

A Case Report on the Use of Korean Medicine Treatment for a Patient with Dysarthria Caused by Progressive Bulbar Palsy (PBP)

Jae Hyun Ahn, Sung Yoon Kim, Jun Hyeong Park, Jeongjae Cho,
Do Young Choi, Seung Hoon Lee and Jae Dong Lee*

Department of Acupuncture & Moxibustion Medicine, College of Korean
Medicine, Kyung Hee University



[Abstract]

Objectives : To introduce a rare case of a patient with Progressive Bulbar Palsy (PBP) and suggest the possibility of treatment using electroacupuncture and Korean Medicine.

Methods : A 61-year-old man with PBP, complaining of dysarthria, was treated with electroacupuncture and Korean Medicine, from June 16 to July 10, 2015. Improvements in symptoms were measured using the speech mechanism screening test (SMST), measurement of tongue and orbicularis oris motility, and speech handicap index (SHI).

Results : The scores of SMST, motility of tongue and orbicularis oris showed a tendency for gradual improvement with 25 days of Korean Medicine Treatment after admission, but conversation was still impossible. In SHI scores, one point increased in the speech domain and one point decreased in the psycho-social domain, and three points increased in the other domain.

Conclusion : Electroacupuncture and Korean Medicine Treatment improved dysarthria caused by PBP, but not completely. Korean Medicine Treatment seems effective in the management of accompanying symptoms such as black hairy tongue, dry mouth, and general condition. The symptoms of PBP are similar to those of amyotrophic lateral sclerosis and there is controversy regarding the classification of PBP. The most important aspect of treating a patient with PBP is an early diagnosis and devising appropriate rehabilitation strategies.

Key words :

Progressive Bulbar
Palsy, PBP;
Electroacupuncture;
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* Corresponding author : Department of Acupuncture & Moxibustion Medicine, Kyung Hee
University Korean Medicine Hospital, 23, Kyunghedae-ro, Dongdaemun-gu, Seoul, 02447,
Republic of Korea
Tel : +82-2-958-9202 E-mail : ljdacu@khmc.or.kr

I. Introduction

Progressive Bulbar Palsy (PBP) is a motor neuron disease that attacks the nerves supplying the bulbar muscles. It is characterized by selective degeneration of motor neurons of the lower brainstem¹. This specifically involves the glossopharyngeal nerve (IX), vagus nerve (X), and hypoglossal nerve (XII)². PBP produces symptoms such as dysarthria, dysphagia, atrophy and fasciculations of the musculature of the tongue, and excessive accumulation of secretions³. Effective treatment is not yet known and the prognosis of PBP is poor. Patients commonly die due to aspiration pneumonia and malnutrition 2-4 years from the commencement of symptoms⁴.

There have been four case reports³⁻⁶ of PBP introducing the disease itself, one case report⁷ of analgesic treatment applied to a PBP patient complaining of masseter spasticity, and one case report⁸ of using Korean Medicine to treat PBP. Thus, there have been only a few case reports dealing with PBP, and there have been no reports of treating dysarthria as one of the representative symptoms of PBP. In this paper, we report the results of applying electroacupuncture and Korean Medicine treatment to a patient suffering from dysarthria caused by PBP.

II. Case study

1. Subject of study

1) Patient

Hong ○ ○. Man, 61 years old
(Height: 170cm, Weight: 55kg)

2) Symptoms

- (1) Dysarthria
- (2) Dysphagia

3) Onset

2012, July.

4) Past medical history

- (1) Left cataract operation (2012, June)

5) Family history

None

6) Present history

- (1) 2010: Blurred vision and bleary eyes.
- (2) June, 2012: Left eye was operated upon for cataract and the symptoms deteriorated.
- (3) July, 2012: Dysarthria, dysphagia and paralysis of tongue suddenly appeared after a nap.
- (4) August, 2012: The patient visited a neurosurgery specialist and magnetic resonance imaging (MRI) of the brain was entirely normal.
- (5) August, 2013: Over time, the symptoms worsened and he visited a neurology department. Brain MRI and thyroid function tests were normal. After electrodiagnostic examination of his whole body including the tongue, PBP, a motor neuron disease, was diagnosed.
- (6) September, 2013: He visited another neurology specialist. After electromyography of the right side, he was diagnosed as not having amyotrophic lateral sclerosis (ALS). He started taking medication to increase peripheral circulation and follow up electromyographic examinations were performed every 12 months until he visited our hospital.
- (7) June 15, 2015: The symptoms continued to worsen and he received admission treatment in a hospital of Korean Medicine.

7) Findings at first visit

- (1) Dysarthria

Almost no spontaneous movement of the tongue was observed, and fasciculation of the tongue was accompanied. Conversation was impossible. However, the sensations of tongue and lips were normal. There was difficulty in puckering lips.

- (2) Four diagnostic examinations of Korean Medicine
- ① Physical appearance: Darkish complexion, skinny body type.
 - ② Digestion: Good condition
 - ③ Stool: Once a day, good condition
 - ④ Urination: Good condition
 - ⑤ Sleep: Profuse dreaming
 - ⑥ Emotion: Stress, anxiety
 - ⑦ Cold and heat: Upper body heat
 - ⑧ Sweat: No abnormal findings
 - ⑨ Mouth and tongue: Dry mouth
 - ⑩ Tongue diagnosis: Black hairy tongue, sticky saliva
 - ⑪ Pulse diagnosis: Floating pulse

8) Examination results

(1) Neurological examination

Mental state: Alert

Biceps reflex: Normal

Knee reflex: Normal

Babinski sign: Normal

Ankle clonus: Normal

Cranial nerve examination: The functions of olfactory nerve (I), optic nerve (II), oculomotor nerve (III), trochlear nerve (IV), trigeminal nerve (V), abducens nerve (VI), facial nerve (VII), vestibulocochlear nerve (VIII), and accessory nerve (XI) were intact, and functions of glossopharyngeal nerve (IX), vagus nerve (X), and hypoglossal nerve (XII) were not intact (Table 1).

(2) Electrodiagnostic examination

① Electromyography (EMG)

(August 7, 2013)

Multifocal denervation of the bilateral tongue, masseter, cervical paraspinalis, and the left flexor carpi ulnaris were suggested.

(May 4, 2015)

Monopolar needle EMG of right first dorsal interosseus, tongue, masseter, C5, C7, T1, T5, T7 muscles showed mild to moderate degree denervation potentials.

Table 1. Cranial Nerve Examination

Cranial nerve	Function
Olfactory (I)	Intact
Optic (II)	Intact
Oculomotor (III)	Intact
Trochlear (IV)	Intact
Trigeminal (V)	Intact
Abducens (VI)	Intact
Facial (VII)	Only the movement of the mouth was decreased and other parts of the face were intact
Vestibulocochlear (VIII)	Intact
Glossopharyngeal (IX)	Gag reflex
Vagus (X)	Dysphagia, hoarseness
Accessory (XI)	Intact
Hypoglossal (XII)	Degradation of the tongue and fasciculation were observed

EMG of right dorsal interosseus, tongue, and masseter muscles showed giant motor unit action potential (MUAPs) with reduced recruitments.

EMG of right biceps brachii, vastus lateralis, tibialis anterior, L2, L3, L4, L5, and S1 paraspinal muscles were normal.

There was electrophysiologic evidence of denervation and reinnervation potentials in the right craniobulbar segments and denervation potentials in thoracic paraspinal muscles. There was also electrophysiologic evidence of cervical radiculopathy (C8).

② Repetitive nerve stimulation

(August 30, 2013)

Repetitive nerve stimulation (2, 3, 5, 30 hertz) was performed in the ulnar, spinal accessory, and facial nerves. The recordings were performed on the abductor digiti minimi, trapezius, and orbicularis oculi. There was no equivocal electrophysiologic dysfunction.

③ Nerve Conduction Study (NCS)

(May 4, 2015)

Right median, ulnar motor, sensory, F responses were normal.

Right posterior tibial, peroneal motor, F responses were normal.

Right sural sensory response was normal.

Bilateral soleus H-reflexes were normal.

(3) Other examinations

Antinuclear Antibody (ANA) Test: Negative

Kennedy disease genetic analysis: Negative

2. Treatment method

1) Electroacupuncture treatment

Electroacupuncture treatment was conducted by a resident, training at the Oriental Medicine Hospital of Kyung Hee University. The treatment was performed from June 16 to July 10, 2015 for a total of 25 days, using single-use stainless steel 0.25 × 40 millimeter needles (DongBang acupuncture Inc, Korea). The patient lied down supine and the acupoints Sugu (GV26) and Seungjang (CV24) were used to target orbicularis oris muscle, and two points 5 millimeter beside Yeomcheon (CV23) were used to target genioglossus muscle. The needle was inserted in Sugu (GV26) and Seungjang (CV24) 15 millimeter obliquely, and Yeomcheon (CV23) 20 millimeter perpendicularly. Two-hertz electroacupuncture (STN-111, Stratek, Korea) treatment was performed once a day for 30 minutes.

2) Speech therapy

Speech therapy was performed once a day for 30

minutes from June 17 to July 10 by a speech therapist.

3) Dysphagia treatment

Dysphagia rehabilitation and electrical stimulation were performed once a day from June 25 to July 10.

4) Acupuncture treatment

Ten acupuncture needles using single-use stainless steel 0.25 × 40 millimeter needles (DongBang acupuncture Inc, Korea) were inserted 1 millimeter in posterior cervical muscles and the upper trapezius muscle including Pungji (GB20), Pungbu (GV16), Gyeonjeong (GB21), Daechu (GV14) once a day from June 16 to July 10 for 20 minutes.

5) Pharmacopuncture treatment

After checking for allergic reactions using a skin test, a single dose of 1:30,000, 0.1cc of bee-venom pharmacopuncture was performed using a single-use syringe was administered in the posterior neck and back regions, once a day from June 16 to July 10.

6) Cupping treatment

Cupping treatment was administered on posterior neck and back region once a day for 5 minutes from June 16 to July 10.

7) Herbal medicine treatment

Prescription named ‘補心瀉火清肝湯’ (Bosimsahwacheonggan-tang) was administered 2 hours after breakfast and lunch and ‘荊防瀉白散’ (Hyeongbangsabaeksan) 2 hours after each dinner (Table 2).

Table 2. Herbal Medicine Treatment

Herbal medicine	Compositions of herbal medicine
補心瀉火清肝湯 Bosimsahwacheonggan-tang	<i>Longanae arillus</i> 12g, <i>Acori graminei rhizoma</i> 8g, <i>Zizyphi spinosae semen</i> 8g, <i>Crataegi fructus</i> 8g, <i>Pinelliae rhizoma</i> 6g, <i>Citri pericarpium</i> 6g, <i>Hoelen</i> 6g, <i>Bambusae caulis in taeniam</i> 4g, <i>Aurantii immaturus fructus</i> 4g, <i>Gardeniae frutus</i> 3g, <i>Moutan radidis cortex</i> 3g, <i>Fossilia ossis mastodi</i> 3g, <i>Ostreae concha</i> 3g, <i>Glycyrrhizae radix</i> 2g, <i>Zingiberis rhizoma crudus</i> 10g, <i>Jujubae fructus</i> 6g
荊防瀉白散 Hyeongbangsabaeksan	<i>Rehmanniae radix</i> 12g, <i>Hoelen</i> 8g, <i>Alismatis rhizoma</i> 8g, <i>Anemarrhenae rhizoma</i> 4g, <i>Gypsum</i> 4g, <i>Osterici radix</i> 4g, <i>Angelicae pubescentis radix</i> 4g, <i>Schizonepetae spica</i> 4g, <i>Saposhnikoviae radix</i> 4g, <i>Arctii fructus semen</i> 2g

8) Western Medicine treatment

Western Medicine was prescribed since September, 2013. Buspirone HCl 10 milligram, *Ginkgo biloba* leaf extract 40 milligram, and nicergoline 30 milligram were administered after breakfast and dinner, ascorbic acid 1000 milligram, and toco-pherol 400 international units after every meal.

3. Assessment methods

1) Speech Mechanism Screening test (SMST)

SMST⁹⁾ consists of three categories. Among them, only the items evaluating the structure and function of the articulators were evaluated because the patient had problems with articulation. In total, 13 items for evaluating structure and 17 items for evaluating function, divided into face, lips, tongue, jaw and tooth, soft and hard palate, pharynx, and respiration area. All items were evaluated using audiovisual observation. Each item was rated on a scale of zero to two, with zero indicating 'serious abnormality', one indicating 'slightly abnormal', and two indicating 'normal'. The total score of the structural items was 26 points and the total score of the functional items was 34 points. The evaluation was performed on June 16, before treatment, and on July 8, after 23 days of treatment.

2) Tongue motility

Tongue motility evaluation was based on Bachher's seven methods¹⁰⁾ for evaluating tongue motility, to see the recovery process after partial glossectomy. Seven movements of the tongue (protrusion, to upper teeth, to palate, curl back, lateralize, elevate, and depress) were evaluated. In Bachher's research, the evaluator scored three grades of poor, fair, and excellent, but in this study, we described lengths and subjective expressions to see the specific changes in each movement. The method of measuring the protrusion distance of the tongue was based on the method of Lee et al¹¹⁾ which evaluated the tongue mobility in

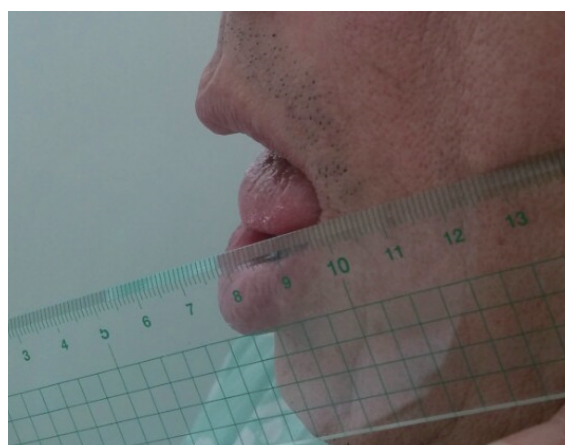


Fig. 1. Protrusion distance measured from the corner of the mouth to the tongue tip

patients with ankyloglossia. In their research, the evaluator measured the distance from the edge of the upper lip to the tip of the tongue after protruding the tongue forward as far as possible. However, the patient was unable to protrude his tongue with the mouth shut, so the distance was measured from the corner of the mouth to the tip of the tongue (Fig. 1). The evaluation was performed every 3–4 days, and the patient was placed in front of a wall in a standing posture.

3) Orbicularis oris motility

To evaluate the motility of orbicularis oris, the length between the two corners of the lips was measured and the patient was asked to encircle his mouth. The evaluation was performed every 3–4 days, and the patient was placed in front of a wall in a standing posture (Fig. 2).



Fig. 2. Measurement of the length between two corners of the lip while encircling the lips

Table 4. Measurement of Tongue Motility

Tongue motility	June 15	June 19	June 22	June 25	June 29	July 3	July 8
Protrusion	1.5cm	1.7cm	1.8cm	1.9cm	2.2cm	1.7cm	1.8cm
To upper teeth	unable	Able to touch the inner part of the upper teeth					
To palate	unable	See Fig2					
Curl back	unable	unable	unable	unable	unable	unable	unable
Lateralize	unable	Able to touch the right corner of the mouth, but unable to lateralize to the left			Able to touch the right corner of the mouth, and lateralize a little to the left		
Elevate	unable	unable	a little	a little	a little	a little	a little
Depress	unable	unable	unable	unable	unable	unable	unable

4) Speech Handicap Index (SHI)
(Appendix 1)

SHI was developed by Rinkel et al¹²⁾ and this 30-item speech questionnaire was modeled after the Voice Handicap Index (VHI) designed by Jacobson¹³⁾ It consists of 14 items of speech domain, 14 of psycho-social domain, and two other items. The answer to each question is to choose between zero (no), one (few), two (occasionally), three (often), and four (always). The higher the score, the higher is the discomfort felt by the patient. It is a questionnaire that can help understand the discomfort of and changes in patients with speech disorders.

4. Assessment results

1) Speech Mechanism Screening test (SMST)

In the initial evaluation, pronunciation and accuracy were decreased due to the problems of the articulation. Especially, the range of the movements of the tongue (protrusion, laterality, and elevation) and lip closure were decreased. The structural score improved from 19 to 23, and the functional score improved from six to 14 (Table 3).

Table 3. Structural and functional scores of the articulators

Total score	June 16	July 8
Structure (26)	19	23
Function (34)	6	14

Lip closure time increased from within 1 minute to more than 6 minutes, and the control ability of the muscle tone of the tongue increased during protrusion.

2) Tongue motility

The protrusion distance increased from 1.5cm to 2.2cm, but this improvement was not maintained. The ability to touch the upper teeth and palate, lateralize, and elevate was absent at the initial evaluation but became possible through treatment. Curling back and depressing the tongue was impossible until the end of the treatment (Table 4, Fig. 3).

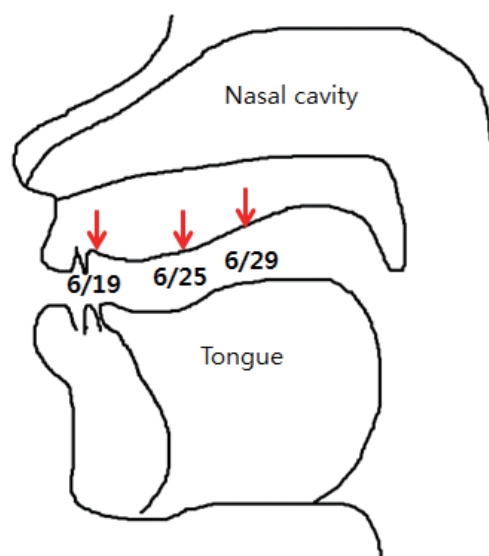


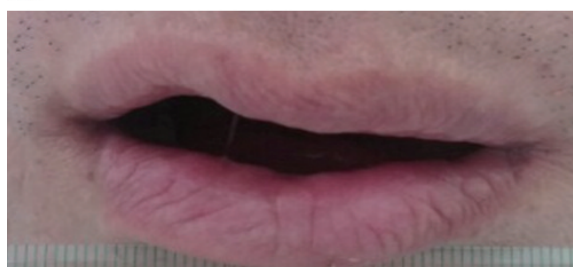
Fig. 3. The site of the tongue tip having the patient perform tongue to palate

Table 5. Length between the two corners of the lips while encircling

Orbicularis oris motility	June 15	June 19	June 22	June 25	June 29	July 3	July 8
	5.5cm	5.2cm	5cm	4.8cm	4.5cm	4.9cm	4.8cm



(A)



(B)

Fig. 4. Increased motility of orbicularis oris muscle

(A) 5.5cm at June 15

(B) 4.8cm at July 8

3) Orbicularis oris motility

The length between the two corners of the lips decreased from 5.5cm to 4.8cm, which implies improvement of the orbicularis oris (Table 5, Fig. 4).

4) Speech Handicap Index (SHI)

One point increased in the speech domain and one point decreased in the psycho-social domain. The score of the item “My speech problem causes me to lose income”, which is an objective index, increased from one point to four points, and the total score increased from 90 points to 93 points (Table 6).

5) Changes in other symptoms

Black hairy tongue, dry mouth, and general condition were improved. Breathing while lying supine was a little easier and according to the patient’s subjective expression, swallowing improved by

Table 6. Score of Speech Handicap Index

Domain	Jun 15	Jul 9
speech domain	40	41
psycho-social domain	46	45
Etc.	4	7
total score	90	93

20% due to the improvement of the tongue motility.

6) Follow up after discharge

Follow up was performed via a phone call five months after discharge. The grasping power of both hands and muscle strength of the limbs weakened and respiration worsened slightly. EMG was performed several times, but the patient was still not diagnosed of ALS.

III. Discussion

1. Diagnosis process

In neurological examination, no upper neuron signs were observed clinically. The functions of the peripheral nerves, glossopharyngeal nerve (IX), vagus nerve (X), and hypoglossal nerve (XII) were decreased. This is consistent with the characteristics of PBP. Electrodiagnostic examination showed normal EMG and NCS of the limbs and denervation potentials in craniobulbar segments, which implies that the patient had pure bulbar symptoms. Myasthenia gravis was ruled out by the results of repetitive nerve stimulation, though the patient had blurred vision and bleary eyes at the first time.

The examination results showed that the symptoms had only lower motor neuron bulbar involvement.

2. Therapeutic significance

This patient had disorders of motility of the tongue and encircling the mouth. Therefore, genioglossus, which is responsible for depressing and protruding the tongue, and orbicularis oris, which is responsible for circling the mouth, were the targets of electroacupuncture treatment. Based on the results of a study by Abdulkhakeem¹⁴⁾, which indicated that a short-time, low-frequency electrical stimulation positively affected the speed and accuracy of motor axonal regeneration, 2-hertz electroacupuncture treatment was performed for 30 minutes.

Speech therapy is recommended by speech therapists to ameliorate language impairment due to motor neuron disease¹⁵⁾. Therefore, speech therapy was performed. In addition, dysphagia treatment was administered because food intake was low and weight loss occurred due to swallowing disorder.

Decreased motility of the tongue and mouth caused excessive use of muscles in the neck and upper shoulder, resulting in increased tension. In order to relieve the muscles and to help the flow of qi and blood, acupuncture, pharmacopuncture, and cupping treatment were performed in the posterior neck and back region.

Herbal medicine bosimsahwacheonggan-tang was prescribed to relieve stress and anxiety, and hyeongbangsabaeksan was prescribed according to the soyangin constitution to cure upper body heat and black hairy tongue.

Western Medicine was administered for increasing peripheral circulation and decreasing anxiety disorders.

The patient showed increase in the structural and functional scores of the articulators in SMST, and the motility of tongue and orbicularis oris muscle after Korean Medicine Treatment. Although the emotional stability was high in the treatment process, only one point decreased in the psycho-social domain. The second evaluation was performed right after the recommendation of a neurologist that there was no possibility of im-

provement, and this might have had an effect on the score. In addition, black hairy tongue, dry mouth, and general condition improved after 25 days of treatment.

3. Disease Review (Compared to ALS)

The symptoms of PBP are similar to those of ALS, and there is controversy regarding the classification of PBP. To be diagnosed with ALS, patients must have signs and symptoms of both upper and lower motor neuron damage that cannot be attributed to other causes. Unless patients with PBP have evidence of both upper and lower motor neuron signs, they are not considered as probable ALS according to the El Escorial criteria (EEC)¹⁶⁾. Patients with only lower motor neuron bulbar involvement are hard to classify.

This patient could not be diagnosed as having ALS because there was no upper motor neuron injury or limb muscle weakness. However, some neurologists consider PBP as a subset of ALS that always evolves into ALS after a few months or more¹⁷⁾. In a study by Chafic et al¹⁾, they reviewed patients with PBP who had normal limb EMG and only pure bulbar symptoms, and found that almost all patients with PBP developed into ALS.

Due to the potential for PBP to develop into ALS, it is important to make an early diagnosis and appropriate rehabilitation for patients with only bulbar palsy. This is also emphasized in two case reports^{3,4)} of PBP.

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Appendix 1. Speech Handicap Index (SHI)

Item

1. **My speech makes it difficult for people to understand me**
2. I run out of air when I speak
3. **The intelligibility of my speech varies throughout the day**
4. My speech makes me feel incompetent
5. **People ask me why I'm hard to understand**
6. **I feel annoyed when people ask me to repeat**
7. I avoid using the phone
8. I'm tense when talking to others because of my speech
9. **My articulation is unclear**
10. **People have difficulty understanding me in a noisy room**
11. I tend to avoid groups of people because of my speech
12. People seem irritated with my speech
13. **People ask me to repeat myself when speaking face-to-face**
14. I speak with friends and neighbors or relatives less often because of my speech
15. **I feel as though I have to strain to speak**
16. I find other people don't understand my speaking problem
17. My speaking difficulties restrict my personal and social life
18. **The intelligibility is unpredictable**
19. I feel left out of conversations because of my speech
20. **I use a great deal of effort to speak**
21. **My speech is worse in the evening**
22. My speech problem causes me to lose income *
23. I try to change my speech to sound different *
24. My speech problem upsets me
25. I am less outgoing because of my speech problem
26. **My family has difficulty understanding me when I call them throughout the house**
27. My speech makes me feel handicapped
28. **I have difficulties to continue a conversation because of my speech**
29. I feel embarrassed when people ask me to repeat
30. I'm ashamed of my speech problem

How do you rate your own speech at this moment please circle the right answer?

Excellent(0) Good(30) Average(70) Bad(100)

Questions in **bold** are the one used to calculate scores of speech domain

Questions not in bold (except with*) were used to calculate scores of psycho-social domain
