

Secondary Trigeminal Neuralgia Caused by a Tentorial Dural Arteriovenous Fistula: A Case Report and Literature Review

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Abstract: Trigeminal neuralgia caused by a tentorial dural arteriovenous fistula has rarely been reported. The mechanism underlying the development of trigeminal neuralgia has been reported to be the compression of the root entry zone by the arterialized draining vein. However, the cases showing refractory or recurrent neuralgia could imply the presence of another underlying mechanism. We herein describe a case in which the arterialized draining vein and the superior cerebellar artery co-compressed the root entry zone of the trigeminal nerve. This case shows another pathophysiology of secondary trigeminal neuralgia caused by a tentorial dural arteriovenous fistula. Direct surgery should be considered for the treatment of such cases.

Key words: Trigeminal Neuralgia, Dural Arteriovenous Fistula, Tentorium, Superior Cerebellar Artery, Microvascular Decompression

Introduction

Trigeminal neuralgia caused by a tentorial dural arteriovenous fistula (DAVF) is extremely rare. Only 10 cases have been previously reported.¹⁾⁻¹⁰⁾ Some of these cases were refractory to the initial therapy for a tentorial DAVF or were recurrent. In such rare clinical entities, the optimal treatment strategy is unclear. We herein describe a case of secondary trigeminal neuralgia caused by a tentorial DAVF, discuss the pathophysiology of the case and advocate an optimal therapeutic strategy involving microvascular decompression (MVD).

Case report

A 57-year-old male had a one-year history of suffering from typical trigeminal neuralgia, consisting of paroxysmal lancinating facial pain in the area of the third division of the left trigeminal nerve. He described the pain as being sharp and like an electrical impulse,

lasting from a few seconds to minutes. The trigger zone was in the left mental foramen. The patient also complained of pulsatile tinnitus in the left ear. The symptoms were initially well-controlled with the administration of carbamazepine; however, he suffered from severe drug eruptions and quit taking the medication, resulting in recurrence of the trigeminal neuralgia.

Thin-cut magnetic resonance imaging (MRI) revealed that an enlarged left petrosal vein was compressing the left trigeminal nerve in the cerebellopontine cistern, and there were normal vessels running between the petrosal vein and root entry zone (Fig. 1).

Cerebral angiography revealed a DAVF in the petro-tentorial region. The DAVF was fed by the posterior branches of left middle meningeal artery and the left tentorial artery. Venous drainage was occurring through an enlarged left petrosal vein and the bilateral posterior mesencephalic veins, into the straight sinus (Fig. 2).

Initially, endovascular surgery was carried out with N-butyl cyanoacrylate, and the main feeders of the

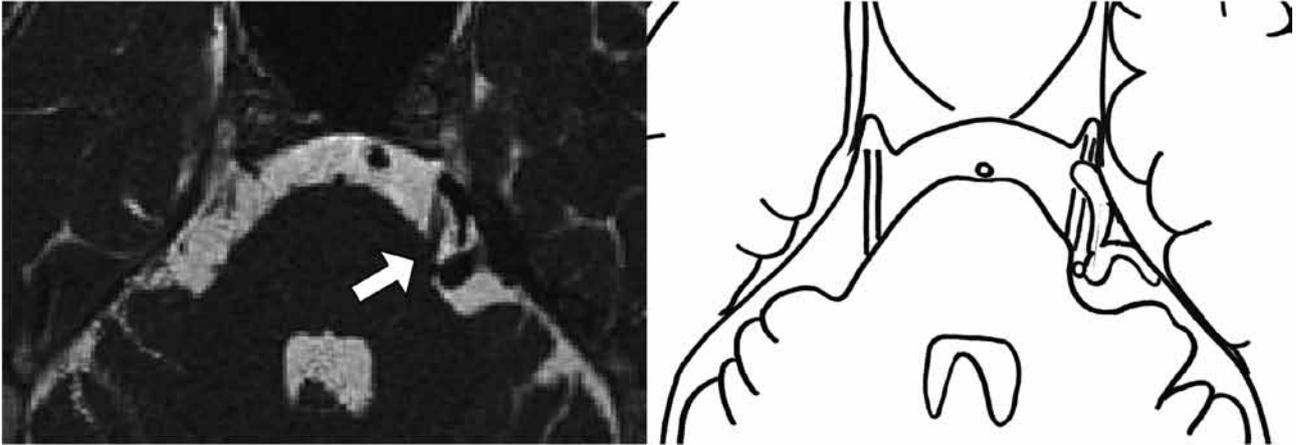


Fig. 1. T2-weighted MR axial images demonstrated thick linear flow voids adjacent to the left trigeminal nerve in the prepontine cistern and the SCA running nearby to the REZ (arrow).



Fig. 2. Left external carotid angiography showed a dural AVF in the petrotentorial region. The dural AVF was supplied by the posterior branches of the left middle meningeal artery (A: anterior-posterior view, B: lateral view). Left internal carotid angiography showed the feeder running from the left tentorial artery (C: lateral view). Postoperative left common carotid angiography showed complete obliteration of the fistula (D: lateral view).

middle meningeal artery were successfully embolized. Angiography after the embolization demonstrated almost total occlusion of the fistula; however, the patient had no relief of the trigeminal neuralgia. On the following day, a craniotomy was performed via the lateral suboccipital approach. An enlarged vein running along the trigeminal nerve was identified in the rostral left cerebellopontine angle. An abnormal proliferation of the small feeding arteries was observed on the dural wall. The draining vein appeared blue, but

was arterialized. Further observation revealed that the superior cerebellar artery (SCA) was also compressing the root entry zone of the trigeminal nerve just below the arterialized petrosal vein. The petrosal vein and the SCA were mobilized, and we observed indentations on the root entry zone of the trigeminal nerve (Fig. 3). The artery was secured on the dural wall with a prosthesis (teflon felt and fibrin glue) (Fig. 3). Postoperatively, the patient was immediately free from the trigeminal neuralgia.

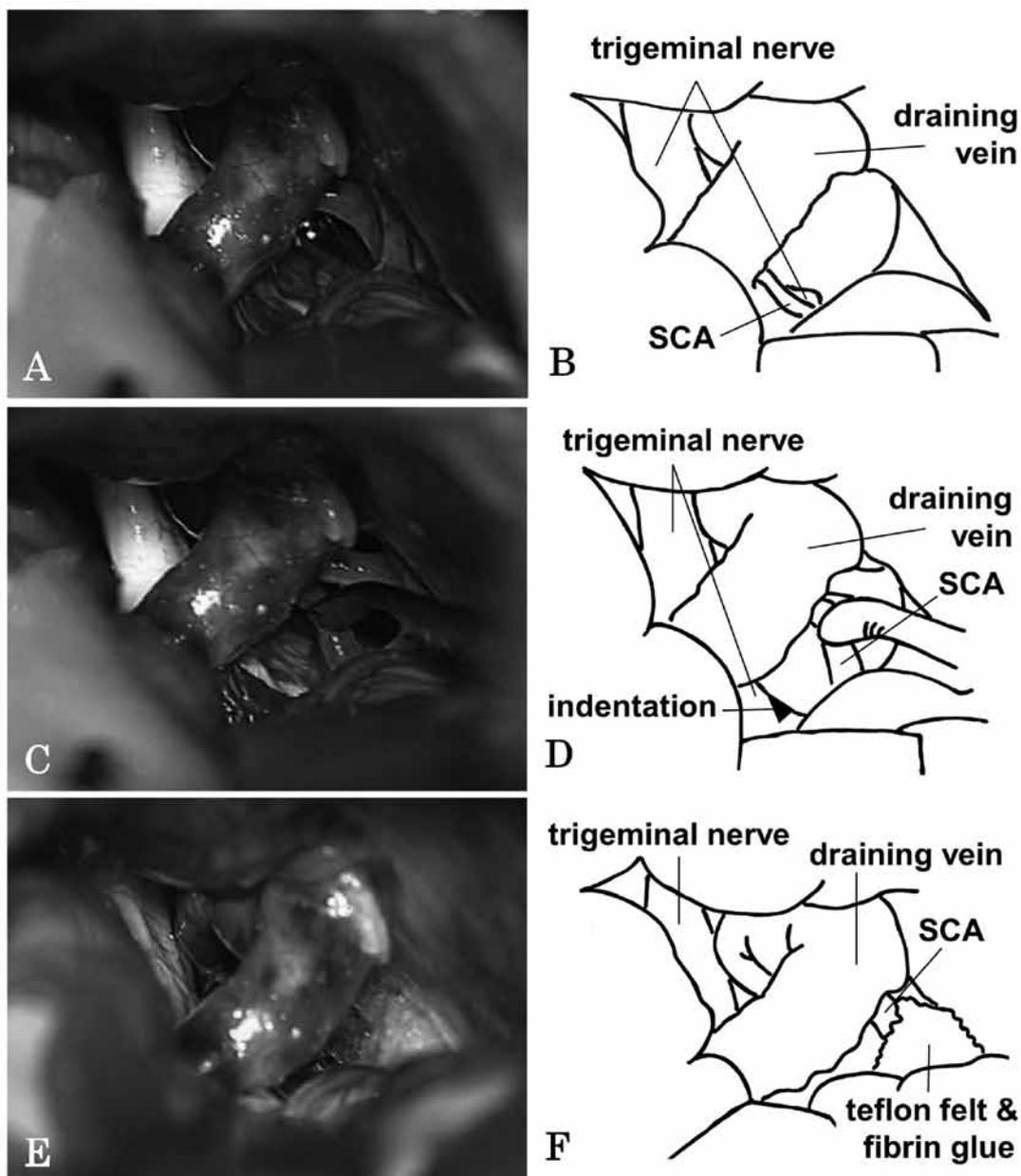


Fig. 3. Intraoperative photographs (A,C,E) and corresponding illustrations (B,D,F). The trigeminal nerve was compressed by the hypertrophic draining vein. The SCA running just below the draining vein was revealed (A,B). The SCA was mobilized, and the compression of the trigeminal nerve was relieved (C,D). The SCA was secured on the dural wall with teflon felt and fibrin glue (E,F).

Table 1 Reported cases of trigeminal neuralgia caused by tentorial dural arteriovenous fistula

	Initial Therapy	Outcome	Reccurrence	Secondary Therapy	Outcome	Reccurrence	Third Therapy	Outcome
Du et al.	Endovascular	CR	—					
Tomak et al.	Endovascular	CR	—					
Ito et al.	Endovascular	CR	+	Endovascular	NC	—	MVD	CR
Ott et al.	Endovascular	NC	—	Endovascular	CR	—		
Mendelowitsch et al.	Endovascular	CR	+	Surgery for DAVF	CR	—		
Seo et al.	Radiation	NC	—	Endovascular	CR	—		
Matsushige et al.	Radiation	CR	—					
Lucas et al.	Surgery for DAVF	CR	—					
Rahme et al.	Surgery for DAVF	CR	—					
Harders et al.	MVD	CR	+	Surgery for DAVF	CR	+	Surgery for DAVF	CR
Present case	TAE + MVD	CR	—					

CR: complete remission; NC: no change; MVD: microvascular decompression; TAE: transarterial embolization

Discussion

The pathophysiology of trigeminal neuralgia was not well understood. Therefore, most trigeminal neuralgias were considered idiopathic diseases. Later, the “neurovascular compression theory” was reported by Jannetta.¹¹⁾ Trigeminal neuralgia secondary to structural anomalies is called “secondary trigeminal neuralgia.” This condition accounts for approximately 2% of all cases of trigeminal neuralgia¹²⁾ and 0.2-1.5% of all of trigeminal neuralgias are associated with vascular malformations.¹³⁾

Only 10 previous cases of trigeminal neuralgia caused by a tentorial DAVF have been reported (Table 1).¹⁾⁻¹⁰⁾ Most tentorial DAVFs drain into the petrosal veins, mesencephalic veins and cerebellar veins.¹⁴⁾ Neurovascular compression by the enlarged draining veins is considered to be the cause of secondary trigeminal neuralgia due to a tentorial DAVF. In all of the previous cases, the main therapeutic strategy for tentorial DAVF-induced trigeminal neuralgia was the treatment of the tentorial DAVF as the primary lesion by direct surgery, endovascular surgery or stereotactic radiosurgery.¹⁾⁻¹⁰⁾ The compression of the

trigeminal nerve by the enlarged draining veins was actually confirmed during surgery in four out of the 10 cases.^{3), 4), 6), 8)} However the clinical outcome was not always favorable; the trigeminal neuralgia was not relieved in three cases^{7), 9), 10)} and the neuralgia recurred in three cases.^{2), 3), 6)} These reports suggested that another pathophysiological mechanism was underlying the neuralgia.

Our present case showed the presence of co-compression on the root entry zone of the trigeminal nerve by the SCA and the arterialized draining vein; providing evidence of another pathophysiology of the secondary trigeminal neuralgia caused by the tentorial DAVF. Four out of seven previously reported cases treated by endovascular or radiation therapy as initial therapy required multiple therapies.^{1), 3), 5)-7), 9), 10)} There was one recurrent case after MVD who was treated with translocation of the draining vein only.²⁾ On the other hand, other MVD cases performed with translocation of the SCA avoided recurrence.³⁾ Therefore, we advocate not only treatment of the tentorial DAVF by direct surgery, endovascular surgery or stereotactic radiosurgery, but also subsequent microvascular decompression, especially in refractory or recurrent cases.

Conclusion

We herein reported a case of tentorial DVAF-induced trigeminal neuralgia that was refractory to the initial endovascular modality. The co-compression of the root entry zone of the trigeminal nerve by the SCA and the arterialized draining vein may be another pathophysiology of the secondary trigeminal neuralgia. Direct surgery should therefore be considered for the treatment of such cases.

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(平成 24. 12. 25 受付, 平成 25. 4. 1 受理)