Clinical Case Report on Electroacupuncture Combined with Warm Acupuncture for the Treatment of Painful Ophthalmoplegia (Tolosa-hunt Syndrome)

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Abstract: Tolosa-Hunt Syndrome (THS) is a rare and complex clinical syndrome characterized by severe headache, orbital pain, and multiple cranial nerve palsies. The precise etiology of THS remains unclear, though it is hypothesized to be related to immune system deficiencies, viral infections, or nonspecific inflammation. Due to its low incidence, THS is infrequently encountered in clinical practice. This case report presents a 39-year-old female patient with THS who was successfully treated with a combination of electroacupuncture and warm acupuncture. The patient's symptoms included left eyelid ptosis, left-sided headache, and reduced eye movement, which initially responded to high-dose steroid therapy but recurred upon cessation. After discontinuing steroid treatment, the patient received comprehensive acupuncture therapy aimed at soothing the liver and gallbladder, tonifying Qi, activating blood, and unblocking channels to relieve pain. Over the course of more than 30 acupuncture sessions, the patient's symptoms significantly improved, with complete resolution of pain and restoration of eye function. This case highlights the potential of acupuncture as an effective alternative treatment for THS, providing valuable insights for the management of this rare condition. Future research should further explore the mechanisms and efficacy of acupuncture in treating THS and other cranial nerve disorders.

Keywords: Tolosa-hunt Syndrome; Warm Acupuncture; Electroacupuncture

1. Introduction

Painful ophthalmoplegia, also known as Tolosa-Hunt syndrome (THS), is a rare and complicated clinical syndrome, characterized by severe headache, orbital pain and multiple pairs of cranial nerve paralysis. The specific pathogenesis of THS is not completely clear, but it is speculated that it is related to immune system defect, virus infection or nonspecific inflammation. Because the clinical incidence of this disease is low, it is rare in actual medical work. At present, high-dose glucocorticoid is an accepted treatment for this disease, but there is no uniform standard for the specific dose and course of use of hormone, and it is not uncommon to report the side effects caused by long-term use of hormone [1]. Acupuncture treatment of traditional Chinese medicine is widely used in various diseases because of its green, innocuity, simplicity and low cost, but there are still few reports about this disease. The author has been a doctor for 7 years, only one case has been encountered in clinic, and he has been successfully cured by acupuncture treatment of traditional Chinese medicine. This case is reported in detail as follows, in order to provide valuable reference for the diagnosis and treatment of related diseases.

2. Case Information

The patient, Ms. Xu XX, a 39-year-old female, first presented on June 1, 2021. Chief complaint: Left eyelid ptosis accompanied by left-sided headache for one month.

2.1 History and Examination

One month prior to the initial visit, the patient experienced sudden onset of left upper eyelid ptosis without any obvious cause, the onset time varies from several minutes to several hours every day, and the nature of the pain is persistent swelling and pain, with light day and heavy

night, dizziness, decreased vision in the left eye, diplopia and inability to turn the eyeball inward. No nausea and vomiting, no chest tightness, no discomfort such as fever and aversion to cold. The patient was diagnosed as "painful ophthalmoplegia" in the Department of Neurology of Quzhou People's Hospital, and received high-dose (500mg) methylprednisolone needle shock and sequential reduction treatment for 20 days. After hormone therapy, the pain is obviously relieved and the frequency of attack is reduced, but it is repeated again after drug withdrawal. When the symptoms are serious, ptosis can completely cover the eyeball, and the frequency of pain attack increases, accompanied by numbness in the left head and face, which affects sleep and leads to depression and anxiety. Because of the patient's physique and other comprehensive reasons, he could not use hormones for a long time, so he sought Chinese medicine treatment. The patient was in good health in the past, and denied the history of hypertension, diabetes, coronary heart disease and other infectious diseases such as hepatitis and tuberculosis.

2.2 Physical Examination

Mental status: Clear, with signs of nervousness and anxiety. The speech response is not fluent, no abnormal stars are found in heart and lung percussion and auscultation, the abdomen is soft, there is no tenderness and rebound pain in the whole abdomen, the liver and spleen are not touched, and Murphy's sign (-) and bowel sounds are normal. Left eyelid ptosis, blurred vision with diplopia, left pupil dilation to approximately 4 mm, sluggish light reflex, inability to adduct, elevate, or depress the left eyeball, but could abduct. No nystagmus was observed. Limb muscle strength and tone were normal, with no difference in depth and compound sensation, and the finger nose test and heel, knee and tibia test were negative and no pathological signs elicited. Brain MRI suggested abnormalities in the cavernous sinus area. The patient reported poor sleep and appetite, with normal bowel movements. Tongue examination revealed a dark red tongue with little coating, and the pulse was wiry, thin, and rapid.

2.3 Treatment Process

After 20 days of hormone shock treatment, the patient had discontinued steroid treatment and continued oral administration of mecobalamin tablets (0.5 mg tid) and vitamin B1 tablets (10 mg tid) to nourish the nerves. The TCM acupuncture treatment followed the principles of "soothing the liver and gallbladder, tonifying Qi and activating blood, and unblocking the channels to relieve pain." The selected acupuncture points included Yangbai, Zanzhu, Tongziliao, Taiyang, Xiaguan, Jingming, Shuaigu, Touwei, Fengchi, Yanglingquan, Zusanli, Guangming, Hegu, and Taichong. The operational method was as follows:

(1) Using Huatuo-brand acupuncture needles (specifications: 0.3 mm × 40 mm, 0.25 mm × 25 mm), after disinfecting the acupoints, Jingming was needled with a 0.25 mm × 25 mm filiform needle without lifting, thrusting, or twisting. Fengchi was obliquely needled towards the opposite eye corner with a 0.3 mm × 40 mm needle, achieving deqi with a sensation of soreness and distention radiating to the lateral head and forehead. The remaining points were needled conventionally, with needles retained for 30 minutes.

(2) Alternating usage of electroacupuncture with sparse-dense waves on Yangbai-Tongziliao and Taiyang-Xiaguan. Zusanli received warm acupuncture for approximately 20 minutes.

(3) Continuous acupuncture treatment for 6 days followed by a 1-day rest, with symptoms gradually improving after 2 weeks. The treatment was continued on alternate days for a total of over 30 sessions to consolidate the efficacy.

Herbal medicine "Dan Zhi Xiao Yao San" was modified and administered, including: Moutan Cortex 10g, Gardenia 10g, White Peony 30g, Angelica 10g, Poria 12g, Atractylodes Macrocephala 12g, Bupleurum 10g, Mint 6g (added last), Glycyrrhiza 6g, Rehmannia 10g, Anemarrhena 10g, Fushen 20g, and Chuanxiong 10g.

2.4 Outcome

Following the treatment, the patient's left temporal and orbital pain disappeared. The left upper eyelid could elevate, the range of motion is basically the same as the right side, but the opening and closing speed of eyelids is slower than that on the healthy side. And eye movement returned to normal. The patient reported improved vision with the disappearance of diplopia. No further ophthalmologic examination was conducted, and follow-up

indicated no recurrence of symptoms.

3. Discussion

In 1954, Tolosa reported a patient for the first time: the main symptom was periorbital pain of on one side of the orbit with cranial nerve paralysis of the third, fourth and sixth sides [2]. Six years later, Hunt reported six cases of this disease, including one patient treated by surgery, and put forward the diagnostic criteria of this disease at the earliest [3,4]. This case was first reported by Gao Yingbi and Zhang Fanghua in China in 1979 [5].The etiology of THS remains unclear. Currently, it is believed to be associated with immune deficiencies, viral infections, or nonspecific inflammation. The main pathogenesis theories include [6]: nonspecific granulomatous inflammation of the dura mater around the internal carotid artery, cavernous sinus segment, and superior orbital fissure; sphenoid sinusitis; and hypersensitivity inflammation. Among them, according to the revisions of the International Headache Association, it is unanimously believed that the disease is caused by cavernous sinus, supraorbital fissure or non-specific granulomatous inflammation of orbital apex. With the development of nuclear magnetic resonance technology, the detection rate of MRI positive imaging changes is increasing, which provides evidence for more reliable collection of nonspecific inflammatory granulomas in cavernous sinus and other parts. However, because there is no complete standard and unified imaging diagnosis at present, even if the MRI results of the head are negative, the possibility of this specific inflammatory granuloma can not be completely ruled out [7], so the clinical signs and symptoms are still of great significance in the diagnosis of this disease.

THS presents with primary clinical symptoms such as unilateral eye pain, typically centered in the orbit and retro-orbital regions, possibly radiating to the forehead, temple, and occipital regions. The cavernous sinus is located in the middle cranial fossa, between the medial meninges and the lateral endosperm on both sides of the sella turcica, one on the left and one on the right. The fibrous trabeculae in paranasal sinus make its tissue structure similar to sponge, and there are many small cavities that are interconnected. The oculomotor nerve, abducent nerve, trochlear nerve and the first and second branches of the trigeminal nerve pass through it. Its anatomical structure determines the characteristics of injury after pathological changes. Cranial nerve palsy primarily affects the oculomotor, trochlear, trigeminal, and abducens nerves, manifesting as eyelid ptosis, strabismus, diplopia, pupil dilation, absent reflexes, and decreased vision. Some patients may develop local circulatory disturbances due to cavernous sinus inflammation, resulting in symptoms like optic disc edema, proptosis, conjunctival congestion, eyelid edema, or orbital hemorrhage [8]. Related research [9,10] shows that although THS patients are characterized by inflammatory diseases and hormone therapy is effective, the quantitative indexes of granulocyte, monocyte and protein in cerebrospinal fluid are mostly slightly increased, which has no specific diagnostic significance. Imaging examination, particularly high-resolution MRI scans, can significantly enhance the diagnostic accuracy of THS by identifying lesions in the cavernous sinus and superior orbital fissure areas. Although THS is responsive to steroid therapy, many patients experience relapse after treatment [11], and long-term steroid use can lead to side effects and other health risks. Therefore, finding effective alternative treatments is crucial.

Modern medical research indicates that acupuncture signals are fed back from the body surface to the nerve center, they pass through multi-level neuronal activities such as the cerebral cortex, brain stem and spinal cord, and at the same time stimulate the body to secrete analgesic substances such as serotonin and acetylcholine. As a non-pharmacological treatment, it can effectively stimulate the release of endogenous opioids such as endorphins at the connections between spinal cord and hypothalamic neurons, thereby suppressing pain responses and alleviating chronic pain conditions [12], including THS. Endorphins are effective endogenous analgesics. Acupuncture can also regulate the function of nervous system, improves local blood circulation, and reduce inflammatory reaction through various mechanisms.

In traditional Chinese medicine, THS is often categorized under "sideways glance (Partial Vision)" syndromes, primarily due to qi stagnation and blood stasis in the head and facial meridians, leading to impaired qi and blood flow, blocked channels, and malnourished muscles, resulting in localized atrophy and dysfunction.

Acupuncture treatment for THS, employing specific acupoints combined with electroacupuncture and warm acupuncture, aims to soothe the liver and gallbladder, tonify qi and activate blood, and unblock channels to relieve pain. This approach effectively improves local blood supply and nerve conduction, alleviating the patient's pain and cranial nerve palsy symptoms. At the same time, because the analgesic effect of acupuncture has a slow onset and sustained effect, the body can not get the positive feedback of analgesic regulation at the moment of acupuncture, but it takes enough time and stimulation to reach the analgesic threshold. It usually takes several or even dozens of consecutive treatments to achieve the desired effect, so enough patience is needed in clinical treatment. However, long-term acupuncture can also lead to stress fatigue of the body's nervous system, so it is necessary to pause intermittently during treatment to restore the body's sensitivity to acupuncture reaction. According to the report [13], THS is not hormone shock, but is treated according to syndrome differentiation of traditional Chinese medicine, and it still has a clear curative effect by treating the liver and expelling wind, eliminating phlegm and dredging collaterals, and reusing insect drugs and drugs entering the liver meridian.

In this case, the patient's symptoms significantly improved after over 30 acupuncture sessions, with headache and orbital pain disappearing, eyelid function restoring, and vision partially recovering. The combined treatment with the herbal formula "Dan Zhi Xiao Yao San" further consolidated the acupuncture efficacy, avoiding the side effects and recurrence risks associated with steroid therapy.

The specific acupuncture treatment strategy included selecting acupoints such as Zanzhu, Touwei, Shuaigu, Yangbai, Taiyang, Tongziliao, Xiaguan, etc., and applying sparse-dense wave electroacupuncture to improve local blood supply and muscle nourishment while promoting the release of neurotransmitters like endorphins to inhibit pain responses. Distant acupoints were selected based on channel theory, diagnosing this condition as a Shaoyang channel syndrome, using Fengchi, Yanglingquan, and Guangming from the Shaoyang channel to soothe the liver and gallbladder. Fengchi, as a meeting point of the hand and foot Shaoyang channels, has the functions of dispelling wind and clearing heat. Yanglingquan, the He-Sea point of the Gallbladder channel, can alleviate anxiety. Guangming, the Luo-Connecting point of the Gallbladder channel, has the function of improving vision. The patient showed signs of qi and blood deficiency despite the presence of yin deficiency signs, necessitating the use of warm acupuncture at Zusanli to strengthen qi and blood. Jingming and Zhaohai, as points associated with multiple channels, play roles in eyelid opening and closing. Hegu and Taichong, paired as the "Four Gates," harmonize yin and yang, regulate qi and blood, and relieve depression and pain, with slightly heavier stimulation to ensure effective acupuncture stimulus and maintain a positive dose-response relationship.

4. Conclusion

Electroacupuncture combined with warm acupuncture has demonstrated significant efficacy in treating THS, providing an effective alternative therapy for patients who are concerned about or unsuitable for steroid treatment. Future research should further explore the mechanisms and clinical efficacy of acupuncture in the treatment of THS and other cranial nerve diseases.

Reference

[1] Ishikawa R, Ishikawa Y, Kubota T, et al. A case of delirium in Tolosa-Hunt syndrome during corticosteroid therapy. JA Clin Rep, 2018, 4(1):58.

[2] Tolosa EJ. Pefiartefitic lesions of carotid siphon with clinical features of carotid infraclinoid aneurysm. Neurol Neurosurg Psychiatry. 1954. 17: 300-302.

[3] Hunt WE. Tolosa-Hunt syndrome: one case of painful ophthalmoplegia. J Neurosurg, 1976, 44: 544-549.

[4] Hunt WE, Meagher JN, Lefever HE, et al. Painful opthalmoplegia. Its relation to indolent inflammation of the carvernous sinus. Neurology, 1961, 11: 56-62.

[5] Gao Yingbi, Zhang Fanghua. Painful ophthalmoplegia syndrome (report of 8 cases). Chinese Journal of Ophthalmology, 1979, 15 (1): 49-51.

[6] Xie Ruiman. Practical Neuro-Ophthalmology. Shanghai: Shanghai Scientific and Technical Literature Publishing House, 2004: 325-325.

[7] Jarholm JA, Faiz KW, Nysted T, et al. Orbital pain, ophthalmoplegia, and

oligoclonal bands in the cerebrospinal fluid:A case report of tolosa-hunt syndrome. Headache, 2018, 58(5): 758-760.
[8] Zhou Baiyu, Liu Xiaohui, Zhang Yanxia. Research progress on painful ophthalmoplegia . International Journal of Ophthalmology, 2012, 12(09): 1683-1685.
[9] Arnold M. Headache classification committee of the international headache society (IHS) the international classification of headache disorders, 3rd edition. Cephalalgia, 2018, 38(1): 1-211.
[10] Wei Chuanqiao, Chen Chunfu, Cui Xiaochen, et al. Clinical analysis of 62 patients with Tolosa-Hunt syndrome. Journal of Clinical Neurology, 2019, 32(3): 215-218.
[11] Cao Junfeng. Clinical diagnostic considerations of painful ophthalmoplegia. Guide of China Medicine, 2017, 15(13): 62-63.
[12] Liu Ling, Zhang Renliang, Chen Guanghui. Clinical features, diagnosis, and treatment of Tolosa-Hunt syndrome. Journal of Clinical Neurology, 2004(05): 383-385.
[13] Mao Yijia, Zhang Zongli, Hou Ronghui, et al. A case of painful ophthalmoplegia syndrome treated by Zhang Zongli. new traditional chinese medicine, 2019, 51(4): 323-324.

