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CASE REPORT

CT-Guided Thermocoagulation of the Pterygopalatine Ganglion for Refractory Trigeminal Autonomic Cephalalgia

Ying Ma · Shuangshuang Xu · Xiaolan Liu ·
Zhangtian Xia · Wei Zhao · Bing Huang

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ABSTRACT

Introduction: Trigeminal autonomic cephalalgia (TAC) is a type of one-sided cerebral painful headache, with attacks regularly accompanied by autonomic responses, such as tearing, runny nose, panic, nausea and vomiting on the affected side. Currently, the most common treatment strategies are drugs, nerve grafts and surgery. Clinical understanding of TACs is limited. Here, we report the case of thermocoagulation treatment of the pterygopalatine ganglion in an uncommon TAC under local anesthesia.
Case Presentation: A rare case of TAC was treated with computed tomography (CT)-guided thermocoagulation within the pterygopalatine ganglion. Pain and autonomic signs were relieved immediately after surgery, with the patent retaining only slight numbness on the left side of the face. This numbness completely resolved at 6 months of follow-up and there was no recurrence.
Discussion: Trigeminal autonomic cephalalgia seriously affects the patient's quality of life, but clinical understanding is limited. In the case reported here, we performed CT-guided thermocoagulation of the pterygopalatine ganglion at 90 °C for 180 s for treatment of a trigeminal autonomic headache. To our knowledge, this is the first report of using thermocoagulation at 90 °C to treat the pterygopalatine ganglion. We found that this strategy results in fewer side effects and is a more cost-effective treatment for such patients than other options.
Conclusion: Computed tomography-guided thermocoagulation of the pterygopalatine ganglion at 90 °C for 180 s for treatment of trigeminal autonomic headache is a safe and economical treatment option.

Keywords: Trigeminal autonomic cephalalgia; Pterygopalatine fossa; Pterygopalatine ganglion; Thermocoagulation

Y. Ma · S. Xu · X. Liu ·
Z. Xia · W. Zhao · B. Huang (✉)
Department of Anesthesiology and Pain Medicine, The Affiliated Hospital of Jiaxing University, No. 1882 Zhong Huan South Road, Jiaxing 314000, Zhejiang, China
e-mail: 00181727@zjxu.edu.cn

Key Summary Points

Trigeminal autonomic cephalalgia (TAC) is a rare type of primary unilateral headache.

The pathophysiological mechanisms of TAC are still unclear, and drugs are the preferred first-line prophylactic treatment, but these have significant side effects; clinical understanding and treatment options are currently relatively limited.

We report an exceptional case of TAC in which pharmacological treatment and nerve blocks were ineffective and finally radiofrequency treatment of the pterygopalatine ganglion was used.

Following radiofrequency treatment, the patient's pain score rapidly decreased to zero, and there had been no recurrence at 6 months of follow-up, as well as no significant complications or adverse effects.

This technique can provide safe and economical treatment for patients with painful TAC.

INTRODUCTION

Trigeminal autonomic cephalalgias (TACs) are a group of primary headaches characterized by paroxysmal, fluctuating unilateral headaches distributed along the trigeminal nerve, with pain often located in the orbital, forehead and temporal regions, accompanied by clinical manifestations, such as ipsilateral autonomic dysfunction including, for example, lacrimation and bulbar conjunctival congestion. TACS are grouped into four distinct primary headache types: cluster headache, paroxysmal migraine, persistent migraine and unilateral transient persistent neuralgia-like headache with conjunctival congestion and tearing [1]. TAC appeared for the first time as an independent headache type in the 2013 International Classification of Headache, version 3β [2]. Currently, the most common treatment strategies are drugs, nerve grafts and surgery [3, 4]. Here, we report a case of standard radiofrequency treatment of the pterygopalatine ganglion (SPG) in a uncommon TAC performed under local anesthesia, a strategy that has fewer side effects and is more cost-effective than other treatments for such patients.

Computed tomography (CT)-guided radiofrequency treatment of the pterygopalatine ganglion have been reviewed and approved the Medical Ethics Committee of The First Hospital of Jiaxing (LS2022-XJS-011-02). Written informed consent was obtained from the patient for publication of this case report and any accompanying images. This study was performed in accordance with the Helsinki Declaration of 1964 and its later amendments.

CASE PRESENTATION

Medical History

The patient, a 44-year-old women, was admitted to the hospital with "recurrent attacks of left-sided cephalofacial pain for more than 29 years, aggravated for 1 month". She said her first headache attack was at age 15 years. The pain was located on the left side of the head and face, mainly in the periorbital and temporal areas, with severe pain accompanied by lacrimation and runny nose on the affected side, and profuse sweating. There had been an onset of pain on the left side of the head and face that had worsened 1 month previously, affecting sleep, with attacks ranging in frequency from one to several times a day, with each attack lasting between 3 and 4 h. The pain could be relieved by forceful defecation and vomiting. During the attack, oral antipyretic and analgesic drugs were ineffective, while the symptoms were slightly relieved by intravenous drip of mannitol and dexamethasone in the local hospital. In December 2021, she was in unbearable pain, and as the local hospital could provide no effective pain relief, she came to our hospital for treatment. On admission, the pain was located in the periorbital and temporal regions, with no touch-evoked pain and a numerical rating scale (NRS) score of 6 at rest and 9–10 at outbreak.

The erythrocyte sedimentation rate was 32 mm/h. Chest CT showed a ground glass nodule in the upper lobe of the right lung; paranasal sinus CT showed left maxillary sinusitis; ultrasound showed no abnormality in the liver, gallbladder, pancreas, spleen and kidney; trigeminal nerve MRI showed that the left superior cerebellar artery interacted with the trigeminal nerve. Based on these imaging findings she was then diagnosed with trigeminal autonomic headache, pulmonary nodule (right side) and maxillary sinusitis (left side).

Treatment

Upon admission, she was given intermittent oxygen. A cardiothoracic surgeon was consulted who, based on the patient's symptoms and signs, recommended that no specific treatment was needed for the pulmonary nodule and that regular review was sufficient. In contrast, the main symptom of maxillary sinusitis, which was a yellow purulent nasal discharge, especially when the patent was in the supine position, also causing throat discomfort. The patient presented with lacrimation and runny nose only during headache attacks, with immediate relief of post-attack pain and no recurrence. As these symptoms were possibly unrelated to the pulmonary nodule and maxillary sinusitis, she was prescribed oral gabapentin 0.3 g twice daily and a left stellate ganglion block (2% lidocaine + meclizine 1 mg diluted to 10 ml in saline). This treatment provided slight relief of symptoms and a NRS score of 4–5. On the third night, she reported a sudden worsening of pain, with an NRS score of 9–10 for pain and slight relief of symptoms after forceful defecation and vomiting. Consultation with the patient led to a decision to perform thermocoagulation treatment of her pterygopalatine ganglion under CT guidance. The patient agreed with this treatment and signed an informed consent form.

CT-Guided Thermocoagulation of the Pterygopalatine Ganglion

The patient abstained from drinking and eating before surgery. In the operating room, the patient's vital signs were monitored from the beginning. She was given an intravenous infusion and inhaled oxygen through a nasal catheter. She then she lay on her back on the CT stage with a thin pillow under her shoulder, a positioning grid on the right side of her face, with her head tilted backwards at 20°. Positioning grids were placed on the skin of the face below the outer canthus of the affected eye, and lead shields were placed on the eye and neck for protection from radiation. Head localization images were taken in paranasal sinus mode, and then the zygomatic face was scanned in a semi-coronal position with a layer thickness of 2–3 mm. The scan baseline was parallel to the line of the external auditory foramen (midpoint of the line between the chin prominence and the cuspid), and the upper edge of the scan frame reached the upper edge of the zygomatic arch [1]. The level containing the internal orifice of the foramen ovale was selected as the puncture level, and the puncture path was designed at this level: a straight line was drawn from the pterygopalatine fossa immediately above the tip of the coronoid process toward the soft tissues of the face, and the point where

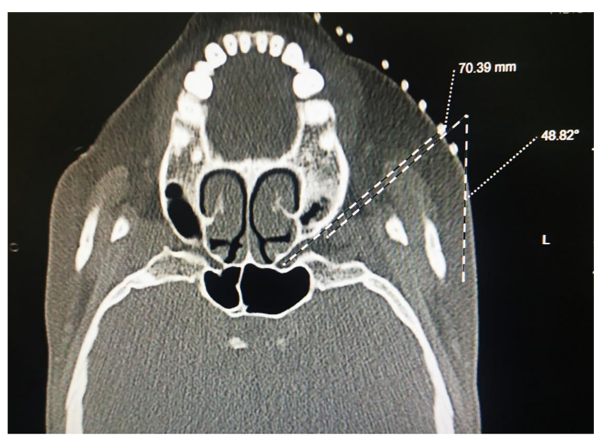

Fig. 1 Left pterygopalatine fossa puncture mid-diameter design: puncture depth was 7.39 cm, and puncture angle (angle between puncture direction and sagittal plane) wss 48.82°

Fig. 2 Computed tomography guided puncture to the pterygopalatine fossa to locate the pterygopalatine ganglion (level of the internal orifice of the foramen ovale)

the line intersected with the skin was chose as the puncture point. The puncture depth (distance from the pterygopalatine fossa to the puncture point) and the puncture angle (angle between the puncture direction and the sagittal plane) were measured with the software of the CT tool (Fig. 1). After applying local anesthesia (2% lidocaine) to the puncture site, a 7-gauge radiofreqency needle (length: 10 cm, exposed end: 10 mm) was used to puncture into the pterygopalatine fossa under CT guidance according to the designed puncture path (Fig. 2). Stimulation with a high-frequency (50 Hz) current (0.5 mA) induced soreness and distension in the nasal cavity, orbit, deep in the external auditory canal and even in the occipital region, while low-frequency (2 Hz) current (1 mA) stimulation failed to induce rhythmic shaking of the upper lip. Stimulation was followed by thermocoagulation at 90 °C for 180 s. At the end of the procedure, a band aid was applied locally to the puncture site after needle extraction, and the patient was returned to the ward.

Result and Follow-Up

On postoperative day 1, she reported significant improvement in the paroxysmal head and facial pain, and there was no swelling of the left side of the face, no nausea and vomiting; the NRS score was 2. After 4 days of postoperative observation, the pain did not recur and there was only slight numbness in the left palate; the Visual Analogue Scale/Score (VAS) was 0. She was followed up at monthly intervals for the first 3 months postoperative at which times she reported no recurrence of pain. At 6 months postoperative, and the pain had not recurred and the numbness of the left palate was completely relieved. The patient and her family were very satisfied with the results of the treatment.

DISCUSSION

Trigeminal autonomic cephalalgias are a group of primary headaches with paroxysmal, fluctuating unilateral headaches distributed along the trigeminal nerve, with pain often located in the orbital, forehead and temporal regions, accompanied by clinical manifestations, such as ipsilateral autonomic dysfunction, including lacrimation and bulbar conjunctival congestion [1]. The most characteristic feature of TACs is that pain attacks are accompanied by symptoms of autonomic dysfunction, and the SPG, as the largest parasympathetic ganglion in the head and neck, is closely associated with the appearance of this symptom. It is considered that the pathogenesis of TACs is the action of the trigeminal autonomic reflex and parasympathetic activation: the trigeminal-autonomic reflex is a reflex pathway consisting of the trigeminal nerve and the facial cranial nerve through the brainstem connection between the suprasalivary nucleus and the parasympathetic outflow of the SPG [5]. Pains due to TACs is characterized by symptoms of autonomic dysfunction and, in addition to unilateral severe or very severe pain, the location of the pain often spreads to the periorbital area, the back of the eye, the temporal area, the nasal cavity, the maxilla and, occasionally, to the forehead, neck, ear, jaw, viscera and other areas. The characteristic feature of the attack is that the pain is accompanied by symptoms of autonomic dysfunction, such as lacrimation, runny nose, frontal and facial sweating and pupil narrowing. The patient is hyperactive and often unable to calm down, and even has palpitations, nausea and vomiting.

The SPG is located in the pterygopalatine fossa in the lateral wall of the nasal cavity at the level of the middle turbinate. The pterygopalatine fossa is a triangular or heart-shaped structure that is 2 cm high and 1 cm wide, located adjacent to the maxillary sinus anteriorly, the pterygoid sinus superiorly, the medial plate of the pterygoid process posteriorly and the vertical plate of the palate medially. The SPG consists of sensory, parasympathetic and sympathetic nerves. Of these, the sensory nerve originates from the pterygopalatine nerve of the maxillary branch of the trigeminal nerve and is located above the SPG; the parasympathetic nerve originates from the Iambda nerve of the facial nerve and is located posterior to the SPG; and the sympathetic nerve originates from the Iambda deep nerve of the superior cervical plexus and is located posteriorly below the pterygopalatine SPG. The sensory nerve innervates the eye, orbit, upper part of the skull, pterygoid sinus, septal sinus and lacrimal gland, respectively; the parasympathetic nerve innervates the mucosa and glands of the nasal cavity, the glands of the hard palate, soft palate, uvula and tonsils; and the sympathetic nerve innervates the glands of the pharynx. In addition, the SPG sends out many tiny nerve branches to interconnect with the small occipital nerve, cervical nerve cutaneous branch, tympanic nerve, ear ganglion and vagus nerve to induce pain or discomfort in the head, neck, ear, jaw and viscera. The associations of these nerve branches also explains the pain attack with nausea and vomiting and strong bowel movement in this patient, and can be used as a basis for electrical stimulation to determine whether the radiofrequency needle is close to the SPG.

Although the pterygopalatine fossa can be clearly visualized by transverse cranial CT, the zygomatic arch at the same level obstructed the needle route, so we borrowed the round hole puncture technique used in the CT-guided percutaneous subzygomatic approach [6] and used a half-coronal scan so that the pterygopalatine fossa and the inferior edge of the zygomatic arch were at the same level. This strategy avoided the obstruction of the zygomatic arch and realized the in-plane puncture technique of the subzygomatic approach to the pterygopalatine fossa, making the whole operation precise and intuitive (Fig. 2).

When performing radiofrequency ablation of the pterygopalatine ganglion in the pterygopalatine fossa, high- and low-frequency electrophysiological tests should be performed first to prevent damage to the V2 branch of the trigeminal nerve in the external orifice of the foramen magnum from causing numbness in the innervation area of this branch. In the present case, the patient was tested by 50-Hz and 2-Hz sensorimotor electrostimulation prior to radiofrequency ablation, which effectively prevented the injury to the maxillary branch of the trigeminal nerve. There was no numbness and hypesthesia in the zygomatic face and upper lip and nasal flank after the operation, but there was obvious hypesthesia in the affected palate, which should be related to the large palatine nerve connected with the SPG.

Most researchers believe that the choice of pulsed radiofrequency mode (working voltage: 45 V; working tip temperature: < 42 °C) only interferes with the conduction function of the nerve but does not destroy the integrity of the nerve structure; as such, it has little effect on the patient and also has limited efficacy [7]. However, information in the literature on the effect of pulsed radiofrequency for the treatment of neuralgia is not very satisfactory, with reports that after treatment each patient requires analgesic drugs, which cannot achieve complete pain relief and the recurrence rate is high [8]. In contrast, the efficacy of using the classical continuous radiofrequency thermocoagulation modality is well established, although it may leave a longer period of hyperalgesia or loss of muscle strength in the area of the original pain area due to the destruction of the neural structure by thermocoagulation. There is no uniform standard for the choice of temperature and duration of continuous radiofrequency thermocoagulation. Some studies have suggested that radiofrequency thermocoagulation at a temperature of > 90 °C has more complications, such as facial numbness [9]. In recent years, our department has directly adopted a thermocoagulation procedure of 90 °C and 120 s continuous radiofrequency. The safety and effectiveness of this approach have been confirmed.

CONCLUSION

There are very few treatment options for TACs. In our case, after giving the patient several stellate nerve blocks with less than satisfactory results, we obtained the patient's consent to go forward with CT-guided radiofrequency treatment of the SPG. Following this procedure, the patient's pain score rapidly decreased to 0, and there was no reccurence at 3 months of follow-up. Also, there were no obvious complications or adverse effects. Taken together, these results indicate that CT-guided SPG standard radiofrequency treatment can be expected to be an effective treatment for TACs.

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Author Contributions. Ying Ma: the study concept and design, drafting the manuscript. Shuangshuang Xu: acquisition of data, collection patient information. Xiaolan Liu: acquisition of data, postoperative follow-up. Zhangtian Xia and Wei Zhao: analysis and interpretation of data. Bing Huang: project leader, technical support, approval of the final version to be published.

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

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