Online August 18, 2021
Journal of Neuroendovascular Therapy 2022; 16: 170–174 DOI: 10.5797/jnet.cr.2021-0071

Case Report

Trigeminal Neuralgia Caused by Dural Arteriovenous Fistula of the Transverse-Sigmoid Sinus without Vessel Compression at Root Entry Zone

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Objective: The vessel compression at the root entry zone (REZ) of trigeminal nerve is a common cause of trigeminal neuralgia (TN). We report a rare case of TN caused by dural arteriovenous fistula (DAVF) of the transverse-sigmoid sinus without vessel compression at REZ.

Case Presentation: A 45-year-old woman presented with right side tinnitus and was diagnosed as a DAVF of the right transverse-sigmoid sinus (Borden Type I). After that, the facial pain in the right maxillary nerve area appeared and was getting worse. DSA revealed an enlargement of the artery of foramen rotundum (AFR) as one of the feeding arteries. MRI revealed no evidence of vascular compression at REZ. The patient was treated with transarterial embolization (TAE) with Onyx via the branches of the middle meningeal artery (MMA) and occipital artery (OA). The AFR decreased in size and the facial pain was improved. However, the DAVF and the facial pain were recurred. Finally, the DAVF was completely embolized with transvenous embolization (TVE). During 1-year follow-up period, the patient remained free of pain without recurrence.

Conclusion: The compression of the maxillary nerve by the AFR might result in TN, because the pain diminished after shrinkage of the AFR by the endovascular treatment.

Keywords ▶ artery of foramen rotundum, dural arteriovenous fistula, trigeminal neuralgia

Introduction

Trigeminal neuralgia (TN) is one of the representative diseases which cause facial pain. The annual incidence rate of TN in the United States is reported 5.9 per 100000 women and 3.4 per 100000 men.[1)] Classic TN occurs as a result of vessel compression at the root entry zone (REZ) of the trigeminal nerve and has been treated surgically with microvascular decompression.[2)] TN also is induced by other neurovascular diseases such as cerebello-pontine angle tumors, brain stem infarction, cerebral aneurysms, and vascular malformations.[3,4)] Among them, vascular malformations have been reported to be associated with 0.2–1.5% of TN.[5)] We report a rare case of TN caused by a dural arteriovenous fistula (DAVF) of the transverse-sigmoid sinus without vessel compression at REZ.

Case Presentation

A 45-year-old female presented with 1-month history of the right pulsatile tinnitus. MRI revealed a DAVF of the right transverse-sigmoid sinus. The pulsatile tinnitus was gradually improved; however, the facial pain in maxillary nerve area appeared. The pain was a paroxysmal pain along with a constant pain in the background. It got gradually worse in spite of medication with pregabalin. DSA showed multiple feeding arteries including tentorial artery, artery of foramen rotundum (AFR), branch of inferolateral trunk (ILT), middle meningeal artery (MMA), posterior auricular artery, and

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Received: April 27, 2021; Accepted: June 11, 2021
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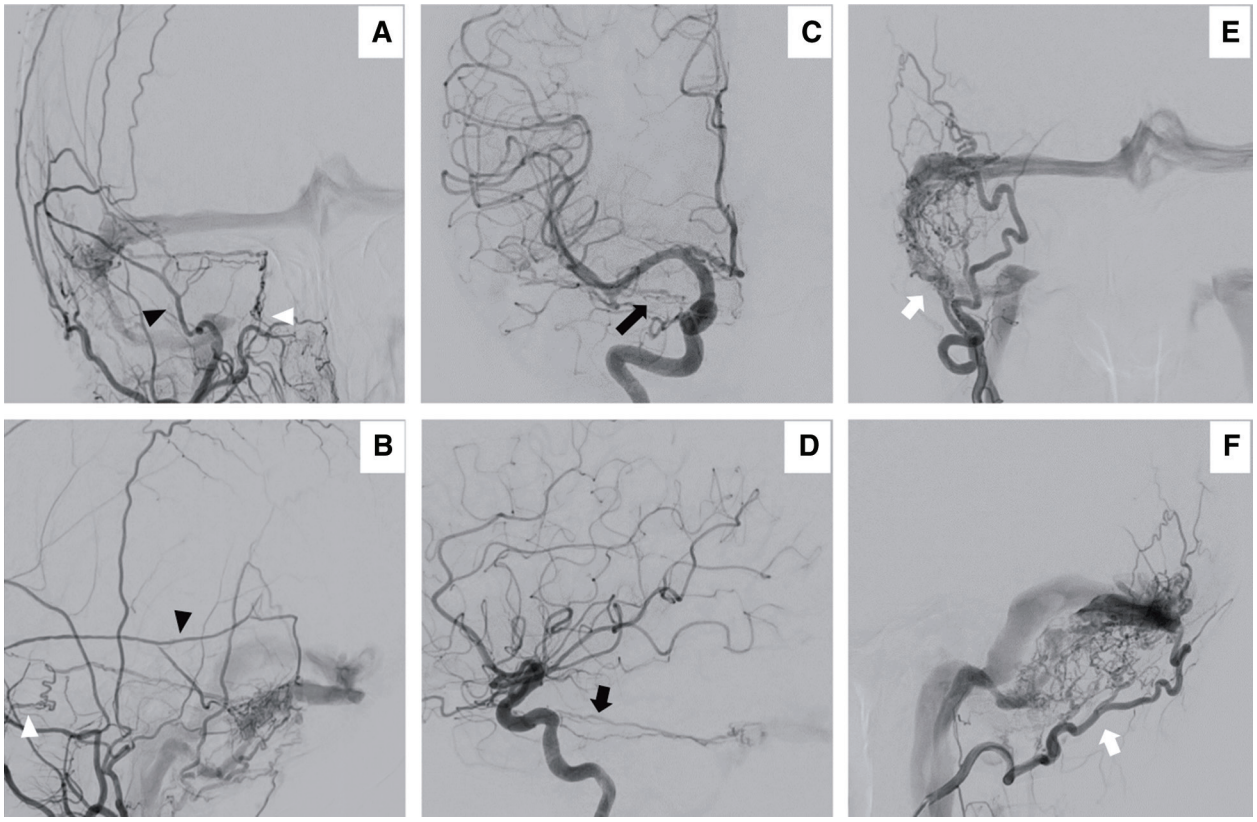

Fig. 1 External carotid angiograms (**A** and **B**) showing the petrosquamous branch of MMA (black arrowhead) and AFR (white arrowhead) as the feeding arteries of the dural arteriovenous fistula. Internal carotid angiograms (**C** and **D**) showing the tentorial artery and the ILT (black arrows) as the feeding arteries. Selective occipital angiograms (**E** and **F**) showing many feeding branches from the OA (white arrows). The shunt was drained into the transverse-sigmoid sinus anterogradely and retrogradely without cortical reflux. AFR: artery of foramen rotundum; ILT: inferolateral trunk; MMA: middle meningeal artery; OA: occipital artery

occipital artery (OA; **Fig. 1**). The shunt was drained into the right transverse-sigmoid sinus anterogradely and retrogradely without cortical reflux. The DAVF was classified as Borden Type I and Cognard Type IIa. Transarterial embolization (TAE) using Onyx (Medtronic, Minneapolis, MN, USA) was performed. Onyx 18 was injected from the branch of the OA and petrosquamous branch of MMA. Although the small shunt from the OA remained, the feeding arteries via tentorium cerebelli including the AFR, branches of ILT, and tentorial artery were not opacified after the TAE (**Fig. 2**). The facial pain improved after TAE. However, the facial pain recurred 3 months later and MRI revealed the recurrence of the DAVF. MRI fast imaging employing steady-state acquisition (FIESTA) images showed no evidence of vessel compression at the REZ (**Fig. 3**). DSA revealed the recurrence of the feeding arteries via the tentorial edge including the AFR. Transvenous embolization (TVE) was performed and the right sigmoid sinus was packed with platinum coils via contra-lateral internal jugular vein, preserving the right vein of Labbé (**Fig. 4A** and **4B**). The DAVF completely diminished after TVE (**Fig. 4C**–**4F**). The facial pain was worsened for a few days and finally disappeared. During 1-year follow-up period, the patient remained free of pain without recurrence.

Discussion

Vascular compression at the REZ of the trigeminal nerve is thought to be one of the etiologies of the TN in most of patients. Several neurovascular diseases including tumors, cerebral aneurysms, and vascular malformations are reported as a cause of TN.[3,4] Brown et al. reported two cases and reviewed 31 papers about TN caused by DAVF.[6] In 33 of the total of 49 patients in their reports, 22 (67%) were males and 11 (33%) were females. The age of the patients ranged from 30 to 77 years. In 48 of the 49 cases, the location of DAVF was specified; tentorial sinus in 34, cerebello-pontine angle in 3, foramen magnum in 3, superior petrosal sinus in 2, transverse-sigmoid sinus in 2, cavernous sinus in 2, inferior petrosal sinus in 1, and posterior

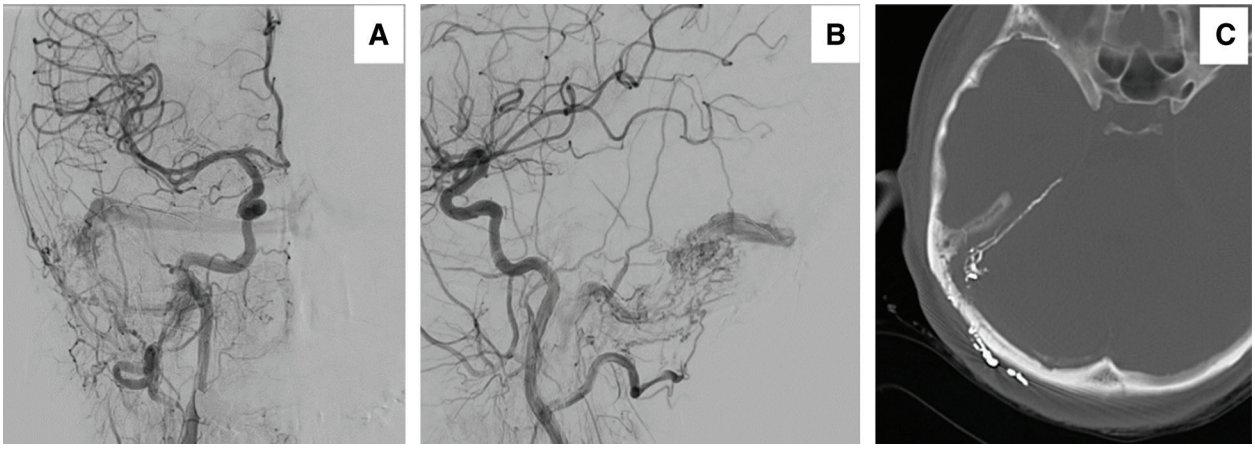

Fig. 2 The right common carotid angiograms (**A** and **B**) after the TAE. Note the AFR, tentorial artery, and ILT were not opacified after TAE. Bone window head plain CT (**C**) showing the distribution of Onyx. AFR: artery of foramen rotundum; ILT: inferolateral trunk; TAE: transarterial embolization

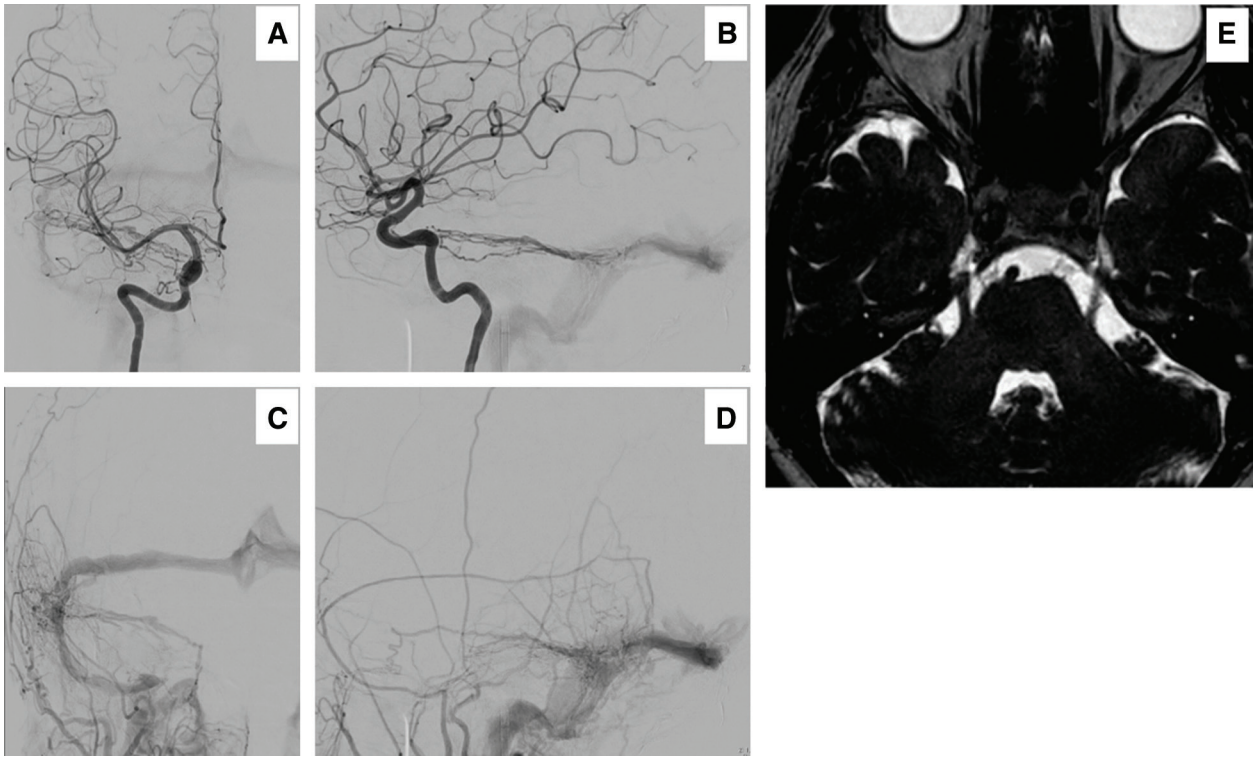

Fig. 3 The right internal carotid (**A** and **B**) and the right external carotid (**C** and **D**) angiograms 3 months after the first treatment showing the recurrence of the DAVF. MRI FIESTA image (**E**) shows no evidence of vessel compression at the REZ. DAVF: dural arteriovenous fistula; FIESTA: fast imaging employing steady-state acquisition; REZ: root entry zone

fossa in 1 case. In 44 cases, 34 (93%) treated with endovascular treatment, 15 with surgical treatment, and 4 with radiosurgery. As a result, complete or eventual relief of the facial pain was achieved in 41 cases. In most cases, arterialized draining vein caused the vascular compressions at the REZ. However, in some cases, the vascular compression was not located at the REZ as our case. Du et al. reported a case of TN with a DAVF in Meckel's cave.[5] In their case, the vascular compression at the gasserian ganglion resulted in TN. However, in our case, FIESTA images showed no evidence of vessel compression at the REZ and Meckel's cave.

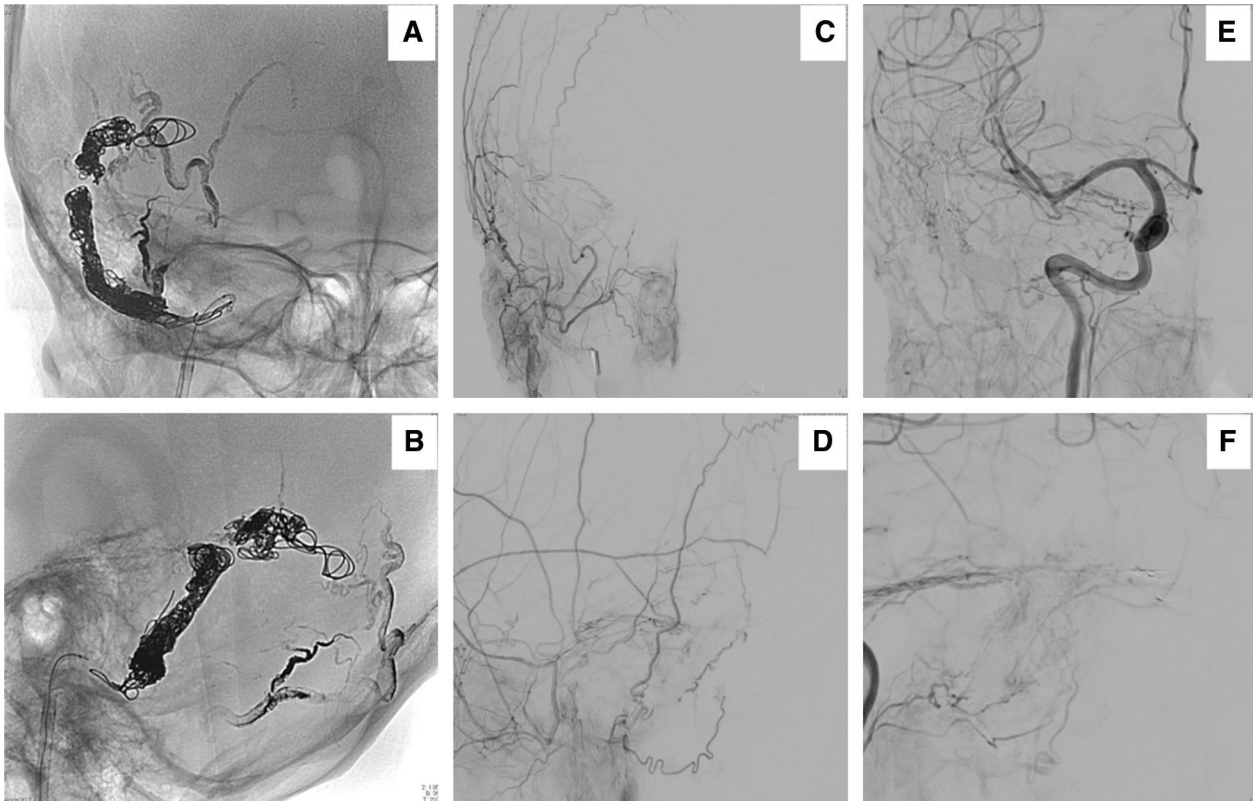

Fig. 4 Anteroposterior view (**A**) and lateral view (**B**) of the postoperative craniograms showing the platinum coils and Onyx cast. Anteroposterior views (**C** and **E**) and lateral views (**D** and **F**) of the external carotid (**C** and **D**) and internal carotid (**E** and **F**) angiograms after the TVE. Note the complete obliteration of the DAVF. DAVF: dural arteriovenous fistula; TVE: transvenous embolization

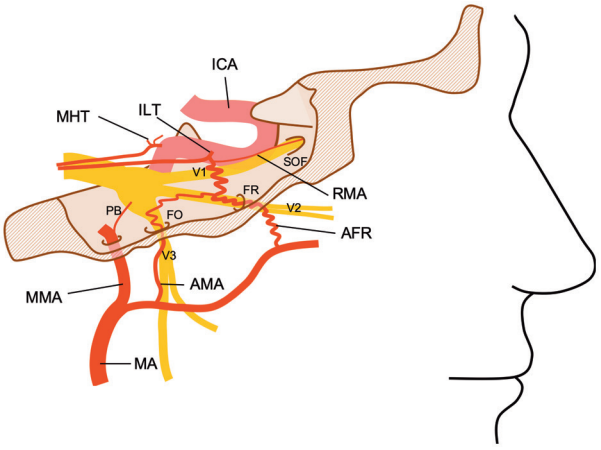

Fig. 5 Illustration (right lateral view) shows the relationship between the trigeminal nerve and branches of the external and internal carotid artery. AFR: artery of foramen rotundum; AMA: accessory meningeal artery; FO: foramen ovale; FR: foramen rotundum; ICA: internal carotid artery; ILT: inferolateral trunk; MA: maxillary artery; MHT: meningohypophyseal trunk; MMA: middle meningeal artery; PB: petrosal branch; RMA: recurrent meningeal artery; SOF: superior orbital fissure

The AFR is a branch of the 3rd segment of maxillary artery which runs along the posterosuperior side of the maxillary nerve and enters the foramen rotundum and can anastomose to the ILT of internal carotid artery (**Fig. 5**).[7] Although it is hard to detect the AFR in angiography in usual condition, it can be detected as a feeder of arteriovenous shunts. Liu et al. investigated the size of the foramen rotundum and the foramen ovale in 21 consecutive TN patients with no vascular compression during surgery and 30 healthy volunteers.[8] They reported that the foramen ovale in the affected side has narrower in size and higher in aspect ratio than the contralateral side. The foramen rotundum in the affected side also tends to be narrower in size compared with the contralateral side. Their results suggested that the narrow skull foramen had an etiological importance in TN patients with no vascular compression at REZ. In our case, there was no significant difference in the size of either foramina rotunda (2.9 mm) (**Fig. 6A** and **6B**). In addition, we confirmed the enlarged AFR passing through the foramen rotundum (**Fig. 6C**). It may suggest the relatively narrow foramen rotundum, causing TN in the maxillary nerve area.

The pain of TN has several features such as sudden onset, brief period, electric shock-like, and recurrent. Symptomatically, TN can be divided in two types. Type 1

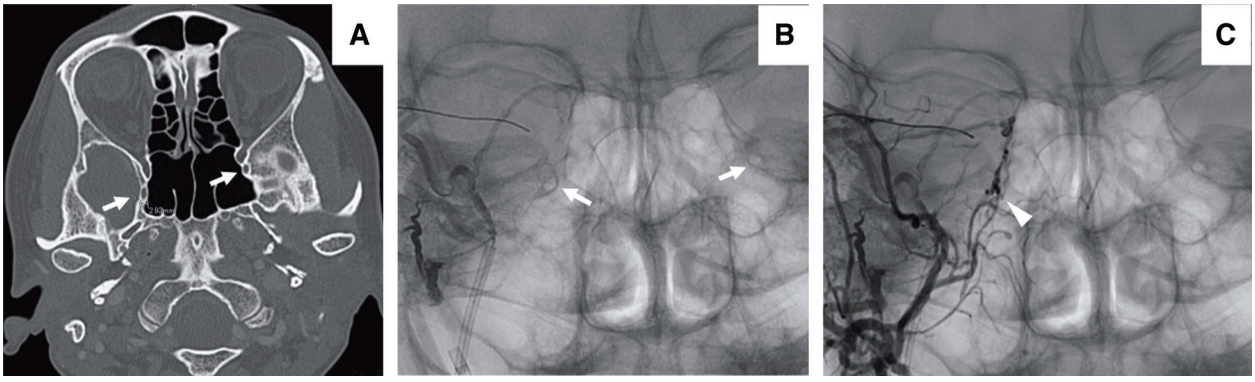

Fig. 6 The bone window of head CT (**A**) and anteroposterior view of the craniogram (**B**) showing the bilateral foramina rotunda (white arrows). There was no significant difference in the size of either foramina rotunda (2.9 mm). Anteroposterior views of the external carotid angiogram (**C**) showing the AFR passing through the foramen rotundum (white arrowhead). AFR: artery of foramen rotundum

has paroxysmal pain alone and Type 2 has continuous pain in the background in addition to paroxysmal pain.[9] Our patient had Type 2 pain rather than Type 1. The pain was transiently worsened for several days after TVE. In coil embolization for intracranial aneurysms, the development of transient headache has been reported. The headache is speculated to be caused by the extension of intracranial arteries by coil mass and the local inflammation due to thrombosis in the aneurysms.[10] Although it is difficult to prove the thrombosis of AFR or perivascular inflammation directly, the local inflammation due to thrombosis may result in a transient worsening of the facial pain in our case.

Conclusion

The maxillary nerve compression by the AFR might result in TN in our case with the DAVF of the transverse-sigmoid sinus. Resolution of the vascular compression after endovascular treatment might contribute to the cure for the facial pain.

Disclosure Statement

The authors declare that they have no conflicts of interest.

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