

博士学位論文

眼窩吹き抜け骨折の術後残存複視に対する
視能訓練の効果

近畿大学大学院
医学研究科医学系専攻
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Doctoral Dissertation

**Effects of Orthoptic Training for Residual Diplopia after
Surgical Repair of Blowout Fractures**

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同意書

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論文題目

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Effects of orthoptic training for residual diplopia after surgical repair of blowout fractures

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Abstract

Purpose To investigate the effects of orthoptic training for residual diplopia after blowout fracture (BOF) surgery.

Methods We retrospectively reviewed the medical records of 14 (average age, 22.9 ± 13.1 years) patients with residual diplopia, who had undergone orthoptic training after BOF surgery at the Department of Ophthalmology, Kindai University Hospital, between August 2013 and September 2019. The orthoptic training included exercises for eye movement, convergence, and fusional area expansion. We assessed the training effects by scoring patients' Hess screen (Hess) test results and fields of binocular single vision (BSV). The scores obtained before/after surgery and after training were compared. We also investigated the factors that influenced patients' BSV scores after training. $p < 0.05$ was considered statistically significant.

Results The respective pre- and postoperative and after-training average scores were 7.0 ± 5.3 , 5.4 ± 4.3 , and 2.5 ± 3.2 points for Hess and 50.0 ± 41.3 , 48.2 ± 35.9 , and 89.4 ± 14.0 points for BSV. Neither Hess nor BSV score showed a significant difference before and after surgery ($p > 0.05$, the Steel–Dwass test). Compared to the postoperative (i.e., before training) scores, both Hess and BSV scores significantly improved after training ($p < 0.05$ for Hess, $p < 0.01$ for BSV; the Steel–Dwass test).

Conclusion Orthoptic training appeared effective in resolving residual diplopia after BOF surgery by improving patients' ocular motility and expanding the BSV field.

Keywords Blowout fracture · Orthoptic training · Diplopia · Ocular motility · Field of binocular single vision

Key messages

- Even after orbital surgery, new or residual diplopia occurs in 37%–52% of the patients with blowout fractures. Besides observation, strabismus surgery, prism therapy, and orthoptics are the treatment options for residual diplopia.
- We investigate the effects of orthoptic training in patients with residual diplopia after surgical repair of BOF by comparing patients' Hess and BSV scores at 3 time points: before and after orbital surgery, and after orthoptic training. While about half of the patients could not obtain improved Hess and BSV scores after surgery, both scores were improved and diplopia disappeared in all the patients after orthoptic training.
- Orthoptic training following BOF surgery appears effective in resolving postoperative diplopia and ocular motility limitations that could not be achieved by surgery alone.

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Introduction

Blowout fracture (BOF) is a condition in which the intraorbital pressure rises due to a blunt trauma on the eye and results in distorted orbital wall and fractures. The consequent herniation of adipose and soft tissues such as the extraocular muscles (EOMs) and connective weave into the paranasal sinuses causes mechanical restrictions of eye movements [1, 2]. While management of BOF and timing of surgery remain controversial, general consensus suggests persistent diplopia as one of the indications for surgical intervention [3]. The incidence rate of diplopia in BOF varies from 36 to 86% [4]. Diplopia narrows patient's field of binocular single vision (BSV), and impairment of activities of daily living (ADL) occurs [5]. Reportedly, the severity of patient's ADL impairment is associated with the size of the BSV field, direction of ocular motility limitations, and presence of primary gaze diplopia [6]. Therefore, treatment for BOF aims for a wide field of BSV in the primary eye position besides orbital reconstruction.

Even after orbital reconstruction, new or residual diplopia could still affect 37–52% of patients [7]. Besides observation, measures for diplopia include strabismus surgery and conservative treatments such as prism and orthoptic therapy. Although the choice between observation and surgery for diplopia has been previously discussed [8], to our knowledge, the potential therapeutic effects of orthoptic training performed following BOF surgery have not been investigated. Surgical repair combined with orthoptic training could provide a treatment modality that leads to early acquisition of a wide field of BSV and thus better improves patients' impairment of ADL without further delay and complications.

In this study, we aimed to assess the effects of orthoptic training performed after surgical repair of BOF and to investigate the factors in patients' BSV scores after training.

Materials and methods

Subjects

After obtaining approval from the Kindai University Faculty of Medicine Institutional Review Board, we retrospectively reviewed the medical records of 14 patients (11 males; age range, 12–57 years, average, 22.9 ± 13.1 years) who had received orthoptic training after BOF surgery between August 2013 and September 2019 at our department. Table 1 shows the patients' characteristics. Patients with postoperative diplopia and ocular motility disorders

confirmed using a cover test, self-reported disturbances in daily activities, and strong willingness for training were qualified for orthoptic training. In addition, all the included patients had reconstructed fractures confirmed by computed tomography and no EOM incarceration. Depending on the severity of patients' motility limitations, orthoptic training for eye movements, convergence, and fusional area expansion was performed. Training ended when the patient acquired 30° BSV in all directions or when the patient claimed no more disturbances in daily activities.

The orthoptic training

I. Eye movement exercise

In this exercise, the head was in a fixed position, and the unaffected eye was occluded. The affected eye was examined in a distance of 50 cm for its range of ductions. An accommodative target was presented slightly inside the measured range, and saccadic eye movements in 8 directions (up, down, left, right, and oblique) were performed. The exercise lasted for 5 min. The movement in the direction showing strong restriction was reinforced with repeated exercises.

II. Convergence exercise

This exercise used a convergence training card with a series of dots lining in the center of the card. The patient was first asked to find a position in which he or she could converge easily and to hold one end of the training card to his or her nose. The patient was then told to look along the line at the dot furthest away and to fixate on the dot. The patient was reminded that he or she should be seeing two lines join at the dot before moving the focus to the next dot which was one dot closer to him or her. Like this, the patient would fixate on the closer dots in sequence. The patient was instructed to change the focus back to the previous dot if he or she saw the dot double. After fixating on the dot closest to his or her nose, the patient continued to change the focus back to the dot one further away. When the patient completed fixating on the dot furthest away, 1 round of the exercise was completed. One exercise session usually had 5 rounds, but it could be flexible depending on the patient's condition.

III. Exercise for fusional area expansion

In this exercise, an accommodative target was presented at a viewing distance of 50 cm. After confirming fusion in the primary position, the trainer moved the target slowly toward the direction showing anomalous BSV with caution given not to blur the target. If patient's fusion broke (diplopia occurred), the target would be moved back into the range

Table 1 Patients' characteristics and clinical data

Patient	Age(y)	Gender	Injury cause	Fracture location	Orthoptic exercises	Training period (days)	Period between injury and surgery (days)	Period between surgery and training (days)	Preoperative HESS score	Postoperative HESS score	HESS score after training	Preoperative BSV score	Postoperative BSV score	BSV score after training
1	20	M	Traffic	Floor/medial	Saccadic eye movement	20	97	6	14	14	11	30	0	50
2	36	M	Daily activity/insider	Floor	Saccadic eye movement Fusional area expansion	21	7	1	7	8	6	80	73	86
3	12	F	Sports	Floor	Saccadic eye movement Convergence Fusional area expansion	48	11	8	6	0	0	60	82	98
4	44	M	Fall/Slip	Floor	Saccadic eye movement Convergence Fusional area expansion	18	9	7	7	7	4	0	51	68
5	57	M	Assault	Floor	Saccadic eye movement Convergence Fusional area expansion	34	9	6	4	7	3	91	86	94
6	17	F	Fall/Slip	Floor	Saccadic eye movement Convergence Fusional area expansion	35	44	2	2	2	0	81	87	98

Table 1 (continued)

Patient	Age(y)	Gender	Injury cause	Fracture location	Orthoptic exercises	Training period (days)	Period between injury and surgery (days)	Period between surgery and training (days)	Preoperative HESS score	Postoperative HESS score	HESS score after training	Preoperative BSV score	Postoperative BSV score	BSV score after training
7	27	M	Sports	Floor	Saccadic eye movement Convergence Fusional area expansion	47	16	3	2	3	1	95	73	95
8	12	M	Sports	Floor	Saccadic eye movement smooth pursuit eye movement Convergence Fusional area expansion	52	1	1	18	10	3	0	0	94
9	17	M	Sports	Floor	Saccadic eye movement Convergence Fusional area expansion	26	9	3	3	4	1	92	0	96
10	18	F	Traffic	Floor	Saccadic eye movement Convergence Fusional area expansion	19	9	2	0	0	0	100	94	100

Table 1 (continued)

Patient	Age(y)	Gender	Injury cause	Fracture location	Orthoptic exercises	Training period (days)	Period between injury and surgery (days)	Period between surgery and training (days)	Preoperative HESS score	Postoperative HESS score	HESS score after training	Preoperative BSV score	Postoperative BSV score	BSV score after training
11	16	M	Sports	Floor	Saccadic eye movement Convergence Fusional area expansion	65	0	12	10	1	1	0	22	87
12	15	M	Daily activity insider	Floor	Saccadic eye movement Convergence Fusional area expansion	36	9	2	14	9	5	0	41	90
13	14	M	Sports	Floor	Saccadic eye movement Convergence Fusional area expansion	24	2	11	8	8	0	9	9	95
14	15	M	Sports	Floor	Saccadic eye movement Convergence Fusional area expansion	24	6	5	3	2	0	63	57	100

where BSV was possible, and the patient maintained and stabilized the fusion for 5 to 10 s. The fusional area could be enlarged with repeated exercises. At the end of the exercise, the eye returned to the primary position with stabilized fusion. Each exercise lasted for 3 min, and the duration could be flexible depending on the patient's condition.

Evaluating the effects of the orthoptic training

To evaluate the effects of the orthoptic training, modified scoring methods for patients' Hess screen test results and fields of BSV were employed [9]. The former assessed ocular motility limitations and the later quantified diplopia. Patients' Hess and BSV scores were obtained and compared at 3 time points: before surgery, after surgery (i.e., before training), and after training.

Scoring the Hess result

The Hess screen test records the direction and amount of muscle limitations. The recorded underactions of the 6 EOMs from the normal 30° range on the chart were scored for the affected eye. Each square that represented 5° on the chart was scored as 1 point, and the sum of the 6 scores for the 6 underacting EOMs represented the patient's Hess score. Figure 1 shows the Hess score calculation for patient no. 2.

Scoring the BSV field

In this study, we used the Bagolini striated glasses test to determine patient's fusional range. Wearing the Bagolini striated glasses, the patient was first asked to describe the fusion pattern in the primary position. After confirming the primary gaze fusion, the examiner then directed the light to the right, left, up, down, and oblique directions until patient's fusion broke. The obtained fusional range was regarded as the extent of the patient's BSV field and was scored. A full range would have a score of 100 points with 20 points for primary gaze fusion and 1 point for every 5° in the 8 directions. Figure 2 shows the BSV result and score calculation for patient no. 2.

The factors influencing the BSV score after training

To determine the factors for patients' BSV scores after training, patient's age at the time of injury, the period between injury and surgery, the period between surgery and training, and the BSV score before training were investigated.

Statistical analysis

The Steel–Dwass test was used to compare the preoperative, postoperative, and after-training Hess and BSV scores. Mann–Whitney's *U* test and multiple regression analysis were respectively performed to analyze the BSV score improvement and the influential factors for the BSV score after training.

Results

The period between injury and surgery ranged from 0 to 97 days (average, 16.4 ± 24.6 ; Table 1) and 10 (71.4%) of the 14 patients underwent surgery within 10 days after injury. The period between surgery and the initiation of the training ranged from 1 to 12 days (average, 4.9 ± 3.5), and 11 (78.6%) patients started training within 7 days after surgery. The training period ranged from 18 to 65 days (average, 33.5 ± 14.0).

The Hess scores

The respective pre- and postoperative Hess scores (average) ranged from 0 to 18 (7.0 ± 5.3) and 0 to 14 (5.4 ± 4.3) points. Of 14, 5 (35.7%), 4 (28.6%), and 5 (35.7%) patients had improved, worse, and equivalent postoperative scores, respectively (Fig. 3). The difference was 2 ± 4 points, and the difference was not statistically significant ($p > -0.05$, the Steel–Dwass test). The Hess scores after orthoptic training ranged from 0 to 11 (2.5 ± 3.2) points, and 11 (78.6%) and 3 (21.4%) patients had improved and equivalent scores, respectively. A significant improvement of 3 ± 2 points after training was observed ($p < 0.05$, the Steel–Dwass test).

The BSV scores

The respective pre- and postoperative BSV scores (average) ranged from 0 to 100 (50.0 ± 41.3) and 0 to 94 (48.2 ± 35.9) points. Compared to the preoperative scores, 5 (35.7%), 7 (50.0%), and 2 (14.3%) patients respectively had improved, worse, and equivalent BSV scores after surgery (Fig. 4). The average difference was -2 ± 34 points, and the difference was not statistically significant ($p > -0.05$, the Steel–Dwass test). The BSV scores after training ranged from 50 to 100 (average, 89.4 ± 14.0) points. With the worst score at 50, all 14 (100%) patients had improved scores. A significant improvement of 41 ± 33 points was observed after training ($p < 0.01$, the Steel–Dwass test).

Eyes with BSV fields of 30° in all directions (an equivalent BSV score of about 68 points) are said to be free from ADL disturbances. In this study, 6 (42.9%) of the 14 patients

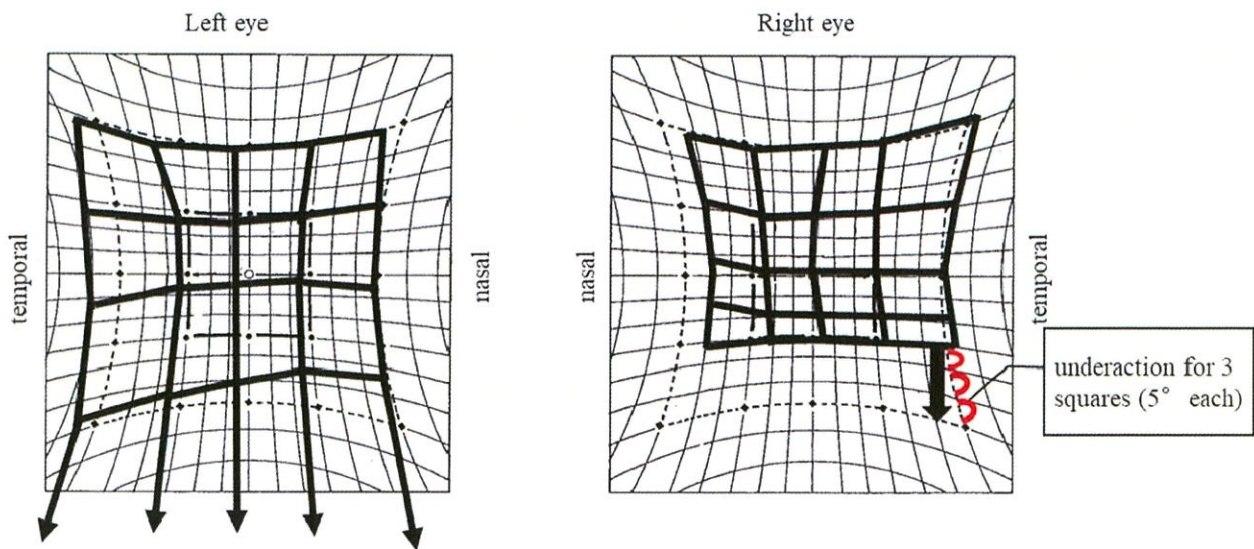


Fig. 1 Calculation of the preoperative Hess score for patient no. 2. The inferior rectus muscle and superior oblique muscle showed underaction for 3 squares (5° for each square and each square was scored as 1 point), and thus, 3 points were scored for each of the 2

EOMs. Similarly, the medial rectus muscle showed underaction for 1 square and 1 point was scored. The Hess score for patient no. 2 was $(3+3+1)$ points

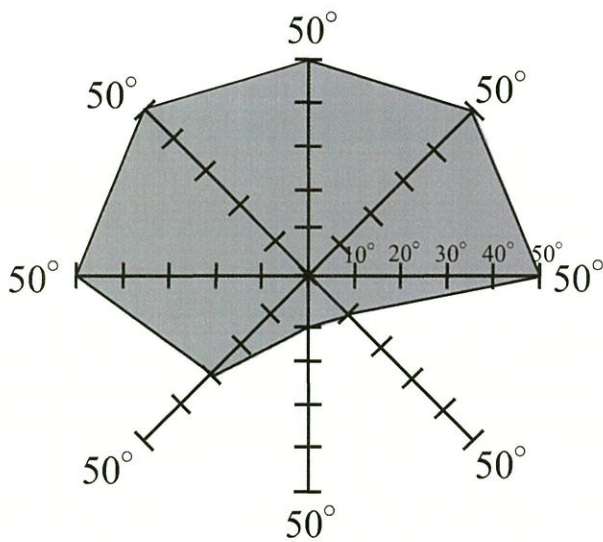


Fig. 2 The BSV score calculation for patient no. 2. The primary eye position with the most significant impact on patients' daily life was scored as 20 points. One point was scored for every 5° in the 8 directions. The field of BSV seen here was given a BSV score of 80 (Following the primary gaze, starting clockwise from up gaze: $20+10+10+10+2+2+2+6+10+10$) points

had a postoperative BSV score > 68 points (range, 73–94, average, 83.0 ± 8.3 ; Fig. 4), and an average improvement of 12.7 ± 5.8 points was further achieved after training within a period of 19–48 days (average, 34.0 ± 12.3). On the other hand, the other 8 patients with a postoperative

BSV score < 68 points (range, 0–57; average, 23.0 ± 24.0) achieved an average improvement of 62.5 ± 28.0 points after training within a period of 18–65 days (average, 33.1 ± 16.9). Compared to the patients with a BSV score > 68 before training, those with a BSV score < 68 before training could achieve a significantly greater BSV improvement after training ($p < 0.01$, Mann–Whitney's U test), and the required training period did not significantly differ between these two subgroups ($p = 0.8$, Mann–Whitney's U test). After training, all the patients had a BSV score higher than 68 points except the patient with the worse BSV score of 50 points (Table 1).

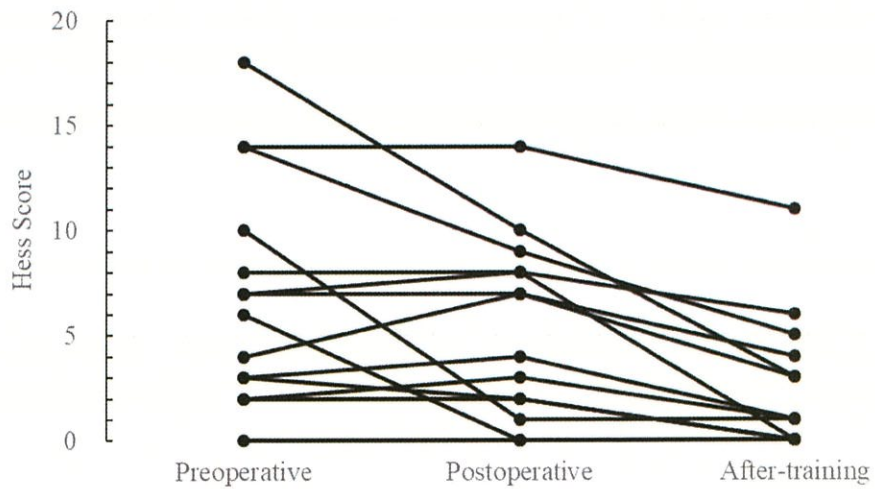
Influential factors for the BSV score after orthoptic training

Among the factors investigated (the age at the time of injury, the period between injury and surgery, the period between surgery and training, and the BSV score before training), the period between injury and surgery was found to be significantly associated with the BSV score after training. The shorter the period, the higher the after-training BSV score was ($p < 0.01$, multiple regression analysis).

Discussion

We have investigated the effects of orthoptic training in patients with residual diplopia after surgical repair of BOF. While about half of the patients could not obtain improved Hess and BSV scores after surgery, both scores were

Fig. 3 Patients' Hess scores assessed at the 3 time points. A lower Hess score indicates a less severe underaction, and 0 point indicates no abnormality in eye movement. The interval between the preoperative and postoperative points ranged from 0 to 97 days (average, 16.4 ± 26.4), and the interval between the postoperative and after-training points ranged from 21 to 77 days (average, 38.4 ± 15.8)



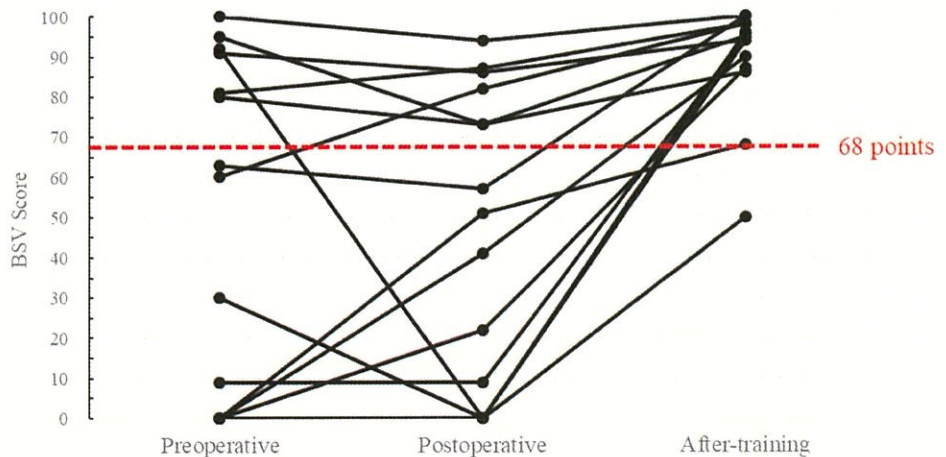
improved, and diplopia disappeared in all the patients after training. This shows the effects of postoperative orthoptic training in helping patients improve their Hess and BSV scores and resolve residual diplopia that could not be accomplished by surgery alone.

The decision for BOF surgery that aims to improve BSV and minimize tissue damages is often made based on CT findings and subjective signs such as persistent diplopia and ocular motility limitations. Early surgical interventions within 6–10 days [10] and 2 weeks [2, 11, 12] have been advocated, and surgery delay for more than 2 weeks is said to be associated with higher incidence of postoperative diplopia [7]. Particularly in pediatric patients (< 18 years of age) with BOF, surgery delay could allow the EOM damages to advance fast and result in poor prognosis of postoperative diplopia [13, 14]. In this study, 11 (78.5%) of the 14 patients underwent surgery within 2 weeks after injury, and orthoptic training was performed within 5 days after surgery in 57% of the patients. The current result also showed that the time of surgery was significantly associated with patient's BSV

score after orthoptic training. We therefore considered that the relatively early surgery and orthoptic training might have helped patients improve their effective BSV and enforce the improved fusion in daily activities, and that led to the acquisition of a broader BSV field.

Although residual diplopia may improve over time with a reported average of 5.9 months [14] and observation is often the standard of care, the symptom may be prolonged, and patients may suffer from sequelae of binocular dysfunction [15]. Loba and colleagues found that while residual diplopia after orbital surgery resolved or did not appear bothersome in the majority (58.6%) of the patients after a period of 6–12 months, significant diplopia remained in more than 30% of the patients who eventually required prism treatment or strabismus surgery [7]. Another study also reported that residual diplopia disappeared over an average period of 6.5 months after orbital surgery in 73.9% of the patients, but 17.8% of the patients required additional strabismus surgery [16]. Biesman observed that patients' ADL were interfered for at least 6 months in more than 30% of the patients who

Fig. 4 Patients' BSV scores assessed at the 3 time points. A higher BSV score indicates a wider field of BSV. A fusional area that extends to 50° in all 8 directions and primary position would score 100 points, and 0 point indicates no existing BSV. The interval between the preoperative and postoperative points ranged from 0 to 97 days (average, 16.4 ± 26.4), and the interval between the postoperative and after-training points ranged from 21 to 77 days (average, 38.4 ± 15.8)



remained diplopic after surgical repair of BOF [4]. Other studies showed that residual diplopia present in at least one field of gaze was observed in more than half of the surgical patients even after improvement was made [3], and that 80% of the pediatric patients could recover from diplopia 1 year after surgical repair [13]. Compared to these previous results, with a maximum training period of 77 days, all the patients' residual diplopia and ADL impairment disappeared in this study. Moreover, strabismus surgery or prism therapy will be necessary if residual diplopia does not spontaneously disappear. Strabismus surgery has the risk of further damaging the EOMs and causing scarring that might affect the soft tissues. Besides, predicting the desired correction with the EOMs initially damaged from the fractures becomes an even greater challenge for surgeons. As for prism therapy, Ceylan reported that diplopia in patients with slight motility disorder responds well to prism therapy [16]. Although prism therapy may be effective in resolving primary gaze diplopia, to solve the disturbances of ADL would require a broader field of BSV in all directions.

Orthoptic training is a form of visual rehabilitation using various targeted exercises. The exercises used in this study included eye movement exercise that promotes extension and contraction of the damaged EOMs to improve patient's ocular motility, convergence exercise to enhance patient's BSV aiming for a recovery to patient's BSV capacity before injury, and exercise for fusional area expansion that facilitates fusion. A fusional area that improved to 35° at down gaze after 6 months of orthoptic training has been reported [17]. In this study, both BSV and Hess scores were improved after training but not postoperatively. We considered that the training had prevented muscle adhesion with saccadic eye movements and provided continuous stimulation to the damaged EOMs. The improved ocular motility resulted in the improved Hess score. With the training, residual diplopia resolved in all the patients in this study, and no further measures were required. In patients with significant residual diplopia that seriously compromises their quality of life, non-invasive orthoptic training can help patients improve their ocular motility and quality of life without the risk of causing further complications over a relatively short period of time.

As the gold standard, diplopia is usually measured using a BSV test with a Goldmann perimeter. In this study, we used a BSV test with a Bagolini striated glasses test instead. With Goldmann perimetry, the examiner cannot confirm the state of the examinee's eye movement during measurement. Furthermore, since binocular separation is not performed, determination of the presence of suppression will be impossible and that could have some impact on the accurate assessment of BSV. We therefore used a BSV test with a Bagolini striated glasses test for quantifying diplopia.

Although the age at injury was not a significant factor for after-training BSV score in this study, 9 (64.3%) of the 14 subjects were under the age of 20. Reportedly, the incidents of BOF have been increasing among children under the age of 17 [18]. With a stronger desire for an early return to social activities, young people tended to have an aggressive attitude toward orthoptic training. This could also have contributed to the good training outcomes in this study.

This study however has some limitations including the lack of a control group treated with BOF surgery only, the retrospective nature of the study, and the lack of a uniform period for assessment of the BSV and Hess scores.

In conclusion, orthoptic training following BOF surgery appears effective in resolving postoperative diplopia and ocular motility limitations that could not be achieved by surgery alone. For patients with BOF to achieve the ultimate goal of an early return to the society, the potentials of postoperative orthoptic training that improves patients' impairment of ADL should be considered.

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Author contributions Ikumi Narita drafted and confirmed all the versions of the manuscript. Fumi Tanabe collected the data. Yukari Shiraishi interpreted the statistical analysis. Akemi Wakayama drafted the manuscript and contributed to the data analysis and interpretation. Shunji Kusaka supervised the study design and reviewed the final version of the manuscript. All the authors read and approved the final manuscript.

Data availability The datasets used and analyzed in the current study are available from the corresponding author upon reasonable request.

Code availability Not applicable.

Declarations

Ethics approval All procedures performed in this study were in accordance with the ethical standards of the Institutional Review Board of Kindai University Faculty of Medicine and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Consent to participate Verbal consent was obtained from all the patients or parents because this retrospective study contained no potentially identifying images, personal or clinical details of the participants.

Consent for publication Not applicable.

Conflict of interest The authors declare no competing interests.

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