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Trigeminal neuralgia due to arterialization of the superior petrosal vein in the context of dural or cerebral arteriovenous shunt

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ABSTRACT

Objective: Trigeminal neuralgia (TN) is a rare revealing symptom of dural or cerebral arteriovenous (AV) shunt. Few isolated cases have described the association between TN and arteriovenous shunt. We presented our experience concerning cerebral AV shunt revealed by a TN.
Methods: Between 2003 and 2013, 548 brain AV malformations (bAVM) and 268 dural AV fistulas (dAVF) were treated and a retrospective analysis identified 10 consecutive cases with TN. We reviewed their clinical and angiographic data and as well as their outcome after endovascular treatment.
Results: Among the 10 patients, 5 presented a bAVM and 5 a dAVF. TN was contralateral to the AV shunt in 2 cases (20%). The superior petrosal vein (SPV) was arterialized in all cases. An ectasia of the SPV which could explain a compression of the trigeminal nerve was found in two cases only (20%). It seems that a venous hyperpressure in the ponto-trigeminal vein (tributary of the SPV) is responsible of the TN, rather than a compressive mechanism.
Conclusions: Contrary to other publications on this topic, we believe that the venous reflux into tributaries of the superior petrous vein (and in particular the ponto-trigeminal vein) seems to be the first mechanism of the TN rather that the nerve compression.

1. Introduction

Trigeminal neuralgia (TN) is frequently idiopathic and, in few cases, the revealing symptom of a cerebral pathology [1]. Dural and cerebral arteriovenous shunts are one of these pathologies but this circumstance is very rare [2]. The association between trigeminal neuralgia and arteriovenous shunt of cerebrum or dura has been reported in few cases only [3–16]. Isolated cases reported good pain reversibility after embolization [4,7,8,10,13,15], surgery [2,5,6] or radiotherapy [3,11,16].

We report our cases of symptomatic trigeminal neuralgia secondary to brain arteriovenous malformation (bAVM) or dural arteriovenous fistula (DAVF) in order to understand the pathophysiology, the structure that compresses the nerve (in case of compression) and factors influencing the presence of the neuralgia.

2. Materials and methods

2.1. Patient demographics

From a prospectively gathered registry, we identified 548 patients with cerebral AVM and 268 patients with dural arteriovenous fistulas who were treated in our institution between 2003 and 2013.

In this study, we retrospectively reviewed data from all the patients who suffered from trigeminal neuralgia, and we identified 10 consecutive cases. Demographic data were recorded for each patient including age, sex, clinical presentation and angiographic characteristics of the AV shunt.

2.2. Trigeminal neuralgia characteristics

Each patient had a pre-treatment consultation with a neurologist who assessed the following characteristics of the trigeminal neuralgia: trigeminal territory, presence of a trigger zone, corneal reflex, potential hypoesthesia associated to the neuralgia, side of the neurological symptom, duration of the symptoms and the ongoing medication.

Abbreviations: TN, trigeminal neuralgia; bAVM, brain arteriovenous malformation; dAVF, dural arteriovenous fistula; AV, arteriovenous; DREZ, dorsal root entry zone.

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Table 1
Demographic data and trigeminal neuralgia description.

Variable	$n = 10$
Male	8 (80%)
Mean age (range)	37 (25–56)
Trigeminal territory	
V1	3 (30%)
V2	1 (10%)
V3	6 (60%)
Trigger zone	6 (60%)
Loss of corneal reflex	2 (20%)
Associated hypoesthesia	2 (20%)
Side	
Homolateral to the shunt	8 (80%)
Contralateral to the shunt	2 (20%)
Duration of symptoms (months)	25.9 (1–120)
Treatment	
No	3 (30%)
Carbamazepine	5 (50%)
Gabapentine	2 (20%)

2.3. Arteriovenous shunt anatomy

Each patient had a radiologic assessment with cerebral magnetic resonance imaging (MRI) and six-vessels cerebral digital subtraction angiography (DSA). Two distinct pathologies were analyzed: brain arteriovenous malformation and dural arteriovenous fistula. In all cases, we noted angioarchitecture landmarks to understand the relation between the underlying vascular disease and the trigeminal neuralgia. We insisted on the analysis of the mechanisms involved in the trigeminal impairment (compression or venous reflux) and the vascular structures involved.

2.4. Endovascular therapy

All patients were treated by endovascular embolization performed under general anesthesia. After cerebral MRI and DSA were performed, the appropriate working projections for catheterization of the arterial supply of the AV shunt were recorded. Treatment options included parent artery occlusion or nidus embolization by cyanoacrylate synthetic glue (*Glubran, GEM, Viareggio, Italy*) or Onyx (*ev3 Neurovascular, Irvine, CA, USA*).

2.5. Clinical evolution

Follow-up started at the time of the last embolization session and finished with the last visit or angiography. Angiographic follow-up after complete exclusion of an AVM consisted of one year and long term (5 years) DSA. For dAVF, only one control angiography was performed 6 months after the treatment to confirm the occlusion of the AV shunt. Neurological evaluation regarding the trigeminal neuralgia evolution was made between 6 and 12 months after the treatment. The presence of residual pain, its type and the prescription of an analgesic treatment were assessed.

3. Results

3.1. Clinical presentation

Between 2003 and 2013, 10 patients (8 [80%] men, mean age = 37 years old) with dural or cerebral AV shunts revealed by a trigeminal neuralgia were treated in our department. Patient baseline data and neuralgia types are described in Table 1.

The facial pain was in the ophthalmic division (V1) of the TN in 3 cases (30%), the maxillary (V2) in 1 case (10%) and in the mandibular (V3) in 6 cases (60%). A trigger zone was noted in 6 cases (60%), particularly in V3 neuralgia, a loss of corneal reflex in 2 cases (20%) and an associated hypoesthesia in 2 other cases (20%). The symptoms were homolateral of the AV shunt in 80% of cases. The mean duration of symptoms was 26 months. Seven patients presented a drug-resistant pain and the three others had no specific analgesic medication. Five patients were treated by carbamazepine (from 800 to 1200 mg/day) and 2 patients received gabapentine (1200 mg/day) without pain relief.

3.2. Angio-architecture of brain arteriovenous malformations

Five patients presented a bAVM with an identifiable nidus, which was located in the cerebellar hemisphere in 4 cases and in the vermis in one case. The nidus size was less than 30 mm in 2 cases and between 30 and 60 mm in 3 cases. Four nidi were diffuse and one was compact. The Spetzler–Martin grade [17] was 3 in 4 cases and 4 in 1 case. All the AVMs presented multiple arterial supplies (2–5), at least one of the superior cerebral arteries (SCA) was involved in every case and antero-inferior cerebellar arteries (AICA) were involved in 4 cases as well as the postero-inferior cerebellar arteries (PICA). Flow-related arterial aneurysm was noted in 3 cases but without relation to the pons or the trigeminal nerve pathway. The venous drainage always involved the superior petrosal vein (Dandy's vein). In 1 case of right cerebellar hemisphere AVM, the venous drainage impaired the lateral recess vein then the superior petrosal with large ectasia, which compressed the trigeminal nerve DREZ medially. In another case presenting a right-sided TN (Fig. 1), a large cerebellar hemisphere AVM located in the left side had a venous drainage by the left SPV, the left ponto-mesencephalic vein, the left basal vein with large reflux in the right SPV by the right ponto-mesencephalic vein and causing a contralateral TN. The remaining three cases had a venous drainage by the SPV without reflux but with hyperpressure in tributaries of this vein.

3.3. Anatomy of dural arteriovenous fistulas

The five other patients presented dural arteriovenous fistulas. The shunt was in the cerebellopontine cistern in 3 cases, the foramen magnum in 1 case and at the free margin of the tentorium in one case. The number of arterial feeders ranged between 2 and 5 with participation of the petrous branch of the middle meningeal artery and clival branches of meningo-hypophyseal trunk in the majority of the cases. Two fistulas had recruited feeders by the AICA and made then duro-pial fistulas. The three other cases were duro-arachnoidal fistulas. According to the classification of Lariboisière [18], 3 were grade 3 and 2 grade 4. The superior petrosal vein was arterialized in all the cases. In one case, the arterialized superior petrosal vein presented a large ectasia (Fig. 2) that created a major compression of the cisternal portion of the trigeminal nerve. Cystic rearrangements could be seen on pre-operative MR imaging. In another case with contralateral TN, a large venous reflux in the SPV could be seen. In the remaining cases, the venous drainage of the fistula increased venous pressure in the SPV.

3.4. Treatment of the pathology

All the patients were treated by endovascular therapy as a stand-alone treatment modality. All dAVF were cured after one embolization session and 6 month angiographic follow-up showed the durability of the exclusion of these fistulas. Four fistulas were treated with Onyx injection and the other case with glue. One patient presented per-procedural hemorrhage due to venous perforation and a transient cerebellar dysmetria was deplored. Thirteen embolization sessions were performed to treat the 5 patients with AVM (overall: 2.6 embolizations per patient; range: 2–5), and 25 arterial pedicles were navigated to catheterize the AVM nidus. Glue was used in 8 sessions and Onyx in 5. Three AVM nidi were