

Case Report

Rehabilitation practice for external ophthalmoplegia including voluntary training for patients with medial longitudinal fasciculus syndrome

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ABSTRACT

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Introduction: This report presents a case of external ophthalmoplegia caused by medial longitudinal fasciculus (MLF) syndrome. The patient underwent oculomotor rehabilitation by an occupational therapist during hospitalization and voluntary training supervised by the occupational therapist after discharge.

Case: The patient presented with MLF syndrome due to bridge infarction. The left eye had a pronounced adduction disorder, and diplopia was observed in the median vision, resulting in severe discomfort in daily life. During the hospitalization, the patient underwent eye movement rehabilitation led by an occupational therapist that included pursuit, fixation, saccades, and convergence, and after discharge from the hospital, the patient underwent two sets of voluntary training for 10 min daily to induce pursuit, fixation, and convergence under the guidance of the occupational therapist. As a result, the angle of squint, degree of diplopia, and degree of inconvenience in daily life improved.

Discussion: Eye movement rehabilitation, including voluntary training, improved external ophthalmoplegia.

Key words: external ophthalmoplegia, MLF syndrome, voluntary training, rehabilitation

Introduction

External ophthalmoplegia affects patients' level of independence and requires intervention by rehabilitation professionals [1]. In reports on rehabilitation for external ophthalmoplegia, the usefulness of ocular movement rehabilitation using a computer program [2–4], and repetitive facilitative therapy, pursuit, and convergence by the labyrinthine eye reflex of Kawahira et al. [5] have been reported [6, 7]. The authors developed an eye movement rehearsal program that does not require a special environment and is easy to implement in clinical practice and reported that it is effective in improving external ophthalmoplegia in patients with brain injury [8, 9]. However, no previous report evaluated the effectiveness of voluntary training for patients.

This report presents the positive results of eye movement rehearsal by an occupational therapist during hospitalization and voluntary training by an occupational therapist after discharge from the hospital for a patient who presented with external ophthalmoplegia due to MLF syndrome. This study was approved by the ethics committee of the institution to which the patient belonged (date of approval: July 24, 2019, No. 477), and written informed consent was obtained from the patient.

Case

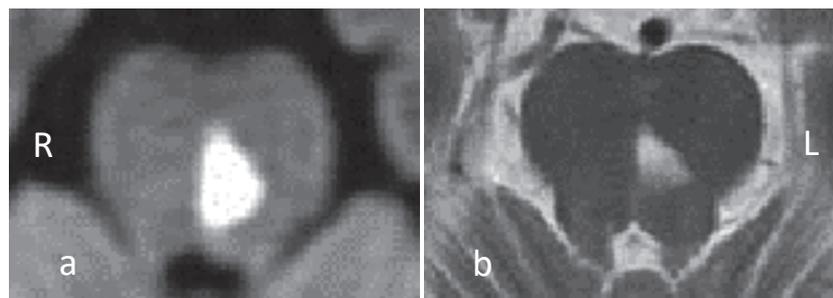
A right-handed man in his 60s was diagnosed with brainstem infarction (left median and dorsal bridge), and the disability was medial longitudinal fasciculus (MLF) syndrome. The patient had a history of double vision, and magnetic resonance imaging (MRI) revealed brain stem infarction (Figure 1a). He was admitted to our hospital on the same day, treated conservatively, and started occupational therapy on

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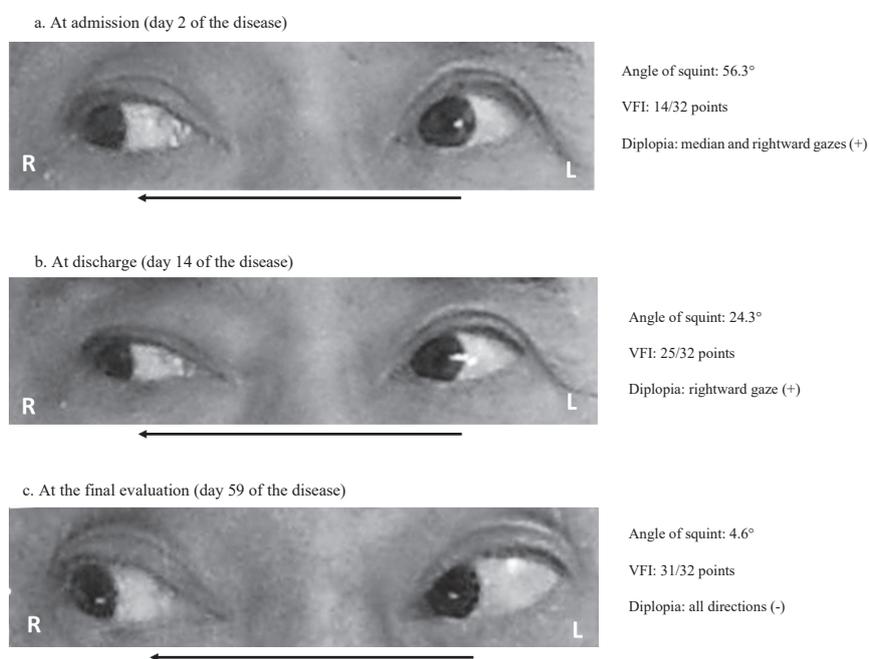


MRI, magnetic resonance imaging.

a. Diffusion-weighted magnetic resonance imaging (MRI) on the day of disease onset showed a high-signal area from the midline of the left bridge to the dorsal side.

b: MRI (T2-weighted image) on day 58 of the disease showed a high-signal area in the midline of the left bridge.

Figure 1. Brain imaging findings of the patient.



VFI, Visual Function Index.

Figure 2. Changes in external eye muscle paralysis over time.

day 2 after disease onset. He had a history of hypertension and atrial fibrillation, lived with his wife only, performed activities of daily living (ADLs) independently, and engaged in many hobbies, such as music and blogging. The patient's chief complaint was "double vision" and he strongly wanted to improve diplopia.

Inpatient evaluation

Consciousness was clear, and there was no dizziness at rest or during movement. He had no motor paralysis, sensory impairment, or dysphagia and scored 29/30 on the Hasegawa Disability Scale. The patient's ADLs

were generally at watchful waiting level at the time of admission but became independent indoors on day 7 of the disease.

In the evaluation of eye movement disorder, the left eye was mildly exotropic in the frontal view. The left eye had pronounced adduction disorder, and diplopia was observed during the median and rightward gazes (Figure 2a). The right eye showed nystagmus during abduction, and the convergence reflex was preserved. To quantify the left eye adduction disorder, the angle of squint was measured during a rightward gaze (left eye adduction). The angle of squint is generally considered normal when it is within 5° . A video camera was set up at 50 cm in front of the patient's face, and the movements

of both eyes during the rightward gaze were filmed. The video was analyzed using the HAS-X Viewer analysis software (developed by DITECT Corporation), and the maximum distance of the rightward gaze was measured. The obtained moving distance was converted from distance to angle using the Hirschberg method [10], and the value of the angle of squint was calculated. The angle of squint was 56.3° on day 2 of the disease.

The authors translated the Visual Function Index (VFI) [11], which has been reported to be useful in assessing the degree of inconvenience in the lives of patients with external ophthalmoplegia, and used it as a test. In the present study, the patient felt strong discomfort in his daily life with a score of 14/32 points for 8 items, excluding the items he does not currently perform, such as driving and cooking (Table 1).

Intervention and progress

1. Eye movement rehabilitation program during hospitalization

A total of 10 interventions were performed by an occupational therapist from days 2 to 14 of the disease after discharge, excluding holidays. The intervention during the hospitalization was an eye movement rehearsal program designed by the authors. This program was developed based on the results of a systematic review [12] of eye movement rehearsal in patients with brain injury and discussions with experts [8]. The content was as follows: the therapist showed a target object 30 cm in front of the patient while in a sitting-up-in-bed position and performed (1) 3 sets of pursuits of the target object in all 8 directions at a speed that the patient could follow, (2) 3 sets of 10-s fixations at the final range of motion in all 8 directions, (3) 3 sets of random movement of the target object in 8 directions to induce saccades, and (4) 3 sets of 10 training sessions to stimulate convergence by slowly bringing the target closer to the patient. During the

training, the program was conducted once for 20 min, and massages around the ocular muscles were performed before and after taking appropriate breaks (Table 2). At the time of discharge from our hospital, the angle of squint was reduced to 24.3°, and the VFI had improved to 25/32 points. The median diplopia had disappeared, but the rightward diplopia remained (Figure 2b).

2. Voluntary training for eye movement rehearsal after discharge from the hospital

After hospital discharge, no outpatient rehearsal was prescribed. Therefore, an occupational therapist prepared a paper-based self-directed training program for eye exercises that could be performed at home and instructed the patient for about 10 min during the last intervention in the hospital. When considering the content of the training, we kept in mind that the training should expand the range of motion of the left eye's adduction, improve the diplopia of the right eye, and increase the load without causing discomfort or fatigue. In the seated position without moving the head and neck, the following exercises were performed: (1) 3 sets of tracking the left eye to the maximum adduction position by blocking the right eye and using the patient's finger as a reference to position his left eye at the maximum adduction position for 10 s, (2) 3 sets of visual fixation of the left eye at the maximum adduction position for 10 s by blocking the right eye and using the patient's own finger as a reference, and (3) 3 sets of convergence for 10 s by the patient moving his finger close to his face at a fast speed in binocular vision. The program was included in the hospitalization program. Induction of saccades included in the inpatient program was excluded because of the difficulty of self-induction. The patient was asked to take breaks as needed while paying attention to the onset of dizziness and to perform self-massage of the orbicularis oculi muscle

Table 1. Changes in Visual Function Index over time.

Item	At admission (day 2 of the disease)	At discharge (day 14 of the disease)	At the final evaluation (day 59 of the disease)
Write letters	2	3	4
Recognize a person's face	3	4	4
Watch TV	1	3	4
Recognize steps and stairs	2	4	4
Perform detailed tasks	1	2	4
Read large characters	2	4	4
Read normal-sized letters	2	3	4
Read small print	1	2	3
Total	14	25	31

Rate each item on the following 5-point scale.

0, Cannot do it at all; 1, Severely inconvenient; 2, Moderately inconvenient; 3, Mildly inconvenient; 4, No inconvenience.

Table 2. Oculomotor rehabilitation program.

	Programs during hospitalization	Self-directed training after discharge
Implementation time	Days 2–14 of the disease	Days 15–59 of the disease
Frequency	20 min each time, 5 times per week	Two sets per day, 7 times weekly (generally within 10 min each time)
Posture	Supine	Sitting posture
Content	<ol style="list-style-type: none"> 1. Pursuit training Encourage the pursuit of objects placed 30 cm above the subject’s face. 2. Fixation training The subject is made to fixate at the maximum range of motion, and maintains that position for 10 s. 3. Convergence training A target object is positioned 30 cm above the subject’s face and is moved at a fast speed close to the face to stimulate the convergence reflex. 4. Saccade training The target object is positioned outside the subject’s field of view, and the subject is made to look at the target object rapidly. 	<ol style="list-style-type: none"> 1. Training to make the left eye look in the direction of adduction The patient blocks the right eye and follows the left eye to the maximum adduction position using his own finger as a marker for 10 s × 3 sets. 2. Fixation training at maximum adduction of the left eye The right eye is blocked, and the left eye is fixed at its maximum adduction range using the finger as a marker for 10 s × 3 sets. 3. Convergence training In binocular vision, the subject moves his or her fingers close to the face at a fast rate and converges for 10 s × 3 sets.
Remarks	<p>*In-hospital trainings 1, 2, and 4 are performed randomly in all 8 directions, including the oblique direction, in the following order: right eye blocking, left eye blocking, and binocular vision. Each set is performed in 8 directions × 3 sets.</p> <p>*Training 3 is performed in binocular vision, 10 times × 3 sets.</p> <p>*Massage of the orbicularis oculi muscle is performed before and after training.</p> <p>Breaks are taken as needed, while monitoring for the presence of dizziness and fatigue.</p>	

before and after training (Table 2). The abovementioned self-directed training sessions were completed twice daily with one training session being completed within 10 min and continued from day 15 to day 59 of the disease. No adverse events, such as dizziness and mood discomfort, were observed. The patient reported that the training made his eyes more mobile.

MR images at the final evaluation are shown in Figure 1b. The angle of squint was reduced to 4.6°, and the VFI had improved to 31/32 points. The diplopia had disappeared in both median and right lateral views (Figure 2c). The patient became independent in performing ADLs, including outdoor activities, and resumed his hobbies of playing music and blogging.

Discussion

MLF syndrome is a transient phenomenon in cerebrovascular disease and often spontaneously resolves with time [13]. Conversely, when there is direct

damage to the dorsal bridge, external ophthalmoplegia may remain as a sequela [14]. In this case, spontaneous healing was expected because of the acute stage of the injury, but there was a possibility that the injury was on the dorsal side of the bridge and that the external ophthalmoplegia would remain as a sequela. Because the patient strongly desired improvement of the external ophthalmoplegia, we did not use compensatory measures but actively introduced oculomotor rehearsal from the early stage of the disease. Finally, the diplopia disappeared, and the degree of inconvenience in daily life markedly improved; we believe that our intervention policy was appropriate.

This eye movement rehearsal program implemented during hospitalization has been reported to improve the angle of squint, degree of diplopia, and degree of inconvenience in daily life of patients with brain injury in the recovery period [9]. In this case, the angle of squint of the left eye in the adduction direction was improved, diplopia disappeared in the median vision,

and the degree of inconvenience in daily life was improved by the intervention during hospitalization. Since this program is a 20-min daily training session, it is highly likely that the patient will not be able to perform it until the horizontal oculomotor neural mechanism has been reconstructed and strengthened. The improvement of the angle of squint and diplopia was caused by the shortening of the external rectus muscle and atrophy of the internal rectus muscle in the left eye.

Self-directed training is necessary to maintain the therapeutic effects of interventions by therapists and acquire new functions and abilities over a long time. Reports on the usefulness of self-directed training in patients with brain injury have described the importance of self-directed training for improving physical functions and abilities, such as interventions using the transfer package for upper limb functions [15] and examined the guidelines for voluntary gait training [16]. Conversely, there has been no previous report on self-directed training in eye movement rehabilitation, and the practical report of this case is a new finding. In this study, we developed a self-directed training program with the principle that it should be performed < 10 min daily to avoid discomfort or fatigue. As a result, we did not observe any of the abovementioned findings and were able to conduct the training. Unlike the limbs, the eyes are always active during wakefulness. Fatigue due to training may cause significant risks in daily life; therefore, the content of training should be carefully considered. Conversely, there is a possibility that the time to improvement can be shortened by increasing the amount of load. The program during the hospitalization included eye movements in all 8 directions, including those in the direction of paralysis, based on previous studies [8, 9]. However, the self-directed training focused only on the direction of paralysis, and the results were favorable. Programs performed during the hospital stay may also yield more efficient interventions if implemented only in the paralysis direction. Based on the results of this case, it is necessary to accumulate cases in the future and study the load and content of the program.

In patients with no paralysis of the limbs and no problems with higher brain functions, such as this case, external ophthalmoplegia is often treated as a follow-up, and outpatient rehabilitation is difficult to continue. We believe that the findings obtained in this study will be helpful for such patients.

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