CASE REPORT



Extracranial Radiofrequency Treatment for Painful Tic Convulsif Syndrome Under Local Anesthesia

Shuangshuang Xu \cdot Xiaolan Liu \cdot Wei Zhao \cdot Ying Ma \cdot Xindan Du \cdot Bing Huang

Received: January 10, 2022 / Accepted: March 14, 2022 / Published online: April 16, 2022 \circledcirc The Author(s) 2022

ABSTRACT

Introduction: Painful tic convulsif syndrome is ipsilateral facial trigeminal neuralgia combined with hemifacial spasm, which is relatively rare in the clinic. Microvascular decompression is currently considered to be an effective treatment. We report extracranial radiofrequency treatment of painful tic convulsif syndrome under local anesthesia, a technique which provides a safer and more economical treatment for this kind of patient.

Case Presentation: We report a case of painful tic convulsif syndrome which was treated with extracranial radiofrequency therapy of the trigeminal nerve and facial nerve. After operations, the symptoms of pain and spasm were relieved immediately, but the right facial

Shuangshuang Xu and Xiaolan Liu contributed equally to this work.

numbness and facial paralysis (House–Brackmann grade III) were left. The facial paralysis was completely relieved after 3 months of follow-up, and there was no recurrence of trigeminal neuralgia or hemifacial spasm after 35 months.

Discussion: Painful tic convulsif syndrome is a combination of ipsilateral facial trigeminal neuralgia and hemifacial spasm, which is relatively rare in the clinic. So far, only one treatment method of microvascular decompression has been reported for the disease. We report the first case of CT-guided extracranial radiofrequency therapy for painful tic convulsif syndrome.

Conclusion: Extracranial radiofrequency therapy can provide safe and economical treatment for patients with painful tic convulsif syndrome.

Keywords: Painful tic convulsif syndrome; Trigeminal neuralgia; Hemifacial spasm; Radiofrequency thermocoagulation; Computed tomography

S. Xu · X. Liu · W. Zhao · Y. Ma · B. Huang (🖂) The Affiliated Hospital of Jiaxing University/The First Hospital of Jiaxing, No. 1882 South Zhonghuan Road, Nanhu District, Jiaxing City, Zhejiang, China e-mail: jxhb999@sina.com

X. Du Hangzhou Red Cross Hospital, Hangzhou, Zhejiang, China

Key Summary Points

A patient with painful tic convulsif syndrome is relatively rare in the clinic.

At present, only one method of microvascular decompression (MVD) has been reported for the treatment of this disease.

We report a case of painful tic convulsif syndrome which was treated with extracranial radiofrequency therapy of the trigeminal nerve and facial nerve. The patient's symptoms were relieved immediately after the surgery.

This technique can provide safe and economical treatment for patients with painful tic convulsif syndrome.

INTRODUCTION

Trigeminal neuralgia (TN) refers to paroxysmal pain in the trigeminal nerve control area [1]. Hemifacial spasm (HFS) is a motor disorder of facial muscles controlled by the facial nerve, which is characterized by irregular and involuntary painless clonus of unilateral facial muscles [2]. When both occur on the ipsilateral side, it is called painful tic convulsif syndrome [3]. Although the clinical manifestations of TN and HFS are different, the cause of both is considered to be the concomitant vascular compression of the nerve root in the prepontine cistern [4]. Microvascular decompression (MVD) is an effective treatment [5], but it requires intracranial operation, and many serious complications may occur, e.g., facial paralysis, hearing loss, cerebrospinal fluid leakage, intracranial infection, and intracranial hemorrhage [6]. We report extracranial radiofrequency therapy for painful tic convulsif syndrome, a technique which provides a safer and more economical treatment for these patients.

Consent for Publication and Compliance with Ethics Guidelines

Trigeminal and facial nerve extracranial radiofrequency therapy techniques have been reviewed and approved by the ethics committee (Medical Ethics Committee of The First Hospital of Jiaxing: 2015-081; LS2019-013). Written informed consent was obtained from the patient for publication of this case report and any accompanying images. Institutional review board approval is not required for this case report.

CASE PRESENTATION

Medical History

A patient in their 70s developed paroxysmal pain in the right mandibular region and lower gum 7 years ago, which could be induced by brushing and eating. Each attack lasted for several minutes, and there was not pain at all during the intermission. The visual analog score (VAS) of pain degree was 7–9. The pain could be relieved by carbamazepine, while relapsed after withdrawal. She came to our hospital and was diagnosed with "primary TN (mandibular branch)". She was treated with "extracranial radiofrequency therapy through the foramen ovale" in 2012 and the pain disappeared after the operation. In 2016, the pain relapsed and gradually worsened. The painful area spread upward from the mandibular region to the innervation area of the right maxillary branch. Carbamazepine could effectively relieve the pain. Since 2018, in addition to the pain on the right side of the face, there had been an involuntary twitch of right facial muscles, which was paroxysmal and first occurred on the right eyelid, then spread to the right cheek. It was difficult for her to open her eyes and the corner of her mouth was tilted to the right, especially when she was nervous. In October 2018, the patient's pain was unbearable and could not be relieved effectively with oral drugs, so she came to our hospital again.

We examined the patient and found that the pain was located in the area dominated by the second and third branches of the right trigeminal nerve, and lips were the "trigger points" of pain. During an attack, involuntary convulsions appeared in her right facial muscles, the right eye was difficult to open, and the mouth was skewed to the right. During the interval, the bilateral forehead wrinkles were symmetrical, eyes closed symmetrically and forcefully, the nasolabial sulcus did not become shallow. When bulging cheeks, there was no air leakage; when showing teeth, the mouth was not crooked, the tongue was in the middle, and the taste and hearing were normal. She suffered from hypertension for over 20 years and took amlodipine orally to control her blood pressure at a normal level.

The trigeminal nerve MRI showed that the bilateral superior cerebellar artery interacted with the trigeminal nerve root, and the right facial nerve root interacted with the anterior inferior cerebellar artery (Fig. 1). CT of the head showed subcortical arteriosclerotic encephalopathy. No obvious abnormality was found in other examinations. She was then diagnosed with primary right TN with HFS (right painful tic convulsif syndrome).

Treatment

The patient was given symptomatic treatments, like alleviation of pain, nerve nutrition, etc. After being informed about the treatments of TN and HFS, such as balloon compression, botulinum toxin injections, MVD [5], radiofrequency technology [7–9], and neurostimulation [10], the patient chose extracranial radiofrequency therapy and signed the informed consent form. After exclusion of radiofrequency contraindications, radiofrequency treatment of the foramen rotundum, foramen ovale, and stylomastoid foramen was performed on her respectively (Figs. 2, 3).

Extracranial Radiofrequency Treatment of Trigeminal Nerve [7–9]

After entering the room, the patient's vital signs were monitored. She was given an intravenous infusion and inhaled oxygen through a nasal catheter. Then she lay on her back on the CT stage with a thin pillow under the shoulder, a positioning grid on the right side of the face,

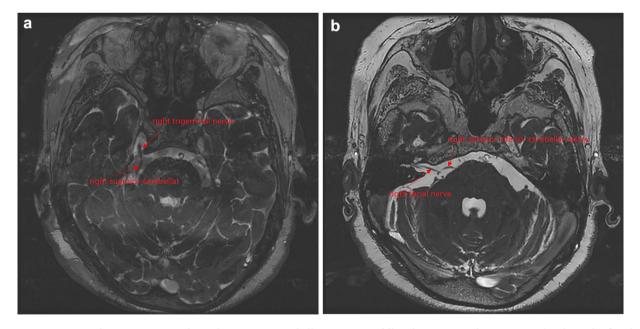


Fig. 1 Trigeminal nerve MRI. a The right superior cerebellar artery straddles the trigeminal nerve root; b The right facial nerve root interacted with the anterior inferior cerebellar artery

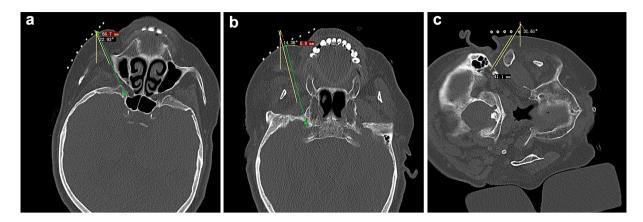


Fig. 2 a Puncture path design for foramen rotundum; b Puncture path design for foramen ovale; c Puncture path design for stylomastoid foramen



Fig. 3 The needle position. a Puncture needle in foramen rotundum; b Puncture needle in foramen ovale; c Puncture needle in stylomastoid foramen

and a wide tape to secure the head. A CT scan of the skull was performed in a semi-coronal position (the scanning baseline was parallel to the line of porus acusticus externus to the middle point of the mental protuberance to the central incisor). We scanned with a thickness of 2 mm, then selected the layers including the lower edge of the zygomatic arch and foramen ovale, and the coronal process of the mandibular and foramen ovale as the puncture layer and designed the puncture paths (Fig. 2a, b). After measuring the angle and depth of the needle with the tool ruler attached to the CT system, we administered local anesthesia with 2% lidocaine at the puncture site. The radiofrequency needle was punctured to the

foramen rotundum and foramen ovale respectively (Fig. 3a, b) and we then performed the electrophysiological tests: high frequency (50 Hz) and low frequency (2 Hz) could induce the abnormal sensation of the skin of the right lips and the rhythmic jitter of the mandible at 0.5 mA or 0.3 V; 0.1 mg of fentanyl citrate Injection was given intravenously and then radiofrequency thermocoagulation was performed at 90 °C for 120 s.

Extracranial Radiofrequency Treatment of Facial Nerve [11]

The patient slept on the CT table with the right side of her face upwards, and a CT positioning

grid was placed in front of the earlobe. The paranasal sinus pattern was selected to take the positioning image of the skull, and the mastoid process was scanned on the axial plane of 3 mm. The CT section with stylomastoid foramen and without temporal bone blocked was then selected as the puncture layer, on which the puncture path was designed: we selected the right stylomastoid foramen as the puncture target, drew a straight line forward from the right stylomastoid foramen with the CT's measurement tool, and the intersection point between the line and the skin was the puncture point. The puncture depth (the distance from the puncture point to the target) and the puncture angle (the angle between the puncture route and the sagittal plane) were measured (Fig. 2c). Local anesthesia was performed with 2% lidocaine at the puncture site, and the No. 7 radiofrequency needle with length of 10 cm and exposed end of 5 mm was punctured to the target under CT guidance (Fig. 3c). The nerve was stimulated with low frequency (2 Hz) current, and 0.4 mA current caused spasmodic twitch of the right facial muscle with the same frequency as that of the electrical stimulation. The patient was given continuous radiofrequency at 65 °C and was asked to puff up her cheeks and close her eyes at the same time, then the radiofrequency was stopped immediately when air leakage occurred and the right eye could not be closed tightly at the 36th second of the treatment.

Result and Follow-up

The TN and HFS disappeared immediately after radiofrequency treatment, but the right facial numbness and House–Brackmann grade III facial paralysis were left. The facial paralysis disappeared completely after 3 months of follow-up, and there was no recurrence of TN or HFS after 35 months.

DISCUSSION

Painful tic convulsif syndrome is a combination of ipsilateral facial TN and HFS, which is relatively rare in the clinic. Owing to the unique clinical manifestations of the disease, the diagnosis is not difficult. Most of the existing case reports were caused by space-occupying lesions in the posterior cranial fossa or lengthy vertebrobasilar artery, which needed craniotomy treatment [3, 5].

Our patient suffered from TN with HFS. CT and MRI examination of the head did not show occupation of the posterior cranial fossa or lengthy vertebrobasilar artery, but the trigeminal nerve root and facial nerve root located in the prepontine cistern were oppressed by the superior cerebellar artery and the anterior inferior cerebellar artery, respectively. Reports of this situation are extremely rare [3–5]. At present, only one method of MVD [5] has been reported for the treatment of this disease.

We developed "extracranial radiofrequency treatment of HFS" [11] and "extracranial non-Gasserian ganglion radiofrequency treatment of TN'' [7–9]: the former only needs one to put a thin radiofrequency needle into the stylomastoid foramen under CT guidance and partially destroy the extracranial facial nerve to eliminate HFS, which can be effective for several years, safe and economical, and solves a difficult problem in the treatment of HFS; the latter needs one to put the radiofrequency needle into the supraorbital foramen, foramen rotundum, or foramen ovale respectively under CT guidance. Radiofrequency is used to treat TN without entering the brain, so complications like intracranial hemorrhage and infection can be avoided. It has also been widely reported that radiofrequency ablation is an excellent treatment for cranial nerve diseases, which has the same efficacy as MVD, but without the risk of invasion and significant complications [12–14].

For this patient, considering her advanced age, we informed her of various alternative treatments like radiofrequency, balloon compression, botulinum toxin injections, MVD, etc., and finally she chose the least risky but effective extracranial radiofrequency technique. After treatment, the patient's syndromes disappeared completely. After 35 months of followup, there was no recurrence of TN or HFS.

CONCLUSION

It is suggested that extracranial radiofrequency therapy can provide safer and more economical treatment than MVD for patients with painful tic convulsif syndrome.

ACKNOWLEDGEMENTS

Funding. Found Program: Zhejiang Social Public Welfare Research and Development Project (LGF20H090021); General Project of Hangzhou Agriculture and Social Development in 2020 (20201203B173); Key Disciplines Jointly Established by Provinces and Cities in Zhejiang Province-Pain Medicine (2019-ss-ttyx); Zhejiang Health Science and technology plan project (2022ZH012). The journal's Rapid Service Fee was funded by the First Hospital of Jiaxing.

Authorship. All named authors meet the International Committee of Medical Journal Editors (ICMJE) criteria for authorship for this article, take responsibility for the integrity of the work as a whole, and have given their approval for this version to be published.

Author Contributions. Shuangshuang Xu, the co-first author, the study concept and design, drafting the manuscript. Xiaolan Liu, the co-first author, drafting and revising the manuscript. Wei Zhao, the second author, acquisition of data, collection patient information. Ying Ma, the third author, acquisition of data, postoperative follow-up. Xindan Du, the fourth author, analysis and interpretation of data. Bing Huang, the corresponding author, project leader, technical support, approval of the final version to be published.

Disclosures. There are no grants, sponsors, funding sources, or direct financial support to disclose. Shuangshuang Xu, Xiaolan Liu, Wei Zhao, Ying Ma, Xindan Du, Bing Huang have nothing to disclose.

Compliance with Ethics Guidelines. Trigeminal and facial nerve extracranial radiofrequency therapy techniques have been reviewed and approved by the ethics committee (Medical Ethics Committee of The First Hospital of Jiaxing: 2015-081; LS2019-013). Written informed consent was obtained from the patient for publication of this case report and any accompanying images. Institutional review board approval is not required for this case report. This study was performed in accordance with the Helsinki Declaration of 1964 and its later amendments.

Data Availability. Data sharing is not applicable to this article as no datasets were generated or analyzed during the current study.

Open Access. This article is licensed under a Creative Commons Attribution-NonCommercial 4.0 International License, which permits any non-commercial use, sharing, adaptation, distribution and reproduction in any medium or format, as long as you give appropriate credit to the original author(s) and the source, provide a link to the Creative Commons licence, and indicate if changes were made. The images or other third party material in this article are included in the article's Creative Commons licence, unless indicated otherwise in a credit line to the material. If material is not included in the article's Creative Commons licence and your intended use is not permitted by statutory regulation or exceeds the permitted use, you will need to obtain permission directly from the copyright holder. To view a copy of this licence, visit http://creativecommons.org/licenses/bync/4.0/.

REFERENCES

- 1. Bendtsen L, Zakrzewska JM, Abbott J, et al. European Academy of Neurology guideline on trigeminal neuralgia. Eur J Neurol. 2019;26(6):831–49.
- 2. Chaudhry N, Srivastava A, Joshi L. Hemifacial spasm: the past, present and future. J Neurol Sci. 2015;356(1–2):27–31.
- 3. Sindou M, Mercier P. Hemifacial spasm associated with other cranial nerve syndromes: literature review. Neurochirurgie. 2018;64(2):101–5.

- 4. Gardner WJ. Concerning the mechanism of trigeminal neuralgia and hemifacial spasm. J Neurosurg. 1962;19(11):947–58.
- Crevier-Sorbo BG, Brock A, Rolston JD. Trigeminal neuralgia plus hemifacial spasm caused by a dilated artery: a case of painful tic convulsif syndrome. Lancet. 2019;394(10211):e36.
- 6. Yu YB. Striving to improve the therapeutic level of microvascular decompression. Chin J Neurosurg. 2016;32(4):325–8.
- Lin H, Cao G, Jin G, et al. Extracranial non-Gasserian ganglion application of radiofrequency thermocoagulation on the mandibular branch of the trigeminal through the foramen ovale for trigeminal neuralgia. Pain Physician. 2021;24(4):e425–32.
- 8. Huang B, Yao M, Feng Z, et al. CT-guided percutaneous infrazygomatic radiofrequency neurolysis through foramen rotundum to treat V2 trigeminal neuralgia. Pain Med. 2014;15(8):1418–28.
- 9. Huang B, Yao M, Liu Q, et al. Personalized needle modification for CT-guided percutaneous infrazy-gomatic radiofrequency ablation of the maxillary

nerve through the foramen rotundum in order to treat V2 trigeminal neuralgia. J Pain Res. 2019;12(9):2321–9.

- 10. Abd-Elsayed A, Grandhi R, Sachdeva H. Effective management of trigeminal neuralgia by neurostimulation. Ochsner J. 2015;15(2):193–5.
- Huang B, Yao M, Chen Q, et al. Awake CT-guided percutaneous stylomastoid foramen puncture and radiofrequency ablation of facial nerve for treatment of hemifacial spasm. J Neurosurg. 2021;32(9): 1–7.
- 12. Abd-Elsayed A, Kreuger L, Wheeler S, et al. Radiofrequency ablation of pericranial nerves for treating headache conditions: a promising option for patients. Ochsner J. 2018;18(1):59–62.
- 13. Abd-Elsayed A, Nguyen S, Fiala K. Radiofrequency ablation for treating headache. Curr Pain Headache Rep. 2019;23(3):18.
- 14. Abd-Elsayed A. The ALblation technique for treating migraine headache. Curr Pain Headache Rep. 2020;24(6):29.