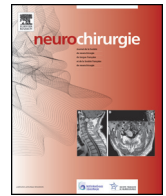




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Short clinical case

Can fifth nerve mapping guide rhizotomy for recurrent trigeminal neuralgia? Case report

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ABSTRACT

Introduction. – Microvascular decompression (MVD) is usually considered the first-line treatment for trigeminal neuralgia (TN) when medical treatments fail. Recurrence is rare and best treatment option is controversial. MVD was proposed as a feasible and effective technique for recurrent TN by many authors. Nevertheless, in a substantial number of cases, not any impingement or deterioration are found intraoperatively and partial selective rhizotomy is then advised. The rhizotomy site is mostly guided by anatomical landmarks, but variations due to scarring and adhesions are common pitfalls in these second surgeries. Intraoperative monitoring is infrequently used during MVD for trigeminal neuralgia. We describe the use of nerve mapping in a case of recurrence, revealing an unexpected rootlet distribution and thus safely guiding partial rhizotomy.

Clinical presentation. – A 53-year-old woman had suffered from bilateral trigeminal neuralgia for 10 years. Symptoms began on the right side. MVD resolved her symptoms but, after a few months, she developed left TN which persisted after left MVD, radiofrequency and radiosurgery. She was referred to our center for a second MVD on the left side. Intraoperative inspection detected no relevant findings, and nerve mapping followed by partial selective rhizotomy was performed. Complete pain relief was achieved. There were no complications.

Conclusion. – Rhizotomy is seldom employed for refractory trigeminal neuralgia. The effects of previous treatments can jeopardize anatomical landmarks. Nerve mapping seems a promising tool to improve results.

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1. Introduction

Trigeminal neuralgia (TN) is the most common facial neuropathy [1]. For patient refractory to medical treatment microvascular decompression (MVD) is usually considered the first choice treatment given its high and durable success rate (80%). Nevertheless, recurrence of TN after successful MVD is reported up to 20% at 10 years [2,3]. Patients with severe symptoms may thus require additional treatments but controversy still exists regarding the best option of therapy [4]. MVD is accounted from many authors as a viable and effective option for retreatment [5]. However, a substantial number of negative intraoperative findings are reported. In

these case many authors advice a partial sensory rhizotomy (PSR) [2,5].

Even though intraoperative neurophysiological monitoring is now a common neurosurgical tool, the relative advantage of its application during MVD is still debated. In the present paper, we describe the use and possible advantages of trigeminal nerve mapping to guide PSR in a case of TN recurrence.

2. Clinical presentation

2.1. Patient data

We describe the case of a 53-year-old woman suffering from bilateral trigeminal neuralgia since 10 years. Symptoms began in the right side. After a first attempt of medical therapy she underwent a right MVD. She gained complete symptoms control in the right side but after a few months she referred left

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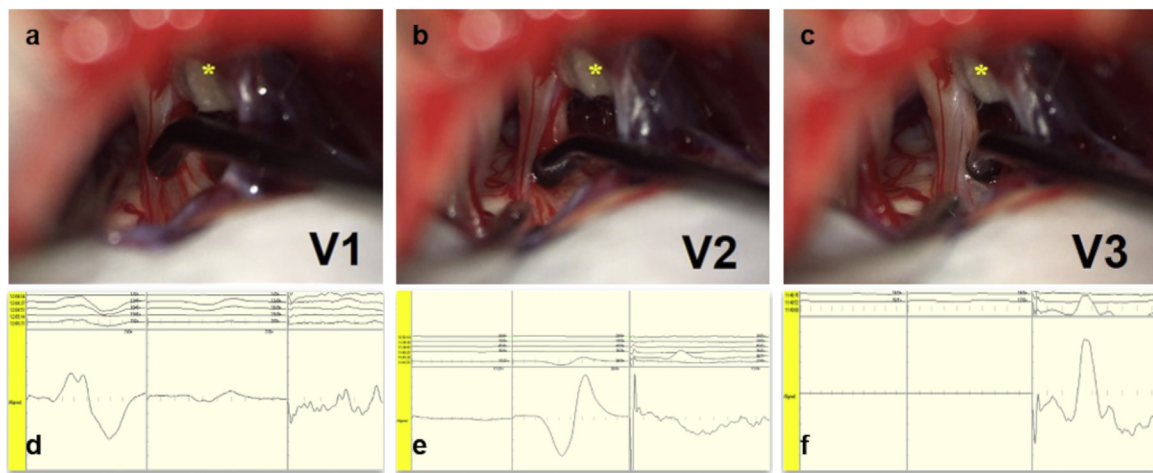


Fig. 1. First mapping of the Vth nerve. Operatory field is seen through a left retro-sigmoid approach (cerebellum is seen in the bottom, tentorium in the right). Images and relative nerve traces show the Vth nerve mapping by mean of concentric bipolar stimulation. First mapping records showed V1 rootlet in a caudal position (A–D), V2 in the middle (B–E) and V3 in the most rostral position (C–F). The Teflon felt is visible next to the upmost and distal part of the nerve (yellow star).

trigeminal neuralgia (II e III branches). Medical therapy definitively failed after 7 years, thus she underwent a left MVD with Teflon interposition. Pain recurred after 1 months (Barrow Neurological Institute – BNI – pain score IV) [6]. Three percutaneous radiofrequency procedures were then performed which again failed in symptoms control. Two consecutive radiosurgery treatments were then attempted without any further clinical improvement.

Finally, the patient was referred to our center for a second MVD procedure on the left side. She was under multiple medical treatment (Carbamazepine 1400 mg/day, Pregabalin 450 mg/day and Baclofen 20 mg/day) with dismal pain control (BNI pain score IV). She furthermore complained drowsiness from the medical therapy and bilateral incomplete facial numbness. Notwithstanding the several and different treatments, along about her clinical history the patient described pain as abrupt and sharp, being evoked by trigger point stimulation while chewing or speaking, then consistent with a persistent trigeminal neuropathy.

Preoperative neuroradiological assessment with MRI showed the Teflon felt and an atrophic fifth nerve in left cerebello-pontine angle. Any clear related vascular conflict was found.

2.2. Surgery

Informed consent was obtained. Surgery was performed through the previous left retrosigmoid approach. Intraoperative findings consisted of a broad scarring of the cerebellopontine angle whose debridement finally show the fifth nerve and a Teflon felt adherent to it. The fifth nerve looked markedly atrophic and discolored. Exploration of the nerve root and root entry zone didn't show any further vascular compression neither arterial nor venous. Even the Teflon felt, which was adherent to the cisternal part of the nerve, didn't seem to generate any contact with the REZ thus not apparently explaining the recurrence of symptoms (Fig. 1).

Since the absence of any relevant finding, we then look for a selective partial rhizotomy. In order to decide which part of the nerve include in the rhizotomy, we performed a direct fifth nerve mapping. Intraoperative neurophysiological mapping of trigeminal branches was carried out by mean of a constant current bipolar concentric stimulator, with 200 μ s pulse width. For sensory nerve action potential (SNAP), trigeminal nerve (pars major) was stimulated at three sites along its circumference with stimulus intensity of 0.2–1.0 mA. For composed motor action potentials (CMAP), motor pars (pars minor) of trigeminal nerve was initially stimulated with intensity of 0.5–1.5 mA. Once a nerve response was

seen, the stimulation intensity could be reduced (< 1.0 mA) to allow a more specific localization. In this case, we were able to measure latency and amplitude of each trigeminal branch SNAP.

Surprisingly, results of mapping showed the sensitive rootlets disposition was almost inverted according to common anatomical descriptions [6], with the V1 rootlet harboring the most caudal part of the nerve and V3 the rostral one (Fig. 1).

We besides went on dissecting and removing the Teflon felt seeking concealed compressions. A repeated exploration of the nerve confirmed the lack of nerve distortion or vascular conflicts. Hence, we rerun nerve mapping with the same settings, finally identifying even the motor branch. Registration of the sensitive rootlets from this second mapping showed a different anatomical pattern, with V3 response arising from the inferior (caudal) portion of the nerve and V1 response from the upper (rostral) one (Fig. 2). Once repeated stimulations confirmed this last outline of responses, we performed a partial internal rhizotomy of the nerve component corresponding to V2 and V3, according to the patient prominent symptoms (Fig. 3).

2.3. Postoperative course

Patient referred complete pain remission (BNI pain score I). The degree of facial numbness was the same as the preoperative evaluation. No postoperative complications were recorded. Follow-up evaluation at six months confirmed bilateral complete pain control without medication (BNI pain score I).

3. Discussion

Microvascular decompression (MVD) has become a widely accepted treatment for TN in patient refractory to medical therapy, but controversy still exists over the best option for patient with recurrent symptoms. Although MVD for recurrent TN (re-MVD) is associated to better results than other techniques, surgery-related risks are reported to be higher than those of the first procedure and the success rate to be lower, with variable results in the literature [5].

While comparing the various series reporting on re-MVD, the most frequent intraoperative findings are new vascular compressions followed by scarring, adhesions or nerve distortion. As well as for our patient, there is a substantial number of cases in which not any evident vascular conflict or nerve alterations are found (range 10–76%). Indeed, in our case, we couldn't identify any

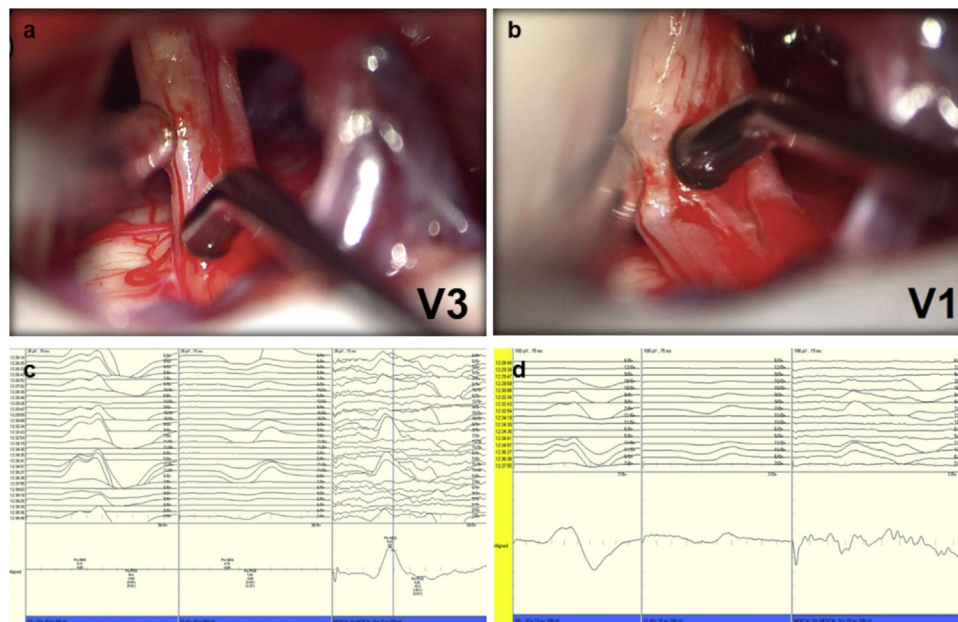


Fig. 2. Second mapping of the Vth nerve. Operatory field is seen through a left retro-sigmoid approach (cerebellum is seen in the bottom, tentorium in the right). Teflon felt has been removed. Images show the Vth nerve mapping by mean of concentric bipolar stimulation. Images and nerve trace show V3 (A–C) and V1 (B–D) response respectively from the caudal (A) and rostral (B) portion of the nerve.

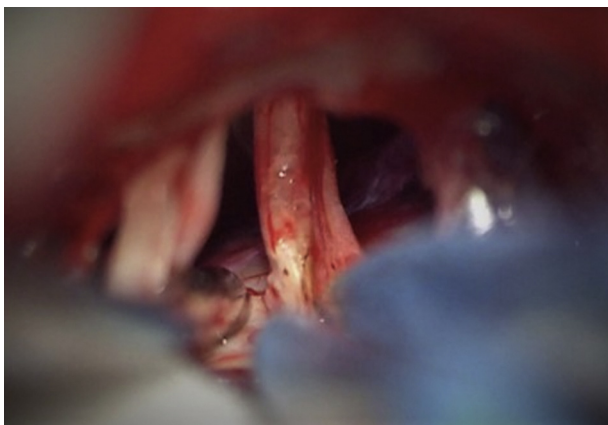


Fig. 3. Rhizotomy. Operatory field is seen through a left retro-sigmoid approach. After repeated mapping, a partial internal V2 and V3 rhizotomy was performed the closest to the pons.

compression able to clearly explain pain recurrence, since even the Teflon felt was distant from the REZ [2,4,8–12]. For those cases with poor intraoperative findings, many authors have reported PSR as a viable and effective treatment. The technical heterogeneity of this ablative procedure and the variable results in pain relief and hypoesthesia, as well as the high risk of impaired corneal reflex, had led to limit the use of this technique over the years [4,13].

Terrier et al. [13] recently published a series of 22 patients in which they performed a PSR tailored according the somatotopy of trigeminal nerve, concluding that limiting the rhizotomy to the ventrolateral two thirds of the nerve would allow to spare corneal sensitivity. Indeed, they focused on anatomical works over the fifth nerve fibers, whose nociceptive portion would harbor preferentially the lateral part of the juxta-pontine portion of the nerve (*pars major*). On the other hand, epicritic and proprioceptive fibers would lay in the intermediate part (*pars intermediaris*) while the motor rootlet in the medial part of the nerve (*pars minor*). Furthermore, they recognize a topographic somatotopy for the *pars major* fibers

with V1 occupying their posterior and medial portion, whereas V2 and V3 the anterior and lateral ones.

In their series, Terrier et al. report intact corneal reflex in all the cases, but a 9% of anesthesia dolorosa of the hemitongue [13]. Unfortunately, their series account for only one patient with previous MVD surgery. Nevertheless, they stressed the relevance of iatrogenic or physiological anatomical inter-individual differences since this would potentially mislead the site of rhizotomy with consequent variability of the expected sensory loss [7,13].

The technique of antidromic nerve mapping potentially allows to exceed these anatomical intervariations. Stechison et al. have already reported this technique in 6 patients undergoing mapping guided rhizotomy for atypical facial pain. Only one out of the 6 patients had formerly undergone a MVD procedure. In their results, intraoperative registrations confirmed the expected anatomical disposition in all cases [14].

While comparing our mapping records to those of the Stechison' series, we found greater latency values than mean latency values of those patients; on the contrary we found similar results to the patient of Stechison' series with previous MDV procedures [14]. These last data could both indicate reliability of the neurophysiological recording as well as possible slight alterations of the V cranial nerve in patients with recurrent TN. Obviously, these findings need validation from a larger series but should these data being confirmed, they could potentially help better understanding the pathophysiology of TN.

The case we report is highly representative of the possible unexpected anatomical discrepancies in re-MVD surgery and for the first time it emphasizes how nerve mapping can provide a reliable information about this variability. Actually, in our patient, repeated mapping before and after Teflon removal showed opposite results about the sensitive fibers disposition. We ascribed these changings to a possible nerve distortion from the interposition material, since the different pattern was recorded once it has been removed. A more valuable explanation conceive a registration of the *pars intermediaris* from the upper part of the nerve during the first mapping mimicking a V3 response. Indeed, the *pars intermediaris* harbors fibers of the epicritic and proprioceptive sensitivity of the three branches [13]. The exact identification of the three rootlets has

therefore allowed a real tailored partial rhizotomy, thus adapting the procedure to fulfill the main symptoms of the patient.

4. Conclusion

While excluding variability factors due to anatomical alterations, nerve mapping can potentially improve outcome results for TN repeated surgeries. Actually, this technique can contribute to reduce the risk of potential severe sensitive complications, which constitute the main concern about TN recurrences treatments.

Validation from a larger case series is necessary, but we found this simple and highly reproducible technique a viable but still underexploited tool in the treatment of this challenging pathology.

Informed consent

Consent to treat clinical data for research was obtained.

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Disclosure of interest

The authors declare that they have no competing interest.

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