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WisdomGale

CASE REPORT

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Intrinsic trigeminal nerve arteriovenous malformation: a rare cause of trigeminal neuralgia

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ABSTRACT

Intrinsic trigeminal nerve (TGN) arteriovenous malformation (AVM)is an exceptional cause of trigeminal neuralgia (TN). Imaging is not only important for visualising the nidus, but should also precise feeding arteries and draining veins, which are very variable in this location. Magnetic resonance imaging provides the spatial resolution required to show the relation of the nidus with the TGN, by using adapted sequences for vascular and neurological structures assessment. Digital subtraction angiography remains the gold-standard imaging technic for showing the angioarchitecture of the AVM, proving its superiority to computed tomography angiography. In this article, we report the case of a 77 years-old woman with TN secondary to an intrinsic TGN AVM with unusual venous drainage, diagnosed by an MRI of the cerebellopontine angle, with the assessment of its angioarchitecture on computed tomography angiography.

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Introduction

Trigeminal neuralgia (TN) is marked by transient, recurrent, and intense facial pain in the territory of the trigeminal nerve (TGN) [1]. It affects 4–13 per 100,000 people annually, with a higher prevalence in women, showing a female-to-male ratio of 1.5–1.7. Most cases occur after age 50, although it can appear in the second and third decades of life, and is rarely diagnosed in childhood [2].

The International Classification of Headache Disorders, Third Edition (ICHD-3) categorizes TN into three types: classical TN, characterized by paroxysmal or continuous pain often due to neurovascular conflicts; secondary TN, resulting from conditions like space-occupying lesions or multiple sclerosis; and idiopathic TN, which has no known cause [3].

While vascular causes, particularly neurovascular conflicts, are the primary culprits, rare causes like brain arteriovenous malformations (AVMs) have also been noted in a few case reports [4].

Research by García-Pastor et al. [5] revealed that 1.3% of all brain AVMs are identified through TN, with 9.8% involving posterior fossa AVMs.

Even rarer, intrinsic AVMs of the TGN occur in about 0.6% of surgical patients [4].

This case report presents TN due to intrinsic AVM in the TGN, featuring unusual venous drainage.

Case Presentation

Patient information and clinical results

A 77-year-old woman presented with a 2-year history of recurrent right hemi-facial pain, described as electric shock-like, affecting the maxillary (V2) and mandibular (V3) nerve territories, consistent with TN. Her pain episodes lasted 10–30 seconds and occurred in clusters over several days. Physical examination revealed normal motor and sensory function of the TGN, with no identifiable trigger zone. After treatment with carbamazepine, the patient experienced a partial response, with significant reductions in both the frequency and intensity of her pain episodes.

Diagnostic evaluation

Due to ongoing residual symptoms, an MRI of the cerebellopontine angle was conducted.

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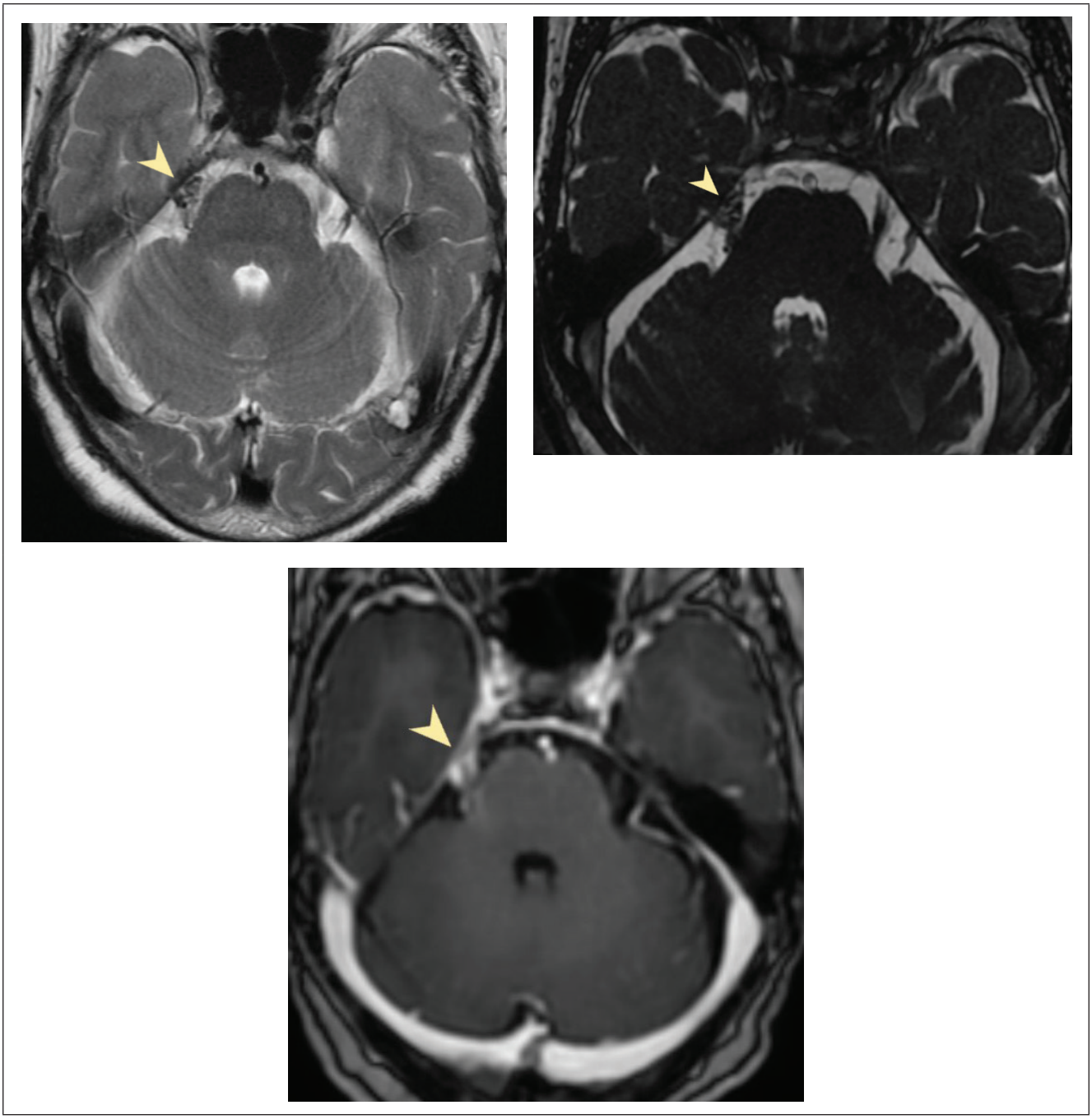


Figure 1. Brain MRI: Cluster of flow-voids around and within the cisternal and cavernous portions of the right trigeminal nerve (Arrow head) on axial T2 weighted-imaging (A), better seen on 3D FIESTA sequence (B), showing a vascular enhancement on axial T1-weighted sequence after gadolinium injection.

The protocol included sagittal T1 sequence, axial T2, Fluid-attenuated inversion recovery, diffusion, and T2 gradient echo (GE) sequences, three-dimensional fast imaging employing steady state acquisition (FIESTA) sequence, three-dimensional time-of-flight (3D TOF) sequence, as well as 3D T1 and axial T1 spin echo (SE) sequences following gadolinium injection.

The T2-weighted sequence (Fig. 1A) revealed flow-void signals surrounding the cisternal and cavernous portions of the right TGN. The three-dimensional FIESTA sequence (Fig. 1B) provided a clearer visualization of the cluster of flow voids within the right TGN, suggesting an intrinsic AVM measuring 7 × 5 mm. The axial T1-weighted sequence post-gadolinium injection (Fig. 1C) showed enhancement of these flow voids, supporting their vascular origin.

Due to the unavailability of conventional and 3D rotational angiography in our facility, we relied

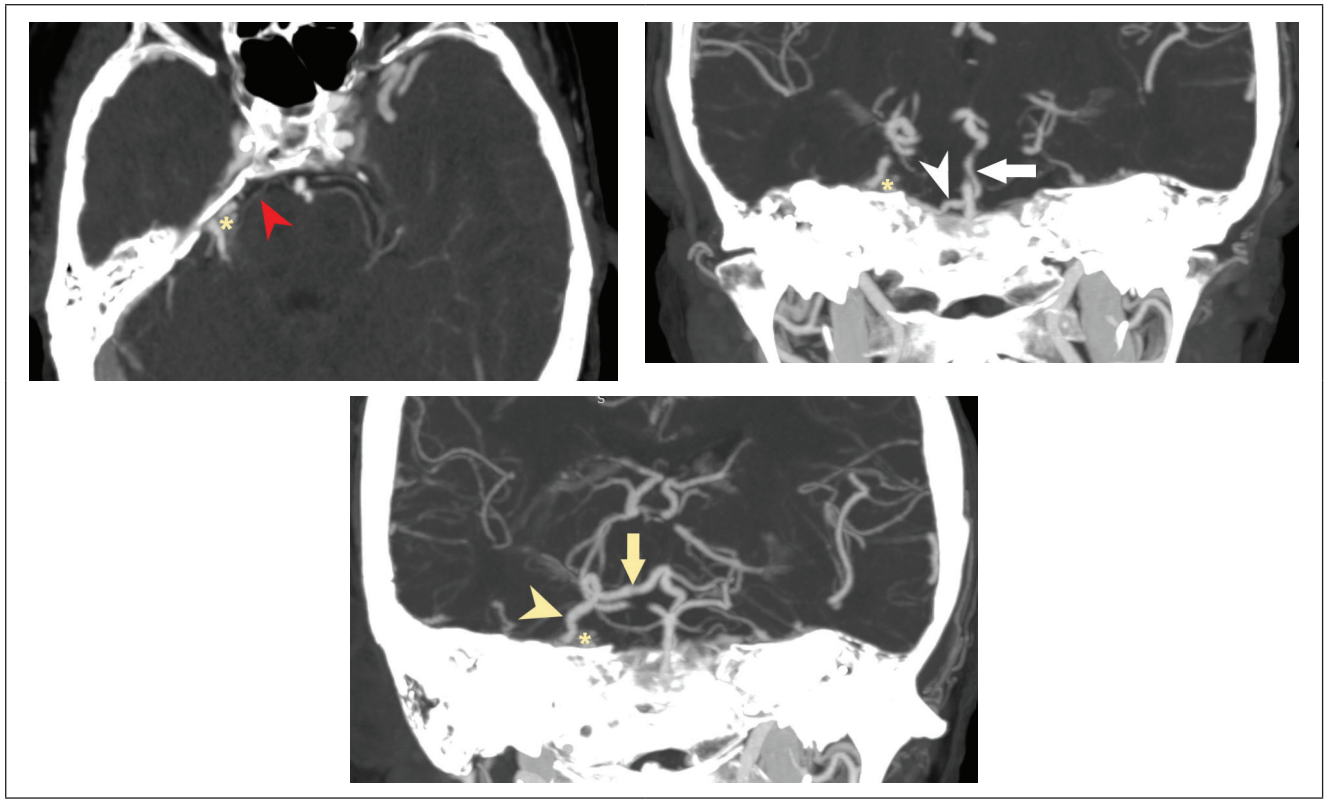


Figure 2. Brain CTA: The AVM (asterisk) is fed by a PA (red arrow head) (A), and drained by a right TPV (White arrow head) to the APMV (White arrow) (B), and by the LMV (Yellow arrow head) to IPV (Yellow arrow) (C).

on computed tomography angiography (CTA) to assess the feeding arteries and draining veins. We performed an acquisition after automatic contrast injection, with the region of interest placed on the brachiocephalic artery.

The AVM was supplied by a pontine artery (Fig. 2A) and drained medially through a right transverse pontine vein (TPV) and the anterior pontomesencephalic vein (APMV) (Fig. 2B), as well as superiorly via the lateral mesencephalic vein (LMV) and the interpeduncular vein (IPV) (Fig. 2C), all of which were significantly enlarged.

Therapeutic intervention

The patient opted against the proposed surgical treatment and chose medical management instead, taking carbamazepine at 800 mg twice daily.

Long-term outcome

The patient has maintained her treatment regimen of carbamazepine at 800 mg twice daily, resulting in a consistent partial response. While she continues to experience some pain episodes, the frequency and intensity remain significantly reduced compared to her initial presentation.

Discussion

TN caused by AVM is rare, accounting for 0.22%–1.78% of primary TN cases [6], and AVMs located in cranial nerves other than the optic nerve remain highly uncommon [7].

According to that, we can present AVMs intrinsic to the TGN as an extremely rare entity with only very few reports known of such cases.

In most reported cases, the feeding artery was a PA [6,8], with drainage mainly through the petrosal vein [4,8–11]. (Table 1).

In our case, the AVM was also fed by a PA, but drained via the TPV to the APMV and the LMV to the interpeduncular vein IPV. Nagai et al. [12] described a similar case of TGN AVM, but the drainage was solely through the TPV into the APMV.

MRI should be conducted for all patients with TN to identify or rule out secondary causes, such as AVMs [13].

The optimal sequences for assessing the posterior fossa and TGN in order to visualise AVM, include 3D TOF MRA and high-resolution T2-weighted imaging (HR T2WI). These must be done in thin slices and in the same section plane, with analysis based on native images in order to properly define

Table1. Feeding and draining vessels reported in the literature of patients with TN secondary to intrinsic TGN AVM.

Patient	Author	Feeding artery	Draining vein
1-5	Edwards et al. [6]	PA, Superior cerebellar artery (SCA) PA, Anterior inferior cerebellar artery (AICA) PA PA PA	Not precised
6	Karibe et al. [4]	SCA	Petrosal vein
7	Anderson et al. [8]	PA, SCA	Basal vein of Rosenthal, Superior petrosal vein
8	Sumioka et al. [9]	AICA	Petrosal vein
9	Ferroli et al. [10]	Not precised	Petrosal vein
10	Choudhri et al. [11]	AICA, Persistant trigeminal artery	Petrosal vein
11	Nagai et al. [12]	PA	TPV, APMV
12	Our case	PA	TPV, APMV, LMV, IPV

the link between the fifth cranial nerve and the AVM [14].

The optimal sequences for assessing the posterior fossa and TGN in order to visualize AVM, include 3D TOF MRA and high-resolution T2-weighted imaging (HR T2WI), with digital subtraction angiography (DSA) remaining the gold standard for detailed visualization of AVM angioarchitecture.

However, it remains difficult to identify all of the feeding arteries and drainage veins, hence the importance of DSA, as it remains the gold-standard imaging technic for AVMs. It provides a detailed view of the angioarchitecture of AVMs, including the feeding arteries, nidus, venous drainage, and any associated aneurysms [15].

Treating an intrinsic AVM is controversial due to the importance of preserving TGN function. While medical therapy with carbamazepine or oxcarbazepine is typically the first line for TN [2], invasive options such as surgical resection, stereotactic radiosurgery, or embolization carry varying risks of nerve function loss [4,7]. Stereotactic radiotherapy offers a promising approach with lower complication risks, though patients remain at risk for hemorrhage during the obliteration period [7]. Microvascular decompression followed by radiosurgery has also been suggested as a treatment strategy for neuralgia [6].

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