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Case Report

Hemimasticatory spasm caused by single venous compression of the root of the trigeminal nerve: An MRI study for a case report and review of literature ☆,☆☆

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ABSTRACT

Hemimasticatory spasm is a very rare disorder of the trigeminal nerve characterized by paroxysmal involuntary contraction of the jaw-closing muscles. Although its cause is not fully known, vascular compression of the trigeminal nerve is thought to be involved. Magnetic resonance imaging (MRI) can indicate continuing vascular compression for hemimasticatory spasm. Here, we report a case of hemimasticatory spasm that was caused by single venous compression of the trigeminal nerve root on MRI and was confirmed by microvascular decompression surgery.

Introduction

Hemimasticatory spasm (HMS) is a very rare disorder of the trigeminal nerve characterized by paroxysmal involuntary contraction of the jaw-closing muscles [1]. Although the cause of HMS is not fully known, vascular compression of the trigeminal nerve is thought to be involved, and HMS falls under the broad umbrella of trigeminal neuralgia [1–6]. Magnetic resonance imaging (MRI), particularly high-resolution MRI, such as 3-dimensional fast imaging employing steady-state acquisition (3D FIESTA) imaging and high-resolution magnetic resonance tomographic angiography (MRTA), can identify the neurovascular relationships at the root entry or exit zone (REZ)

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and other parts of the cranial nerves [4,5], which could lead to the prediction of surgical findings and could aid in the development of a safe protocol for treatment safe. However, all of these cases of HMS in published reports were caused by either arterial or both arterial and venous compression of trigeminal nerve [1–3,7]. To our knowledge, there have been no reports of HMS caused by single venous compression of the trigeminal nerve. Here, we report a case of HMS that was caused by single venous compression of the root of the trigeminal nerve on MRI and was confirmed by microvascular decompression surgery.

Case report

A 40-year-old woman of Yi nationality was admitted to our hospital in October of 2016 due to paroxysmal muscular painful spasms in the left temporal and facial regions, lasting from 10 to 15 seconds, for 2 years. These episodes occurred more than ten times a day. The symptoms were triggered by masticatory movements, and the patient suffered greatly from this condition. No scleroderma or atrophy of the left masseter and temporalis muscles was present. The patient underwent conservative therapy, such as oral carbamazepine and other painkillers; however, the symptoms were not alleviated. Neurological examination was normal, but contractions of the masseter and temporalis muscles were very intense on the left side in episodes of spasm. Needle electromyography(EMG) of the left temporal and masseter muscles showed normal signals during the resting state (Fig. 1A). However, during periods of induced spasms by masticatory movements, the left temporalis and masseter muscles showed sudden bursts of motor potential units (Fig. 1B). EMG demonstrated irregular sudden bursts of high-voltage motor unit potentials in the temporalis and masseter muscles, similar to hemifacial spasms.

The patient underwent MR preoperatively using a 3.0-Tesla MR scanner (SignaHDxt; GE Medical Systems, LLC Waukasha, WI) with a circular polarized transmit-receive head coil. To visualize the neurovascular relationship, we used 3D FIESTA and MR tomographic angiography (MRTA) and a standard brain protocol, including axial T1, axial and coronal T2,T2 Flair sequences to exclude other pathologies that could cause HMS. T1 and T2 sequences were not used in the analysis presented in this article; thus, we have not listed the parameters for those sequences.

3D-FIESTA was performed with the following parameters: TR ms/TE ms = 5.15/2.0, flip angle 15°, matrix = 512 × 512, FOV = 20 cm, section thickness =1 mm, number of signals acquired (NSA) = 1, sampling bandwidth = 20.83 kHz. MRTA was performed with the following parameters: TR ms/TE ms = 19/2.7, flip angle 60°, matrix = 512 × 512, FOV = 16 cm, section thickness = 1mm, number of signals acquired (NSA) = 2, sampling bandwidth = 41.67 kHz.

The original MRI data were loaded onto a workstation (GE, AW 4.6, Sun Microsystems, Palo Alto, CA) to reconstruct the trigeminal nerve and vessels for review. Two experienced neuroradiologists reviewed the MR images. The vessel in contact with the nerve was identified by tracing it back to its parent vessel [8]. 3D FIESTA and MRTA showed a small venous compression of the trigeminal motor nerve (Fig. 2A-C), and this venous enter into the superior petrosal sinus.

During the microvascular decompression(MVD) surgery, the trigeminal motor rootlet was completely elevated and compressed by an underlying small branch of petrosal veins with high tension (Fig. 3); no other offending arteries were found. After microvascular decompression surgery, the spasms of the temporalis and masseter muscles immediately stopped, and 3 months postoperatively, the patient's symptoms had not recurred.

Discussion

HMS is a rare disease characterized by unilateral involuntary paroxysmal brief twitches or longer-lasting spasms of the masseter and temporalis muscles of the affected side, occasionally also involving the medial pterygoid muscle [1,3]. HMS may have no apparent triggers; voluntary movements, such as laughing, chewing, or talking, are the most common triggers of hemimasticatory spasm [8]. The condition can be diagnosed based on clinical manifestations, imaging studies such as MRI of the trigeminal nerve, and EMG [3]. The characteristic EMG findings of HMS include irregular bursts of motor unit potentials (MUPs) that correlate with the involuntary masseter spasms [9]. Our patient showed typical clinical and electrophysiological findings previously attributed to HMS. HMS is predominant in women (female-male ratio, 2:1), with a mean age of 31 years (age range, 15-57 years) [8,9], as observed in our patient.

The cause of HMS is not well known. Some scholar believe [1–4] that HMS may occur as a result of vascular compression of the motor branch of the trigeminal nerve in the infratemporal fossa, which triggers involuntary contractions of the masseter muscles and closure of the lower jaw joints [3,8]. These scholars have also suggested that HMS is considered to be a peripheral entrapment neuropathy. However, compression of other nerves or tumor tissues also remains a possibility, resulting in demyelinating changes of the trigeminal nerve [8,10]. Scleroderma and facial hemiatrophy could result in HMS [8]. Pontine infarction [11] is also able to cause HMS. The condition might also be secondary to biopercular syndrome [12]. All of these theories are possible fits for HMS.

3D FIESTA produces very high-resolution T2-weighted images with excellent contrast between structures, including the cerebrospinal fluid (CSF), the trigeminal nerve and adjacent blood vessels; however, it is difficult to distinguish between arteries and veins [13]. MRTA selectively demonstrates fast-flowing blood and provides visualization of arteries and nerves, but the inherent defect of this technique is that veins are insufficiently visualized [14]. Therefore, a combination of these 2 MRI techniques can be used to accurately visualize the fine anatomical structures at the REZ of the trigeminal nerve and to differentiate vein from artery. Furthermore, it is very important to observe the original thin-slice images continuously. Many scholars [5,6,13,14] have used the 2 sequences to study trigeminal neuralgia. In the reported literature discussing HMS [1-3,7], only Chon et al. [1], combined these 2 MRI techniques. However, that case was caused by arterial com-

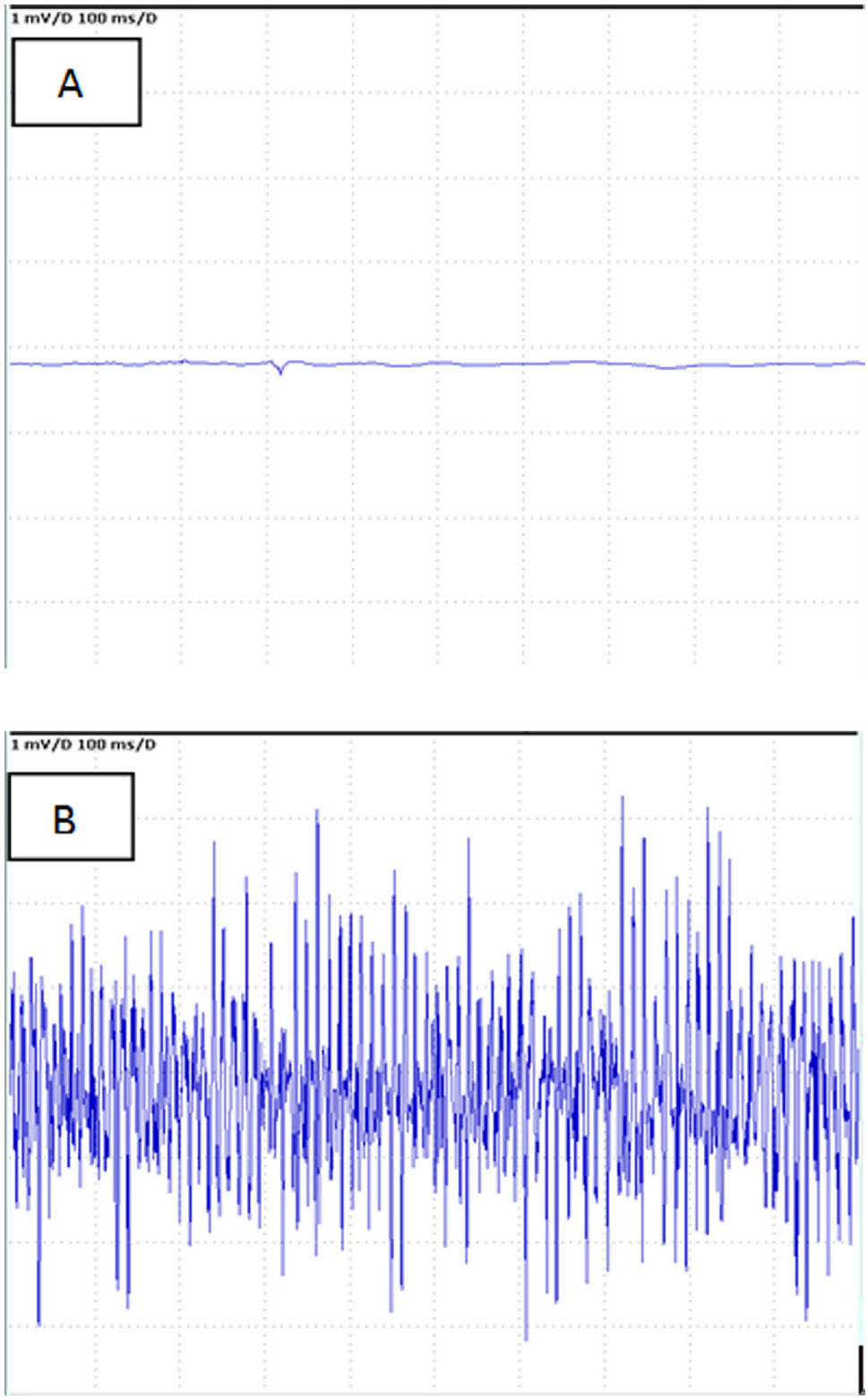

Fig. 1 – Needle electromyography is normal in the left temporalis muscle during the resting state (A); Needle electromyography shows bursts of motor potential units in the left temporalis muscle during periods of involuntary spasm (B).

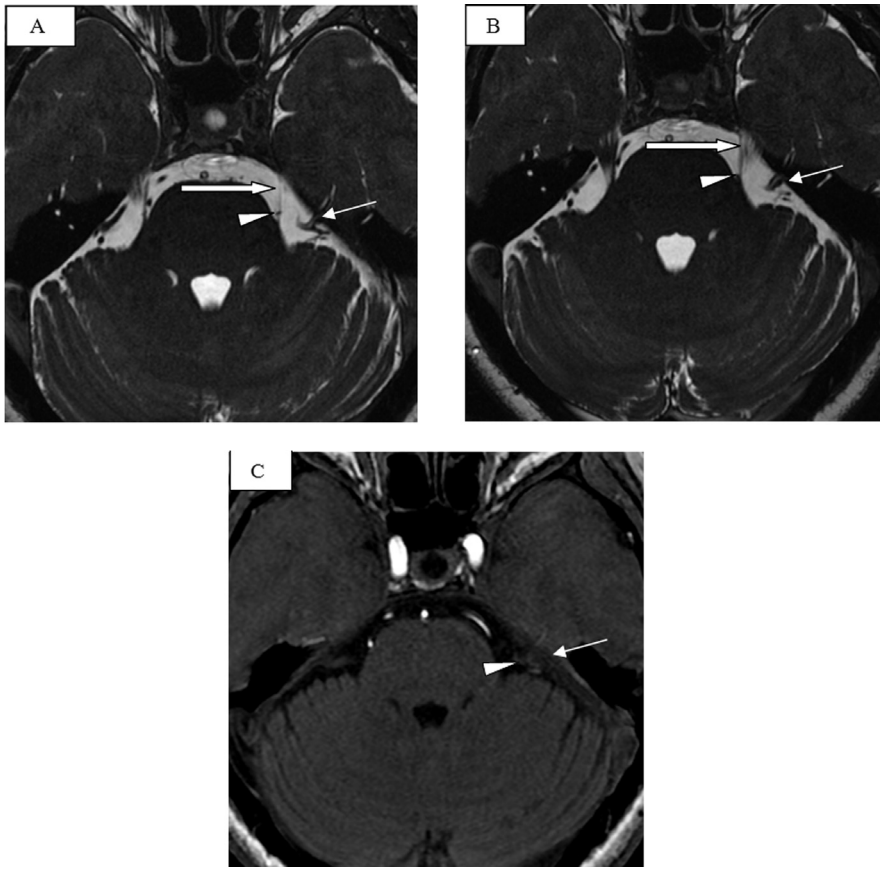

Fig. 2 – Three-dimensional fast imaging employing steady-state acquisition (3D FIESTA) (A), (B) and a 3-dimensional time of flight spoiled gradient (C) show that the motor rootlet (thick arrow) of the trigeminal nerve was compressed by a tortuous vein(triangle), which entered into the superior petrosal sinus(thin arrow). Figure 2A and C are at the same slice.

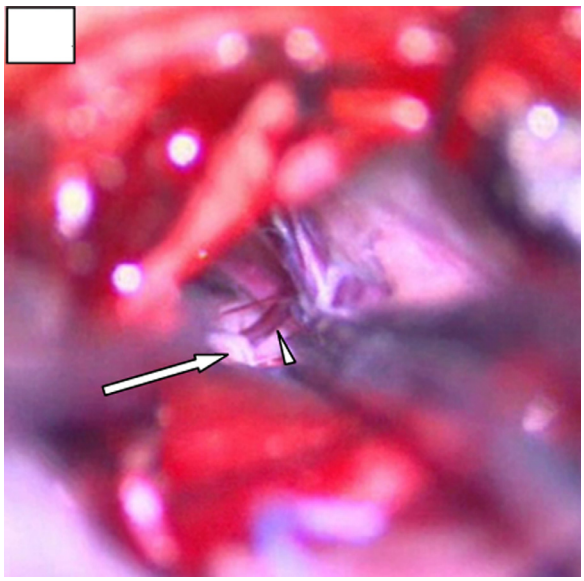

Fig. 3 – During the surgery (3), the cerebellum was gently retracted to expose the trigeminal motor rootlet (thick arrow), which was elevated and compressed by an underlying small branch of the petrosal vein (triangle).

pression, whereas our case was caused by single venous compression.

In this study, axial T1, axial and coronal T2, and T2Flair sequences were also used to exclude other pathologies that could cause HMS, such as brain stem stroke/ischemia and brain stem tumors [3].

Based on the heterogeneous etiologies of HMS, there are also multiple possible treatments according to different etiologies. The treatment options for HMS include the use of oral drugs, injection of botulinum toxin, and surgical treatment. Of the oral drugs, carbamazepine and phenytoin may improve clinical symptoms. However, in this case, the patient's symptoms were not alleviated following oral drug use. Local (temporalis or masseter) injection of botulinum toxin A has been considered the preferred symptomatic treatment [9]. However, recurrences and complications are not uncommon, such as immense costs, adverse effects, and nonresponding patients forming antibodies directed against the toxin [15]. The reported cases have given an impression of the overall effectiveness of vascular decompression for HMS, as was observed in our patient. Therefore, we believe that microvascular decompression is useful for treating HMS. In the future, a larger patient population and a case-control study will be required to validate the effectiveness of this surgical approach.

Conclusions

We report a 40-year-old woman with HMS that was caused by single venous compression of the root of the trigeminal nerve on MRI and was confirmed during microvascular decompression surgery, Following the microvascular decompression surgery, the patient's symptoms never recurred. This report is the first HMS study describing a case with venous compression.

Patient consent

This study was approved by the Ethics Committee of our hospital. Written informed consent for the publication of this case report was obtained from the patient.

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