



Case Report

Comprehensive Korean Medicine Treatment for Benign Essential Blepharospasm in a Patient Responding Poorly to Botulinum Toxin Treatment



Seojung Ha¹, Sung A Kim¹, Suji Lee¹, Sungwoon Choi¹, Sanghoon Lee^{2,*}

¹ Department of Clinical Korean Medicine, Graduate School, Kyung Hee University, Seoul, Korea

² Department of Medical Education, College of Korean Medicine, Kyung Hee University, Seoul, Korea

ABSTRACT

Article history:

Submitted: October 21, 2021

Revised: November 12, 2021

Accepted: November 29, 2021

Keywords:

acupuncture,
benign essential blepharospasm,
herbal medicine

Benign essential blepharospasm (BEB) is a condition/disease which involves involuntary muscle contractions causing the eye to repetitively close, making it impossible for the patient to perform daily activities. This study reports Korean medicine treatment of a rare case BEB in a 34-year-old male patient whose symptoms showed minimal improvement following botulinum toxin injection (the standard treatment). Acupuncture, pharmacopuncture, and herbal medicine treatment for 11 days resulted in clinical improvement for all symptoms as assessed by using the Jankovic Rating Scale, Blepharospasm Disability Index, numerical rating scale, duration of spasms lasting more than 1 hour per day, and number of spasms lasting less than 1 minute per day. This case report suggests that comprehensive Korean medicine treatment could be a treatment option for BEB in patients who do not respond well to botulinum toxin injection.

<https://doi.org/10.13045/jar.2021.00262>
pISSN 2586-288X eISSN 2586-2898

©2022 Korean Acupuncture & Moxibustion Medicine Society. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Benign essential blepharospasm (BEB) is a type of dystonia in which the eyelids are partially or completely closed due to persistent, involuntary spasms of the orbicularis oculi muscles. The prevalence of BEB is estimated to be 14-133 cases per 1,000,000 people and generally occurs between the ages of 50 and 70 years. Symptoms often range from a slight increase in the frequency of blinking to eyelid closing, and in severe cases, it leads to functional loss of vision [1]. The standard treatment for BEB is injection with botulinum toxin. However, because the response to treatment varies widely and antibodies to botulinum toxin are produced, repeated treatments may require an increasing dose. Injection with botulinum toxin can

also cause side effects such as diplopia, blurred vision, and eyelid ptosis [2]. Korean medicine (KM) treatments for BEB such as acupuncture, pharmacopuncture, and herbal medicine can improve symptoms, but published studies are rare. This case report presents the effectiveness of KM treatment in a patient with BEB who did not experience satisfactory improvement following botulinum toxin injections.

Case Report

A 34-year-old Korean man presented at the Korean Medicine Hospital of Kyung Hee University. The patient had a 2-year history of BEB and he presented stereotypically with bilateral, synchronous

*Corresponding author. Sanghoon Lee

Department of Medical Education, College of Korean Medicine, Kyung Hee University, 26 Kyungheedaero, Dongdaemun-gu, Seoul 02447, Korea

E-mail: shlee777@khu.ac.kr

ORCID: Seojung Ha <https://orcid.org/0000-0001-9369-1119>, Sung A Kim <https://orcid.org/0000-0003-0321-2351>, Suji Lee <https://orcid.org/0000-0001-7159-2929>, Sungwoon Choi <https://orcid.org/0000-0002-1510-5441>, Sanghoon Lee <https://orcid.org/0000-0002-1324-6101>

©2022 Korean Acupuncture & Moxibustion Medicine Society. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

spasms with sensory tics, which originated in the eyelids; other facial muscles and neck muscles were not involved. His symptoms included short repeated blinks or persistent closing of his eyes lasting from a few seconds to more than an hour. These spasms could not be suppressed, were exacerbated by stress and bright light, sometimes they were relieved by wearing a hat or sunglasses, and were generally absent whilst pressing the left temporal region hard or sleeping. Moreover, their frequency and severity have increased over the years. The patient had become exhausted due to the condition/disease and had developed a sleep disorder. Due to these characteristics, diagnoses such as facial motor tics and facial myokymia could be excluded.

The patient had visited a neurologist where he received botulinum toxin injections. The patient received 3 injections following which the symptoms improved for 2-3 days, then worsened. The patient was disappointed with the outcome and transferred to another hospital. The patient had no history of any apparent physical or mental impairment. Imaging tests, genetic tests, electromyography, electrical stimulation, and blood tests revealed no abnormal findings. His symptoms worsened without any specific reason, and he was admitted to the Korean Medicine Hospital of Kyung Hee University.

The patient received combined KM treatment including acupuncture, pharmacopuncture, and herbal medicine during hospitalization (July 26, 2021, to August 6, 2021). Acupuncture treatment was performed twice a day for 20 minutes using standardized (0.25 × 40 mm) sterilized stainless steel disposable needles (DongBang Acupuncture Inc., Boryung, South Korea). The acupoints used were BL2, GB14, GV26, LI4, LI11, LR3, ST2, ST36, TE23, EX-HN3, EX-HN4, and EX-HN5. Bee venom (BV) injection (0.1 cm³ dose) was administered once a day using a 1 cm³ disposable 30-gauge syringe (Hwajin Medical Co., Seoul, South Korea) around both eyes, after performing a hypersensitivity skin test to BV. Dried BV (10 mg; Yoomil Garden, Hwasun, South Korea) was diluted with 300 cm³ (1:30,000) of saline (Joongwe Pharmaceuticals, Seoul, South Korea). Sayeoksan decoction consisting of Bupleuri Radix, Paeoniae Radix, Ponciri Fructus Immaturus, and Glycyrrhizae Radix et Rhizoma, was prescribed 3 times a day (Table 1).

Symptoms were assessed at admission and discharge using the Jankovic Rating Scale, the Blepharospasm Disability Index (BSDI), and the numerical rating scale (NRS). The Jankovic Rating Scale consists of 2 subscales (severity and frequency) on a 5-point scale

ranging from 0 to 4, where 0 indicates no symptoms, and 4 indicates the most severe or frequent symptoms. The severity score changed from 3 to 2 and the frequency score changed from 3 to 1 between admission and discharge evaluations (Fig. 1). The BSDI, a disease-specific functional scale developed for self-assessment, consists of 6 items with 5 points ranging from 0 to 4, where 0 indicates no

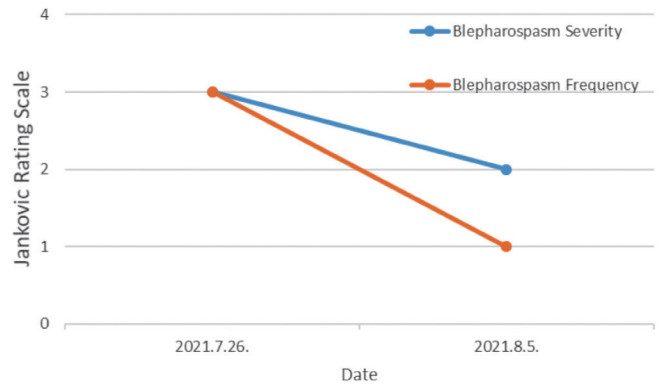


Fig. 1. A reduction in Jankovic Rating Scale scores during hospitalization.

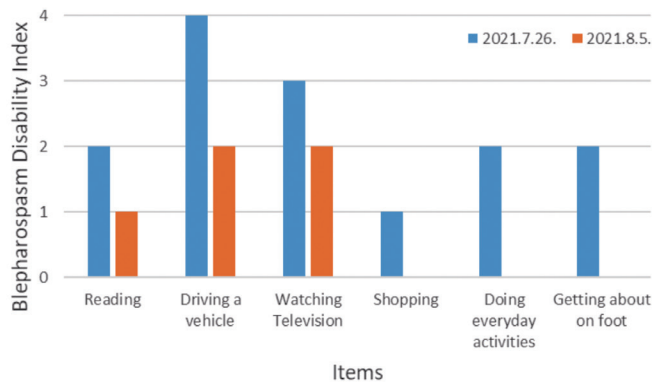


Fig. 2. A reduction in Blepharospasm Disability Index scores during hospitalization.

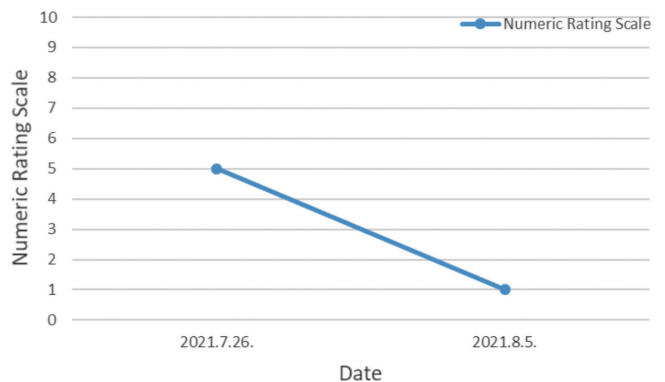


Fig. 3. A reduction in Numeric Rating Scale scores during hospitalization.

Table 1. Components of Sayeoksan Decoction Taken Each Day.

Crude drug name	Amount (g)
Bupleuri Radix	10
Paeoniae Radix	8
Ponciri Fructus Immaturus	4
Glycyrrhizae Radix et Rhizoma	4

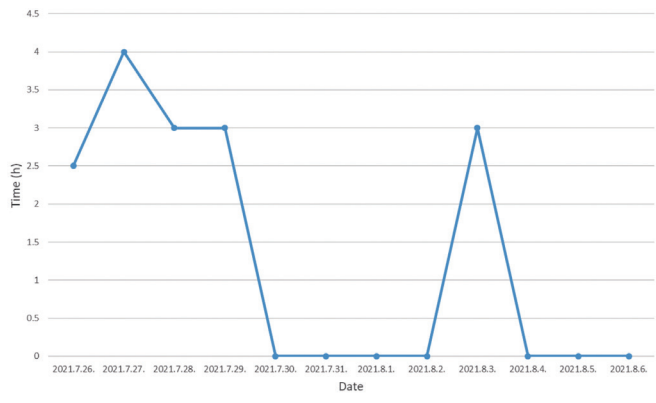


Fig. 4. Changes in the duration of spasms lasting more than 1 hour per day over the hospitalization period.

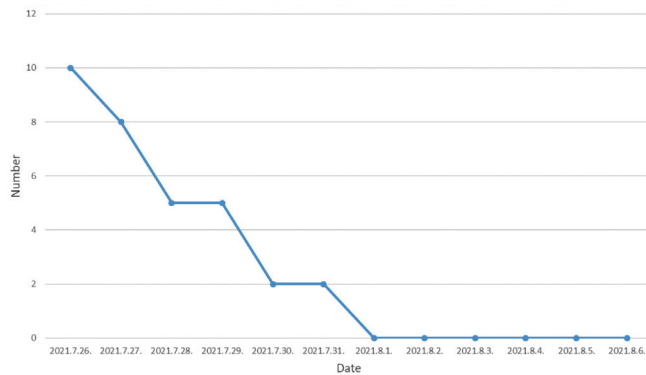


Fig. 5. Changes in the number of spasms lasting less than 1 minute per day over the hospitalization period.

impairment and 4 indicates not condition/disease related. The total BSDI scores decreased from 14 to 5 (Fig. 2). Discomfort due to symptoms was rated on a scale of 0 to 10 on the NRS, where 0 indicates no discomfort, and 10 indicates the greatest discomfort imaginable. The NRS score decreased from 5 to 1 following treatment (Fig. 3).

The duration of spasms (lasting more than 1 hour per day) and the number of spasms (lasting less than 1 minute per day) were recorded at admission then daily as indicators of change in pattern of symptoms. From Day 4 of hospitalization, except on Day 8, spasms lasting more than 1 hour per day were not observed (Fig. 4). The number of spasms lasting less than 1 minute per day decreased from 10 on the day of admission to resolution on Day 7 (Fig. 5).

After discharge, the patient received acupuncture and pharmacopuncture treatment at the outpatient clinic once every 2 weeks and was taking herbal medicine for 3 months. The patient maintained the same improved condition as at the time of discharge which was 3 months ago.

Discussion

The exact pathophysiology of BEB remains unknown, but several environmental risk factors have been reported to cause BEB [3]. These include high levels of urbanization and people working in “white-collar” jobs related to stressful lifestyles. It was reported that 42.6% of patients experience a stressful life event before the onset of BEB symptoms [3]. The standard treatment for BEB is botulinum toxin injection. It works by inhibiting the release of acetylcholine from the neuromuscular junction thereby reducing muscle contraction. However, this treatment can cause many side effects, such as ptosis, diplopia, photophobia, and blurred vision. In addition, most patients over time require increased doses of botulinum toxin to alleviate their symptoms [4].

In traditional KM theory, BEB is usually caused by internal wind produced by long-term blood deficiency and stagnation of the liver and spleen, as well as by malnutrition of the tendons and muscles. Wind, which is characterized by moving and shaking, can attack the eyelids and cause spasms. In this patient, a Sayeoksan decoction was prescribed to relieve mental stress by improving the liver dredging function and to relieve the Qi that is stagnant and does not flow to the extremities. The liver is an organ damaged by anger and stress, and when the liver Qi is stagnant, it can cause stress-related conditions/diseases. The liver’s dredging function helps prevent these conditions/diseases by preventing Qi from stagnating.

As a type of dystonia, BEB has been reported to be related to dopamine deficiency, so dopamine agonists and dopamine absorption inhibitors are used to treat this condition/disease [5,6]. A component present in the bee venom, melittin, is negatively associated with dopamine transmission. In vitro studies have shown that melittin acts directly on the dopamine transporter, reducing dopamine uptake and inhibiting the binding of the antagonist to the dopamine receptor [5,6]. In this case, bee venom was injected to improve microcirculation in both eyes, and acupuncture was used to relax the eyelids. Local acupoints such as BL2, GB14, ST2, TE23, EX-HN3, EX-HN4, and EX-HN5 strengthen Qi-blood, and nourish the eye region. Distal acupoints such as GV26, LI11, LR3 and ST36 restore the movement of Qi-blood and suppress endogenous wind. Bilateral LI4 may enhance therapeutic effects [7].

This study provides meaningful insights into the treatment of BEB using KM treatment. KM treatment is more cost-effective than botulinum toxin for BEB, and it has a better side-effects profile, and better tolerability. However, this report has several limitations in that it is limited to 1 case, and it is difficult to judge the effectiveness of each KM treatment by administering several treatments at the same time. Further studies are needed to support the safety and efficacy of combination therapy with acupuncture, pharmacopuncture, and herbal medicine.

Conflicts of Interest

The authors declare no conflicts of interest.

Ethical Statement

This research did not involve any human or animal experiment.

References

- [1] Hwang CJ, Eftekhari K. Benign Essential Blepharospasm: What We Know and What We Don't. *Int Ophthalmol Clin* 2018;58:11-24.
- [2] Ababneh OH, Cetinkaya A, Kulwin DR. Long-term efficacy and safety of botulinum toxin A injections to treat blepharospasm and hemifacial spasm. *Clin Exp Ophthalmol* 2014;42:254-261.
- [3] Sun Y, Tsai P-J, Chu C-L, Huang W-C, Bee Y-S. Epidemiology of benign essential blepharospasm: A nationwide population-based retrospective study in Taiwan. *PLoS One* 2018;13:e0209558.
- [4] Anwar MS, Zafar H. Efficacy of botulinum toxin in benign essential Blepharospasm: Desirable & undesirable effects. *Pak J Med Sci* 2013;29:1389-1393.
- [5] Keith DJ, Eshleman AJ, Janowsky A. Melittin stimulates fatty acid release through non-phospholipase-mediated mechanisms and interacts with the dopamine transporter and other membrane-spanning proteins. *Eur J Pharmacol* 2011;650:501-510.
- [6] Keith DJ, Wolfrum K, Eshleman AJ, Janowsky A. Melittin initiates dopamine transporter internalization and recycling in transfected HEK-293 cells. *Eur J Pharmacol* 2012;690:13-21.
- [7] Cheng F, Ma C, Wang X, Zhai C, Wang G, Xu X et al. Effect of traditional Chinese medicine formula Sinisan on chronic restraint stress-induced nonalcoholic fatty liver disease: A rat study. *BMC Complementary and Alternative Medicine* 2017;17:203.