**Original Article** 

# Clinical Features of Benign Essential Blepharospasm in Korean Patients

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Purpose: To analyze the clinical features of benign essential blepharospasm in Korean patients.

- **Methods:** Patients diagnosed with benign essential blepharospasm in Kim's Eye Hospital from November 2014 to December 2016 were evaluated using a clinical examination and questionnaire. The questionnaire reviewed personal medical history, demographic factors, risk factors for blepharospasm development, and relieving and aggravating factors.
- **Results:** Of the 101 patients enrolled, 78 (77.2%) were women. The mean age was 64.9 years old. Hypertension was the most common medical disorder (42.6%), followed by diabetes mellitus. The majority of the patients were non-smokers (83.2%) and drank less than a cup of a caffeinated beverage a day (30.7%). Fifty-seven percent of patients reported no stressful events immediately prior to symptom development. Fatigue and stress were aggravating factors in more than 55% of patients; rest was the most common relieving factor (35.6%).
- **Conclusions:** Here, we report the clinical features of benign essential blepharospasm in Korean patients for the first time. The results were consistent with previous reports showing that the majority of benign essential blepharospasm patients are women and non-smokers. In contrast to previous reports though, fatigue and stress were aggravating factors, and the most common relieving factor was rest. No stressful events had immediately preceded the development of blepharospasm in 57.4% of patients. This report may aid in treating and counseling patients with benign essential blepharospasm.
- Key Words: Aggravating factor, Benign essential blepharospasm, Caffeinated beverage, Relieving factor, Smoking

Blepharospasm is characterized by excessive involuntary closure of the bilateral eyelids and is generally caused by orbicularis oculi muscle spasms [1]. Benign essential blepharo-

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spasm is considered a form of focal dystonia. Initially, spasms are mild and infrequent, but gradually progress and become highly disruptive to the patient's daily activities and quality of life [2,3]. The prevalence of benign essential blepharospasm is reported at 12 to 133 per million. Accurately estimating the prevalence is difficult because of underdiagnosis and misdiagnosis; however, it is more common than myasthenia gravis, amyotrophic lateral sclerosis, and Huntington's disease [4].

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Onset of benign essential blepharospasm typically occurs in the fifth to seventh decade of life. Female predominance has been reported [5,6]. Benign essential blepharospasm is associated with a family history of dystonia, head or face trauma, and other neurological movement disorders (such as Parkinson disease and Tourette syndrome) [7,8]. Studies have reported that 24% to 72% of patients experience one or more stressful event prior to the development of symptoms [9,10]. Caffeine use may be inversely associated with the development of blepharospasm, and the strength of the inverse association between blepharospasm and caffeine intake tends to increase with the average number of cups consumed per day [11]. Defazio et al. [12] had reported that smoking reduces the risk of benign essential blepharospasm.

Blepharospasm is worsened by increasing light intensity and movement, such as walking and driving, and may improve with concentration, rest, and talking or humming. Blepharospasm is ameliorated after sleep, in the dark, and often by gazing down. Focal pressure on the temple or shutting the eyes may also help reduce symptoms [13].

Although studies have been conducted on the clinical features of benign essential blepharospasm, none have reported clinical features in Korean patients. Therefore, we investigated the clinical features of benign essential blepharospasm in Korean patients.

## **Materials and Methods**

The institutional review board of Kim's Eye Hospital in Seoul, South Korea, approved this study (2017-11-001). All study methods adhered to the tenets of the Declaration of Helsinki. A total of 101 patients who were diagnosed with benign essential blepharospasm at Kim's Eye Hospital from November 2014 to December 2016 were evaluated retrospectively. Patients were evaluated using a clinical examination and questionnaire. The questionnaire reviewed personal medical history (including neurological or psychiatric medication), demographic factors (age, sex, caffeine and tobacco use, and stressful events prior to benign essential blepharospasm development), and factors that relieve and aggravate symptoms. We presented the patients with a multiple-choice questionnaire comprised of questions from previous studies as well as additional new questions.

Of the 101 patients included in the study group, 78 were women and 23 were men. The average age was  $64.91 \pm 9.6$ vears (range, 41 to 86 years), and 49 patients (48.5%) had a comorbid disease (Table 1). Hypertension was the most common medical disorder (42.6%), followed by diabetes mellitus (10.9%), heart disease (5.0%), and hypothyroidism (2.0%). Eleven patients (10.9%) were diagnosed with insomnia and 7 patients (6.9%) were diagnosed with depressive disorders. Nineteen patients (18.8%) were taking neurological or psychiatric medication. Of the included patients, 57.4% reported no stressful events prior to the development of symptoms, 19.8% reported a family issue such as divorce, 13.9% reported problems at work, 2% reported illness of a family member, 1% reported the death of a parent, 1% reported moving to a new house, 1% reported head trauma, 1% reported a traffic accident, and 1% reported cerebral infarction (Table 2). In addition, 4% of patients reported ocular conditions and related surgery (for example, blepharoplasty, cataract surgery, conjunctivitis).

Comorbid disease	Number (%)
Hypertension	43 (42.6)
Diabetes mellitus	11 (10.9)
Heart disease	5 (5.0)
Hypothyroidism	2 (2.0)
Total	49 (48.5)

 Table 2. Stressful events prior to symptom development in patients

Stressful event	Number (%)
Family issue such as divorce	20 (19.8)
Problems at work	14 (13.9)
Ocular condition and ocular surgery	4 (4.0)
Illness of a family member	2 (2.0)
Death of a parent	1 (1.0)
Moving houses	1 (1.0)
Head trauma	1 (1.0)
Traffic accident	1 (1.0)
Cerebral infarction	1 (1.0)
None	58 (57.4)

The majority of patients (68.3%) reported relieving factors (Table 3). The most common relieving factor was rest (35.6%), followed by concentrating on one's work (12.9%), singing (5%), talking (5%), eating (3%), sleeping (3%), exercising (3%), a massage (2%), walking (2%), and being in a wet or cold environment (2%). Some patients reported relieving factors such as applying lipstick, receiving facial acupuncture, reading a book, mountain climbing, taking medicine, visiting a sauna, and engaging in outdoor activities. The remaining 31.7% of patients reported no relieving factors.

The majority of patients (95%) reported aggravating factors (Table 4). The most common aggravating factors were fatigue (55.4%) and stressful events (46.5%), followed by watching television (27.7%), seeing a bright light (18.9%), having dry eye symptoms (14.9%), illness (10.9%), reading a book (8.9%), walking (5.9%), driving (4.0%), talking (4.0%), and being in a dusty or windy environment (4.0%). Some patients also reported aggravated symptoms in the evening as well as with sleep, eye irritation, drinking alcohol, getting in a car, working with computers, performing housework, and a lack of sleep. The remaining 5% of patients reported no aggravating factors.

Of the included patients, 83.2% never smoked cigarettes,

Table 3. R	elieving	factors of	f benign	essential	blepharospasm	

Relieving factor	Number (%)
Rest	38 (35.6)
Concentrating on one's work	13 (12.9)
Singing	5 (5.0)
Talking	5 (5.0)
Eating	3 (3.0)
Sleep	3 (3.0)
Exercise	3 (3.0)
Massages	2 (2.0)
Walking	2 (2.0)
Wet or cold environments	2 (2.0)
Applying lipstick	1 (1.0)
Facial acupunctures	1 (1.0)
Reading a book	1 (1.0)
Mountain climbing	1 (1.0)
Taking medicine	1 (1.0)
Visiting a sauna	1 (1.0)
Outdoor activities	1 (1.0)
None	32 (31.7)

7.9% reported prior use of cigarettes, and 7.9% were current smokers. In regard to caffeine, 80.2% of patients consumed caffeinated beverages regularly, with 30.7% drinking  $\leq$ 1 cup per day, 28.7% drinking 1 to 2 cups per day, 9.9% drinking 2 to 3 cups per day, and 9.9% drinking >3 cups per day (Table 5). All patients were treated with botulinum toxin, and symptoms improved with its use.

Aggravating factor	Number (%)
Fatigue	56 (55.4)
Stressful events	47 (46.5)
Watching television	28 (27.7)
Seeing a bright light	19 (18.9)
Symptom of dry eye	15 (14.9)
Sick	11 (10.9)
Reading a book	9 (8.9)
Walking	6 (5.9)
Driving	4 (4.0)
Talking	4 (4.0)
Dusty or windy environments	4 (4.0)
Sleep	3 (3.0)
Evening	2 (2.0)
Irritating of the eyes	2 (2.0)
Drinking alcohol	2 (2.0)
Getting in a car	1 (1.0)
Working with computers	1 (1.0)
Performing housework	1 (1.0)
Lack of sleep	1 (1.0)
None	5 (5.0)

Table 4. Aggravating factors of benign essential blepharospasm

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Smoking or caffeine intake	Number (%)
Smoking	
Never smoker	84 (83.2)
Former smoker	8 (7.9)
Current smoker	8 (7.9)
Caffeine intake	
None	20 (19.8)
$\leq 1$ cup per day	31 (30.7)
1–2 cups per day	29 (28.7)
2–3 cups per day	10 (9.9)
>3 cups per day	10 (9.9)

### Discussion

This report is the first on the clinical features of benign essential blepharospasm in Korean patients. Our results corroborate previous findings in benign essential blepharospasm patients with a similar average age of onset and female predominance [5,6].

Previous studies have shown no significant association between benign essential blepharospasm and comorbidities [4,14]. In our results, almost 50% of patients had a comorbid disease, with hypertension being the most common, followed by diabetes mellitus; however, this study did not include a control group to assess the effect of comorbidities. Comorbid diseases and benign essential blepharospasm tend to occur more frequently in older patients and might not be independent of each other. Therefore, further research should include control groups. In our results, 10.9% of patients were diagnosed with insomnia and 6.9% of patients were diagnosed with depressive disorder. In addition, 19% of patients were taking neurological or psychiatric medications, the details of which we could not determine.

In our study, 42.6% of patients reported stressful events prior to symptom development, which is higher than the 24% reported in one study [9] and lower than the 72% reported in an another study [10]. Family issues such as divorce (19.8%) were the most common stressful event, followed by problems at work, illness of a family member, the death of a parent, moving, head trauma, a traffic accident, and cerebral infarction. Some patients reported ocular conditions and surgery (for example, blepharoplasty, cataract surgery, conjunctivitis). Johnson et al. [15] found a significant relationship between major life stressors and the development of benign essential blepharospasm. They reported that symptoms began within 1 year of a major stressful life event in 70% of cases. Unfortunately, we did not investigate the time period between the stressful event and onset of symptoms. Thus, further studies are needed.

In our study, the majority of the patients (68.3%) had relieving factors. The frequencies of relieving factors (also referred to as "sensory tricks") in patients with benign essential blepharospasm vary widely in the literature (17% to 87%) [10,16]. The Movement Disorder Society's classification of dystonia in 2013 indicated sensory tricks as a major sign and component of the clinical phenomenology of benign essential blepharospasm [17,18]. Variations in sensory tricks have been described, for example motor, forcible, imaginary, and reverse sensory tricks. The most common sensory tricks reported for benign essential blepharospasm include using the forefinger and/or thumb to touch the upper eyelid, stretching or rubbing of the eyebrows, eyelids, or forehead, and touching the forehead or chin [10,16,19]. However, in our study, no patients reported the aforementioned sensory tricks; instead, the most common relieving factor was rest, followed by concentrating on one's work, singing, talking, eating, sleeping, exercising, a massage, and walking. Some patients also reported relieving factors such as applying lipstick, facial acupuncture, reading a book, mountain climbing, taking medicine, visiting a sauna, engaging in outdoor activities, and being in a wet or cold environment. All studies on sensory tricks have been conducted with Caucasian patients: hence, this difference may be due to racial differences and the environment. We presented the patients with a multiple-choice questionnaire consisting of questions taken from previous studies related to Caucasian populations as well as additional new questions.

The majority of patients (95%) in the study reported aggravating factors. Anderson et al. [1] reported that bright light triggers or exacerbates symptoms in nearly 80% of patients with benign essential blepharospasm. However, in our study, the most common aggravating factor was fatigue and a stressful event, followed by watching television, seeing a bright light, symptoms of dry eye, illness, reading a book, walking, driving, and talking. Some patients also reported aggravated symptoms in the evening, as well as sleep, irritation of the eyes, drinking alcohol, getting in a car, working with computers, being in a dusty or windy environment, performing housework, and a lack of sleep. Studies on the aggravating factors of benign essential blepharospasm have also only been conducted with Caucasians; hence, these differences may also derive from racial differences and the environment. We presented the patients with a multiple-choice questionnaire consisting of questions taken from previous studies related to Caucasian populations as well as additional new questions. Therefore, more research is needed on the aggravating factors.

We also observed that 80.2% of patients consumed caffeinated beverages regularly. Of these, 30.7% drank  $\leq 1$  cup per day, 28.7% drank 1 to 2 cups per day, 9.9% drank 2 to 3 cups per day, and 9.9% drank >3 cups per day. Defazio et al. [11] had reported a possible protective effect associated with caffeine that increases with the amount consumed. In our study, caffeine use was high, but the percentage of patients who drank several cups of caffeinated beverages per day was low. Because we did not compare coffee consumption to a control group, determining whether this result indicates that caffeine provides a protective effect regarding benign essential blepharospasm is difficult, but we note here that such a correlation is plausible.

We observed that 83.2% of patients had never smoked. Smoking reduces the risk of benign essential blepharospasm [12]. However, Defazio et al. [11] had found no correlation between smoking and benign essential blepharospasm, and no additional studies are available that clarify the effects of smoking. In our study, the high proportion of non-smokers suggests that smoking lowers the risk of benign essential blepharospasm, but further evaluation of the correlation between cigarettes and benign essential blepharospasm is needed.

The main limitation of our study was that it was retrospective and there was not a healthy control group. Further research is needed to evaluate the effects of cigarettes, caffeine, and other factors on benign essential blepharospasm symptoms.

### **Conflict of Interest**

No potential conflict of interest relevant to this article was reported.

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