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Research Article

A Retrospective Study of Trigeminal Neuralgia Cases Caused Solely by Veins

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Abstract

Background: Trigeminal neuralgia (TN) caused solely by veins is relatively rare, and it is more difficult to visualize the responsible vein than the artery using MRI. There has been no comprehensive description of the condition and best practices for diagnosing and surgically treating it. Here, we aimed to visualize the vein causing TN and discuss intraoperative considerations in view of the reported relatively highly frequent recurrence of TN with vein compared to TN with arterial compression.

Methods: To study vein-caused TN, we reviewed 110 cases of microvascular decompression (MVD) from the past 3 years, including seven cases (6.4%) involving veins alone. All patients underwent preoperative three-dimensional magnetic resonance cisternogram/angiogram (3D-MRC/MRA) fusion imaging. Surgery was performed using conventional retrosigmoid approach, and pain level was evaluated using Barrow Neurological Institute (BNI) Pain Intensity Score.

Results: 3D-MRC/MRA helped detect vein involvement in all cases. Surgical findings confirmed compression caused by vein, consistent with preoperative imaging. In six (86%) cases, trigeminal nerve was adherent to arachnoid membrane, and nerve was deformed. After adhesions were dissected and vein was moved, improving nerve deformity, resulting in complete disappearance of pain (BNI grade I) in all six cases. In the single case with no adhesion (14%), transposition improved TN from BNI grade IV to II.

Conclusions: 3DMRC/MRA helped diagnose TN caused by vein compression and aided surgical simulation and decision-making. We recommend to dissect adhesions around trigeminal nerve to improve deformity. Long-term follow-up is necessary because vein-caused TN has a high recurrence risk.

Keywords: Microvascular Decompression; Trigeminal Neuralgia; Vein; Magnetic Resonance Imaging

Introduction

Idiopathic trigeminal neuralgia (TN) has diverse causes, including vascular compression [1,2], brain tumor [3], arteriovenous malformation [4], adhesion without vascular compression [5-8], and deformity of the pyramidal bone [9]. In TN caused by vascular compression, several cases have been reported to be caused by both arteries and veins [10-13]. The number of reported cases of TN caused solely by venous compression are few and there is no comprehensive description of this condition and discussion of best practices for its diagnosis and surgical treatment in the literature.

To address this knowledge gap, we conducted a retrospective study of cases with TN caused by venous and/or arterial compression in our surgery practice. We have been performing 3DMRC/

MRA fusion images as a preoperative diagnosis of TN. There is no difficulty accurately visualizing the anatomical relationship between the trigeminal nerve and the culpable artery, but traditionally, it has been difficult to visualize the culpable vein because of its low flow velocity. However, we were able to visualize the vein based on the identification of the veins around the trigeminal nerve in three dimensions. We took two-dimensional-fast imaging employing steady-state acquisition (FIESTA) images and superimposed them with MRA images to create 3D-MRC/MRA fusion image.

In this report, we describe our efforts to perform accurate preoperative diagnosis of TN caused solely by a vein using 3D-MRC/MRA fusion images. The condition can be treated by coagulation and cutting the vein, or by moving the vein, but both options are controversial. Therefore, we also examined which surgical method would be the most appropriate in practice to treat TN caused by vein compression. In addition, since recurrence is a concern, we examined what to keep in mind and what to concentrate on during the surgery.

Methods and Materials

Retrospective cohort and clinical evaluation

Among the 110 cases of TN surgery in our practice in the past 3 years (April 2019 to March 2022), we saw 7 cases (6.4%), including 2 males and 5 females, that involved a vein as a sole vessel responsible for TN (Table 1). The mean age of the 7 patients was 66.7 ± 11.0 years (48-79). Five (71%) were left-sided, and 2 (29%) were right-sided. To assess pain severity before and after surgery, we used the Barrow Neurological Institute Pain intensity score (BNI) [14]. Three cases (42%) presented with grade V, and four (57%) with grade IV. Four patients (57%) had presented with paroxysmal electric shock pain with a trigger point as in typical TN, and three patients (43%) had persistent pain without a trigger point (atypical facial pain). All preoperative patients were treated with pregabalin (Pfizer Inc. NY, USA) and carbamazepine (Kyowa Pharmaceutical Inc. Tokyo, Japan), surgery was considered for patients who had a decreased response to pain medication.

Three-dimensional-magnetic resonance cisternogram/angiogram fusion images

Three-dimensional-magnetic resonance cisternogram/angiogram fusion (3D-MRC/MRA) was employed as a preoperative neuroimaging study for all cases. The usefulness of 3DMRC/MRA fusion images for preoperative diagnosis of TN has been described previously [15,16], and is briefly described in this study. For MRA, the arteries and veins around the trigeminal nerve were plotted based on Fast Imaging Employing Steady-state Acquisition (2D-FIESTA) images and composed into a 3D volume rendering. Fusion images of 3D magnetic resonance cisternograms and angiograms were reconstructed by a perspective volume-rendering algorithm from the volumetric data sets of magnetic resonance cisternography, obtained by a FIESTA sequence, and coregistered volume-rendered MRA. This imaging technique makes it possible to depict structures, such as blood vessels, nerves, and brain parenchyma, in three-dimensional detail in the cistern, which helps diagnose trigeminal neuralgia and even allows simulating surgery.

Microvascular decompression

Microvascular decompression (MVD) was performed using a retrosigmoid approach in the lateral recumbent position. Intraoperative auditory brainstem response was continuously monitored. After craniotomy and dura opening, the horizontal fissure was peeled off to reach the trigeminal nerve, the arachnoid membrane around the superior petrosal vein was dissected, and the surgical field was expanded to create an adequate working space for safe manipulation. Six of the seven patients (86%) underwent dissection of the adhesion around the trigeminal nerve, and in three of these cases (43%), sufficient decompression could be achieved by dissection alone. After the dissection, transposition was performed, and interposition was performed in one case. In one case (14%), no adhesions were found, so only transposition was performed.

Results

The area affected area of the trigeminal nerve was located in the third branch in 5 cases (71%), and in the second branch in 2 cases (29%). All patients were initially treated with pregabalin and carbamazepine for TN, which was effective in six patients (86%). In all cases (100%), the 3D-MRC/MRA fusion image helped clearly reveal the responsible vein that ran around the trigeminal nerve contacting it. In this study, it was difficult to depict and evaluate the nerve deformity. The subsequent intraoperative inspection confirmed that the preoperative 3DMRC/MRA fusion image identified the culpable vein with high accuracy. The vein most common implicated was the transverse pontine vein with 3 of 7 cases (43%), followed by the superior petrosal vein, 2 cases (29%). In addition, there was one case involving the vein of the cerebello-

pontine fissure (14%), and another one, the pontotrigeminal vein (14%) (Table 2). The site of vascular nerve compression was in the cisternal region (in the mid-third of the root) in 5 cases (71%), and in the root entry zone (REZ) of the nerve, in 2 cases (29%) (Table 2). Postoperatively, the pain completely disappeared (BNI, grade I) in six patients (86%), and improved to BNI grade II in one patient (14%). There were no cases of permanent complications (Table 2). The average postoperative follow-up period was 24.0months at this point (12 months to 36 months).

Illustrative case

A 70-year-old woman presented with electric shock pain in the region of the third branch of the left trigeminal nerve that began to appear 2 years ago, triggered by washing her face. She was diagnosed with TN by her family doctor and was treated with carbamazepine. Recently, the efficacy of carbamazepine had decreased, and the patient was referred to our department for further treatment. She had a medical history of well-treated hypertension. Family history had no findings of note.

Neuroimaging findings, based on 3D-MRC/MRA fusion image, on admission, demonstrated that the left trigeminal nerve was compressed by the vein of the cerebellopontine fissure alone. No affected artery was observed (Figure 1a, b).

The patient was operated on using a left retrosigmoid approach in the right inferior recumbent lateral position. The trigeminal nerve was severely adherent to the thickened arachnoid membrane in the cerebellar tent, from the region of Meckel's cavity to the REZ (Figure 2a). In addition, the trigeminal nerve had a severe deformity, an arched formation, due to adhesions pulling it into the cerebellar tent. Importantly, the vein of the cerebellopontine fissure was seen making contact with the ventral side of the nerve (Figure 2a), the compression site, confirming what was indicated in the preoperative 3D-MRC/MRA fusion image. The deformity of the nerve could not be accurately assessed preoperatively in the fusion image, but, upon retrospective evaluation of the image, the nerve appeared to be slightly retracted to the lateral side (Figure 1b). To improve the deformity of the nerve, the thickened arachnoid membrane around the nerve was dissected. Then, the vein in contact with the nerve was detached from it, pulled out to the cerebellar surface, and adhered to it with fibrin glue (Figure 2b). After confirming the improvement of the trigeminal nerve deformity and reaching sufficient decompression, MVD was completed.

Figure 1: a, b: Preoperative 3D-magnetic resonance cisternogram/angiogram (3D MRC/MRA) fusion images demonstrate the compression of the trigeminal nerve (arrow) ventrally by the vein of the cerebellopontine fissure (arrowhead).

Figure 2: Trigeminal nerve attached to the cerebellar tentorial side is freed by dissection around the nerve and transposition of the vein.

a. Pre-decompression view. The nerve is attached by a thickened arachnoid membrane and is deformed (arrow). Contact on the ventral side of the nerve by vein (arrowhead) is confirmed.

b. Post-decompression view. After dissection around the trigeminal nerve and transposition of the vein (double arrows), the improvement of the nerve deformity was confirmed.

The postoperative course was uneventful. TN disappeared immediately after MVD, and the patient was discharged from our hospital 7 days after the surgery without any new neurological deficit. Two years have passed since the surgery with no apparent complication or recurrence.

Discussion

We are reporting near complete success at fusion image aided surgical alleviation of TN cases for which nerve compression by a single vein is responsible. This is a rare condition and, in contrast to our records, results of past surgical interventions have been mixed. Several studies have demonstrated that veins are only involved in 12.5%-26.8% of TN [17,18], and TN solely caused by vein compression is an even rarer condition, reported in a mere 3.3% to 18.7% of TN cases [2,10,13,19,20]. In our practice, this fraction was 6.4% (7 out of 110 cases), near the lower and of the reported range. Usual TN, in which an artery is a responsible vessel, presents as paroxysmal electric shock pain with a trigger [1,2]. In contrast, it has been reported that 40% of TN, in which a vein is a responsible vessel, presents with the atypical symptom of persistent pain without a trigger [11]. This fraction is consistent with our observation that 43% (3 out of 7) of patients diagnosed with TN caused by venous compression presented with an atypical symptom. It has been reported that results of MVD in cases presenting with atypical symptoms were not as good as those in typical cases and that surgical indication should be considered [5,8]. Cases with facial numbness, which was not represented in our cohort, have been described as having particularly poor outcomes [8]. Using preoperative imaging worked to our advantage, as it clearly detected the vein as the responsible vessel, allowing us to proceed to surgery for patients presenting atypical symptoms, with the intervention resulting in excellent improvement. Our results suggest that surgery should be undertaken based on detailed preoperative imaging, even in cases of atypical symptoms and a vein as the responsible vessel.

Consistent with reports that the two veins most responsible for compression TN were the transverse pontine vein, followed by the superior petrosal vein [10-13], we identified the transverse pontine vein as the most common responsible vessel (43% of cases), followed by the superior petrosal vein (29%). We also saw one case (14%) each where the culpable vein was the vein of cerebellopontine fissure and the pontotrigeminal vein. Typical TN often presents with arterial compression in the REZ [1,2], whereas for cases of venous compression, the locus of compression has been reported to be more common in the cisternal portion than in the REZ [10-13]. For comparison, the compression site was the cisternal portion in 5 of our 7 cases (71%), and at REZ of the nerve in the remaining two cases (29%). Indeed, trigeminal compression by a blood vessel in the cisternal portion may be one of the characteristics of TN in which the vein is the responsible vessel.

During MVD, the horizontal fissure was first dissected as deeply as possible to reach the trigeminal nerve. The maneuver was useful to prevent stretching and damage to the superior petrosal vein. The options of manipulations are performing internal neurolysis, coagulating and cutting the responsible vein, or preserving it and adding either an interposition or performing transposition. Internal neurolysis, a technique that consists of physically dissecting the trigeminal nerve longitudinally and damaging it, has been reported to be effective [18,21]. It has been also reported that small veins can be cut to obtain sufficient decompression and good results without permanent complications if the venous return territory could be first confirmed [13,22]. But unlike an artery, a vein may be difficult to safely dissect and move, especially if small branches of it are distributed on the surface of the brainstem [13,23]. Indeed, the manipulation of the vein risks disabling venous circulation to brainstem areas serving vital functions. On the other hand, the safer alternative, preserving the vein and proceeding with interposition or transposition, carries the risk of recurrence if these interventions prove to be insufficient. In fact, the recurrence rate of TN is higher for cases caused by venous rather than arterial compression [11].

In this study, we decided to preserve the vein in the first intervention and only perform cutting of the vein or internal neurolysis of the trigeminal nerve in case of a recurrence. In our sample, six out of the 7 cases (86%) featured adhesions around the nerve, and in 3 of these 6 cases, the deformity of the nerve was improved after dissection and sufficient decompression could be achieved by dissection alone. In one case, where a branch to the brainstem could not be moved significantly, interposition was added. Among the 3 cases where transposition was performed, one case (14%) did not present any adhesion (it was a case of root compression in the REZ). In our small sample, all of these interventions were successful, suggesting that there may be no significant difference in surgical efficacy between them. This interpretation is consistent with one report that has found no significant difference in the surgical efficacy between transposition, interposition, and vein amputation [20]. However, the efficacy of the various surgical methods needs to be further investigated in samples of sufficient size to reach conclusions with statistical significance.

TN is thought to be caused by compression of the trigeminal nerve by arteries, veins, brain tumors [1,2], arteriovenous mal-

formation [4], adhesion without vascular compression [5,6,8,23], and deformity of the pyramidal bone [9]. However, details of the pathogenesis are yet to be clarified. TN without vascular compression has been reported [5,6,8,13], with surgical findings showing that adhesions with surrounding tissues lead to deformation of the nerve. These symptoms disappear when the adhesions are removed. Ishikawa., *et al*. reported cases of TN without vascular compression and confirmed adhesions around the nerve in all cases [6]. This suggests that thickening and adhesion to the arachnoid membrane restricted the mobility of the nerve root, leading to sliding stress between the trigeminal nerve and the pulsating brainstem and excessive tension at the nerve root [6]. In a previous study, Matsushima et al. found adhesion in 2 of 7 cases (29%) of TN caused by vein alone and confirmed that dissection of the entire trigeminal nerve and cutting of the responsible vein resulted in good outcomes [13]. In the present study, 6 of 7 cases (86%) featured nerve adhesions to the surrounding tissue, which inhibited nerve pulsation and mobility. The adhesions were located along the entire segment of the nerve that spans between the region of Meckel cavity and the REZ of the nerve, and, in all cases, the adhesions were within the cerebellar tentorial side. It was suggested that not only venous compression but also nerve deformity caused by adhesions might be involved in the onset of symptoms [13,17]. In other words, the veins could be caught in the adhesions, leading to nerve compression. In fact, after dissection of the adhesions, transposition was added in two cases and interposition in one case, but it is unclear whether these additional manipulations were necessary for improving symptoms. Further accumulation of cases and detailed studies are needed to clarify the pathogenesis of TN caused by veins.

The 3D MRC/MRA fusion images used to aid preoperative diagnosis made it possible to study the anatomical structure of the trigeminal nerve and its surrounding in detail, demonstrating the usefulness of this technique [15,16]. The method is easy to implement, requires minimal logistic investment (an image processing workstation) and incurs trivial costs (15 minutes of image processing per fusion image), and is highly recommended for routine employment for suspected TN cases. It was thought that veins, because of their slow blood flow, were difficult to capture by conventional MRA without contrast enhancement [8]. Taking advantage of the high resolution of the MRI, we examined 2D-FIESTA in detail to confirm the blood flow in veins as well as arteries and created 3D MRA for fusion images. However, this manual process is far from optimal, leaving room for improvements in the future. Inoue used contrast-enhanced T1 spoiled gradient recalled (SPGR) imaging and reported that the method allowed them to detect the vein responsible for TN [12]. Their results clearly demonstrated that the contrast-enhanced image provided more accurate information about the vein than the unenhanced image from the steady-state acquisition. While we were able to evaluate the vein responsible for TN using MRA without contrast-enhancement, contrast-enhanced MRA should be considered as an alternative for potential improvements. However, our approach is ultimately limited by the spatial resolution and the sensitivity to flow velocity of this MRI technique. In the case of small peripheral veins, it may be difficult to depict the vascular structure due to the reduced velocity of intraluminal blood flow, and the accuracy of volume data obtained in the original image is limited by the spatial resolution of MRI. Thus, to accurately capture and evaluate blood vessels of even smaller and slower blood flows, further investigations of alternative or novel imaging approaches will be necessary.

Conclusions

Using 3D-MRC/MRA fusion images, we were able to clearly depict not only the arteries but also veins that pressed on the trigeminal nerve. The method is easy to implement, incurs minimal costs, and is highly recommended for routine employment for suspected TN cases. By visualizing adhesions around the nerve, this technique was especially useful for the diagnosis of TN in which the vein was the sole vessel responsible for the compression of the nerve. Experience gained in our surgical practice suggests that efforts should be made to dissect the adhesions and improve the mobility and deformity of the nerve. The vein should be preserved as much as possible to prevent adverse consequences of altered venous return. Albeit on a small sample, our MRI-aided surgical intervention was successful in almost completely eliminating TN pain in all patients. These results suggest the technique is highly efficient to diagnose and treat vein-caused TN and greatly improve the quality of life for those patients. We expect further improvement of the technique from future application of contrast enhanced MRA as well as alternative advance imaging methods with higher spatial resolution and improved sensitivity to low blood flow.

Conflict of Interest

There is no Conflict of Interest.

Financial Support and Sponsorship

None.

Compliance with Ethical Standards

All procedures used in this research were approved by the Ethical Committee of International University of Health and Welfare (protocol #; approved on) and conducted in accordance with the ethical standards laid down in the 1964 Declaration of Helsinki and its later amendments. Informed consent was obtained from all patients in writing prior to their participating in the procedures.

Submission Statements

This manuscript is original and has not been submitted elsewhere in part or in whole.

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