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Case report

Dural arteriovenous fistula presenting as trigeminal neuralgia: Case report and literature review

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A R T I C L E I N F O

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A B S T R A C T

Background: Trigeminal neuralgia (TN) secondary to a dural arteriovenous fistula (DAVF) is quite rare, and the goal of treatment is to resolve both the fistula and the pain.
Case presentation: We herein report a case of TN secondary to a DAVF in a 64-year-old woman with a 1-year history of right-sided TN. Brain magnetic resonance imaging and digital subtraction angiography showed a right tentorial DAVF. Interventional embolization was performed, but the pain was not relieved after the operation. Six months later, we performed microvascular decompression of the trigeminal nerve. During the operation, we electrocoagulated the tortuous and dilated malformed vein, which was compressing the trigeminal nerve, to reduce its diameter and mitigate the compression on the cisternal segment of the trigeminal nerve. That patient's pain was relieved postoperatively. In addition, we reviewed the literature of TN caused by DAVF and found a total of 30 cases, 22 of which were treated by interventional embolization. Of these 22 cases, the interventional embolization healed the fistula with pain relief in 14 cases and healed the fistula without pain relief in 8 cases. We found that the venous drainage methods of the 8 cases were all classified into the posterior mesencephalic group.
Conclusions: We believe that this drainage pattern contributes to the more common occurrence of unrelieved pain. For such patients, microvascular decompression can be performed with intra-operative coagulation to narrow the dilated veins until the cisternal segment of the trigeminal nerve is no longer compressed. Satisfactory curative effects can be obtained using this technique.

1. Introduction

Trigeminal neuralgia (TN) is a common neurological disease characterized by severe facial pain that seriously affects patients' quality of life. Vascular compression of the trigeminal root exit zone (REZ) is a common cause of primary TN [1]. The most common causes of secondary TN are epidermoid cyst, meningioma, arteriovenous malformation, and dural arteriovenous fistula (DAVF) [2]. TN secondary to a DAVF is quite rare. In the past 40 years, only 22 relevant literature reports were found in PubMed. In some cases, the pain was still not relieved after embolization to cure the arteriovenous fistula; however, the reason is not clear. The present report describes a patient with a DAVF who underwent transarterial interventional embolization to cure the fistula in the early stage, but the TN remained unrelieved. We subsequently performed microvascular decompression (MVD) of the trigeminal nerve in this patient, and

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Abbreviations

TN	Trigeminal neuralgia
DAVF	Dural arteriovenous fistula
REZ	Root exit zone
SPV	superior petrosal vein
MVD	microvascular decompression
CPA	cerebellopontine angle

the pain disappeared postoperatively. In this report, we focus on describing the findings of patients undergoing MVD, discuss the respective reasons for pain relief and lack of pain relief after embolization to cure arteriovenous fistulas, and discuss the surgical strategies for further MVD in patients without pain relief.

2. Case presentation

The patient was a 64-year-old retired woman from Yichang, Hubei Province with a past medical history of cholecystectomy for cholecystitis 25 years prior. She had no history of hypertension, hyperlipidemia, or diabetes. The patient presented with characteristic electric shock-like, lancinating pain in the right V2 distribution area accompanied by right pulsatile tinnitus. The pain was triggered by innocuous stimuli such as brushing teeth or washing her face over the affected trigeminal dermatomes. She was first treated with oxcarbazepine 450 mg daily for 1 month without significant relief. She was then switched to carbamazepine 200 mg daily, which provided initial control of her symptoms for 2 months. However, the pain gradually worsened, requiring an increase in carbamazepine to 600 mg daily. At this higher dosage, discomfort such as dizziness occurred, and the pain was still uncontrollable.

Magnetic resonance imaging showed multiple vascular flow void signals in the right cisternal segment near the trigeminal nerve. Further cerebral angiography revealed a right arteriovenous fistula that was supplied by the petrous branch of the middle meningeal artery of the right external carotid artery and the posterior meningeal artery branch of the right vertebral artery (Fig. 1A&B). The fistula was located between the tentorial dura mater and drained through the superior petrosal vein (SPV) to the basal vein (Borden class III). Hence, interventional embolization was performed through the enlarged right external carotid artery to the posterior branch of the middle meningeal artery. During the operation, a Marathon microcatheter was used for superselection to the fistula; the onyx spread well and the casting was satisfactory. Following embolization, the patient's pulsatile tinnitus resolved, however TN recurred on postoperative day 3 with identical location and characteristics as her preoperative pain. Her postoperative medical regimen consisted of carbamazepine 400 mg daily and the addition of pregabalin 300 mg daily in an attempt to control her persistent facial pain. According to the patient, this adjustment provided a modest decrease in frequency of painful paroxysms, though her symptoms remained suboptimally controlled. Six months postoperatively, follow-up angiography revealed complete obliteration of the DAVF (Fig. 2).

Therefore, we decided to perform MVD via the retrosigmoid approach for this patient. Preoperative re-examination of the T2-constructive interference in steady state (CISS) high-resolution nuclear magnetic resonance sequence showed that the vascular flow

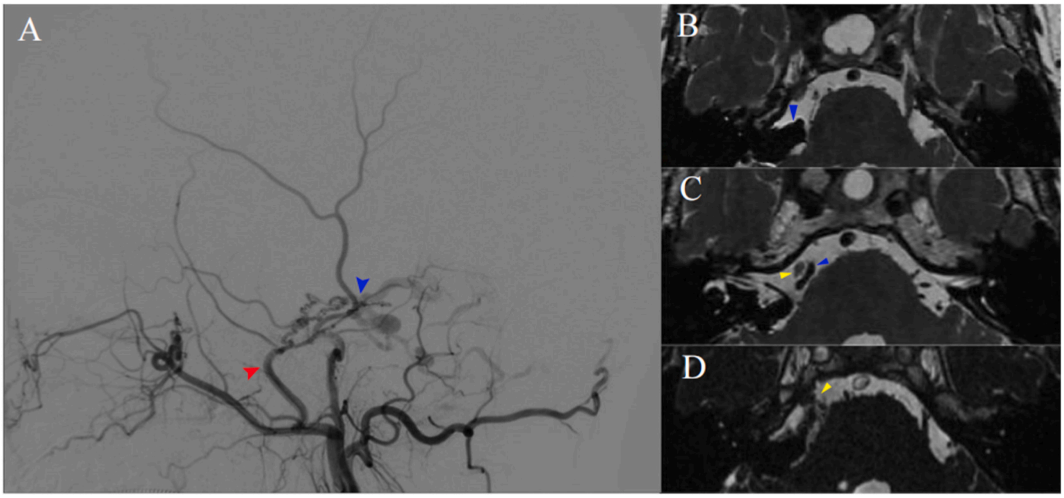

Fig. 1. A. The DSA image of the patient before intervention surgery shows a right external carotid artery angiography. (Red arrow: the branch of the middle meningeal artery; Blue arrow: the location of the arteriovenous fistula opening). **B.** The T2-CISS sequence before embolization showed a huge vascular flow void of the draining vein in the right REZ area. (Blue arrow: the draining vein). **C.** The T2-CISS sequence before MVD surgery showed that the draining vein compresses the cisternal segment of the right trigeminal nerve. (Yellow arrow: the trigeminal nerve; Blue arrow: the draining vein.) **D.** One week after MVD surgery, the draining vein separated from above the main trunk of the trigeminal nerve.

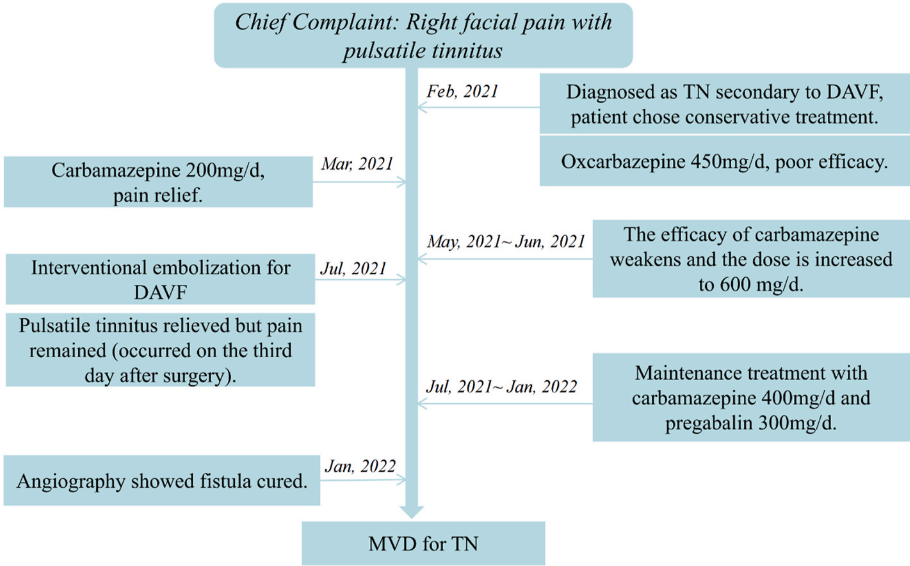

Fig. 2. Clinical course of patients before undergoing MVD.

void signal was significantly reduced compared with that before embolization, but the significantly expanded flow void signal near the trigeminal cisternal segment could still be seen (Fig. 1C). During the operation, the SPV was found to be tortuously dilated and formed a venous lake. The blood vessels showed a post-embolization appearance, were dark blue, and had no arterial pulsation. The pontotrigeminal vein, which joins the SPV from the ventral side, was also tortuously dilated, dark blue in color, and hard in texture, and venous thrombosis was considered. The dilated vein pushed down the trigeminal cisternal segment from the ventral side (Fig. 3A). We attempted to separate the neurovascular vessels with spherical microvascular strippers, but the thrombosed veins were severely sclerotic and difficult to move. Therefore, bipolar coagulation was used to reduce the diameter of the vein in the intermittent 7 W of power so that the trigeminal cistern was no longer compressed. A Teflon pad was then placed between the draining vein and the trigeminal cisternal segment to further open the nerve–vessel space, and the branch of the superior cerebellar artery was found to form compression; therefore, we routinely used a Teflon pad on the responsible blood vessel in the REZ (Fig. 1D). The patient's right facial pain disappeared immediately postoperatively, and she had transient facial numbness that disappeared 3 weeks postoperatively.

3. Literature review

A search of PubMed using varying combinations of the search terms "trigeminal neuralgia," "dural arteriovenous fistula," and "vascular malformation" was undertaken. In each article, its reference list was further reviewed to generate further descriptions of the same cases. We collected and extracted the characteristics of the patients, such as gender and age, the location of the fistula, the drainage vein and the CPA area to which it belongs, surgical approach, the improvement of trigeminal neuralgia, the presence or absence of adverse events, and the results of angiographic follow-up. The classification of the draining veins was determined by referring to and using the venous drainage grouping scheme proposed by Matsushima in 2014. The Borden classification system was used to classify the DAVF because it is easier to apply to case cohorts with varying degrees of descriptive detail than the Cognard classification. When a Cognard type was provided, it was translated into a Borden class using the following scheme: Borden class 1 equivalent to Cognard class 1 or IIa, Borden class 2 equivalent to Cognard class IIb or IIa + b, and Borden class 3 equivalent to Cognard class III, IV, or V. When the manuscript did not provide a Borden or Cognard classification, we applied a Borden class, when able, based on the provided imaging and description using the original Borden classification scheme described in 1995, or referring to the classification provided by the literature review of other articles.

A summary of the literature review is presented in Table 1. A total of 21 articles [4–24] and 30 cases (including the cases provided in this article) were reviewed this time. Of the 20 patients of assigned sex, 10 (50 %) were male and 10 (50 %) were female. Of the 29 patients who described both a direct DAVF treatment and a clinical outcome measure (the 1995 case reported by Borden et al. was excluded for not describing a clinical outcome), 21 (≈72 %) had immediate symptoms of facial pain or delayed complete remission. Of the draining veins corresponding to these 25 patients, 21 (84 %) belonged to the posterior mesencephalic group, 2 cases (8 %) belonged to the anterior pontomesencephalic group, 1 case (4 %) belonged to both the posterior mesencephalic group and the anterior pontomesencephalic group, and 1 case (4 %) belonged to the petrosal group.

We also counted the surgical methods of all cases (Table 2). Of these 29 patients, 5 (≈17 %) underwent microsurgery with postoperative TN relief, and 2 (≈7 %) had pain relief with only Gamma Knife therapy. The remaining 22 patients (≈76 %) underwent some form of endovascular treatment for DAVF. Of these 22 endovascularly treated cases, 19 (≈86 %) underwent transarterial embolization and 3 (≈14 %) transvenous with 1 of these cases representing a combined transarterial and transvenous endovascular

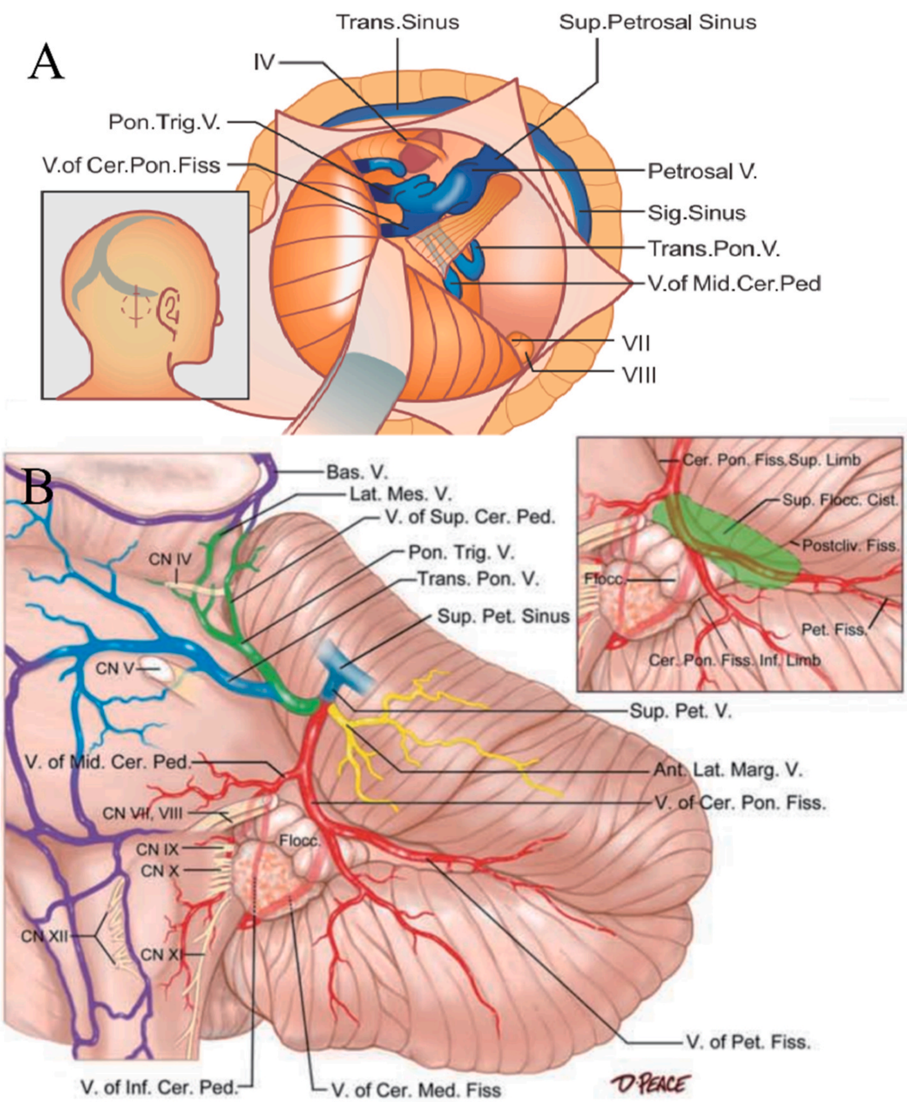

Fig. 3. **A.** Schematic diagram of vascular compression observed during surgery. The tortuous and dilated pontotrigeminal vein pushes the dilated SPV downwards from the ventral side, compressing the trigeminal nerve cistern segment. (Pon.Trig., pontotrigeminal; Cer.Pon., cerebellopontine; Fiss., fissure; Pon., pontine; Mid., middle; Cer., cerebellar; Ped., peduncle; Sig., sigmoid; Sup., superior; Trans., transverse; V., vein.) **B.** Classification of superior petrosal veins cited from Matsushima et al. [3].The largest veins in each of the 4 drainage groups are the vein of the cerebellopontine fissure (red) in the petrosal group, the transverse pontine vein (blue) in the anterior pontomesencephalic group, the pontotrigeminal vein (green) in the posterior mesencephalic group, and the anterolateral marginal vein (yellow) in the tentorial group.

approach. Of these 22 cases, 8 (≈36 %) underwent microsurgery due to unresolved postoperative TN.

4. Discussion

Before 2011, the main treatment of DAVFs was craniotomy. For nearly 10 years, with the advent of new embolization materials and techniques, interventional embolization has become the first choice for the treatment of this disease [25]. The current mainstream view regarding the cause of TN secondary to DAVF is that most of the draining veins of the DAVF near the trigeminal nerve are branches of the SPV, and the steal phenomenon between the arteries and veins causes the pressure of the draining vein to increase and the diameter to expand, resulting in pulsatile compression to the REZ of the trigeminal nerve. In some patients, after the arteriovenous fistula is cured by interventional embolization, the steal phenomenon disappears, the draining vein returns to normal, and no vascular compression forms in the REZ; thus, the patient's pain is relieved [6,17]. However, in other patients, the pain is not relieved after the fistula has been cured by embolization; this occurred in 36.3 % of the cases we reviewed. Why the neuralgia did not disappear after interventional therapy is unclear. The intraoperative findings in our case suggest that although the arterialized pulsation of the vein was relieved by interventional embolization, venous hypertension gradually deteriorated, and the venous thrombosis caused tortuous

Table 1
Literature review of dural arteriovenous fistula causing clinical trigeminal neuralgia.

Num	Author(s), Year	Age (years), sex	Fistula Location	Borden classification	Draining vein	CPA drainage grouping of the draining vein	Treatment	Prognosis	Complication	Angiographic follow-up results
1	Harders et al. et al., 1982 [4]	53, male	transverse sinus-sigmoid sinus	I	SPV	\	Microsurgery	R	\	CR
2	Mendelowitsch et al., 1990 [5]	71, female	tentorium	III	Arachnoid vein in ambient cistern	\	Intraoperative embolization	R	trochlearparalysis	PR
3	Ott et al., 1993 [6]	56, female	tentorium	II	Veins of Rosenthal	posterior mesencephalic group	Transarterial embolization	R	\	PR
4	Borden et al., 1995 [7]	39 , Gender N/S	superior petrosal sinus	III	\	\	Microsurgery	N/S	N/S	N/S
5	Ito et al., 1996 [8]	65, male	Ventral tentorium - petrosum	I	The pontotrigeminal vein	posterior mesencephalic group	Transarterial embolization + Microsurgery	R	\	CR
6	Tomak et al., 2003 [9]	55, female	tentorium	III	Pontine to SPV	posterior mesencephalic group and anterior pontomesencephalic group	Transvenous embolization with coils	R	\	CR
7	Du et al., 2003 [10]	77, female	Mecklecave superior petrosal sinus	III	Meckle intraluminal dilated vein	\	Transarterial embolization/ Transvenous embolization with coils	R	SAH	CR
8	Matsushige et al., 2006 [11]	50, male	Ventral tentorium - petrosum	II	SPV	posterior mesencephalic group	Gamma Knife	R	\	N/S
9	Rahme et al., 2007 [12]	30, male	Ventral tentorium - petrosum	III	SPV	posterior mesencephalic group	Microsurgery	R		CR
10	Lucas et al., 2007 [13]	50, male	transverse sinus-sigmoid sinus	III	SPV	\	Microsurgery	R	\	CR
11	Akhaddar et al., 2010 [14]	42, male	Ventral tentorium - petrosum	I	SPV	posterior mesencephalic group	Microsurgery	R	\	N/S
12	Wakuta et al., 2013 [15]	57, female	Ventral tentorium - petrosum	I	SPV- bilateral posterior midbrain veins	posterior mesencephalic group	Transarterial embolization + Microsurgery	R	\	CR
13	Lu et al., 2013 [16]	58, male	tentorium	III	contralateral SPV	posterior mesencephalic group	Transarterial embolization	R	\	PR
14–18	Robert et al., 2015 [17]	5 patients, Gender N/S	Tentorium*4 foramen magnum*1	III	The pontotrigeminal vein	posterior mesencephalic group	Transarterial embolization	DR	SAH*1	CR
19	Mendes et al., 2016 [18]	53, female	inferior petrosal sinus	II	cerebellopontine fissure vein	petrosal group	Transvenous embolization with coils	R	\	CR
20	Fukutome et al., 2017 [19]	75, female	cavernous sinus	I	posterior cavernous vein	anterior pontomesencephalic group	Transarterial embolization	R	oculomotor paralysis	CR

(*continued on next page*)

Table 1 (*continued*)

Num	Author(s), Year	Age (years), sex	Fistula Location	Borden classification	Draining vein	CPA drainage grouping of the draining vein	Treatment	Prognosis	Complication	Angiographic follow-up results
21	Huang & Yu, 2018 [20]	35, male	foramen magnum area	III	SPV	posterior mesencephalic group	Microsurgery	R	\	CR
22-25	Li et al., 2018 [21]	4 patients, Gender N/S	tentorium	III*3 II*1	\	posterior mesencephalic group	Transarterial embolization + Microsurgery *3 Transarterial embolization *1	R R	\ \	CR CR
26	Jiang et al., 2019 [22]	57, male	tentorium	III	SPV-lateral midbrain vein	posterior mesencephalic group	Transarterial embolization + Microsurgery	R	\	CR
27	Okromelidze et al., 2019 [23]	71, female	tentorium	I	posterior cavernous vein	anterior pontomesencephalic group	Gamma Knife	R	\	PR
28	Brown et al., , 2020 [24]	53, male	tentorium	III	The pontotrigeminal vein-SPV	posterior mesencephalic group	Transarterial embolization	R	Headache	PR
29		72, female	tentorium	I	superior petrosal sinus	posterior mesencephalic group	Transarterial embolization + Microsurgery	NR	\	PR
30	Present report	64, female	Ventral tentorium - petrosum	III	SPV-The pontotrigeminal vein - Basal vein	posterior mesencephalic group	Transarterial embolization + Microsurgery	R	\	CR

SPV, superior petrosal vein; N/S, not specified; R, relief; NR, not relief; DR, delayed relief; SAH, Subarachnoid hemorrhage; CR, complete resolution; PR, partial resolution.

Table 2
Surgical methods statistics of 29 cases.

Borden classification	Microsurgery	Interventional Embolization	Combined microsurgery after interventional embolization	Gamma knife	Total number
I	2	1	3	1	7
II	0	3	0	1	4
III	4	10	5	0	19
Total number	5	14	8	2	29

expansion of the draining vein. Therefore, we believe that the trigeminal nerve remains compressed because of irreversible venous dilatation changes in some cases [8,22]. Xu et al. reviewed 642 cases and found that patients with preoperative sole venous compression of the trigeminal nerve had worse pre- and postoperative pain scores, as well as higher recurrence rates, compared to those with sole arterial compression [26]. Similar findings have been reported in other cohort studies [3,27]. One potential explanation is that the non-pulsatile blood flow pattern and chronic, constant compressive force exerted by dilated veins may elicit a more pronounced neuropathic pain response versus the intermittent pulsations from arterial loops [26]. Therefore, in cases refractory to embolization, radiosurgery, or MVD, the pulsatile nature of the compressive force as well as differential biomechanical properties between muscular arterial versus thin-walled venous structures may play a key role in symptomatology of TN.

We sought to identify factors affecting the efficacy of interventional embolization for TN secondary to a DAVF. For this purpose, we reviewed the literature and determined the Borden class of all cases (Table 1). The Borden classification is widely used in the clinical setting to assess the severity of DAVFs [28]. In Borden class I, the veins drain directly to the venous sinuses and dural veins, and the patient's prognosis is good. In Borden class II, the veins return both to the venous sinuses and back to the pial veins, with the increased flow to the fistula. In Borden class III, the veins only return to the pial veins near the venous sinuses, and the drainage veins arterialize and become tortuous and dilated. The pressure in the deep veins increases and this is combined with retrograde cortical venous drainage [7]. By analyzing the Borden classification of cases treated with interventional embolization in Table 2, we found that among 15 patients with Borden class III, 5 did not experience pain relief after embolization; that all 3 patients with Borden class II experienced pain relief; and that 3 of the 4 patients with Borden class I experienced no pain relief. Therefore, the Borden classification cannot be used to evaluate the embolization efficacy of TN secondary to a DAVF.

The efficacy of embolization may be related to the anatomical relationship between the draining vein and the trigeminal nerve. In 2014, Matsushima proposed a grouping system of venous drainage in the CPA [29], in which three groups can be associated with neurovascular conflict. The first is the posterior mesencephalic group, the second largest draining group in the area. Its main trunk is the pontotrigeminal vein, which drains blood from the dorsal midbrain and adjacent to the superior cerebellar peduncles. The second group is the anterior pontomesencephalic group, the largest branch of which is the transverse pontine vein, which is located at the midpontine level. It anastomoses with the median anterior pontomesencephalic vein, and some of the transverse pontine veins directly join the pontotrigeminal vein. The third group is the petrosal group, which is the largest draining group in the CPA. The largest tributary is the vein of the cerebellopontine fissure, which not only drains the petrosal cerebellar surface but also drains the surrounding areas of the fourth ventricle, the lateral medulla, and the middle and inferior cerebellar peduncles (Fig. 3B). According to the results of our literature review in Table 1, a total of 18 cases of DAVF drained in the posterior mesencephalic group treated by interventional embolization, 8 required microsurgery after embolization, and the non-relief rate was as high as 44 %; 1 case drained in the petrosal group and 2 cases drained in the anterior pontomesencephalic group, with pain relief after embolization. As to why the drainage of the posterior mesencephalic group is prone to poor prognosis, we speculate that the relative position of the draining vein and the trigeminal nerve is a major reason. Since the pontotrigeminal vein flows into the SPV from above, when it expands, it can push the SPV running parallel to the cisternal segment of the trigeminal nerve downward, thereby compressing the trigeminal nerve.

The case reported here is a typical case in which the draining vein belongs to the posterior mesencephalic group, draining from the SPV to the pontotrigeminal vein to the basal vein. After embolization, the arterialized pulsation of the SPV is relieved, but the structural changes of the tortuously expanded SPV itself cannot be immediately improved. Venous thrombosis formed, the texture became hard, and the pushing of the dilated pontotrigeminal vein made it more difficult to separate the malformed venous mass from the trigeminal nerve during the operation.

MVD can be used for patients whose pain is not relieved after interventional embolization. A study by Fukushima et al. found that separating the petrosal vein from the nerve root contributes to pain relief in patients with petrosal vein conflict, regardless of the accompanying artery [30]. Different from what is seen in primary TN surgery, the SPV often exhibits venous lake changes; after embolization, vein thrombosis forms and the texture becomes hard, making the SPV difficult to move and significantly increasing the difficulty of the operation. In the present case, low-frequency electrocoagulation was used to reduce the diameter of the compressing vein, and inserting Teflon to open the nerve–vessel space. However, electrocoagulation of veins may lead to postoperative complications of venous insufficiency, so it needs to be used with caution. Therefore, in addition to electrocoagulation, a safer method is to push the malformed venous trunk with glue away from the trigeminal nerve as much as possible. Since the malformed vein has a certain elasticity, it can be pushed open to a certain extent, but it may also rebound due to elasticity. In this case, it can be fixed with medical bioprotein glue.

5. Limitations

This study is limited by its presentation as a single case report and the inherent constraints of this study design. Case reports represent one of the more descriptive and lower levels of evidence in the evidence-based medicine hierarchy. While valuable for detailing unique clinical scenarios, the findings lack the robust generalizability derived from higher-quality evidence such as prospective clinical trials or meta-analyses. Larger, systematic studies are needed to definitively evaluate optimal management strategies for dural arteriovenous fistulas presenting as trigeminal neuralgia refractory to embolization alone."

6. Conclusions

A DAVF is a rare cause of TN, and the preferred treatment is interventional embolization. In most patients, the fistula and pain are simultaneously cured after embolization, and it is considered that embolization relieves the pulsatile compression of the malformed vein in such cases. However, some patients experience no pain relief after embolization of the fistula; this situation may be related to the drainage of the posterior mesencephalic group. After embolization, thrombosis in the SPV may result in dilation of the SPV, which continues to compress the cisternal segment of the trigeminal nerve. For such patients, further MVD can be performed.

Ethics statement

We hereby confirm that the present study conforms to the ethical standards and guidelines of the journal.

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Additional information

No additional information is available for this paper.

Data availability statement

The authors confirm that the data supporting the findings of this study are available within the article.

CRediT authorship contribution statement

Xuan Dai: Writing – original draft. **Dongyuan Xu:** Writing – original draft. **Keyu Chen:** Formal analysis. **Yuankun Cai:** Investigation. **Zhimin Mei:** Investigation. **Ji Wu:** Formal analysis. **Lei Shen:** Supervision. **Jingyi Yang:** Supervision. **Nanxiang Xiong:** Writing – original draft, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

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