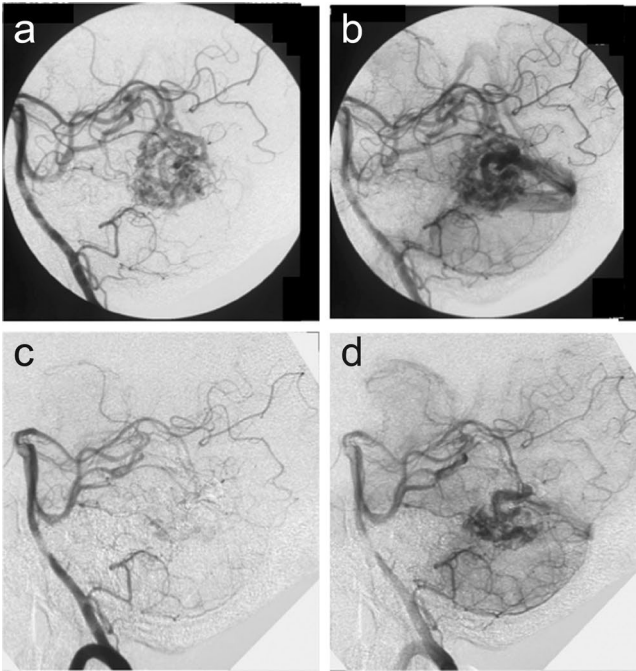


Fig. 1 Lateral views of pre-embolization [early arterial phase (a) and late arterial phase (b)] and those of post-embolization [early arterial phase (c) and late arterial phase (d)]. The flow of nidus was remarkably decreased (c, d) after embolization.

Discussion

TGN has been recognized as one of the most common neurovascular compression syndromes caused by the vascular contact of the trigeminal nerve in its root entry zone with a branch of a superior or anterior inferior cerebellar artery, leading to demyelination of the trigeminal sensory fibers usually within the nerve entry zone. MRI with constructive interference in steady state sequences (CISSs) is reported to be the superior imaging modality for evaluation of TGN.²⁾ MRI-CISS imaging has higher resolution and excellent contrast demonstrating precisely the anatomic detail and abnormal neurovascular relationships responsible for TGN. MRI-CISS is able to identify both arterial and venous compression of the trigeminal nerve. T₂-weighted image (T₂WI) has the same advantage in that it demonstrates excellent contrast of nerves and blood vessels (high-signal) running in the cistern against cerebrospinal fluid (low-signal). We usually make reversed contrast pictures of T₂WI like Fig. 3. Meanwhile, three-dimensional spoiled gradient recalled echo (3D-SPGR) with gadolinium enhancement is also useful to distinguish blood vessels, arteries, and veins (high signal) from nerves (lower signal).

TGN secondary to posterior fossa AVMs has been seldom reported in the literature (Table 1).^{1,3-11)} Most of the cases have been published on a case report basis, and there is not a general agreement about the best method of

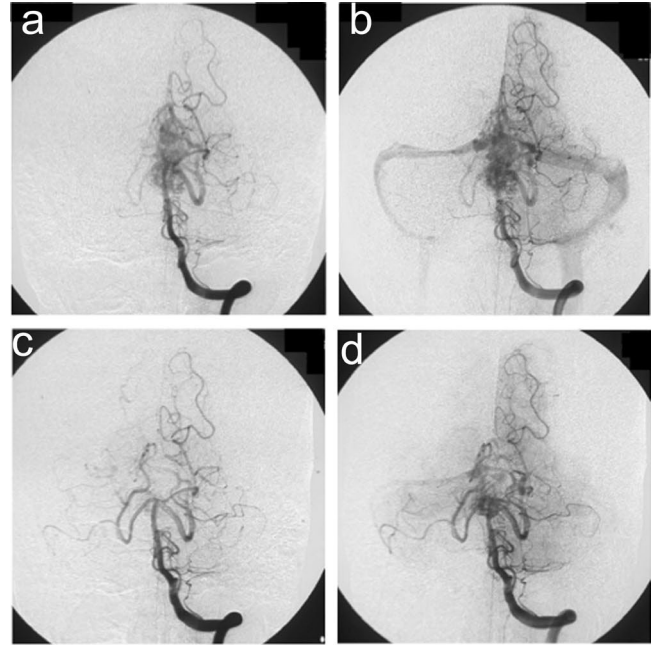


Fig. 2 Frontal views of pre-embolization [early arterial phase (a) and late arterial phase (b)] and those of post-embolization [early arterial phase (c) and late arterial phase (d)]. The flow of nidus was remarkably decreased (c, d) after embolization.

treatment.⁵⁾ In cases of AVM-related TGN, besides medication of carbamazepine or MVD for the root entry zone, treatment options include invasive surgical AVM extirpation, endovascular embolization, and SRS.¹²⁾ Percutaneous puncture of Meckel's cave for glycerol or radio frequency treatment to relieve TGN is contraindicated and may cause catastrophic hemorrhage in this setting.⁶⁾ To decide optimal treatment strategy for each patient, location of the nidus (local at trigeminal nerve or remote), offending vessel (nidus itself, dilated feeding artery, or dilated draining vein), and severity of pain (emergent or durable with medication) are to be considered. Edwards et al.⁴⁾ recommended total microsurgical resection of the AVM with nerve preservation not only for symptom relief but also to eliminate the theoretical risk of hemorrhage. Especially when the nidus is local at trigeminal nerve, surgical complete resection of the nidus is ideal. The relationship between offending vessels and trigeminal nerve would be observed in the same operation field and MVD could be added, if necessary. However, complete resection of the nidus is not always easy. Some authors think that decompression of the trigeminal nerve entry zone is more important. García-Pastor et al.⁵⁾ recommended surgical MVD of the trigeminal nerve as the best treatment even without excision of the AVM. Endovascular embolization usually offers only an adjuvant treatment to reduce the AVM size¹⁾ prior to craniotomy or radiosurgery, but it may relieve TGN

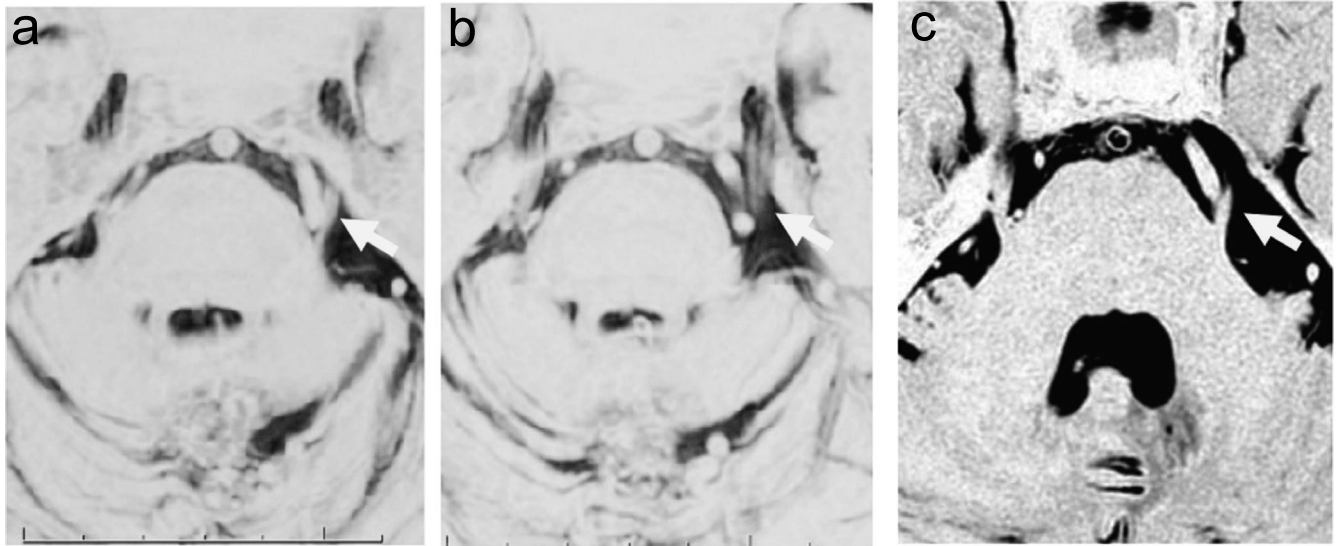


Fig. 3 Axial T₂-weighted magnetic resonance images before embolization [a, b (a little upper slice)] and after embolization and gamma knife radiosurgery (GKRS) (c), 6 years after GKRS (c). The dilated left superior cerebellar artery compressed and bent the left trigeminal nerve before both treatment (a, b). It was normalized in thickness and did not deform the nerve, though it contacted the nerve after embolization and GKRS (c).

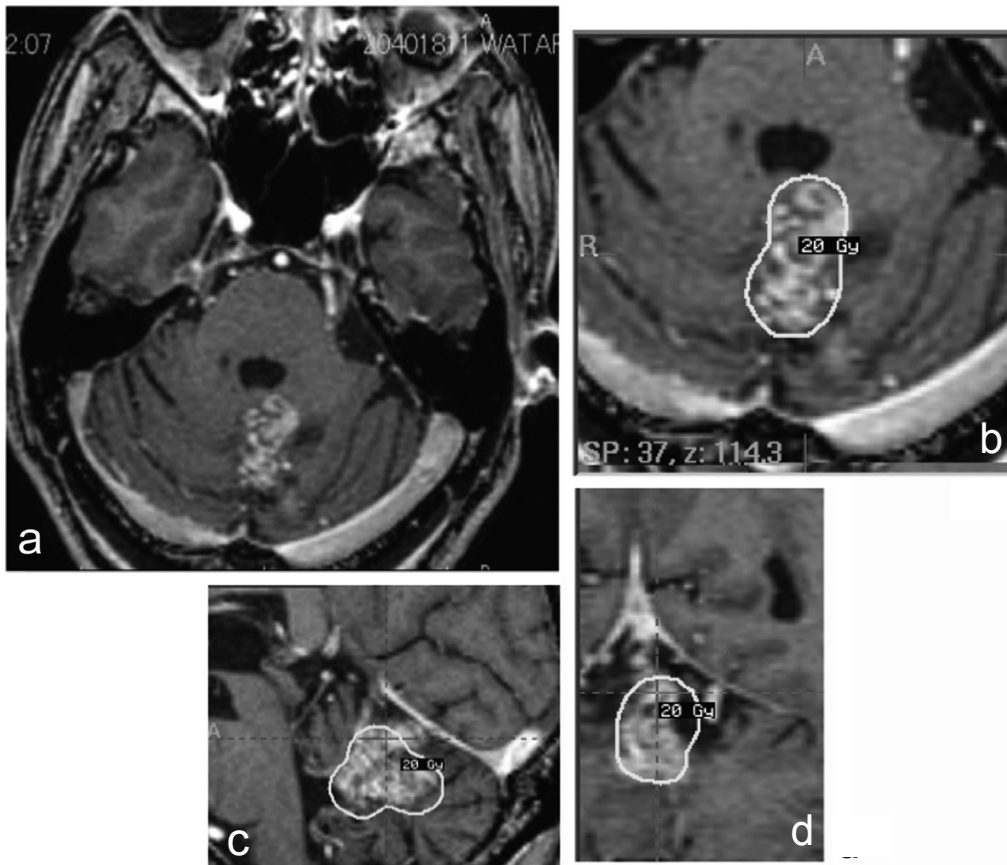


Fig. 4 Axial magnetic resonance images of three-dimensional spoiled gradient recalled echo with gadolinium enhancement (a) showed residual nidus and persistent dilation in the left superior cerebellar artery. Only whole the nidus volume (5.4 ml) was treated with gamma knife radiosurgery with a margin dose of 20 Gy. The isodose curve of 20 Gy is shown in axial (b), sagittal (c), and coronal plane (d) for dose planning.

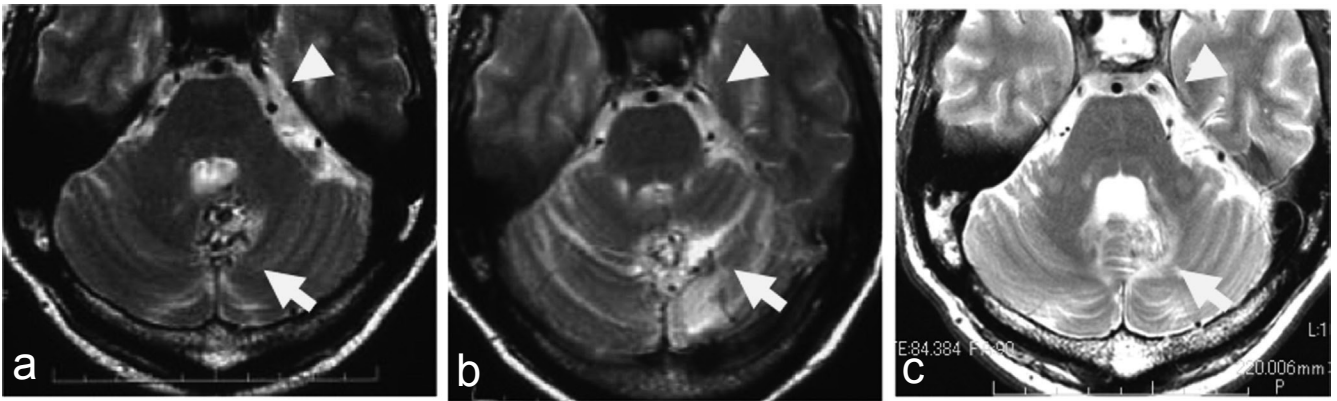


Fig. 5 T₂-weighted axial magnetic resonance images at a: pre-embolization, b: post-embolization (pre-GKRS), and c: 6 years after GKRS. Flow void of the abnormal vessels (a) was decreased after embolization (b) and then disappeared after GKRS (c) (arrows). The flow void of the left superior cerebellar artery was getting thinner after embolization and GKRS in a stepwise manner (arrow heads). GKRS: gamma knife radiosurgery.

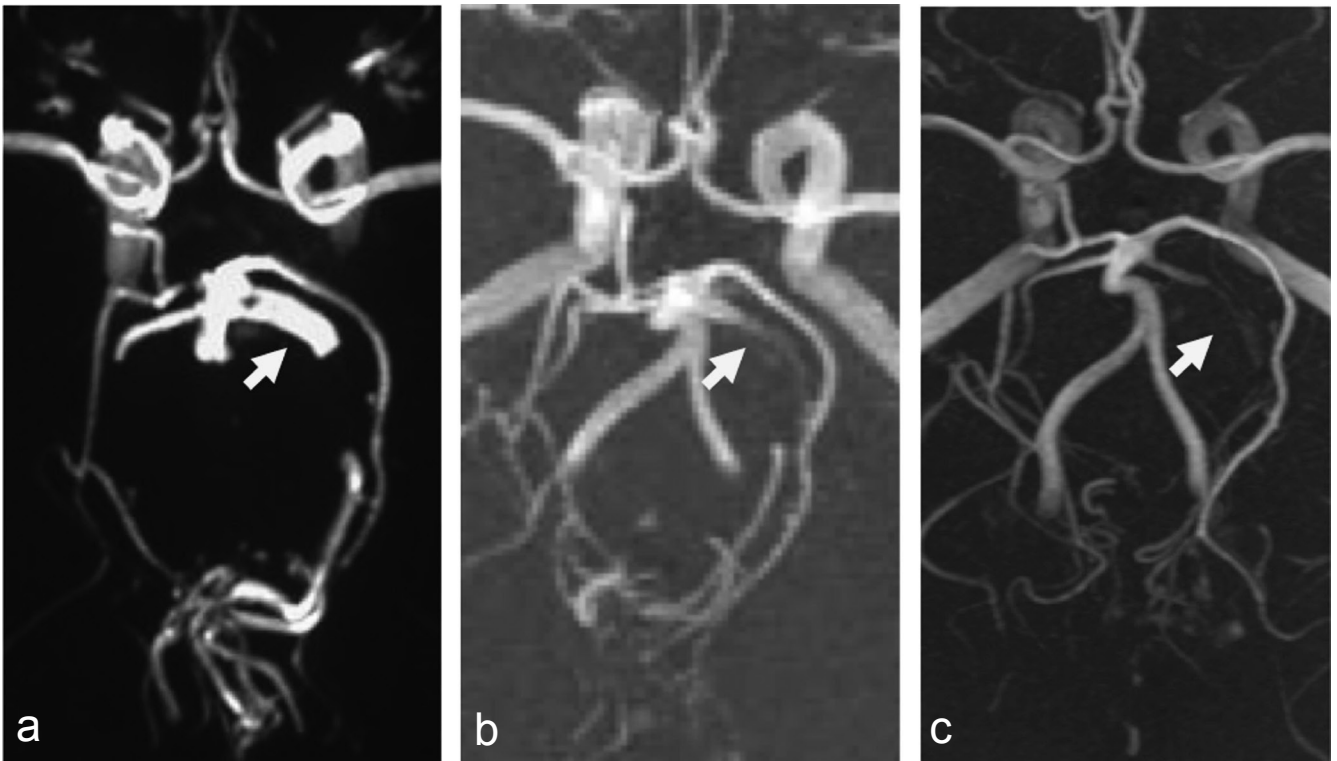


Fig. 6 Basal views of magnetic resonance angiography at a: pre-embolization, b: post-embolization (pre-GKRS), and c: 6 years after GKRS. The dilated left superior cerebellar artery (a) has normalized after embolization and GKRS in a stepwise fashion. GKRS: gamma knife radiosurgery.

quickly and relatively safely. In our present case, embolization relieved the pain quickly, though pain relief was not complete and temporary truncal ataxia occurred as an adverse effect. Wanke et al.,¹¹⁾ Simon et al.,⁹⁾ and Lesley¹⁾ reported similar cases in which embolization of AVM had been effective for pain relief. In the case reported

by Wanke et al.,¹¹⁾ they described that resection of the residual nidus followed embolization. There have been many reports on effectiveness of GKRS for AVM nidus obliteration.¹³⁾ The target of GKRS for AVM is the nidus of AVM. In addition, there have been many reports on effectiveness of GKRS for TGN.¹⁴⁾ The target of GKRS for

Table 1 Reported cases of trigeminal neuralgia associated with AVM (only in MRI era)

AVM location	Authors	No. of cases	Treatment	Pain control	Remarks
Intrinsic TG nerve					
	Karibe et al. (2004) ⁷⁾	1	MVD (then SRS?)	Good	(SRS for the nidus?)
	Anderson et al. 2006 ³⁾	1	SRS on AVM	Good	
	Edwards et al. (2002) ⁴⁾	4	Nidus resection	Good	
		1	Resection + MVD	Good	
	Sumioka et al. (2011) ¹⁰⁾	1	MVD then SRS on AVM	Good	MVD cured pain
CP angle					
	Wanke et al. (2005) ¹¹⁾	1	Emboli. (EVA) then resection	Good	Emboli. cured pain
	García-Pastor et al. (2006) ⁵⁾	2	MVD	Good	No Tx for the nidus itself
		1	Bleeding disturbed MVD	Good*	No Tx for the nidus itself
	Simon et al. (2009) ⁹⁾	1	Emboli. (EVA) (2-times)	Good	2nd emboli. for relapse of pain
Distant (Cbll)					
OA: (veins)	Sato et al. (2003) ⁸⁾	1	SRS then MVD	Good	MVD cured pain
OA: AICA	Garcia-Poster et al. (2006) ⁵⁾	2	MVD	Good	No Tx for the nidus itself
OA: SCA	Lesley (2008) ¹⁾	1	Emboli. (EVA) (2-staged)	Good	
OA: SCA	Present case	1	Emboli. (NBCA) + SRS on AVM	Good	Emboli. and SRS cured pain
Dural (petrotentorial)					
	Ito et al. (1996) ⁶⁾	1	Emboli. (PVA) + surgery	Good	Surgical interruption of the draining vein added

*microcompression of the gasserian ganglia relieved pain. AICA: anterior inferior cerebellar artery, AVM: intracranial arteriovenous malformation, Cbll: cerebellum, CP angle: cerebellopontine angle, EVA: ethlene-vinyl alcohol copolymer, MRI: magnetic resonance imaging, MVD: microvascular decompression, NBCA: n-butyl cyanoacrylate, OA: offending artery, PVA: polyvinyl alcohol particle, SCA: superior cerebellar artery, SRS: stereotactic radiosurgery, TG: trigeminal, Tx: therapy.

TGN is trigeminal root entry zone¹⁴⁾ or root.¹⁵⁾ In our case, complete relief of TGN and complete obliteration of the nidus, which was confirmed only on MRI, was achieved. After embolization and GKRS at the nidus the inner diameter of the SCA became thinner remarkably. Before embolization the right trigeminal nerve was apparently bent by the SCA. After treatment the contact was in less degree and the bending of the nerve was improved. Compression of root entry zone directly and with pulsation was thought to be relieved. In most cases, the latency period between GKRS procedure and obliteration of the nidus was 2 to 3 years.¹³⁾ In our case, TGN was completely relieved within 1½ years after GKRS.

Conclusion

GKRS following intravascular embolization was very effective to relieve TGN, even if the nidus was remote from the affected trigeminal nerve.

Conflicts of Interest Disclosure

The authors have no conflict of interests. YM has declared it to the Japan Neurological Society.

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