TRIGEMINAL NEURALGIA IN A PATIENT WITH A DURAL ARTERIOVENOUS FISTULA IN MECKEL’S CAVE: CASE REPORT

OBJECTIVE AND IMPORTANCE: Trigeminal neuralgia is often the result of vascular compression at the root entry zone of the trigeminal nerve. We report a case of trigeminal neuralgia in a patient with a dural arteriovenous fistula in Meckel’s cave. Endovascular closure of the fistula resulted in elimination of the patient’s pain at the gasserian ganglion level.

CLINICAL PRESENTATION: A 77-year-old woman was referred for treatment of trigeminal neuralgia after failed conservative treatment, including multiple gasserian ganglion blocks. Magnetic resonance imaging of the brain suggested a vascular lesion, and cerebral angiography demonstrated a dural arteriovenous fistula in Meckel’s cave.

INTERVENTION: Endovascular coil embolization was performed, with obliteration of the dural arteriovenous fistula and resolution of facial pain but with decreased sensation in the face.

CONCLUSION: Trigeminal neuralgia may be associated with complex vascular lesions around the base of the brain and along the course of the trigeminal nerve. The evaluation of patients with trigeminal neuralgia should include high-quality, thin-section, magnetic resonance imaging scans, to exclude the possibility of vascular lesions and other structural lesions. In particular, patients who are being evaluated for surgical treatment of trigeminal neuralgia should undergo magnetic resonance imaging, with a focus on the course of the trigeminal nerve.

KEY WORDS: Dural arteriovenous fistula, Embolization, Microvascular surgery, Trigeminal neuralgia

Trigeminal neuralgia resulting from dural arteriovenous fistulae (DAVFs) is rare. Six cases were previously reported (3, 8, 16, 18, 26, 30), all of which involved the trigeminal nerve REZ. The arteriovenous fistula in the case reported by Harders et al. (16) appeared only after the patient underwent a suboccipital craniotomy for microvascular decompression. We report a case of trigeminal neuralgia in which magnetic resonance imaging (MRI) demonstrated the presence of flow voids in the region of the gasserian ganglion. Angiography confirmed an arteriovenous fistula. The fistula was treated with endovascular coil embolization, eliminating the need for open cranial surgery.

CASE REPORT

A 77-year-old woman with a history of right-sided Bell’s palsy and a stroke in the 1970s, which resulted in left facial droop, presented
with pain in her right face in the V3 distribution, radiating to her right jaw, that began in November 1996 after a fall on her face. She exhibited no tinnitus or occipital bruit. The pain was described as sharp and electrical and was exacerbated by talking, chewing, or opening her mouth. Most of the pain resolved after a tooth extraction at that time (1996) but began escalating in September 1998, after which the patient underwent another tooth extraction without relief. These symptoms resolved by December 1998, after administration of gabapentin and acetaminophen/hydrocodone, but recurred in January 2000. The patient subsequently underwent right gasserian nerve blocks in March 2000, July 2000, and November 2000, with good pain relief for 2 months each time. She underwent another gasserian nerve block in April 2001, as well as a tooth extraction in May 2001, but continued to experience pain. The patient was then referred to the senior author. MRI scans obtained in June 2001 demonstrated multiple serpiginous flow voids adjacent to the right trigeminal nerve at Meckel’s cave (Fig. 1). Cerebral angiography (Figs. 2 and 3) performed in August 2001 revealed a DAVF adjacent to the right gasserian ganglion, which drained into a subarachnoid vein that drained into the most anteromedial portion of the superior petrosal sinus. The superior petrosal sinus no longer communicated with the cavernous sinus and drained laterally, with high-grade stenosis downstream. Of note, the DAVF also drained into cortical veins. The right anterior division of the middle meningeal artery, the right accessory meningeal artery, and the meningohypophyseal trunk of the right internal carotid artery fed the DAVF. There was no supply from the posterior circulation. Because the presence of cortical drainage indicates a fistula with a high risk of hemorrhage (4), the patient underwent endovascular coil embolization of her DAVF. Two branches of the middle meningeal artery were embolized with polyvinyl alcohol particles, followed by transvenous embolization of the actual fistula site with six Guglielmi detachable coils, with complete obliteration of the DAVF (Fig. 4). After the embolization procedure, the patient was lethargic and complained of a headache. Head computed tomographic scans revealed diffuse subarachnoid hemorrhage, likely resulting from perforation of a vessel during the embolization procedure, and an increase in ventricular size, indicating hydrocephalus. An external ventricular drain was placed, which resulted in improvement of the patient’s mental status. Because the patient experienced difficulty being weaned from the external ventriculair drain without a decline in her mental status, a ventriculoperitoneal shunt was placed. Subsequently, the patient’s mental status returned to baseline levels. The patient no longer has facial pain, after 14 months of follow-up monitoring. An examination demonstrated decreased sensation in response to light touch and pinpricks throughout the right face.

**DISCUSSION**

Vascular compression of the trigeminal nerve at the REZ is a well-established cause of trigeminal neuralgia (1, 20). Other vascular causes of trigeminal neuralgia, such as vascular malformations and aneurysms, are rare but have been reported (1,
11–13, 18, 19, 21, 31, 37–39, 41–43). Among reported series of patients with trigeminal neuralgia, associated arteriovenous malformations were observed in 0.2 to 1.5% of cases (Table 1) (2, 12, 19, 39, 42). DAVFs causing trigeminal neuralgia are much rarer, with only six previously reported cases (Table 1) (3, 8, 16, 18, 26, 30). Most DAVFs are asymptomatic or present with bruit, headaches, or ophthalmoplegia attributable to arterial steal phenomenon, increased intracranial pressure, venous thrombosis, venous ischemia, venous mass effect, or venous rupture (18).

Neurovascular compression syndromes are typically attributable to compression of cranial nerves at or near their REZs (17). Hamlyn (15) observed that, of 46 patients who underwent posterior fossa surgery for treatment of primary trigeminal neuralgia, 42 had a vessel in contact with the nerve. Of those, 28 had a vessel in contact at the REZ, 12 had a vessel in contact lateral to the REZ (point of contact with the nerve more than one-half of the vessel’s diameter away from the brainstem), and 2 had a vessel in contact at the REZ as well as lateral to it. In comparison, a vessel was in contact with the trigeminal nerve in only 5 of 40 cadavers. Of those vessels, four were in contact at the REZ and one was in contact lateral to it (15). Sindou et al. (39) observed the presence of a contacting vessel for 97% of 579 patients with idiopathic trigeminal neuralgia. The site of contact was at the REZ in 52% of cases, in the middle one-third of the nerve in 54%, and at the exit of the nerve from Meckel’s cave in 10% (more than one site of conflict could be observed for individual patients). Similarly, reported cases of arteriovenous malformations or cavernous malformations causing trigeminal neuralgia for which the

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<tr>
<td>Harders et al., 1982 (16)</td>
<td>DAVF</td>
<td>Lateral and sigmoid sinus</td>
<td>Dilated petrosal vein compressing trigeminal nerve</td>
<td>Microvascular decompression followed by posterior fossa surgery with coagulation of dura, followed by another operation with isolation of lateral and sigmoid sinuses</td>
<td>Recurrence of pain after first two operations and resolution after third, at 4-mo follow-up time; postoperative angiography after third operation showed no DAVF</td>
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<td>Rizzo et al., 1982 (34) (trigeminal neuropathy)</td>
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<td>Mendelowitsch et al., 1990 (26)</td>
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<td>Tentorial</td>
<td>Enlarged draining vein compressing trigeminal REZ</td>
<td>Embolized 6 times, followed by surgery and intraoperative embolization</td>
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<td>Ott et al., 1993 (30)</td>
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<td>Enlarged basilar vein of Rosenthal running along trigeminal REZ</td>
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<td>Awad, 1995 (3)</td>
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<td>Not specified</td>
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<tr>
<td>Borden et al., 1995 (8)</td>
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<td>Superior petrosal sinus</td>
<td>Not specified</td>
<td>Excision of dural nidus and ligation of draining vein</td>
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<tr>
<td>Ito et al., 1996 (18)</td>
<td>DAVF</td>
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<td>Endovascular treatment, followed by surgery 2 mo later for recurrence</td>
<td>Resolution of symptoms at 20-mo follow-up time; postoperative angiography at 20-mo follow-up time showed no DAVF</td>
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<td>Present study</td>
<td>DAVF</td>
<td>Superior petrosal sinus</td>
<td>Gasserian ganglion</td>
<td>Endovascular treatment</td>
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*DAVF, dural arteriovenous fistula; REZ, root entry zone.*
cation of the lesion was specified involved compression of the trigeminal nerve root (at an unspecified location along the root) or the trigeminal nerve REZ (Table 2). There were two exceptions, in which the lesion was located in the intra-axial pons (36) or the intramedullary cervical spinal cord (35).

Previously reported cases of DAVFs causing trigeminal neuralgia all involved the trigeminal nerve REZ, when specified (Table 1). Fehlings and Tucker (14) reported a case of a cavernous angioma compressing the gasserian ganglion at Meckel’s cave and causing trigeminal neuropathy. Rizzo et al. (34) reported a case of an external carotid-cavernous sinus fistula that was thought to cause trigeminal neuropathy via direct compression of the gasserian ganglion, via a steal phenomenon, with the fistula drawing off the ganglionic blood supply, or via chronic venous congestion of Meckel’s cave. In both of those previously reported cases, compression of the gasserian ganglion resulted in trigeminal neuropathy instead of trigeminal neuralgia. In contrast, we report a case of trigeminal neuralgia associated with a vascular malformation, namely, a DAVF, compressing the gasserian ganglion rather than the REZ.

In almost all of the reported cases of vascular malformations associated with trigeminal neuralgia, the vascular malformations were discovered during surgery for microvascular de-

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<tr>
<td>Eisenbrey and Hegarty, 1956 (13)</td>
<td>AVM</td>
<td>Cerebellopontine angle</td>
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<td>Verbiest, 1961 (43)</td>
<td>AVM</td>
<td>Cerebellopontine angle</td>
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<td>Johnson and Salmon, 1968 (21)</td>
<td>AVM</td>
<td>Cerebellopontine angle</td>
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<tr>
<td>Bartlow and Penn, 1975 (7)</td>
<td>Carotid-cavernous sinus fistula</td>
<td>Cerebellopontine angle</td>
</tr>
<tr>
<td>von Rad and Tomow, 1975 (44)</td>
<td>Carotid-cavernous sinus fistula</td>
<td>Cavernous sinus</td>
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<td>Jannetta, 1981 (19)</td>
<td>AVM (1/411)</td>
<td>Not specified</td>
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<tr>
<td>Apfelbaum, 1983 (1)</td>
<td>AVM (3/289)</td>
<td>Not specified</td>
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<td>Szapiro and Sindov, 1986 (41)</td>
<td>Venous angioma (2/100)</td>
<td>Not specified</td>
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<td>Drake et al., 1986 (11)</td>
<td>AVM (3/86)</td>
<td>Cerebellopontine angle</td>
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<td>Silber et al., 1987 (37)</td>
<td>AVM</td>
<td>Cerebellum</td>
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<td>Fehlings and Tucker, 1988 (14)</td>
<td>Cavernous hemangioma</td>
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<td>Nishizawa et al., 1988 (29)</td>
<td>AVM</td>
<td>Cerebellum</td>
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<tr>
<td>Tsukahara et al., 1989 (42)</td>
<td>AVM (3/1257), cryptic angiomas (7/1257)</td>
<td>Not specified</td>
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<tr>
<td>Saito et al., 1989 (35)</td>
<td>Cavernous angioma</td>
<td>C1 spinal cord</td>
</tr>
<tr>
<td>Mendelowitsch et al., 1990 (26)</td>
<td>1) AVM, 2) aneurysm</td>
<td>1) Cerebellopontine angle, 2) parapontine</td>
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<tr>
<td>Kikuchi et al., 1990 (22)</td>
<td>AVM</td>
<td>Cerebellum</td>
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<tr>
<td>Raveau et al., 1992 (33)</td>
<td>Venous angioma</td>
<td>Posterior fossa</td>
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<tr>
<td>Shimpo, 2000 (36)</td>
<td>Cavernous malformation</td>
<td>Pons</td>
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<tr>
<td>Sindou et al., 2002 (39)</td>
<td>AVM (3/579)</td>
<td>Posterior fossa</td>
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<tr>
<td>Peterson et al., 2002 (32)</td>
<td>Venous angiomas (5)</td>
<td>Not specified</td>
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<td>Edwards et al., 2002 (12)</td>
<td>Microarteriovenous malformations (5/141)</td>
<td>Not specified</td>
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* AVM, arteriovenous malformation; REZ, root entry zone.
compression or were discovered with angiography after the patient experienced subarachnoid hemorrhage. If preoperative computed tomographic scans were obtained, then lesions either were overlooked or were simply impossible to detect. Those reports generally predated the widespread use of MRI. In our case, the patient underwent high-resolution MRI only after she had undergone multiple failed gasserian ganglion blocks. The abnormal flow voids observed on MRI scans prompted angiographic evaluation, which revealed a DAVF that was treated endovascularly. Thin-cut (3–4 mm) MRI scans are necessary for accurate delineation of the anatomic features of the gasserian ganglion and any associated vascular abnormalities. Dedicated magnetic resonance angiography may also be useful. It must be noted, however, that even high-resolution MRI may fail to detect DAVFs and clinical findings, such as pulsatile tinnitus, occipital brut, and ophthalmoplegia (18, 40), remain important factors in patient evaluations.

Although some DAVFs are developmental anomalies, most DAVFs are acquired (3). Many DAVFs are associated with an adjacent dural venous sinus thrombosis, which might have caused the DAVF or might be secondary to it. Other acquired DAVFs are associated with trauma or a prior febrile illness. There are three major categories of DAVFs associated with trauma, i.e., 1) those that develop at the site of the injury, 2) those that develop remote from the site of the injury, and 3) those that are incidentally observed during evaluation of the injury. Although the patient in this case experienced a fall before the development of her facial pain, the relationship between the fall and the development of the DAVF is unclear, because imaging was not performed before the injury.

Some have claimed that treatment of DAVFs in the tentorial and torcular regions with transarterial embolization is temporarily beneficial but rarely curative (6, 28). However, Ott et al. (30) reported a case of trigeminal neuralgia caused by a DAVF that was successfully treated with transarterial embolization. In our case, the DAVF causing trigeminal neuralgia was successfully treated with transarterial embolization and venous embolectomy.

Trigeminal neuralgia can be treated with a number of percutaneous procedures that result in damage to the trigeminal nerve. Because our patient’s pain relief was accompanied by sensory loss, we cannot be certain that vascular compression by the DAVF was entirely responsible for her pain. Nevertheless, it would be beneficial for all patients with trigeminal neuralgia who experience failure of conservative treatment to undergo high-resolution MRI with thin cuts, to exclude the possibility of vascular causes and other causes of trigeminal neuralgia, before microvascular decompression.

REFERENCES

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his article is interesting because it reports an exceptional case of trigeminal neuralgia hypothesized to be attributable to a DAVF located in the vicinity of the gasserian ganglion. We cannot be sure that the DAVF was the real cause of the neuralgia. It might be that the multiple failed gasserian ganglion blocks injured some tiny arteries and veins of the dural sheath of Meckel’s cave or of the tentorium. Or perhaps these blocks created thrombosis of some veins or dural sinuses in or around the cavernous sinus, with subsequent development of a DAVF (as classically described). Regardless of the mechanism, the patient needed to be treated for relief of her facial pain and occlusion of the DAVF, to eliminate the risk of hemorrhage from its cerebral drainage.

The pain was relieved with the accurately performed endovascular procedure, which eradicated the DAVF. It would have been interesting for the authors to discuss the mechanism of the sensory loss observed after the embolization procedure. Trigeminal sensory fiber impairment resulting from the endovascular procedure (possibly via ischemia) probably played a role in pain relief, as suggested by the authors.

Marc P. Sindou
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The authors describe an unusual presentation of trigeminal neuralgia. They provide a comprehensive literature review; the reported cases of trigeminal neuralgia produced by vascular malformations (i.e., DAVFs and carotid-cavernous fistulae) are thoroughly detailed in Table 1. The authors also highlight the relationship of the DAVF to the anatomic features of the trigeminal nerve. Overall, the article’s most interesting detail is the description of the contacting vessels’ relationship to Meckel’s cave rather than the classic dorsal root entry zone. Like the authors, we think that magnetic resonance imaging-based screening of patients with trigeminal neuralgia is obligatory. The fact that this patient underwent medical therapy for 4 years before undergoing magnetic resonance imaging supports the authors’ point that high-resolution imaging should be performed for all patients with suspected trigeminal neuralgia as soon as the diagnosis is considered.

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John M. Tew, Jr.
Cincinnati, Ohio

Du et al. present a case of trigeminal neuralgia possibly caused by compression from an adjacent dural arteriovenous fistula (DAVF). As they note, vascular compression causing trigeminal neuralgia most often results from branches of the superior cerebellar artery or from the anterior inferior cerebellar artery. In addition, venous compression may cause trigeminal neuralgia. A high-flow arteriovenous fistula contacting the trigeminal nerve could easily have been responsible for the trigeminal neuralgia in this case. The sensory loss after embolization suggests possible infarction of the trigeminal nerve; infarction of the trigeminal sensory nucleus in the brainstem would have manifested with other clinical signs. The only way to confirm that the DAVF was responsible for the trigeminal neuralgia would have been direct observation, requiring craniotomy. The authors stated that the patient had an unusual history of left facial droop after a cerebrovascular accident and right-sided Bell’s palsy, indicating bilateral facial palsy. This history should be confirmed. We agree with the authors’ statement that medically refractory trigeminal neuralgia warrants high-definition magnetic resonance imaging, and this is our routine practice.

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Michael Sandquist
Portland, Oregon

The authors describe an unusual presentation of trigeminal neuralgia. They provide a comprehensive literature review; the reported cases of trigeminal neuralgia produced by vascular malformations (i.e., DAVFs and carotid-cavernous fistulae) are thoroughly detailed in Table 1. The authors also highlight the relationship of the DAVF to the anatomic features of the trigeminal nerve. Overall, the article’s most interesting detail is the description of the contacting vessels’ relationship to Meckel’s cave rather than the classic dorsal root entry zone. Like the authors, we think that magnetic resonance imaging-based screening of patients with trigeminal neuralgia is obligatory. The fact that this patient underwent medical therapy for 4 years before undergoing magnetic resonance imaging supports the authors’ point that high-resolution imaging should be performed for all patients with suspected trigeminal neuralgia as soon as the diagnosis is considered.

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