Effect of passive limb activation by Functional Electrical Stimulation on wheelchair driving in patients with unilateral spatial neglect: A case study

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Summary  Background/Objective: Limb activation is one of the behavioural interventions to improve unilateral spatial neglect (USN). However, the effect of passive limb activation on activities of daily living (ADL) is not clear. This study examined the effect of passive limb activation by functional electrical stimulation (FES) on wheelchair driving for patients with USN, and to discuss the possibility of application of this treatment to occupational therapy.

Methods: A single subject design-baseline-intervention-baseline (ABA0), was applied to 2 stroke patients with USN. Phase A and A0 consisted of the wheelchair driving task only. Phase B consisted of the wheelchair driving task with FES. Each phase lasted for 2 weeks. The wheelchair driving task was maneuvering on a square passage in the clockwise and counter clockwise conditions for 8 minutes respectively, and four obstacles were set at each side. FES was applied to the affected forearm extensor muscles. Assessor recorded: 1) The distance participants drove wheelchair for 8 minutes, and 2) The number of collisions with obstacles and the wall, for 10 days.

Results: For one participant, the distance of maneuvering significantly increased in phase B (p < .05.), and USN on the cognitive test in the extrapersonal space indicated a tendency to improve after phase B.

Keywords  Activities of daily living; Limb activation; Unilateral spatial neglect; Wheelchair driving
Conclusion: Passive limb activation by FES improved wheelchair driving and cognitive performance for patients with USN. It can be used with instruction from occupational therapists to enhance the performance on ADL.

Introduction

Unilateral spatial neglect (USN) is defined as failure to orient to, respond to or detect stimulation in the affected side (Heilman & Valenstein, 1979). It is a common disorder occurring after the onset of stroke, and its prevalence has been estimated to be approximately 40% in patients with right hemisphere stroke (Diller & Gordon, 1981). Patients with USN have shown to have more significant limitations in activities of daily living (ADL; e.g., eating, dressing, transfers and locomotion) compared with non-neglect patients and commonly require assistance or wardens during their ADL due to their difficulties avoiding risks (Ota et al., 2011; Nijboer, van de Port, Schepers, Post, & Visser-Meily, 2013). Therefore, the improvement of USN is important for independence during the ADL of neglect patients.

Over the past 60 years, several interventions to improve USN, including visual scanning training (VST), prism adaptation (PA), and limb activation have been reported (Luaute´ et al., 2006). PA works on visuo-motor adaptation. Patients with USN wear prismatic goggles to make the patient’s sight move toward the sound side, followed by successfully performing pointing. After taking off the prismatic goggles, improvement of USN was observed during the line bisection task, the cancellation task, drawing task, and reading task (Rossetti et al., 1998), and the improvement was maintained for 2 weeks. However, few studies have reported that passive wrist extension by FES reduced USN on the visual scanning test. In addition, Harding and Riddoch (2009) also reported that FES for the affected forearm extensor reduced USN on the star cancellation test and the baking tray test. In this way, there are several reports that showed the effect of passive limb activation on cognitive tasks; however, the reports showing the effect of passive limb activation on ADL is very few. Although a few reports have suggested the relationship between the neglect on cognitive tasks and ADL, this relationship is not clear because of the variety of factors causing unilateral spatial neglect (Sasaki, Sengoku, Nakashima, Sugama, & Kitajima, 2005). The effects on ADL besides the effects on cognitive tasks are required in the assessment of USN because occupational therapists have to focus on both functional elements and ADL. In addition, USN symptoms occur on ADL in the personal space (e.g., not shaving and wearing clothes), the peripersonal space (e.g., not noticing the dishes), and the space out of reaching (extrapersonal space; e.g., not perceiving and not turning the corner of a corridor) on the affected side. Thus, the assessments for USN in the various spaces are crucial to ameliorate USN effectively.

The purpose of this study was to examine the effect of passive limb activation by FES on ADL, especially wheelchair driving for patients with USN, and to discuss the differences in the effect of passive limb activation on features of USN for application to occupational therapy.

The study protocol was approved by the Ethics Committee of Nagoya University (Ref. no: 13-608) and the Kamiida Rehabilitation Hospital.

Methods

Participants

The participants were recruited from a Rehabilitation Hospital and screened by the following inclusion and exclusion criteria. Inclusion criteria were (a) stroke patients who exhibited USN in any of the following: the line bisection task.
or the star cancellation task or the copying of a flower from subtests of the Behavioural Inattention Test (BIT; Ishiai, 1999; Wilson, Cockburn, & Halligan, 1987), (b) right cerebrovascular accident (left hemiplegia), (c) patients who could drive a manual wheelchair more than 8 minutes, (d) patients who were 1 to 8 months post the onset of stroke (recovery phase), and (e) patients who were aged 20 to 80 years old. Exclusion criteria were (a) patients with diseases for which FES is contraindicated, (b) patients who did not agree to take part in the study, and (c) patients who had scores less than 23 on the Mini-Mental State Examination (MMSE; Folstein, M.F, Folstein, S.E, & McHugh, 1975).

Three patients with stroke showing USN participated in the study. In regard to applying FES to the participants, permission was obtained from the medical doctors in charge. All participants had USN as described by the assessment mentioned in the inclusion criteria and agreed to participate in this research with written informed consent. One participant dropped out from the study due to the discharge. Characteristics of 2 participants are shown in Table 1.

### Apparatus

The IVES+® (manufactured by OG Giken Co., Ltd., Okayama, JAPAN) was used as FES apparatus. FES was applied to the affected forearm extensor muscles for inducing passive movement with the programming set interval of 5 seconds in stimulation and resting alternately. The frequency of the stimulated pulse was 20 Hz, and the intensity of the stimulation was set to produce a muscular contraction. The FES was started just before the wheelchair driving task and was continued throughout the task. During the resting time of the wheelchair driving task, the FES was stopped.

### The wheelchair maneuvering task

The driving course was set referring to earlier researches (Jacquin-Courtois, Rode, Pisella, Boisson, & Rossetti, 2008; Qiang, Sonoda, Suzuki, Okamoto, & Saitoh, 2005). The course was a square passage, with lengths of a short side of 12 m and a long side of 15 m in the hospital. Four round chairs, 450 mm in height and 380 mm in diameter, were set at each side as obstacles (Fig. 1). The shift direction of the obstacles alternated between right and left. This task was carried out in two different conditions, clockwise (CW) and counter clockwise (CCW). The participants were instructed to maneuver the wheelchair through the course as fast as possible without hitting any of the chairs and walls for 8 minutes in each trial.

| Participants | Clinical data 1 | Clinical data 2 |
|--------------|----------------|----------------|
| Age (years)  | 62             | 58             |
| Sex          | Female         | Male           |
| Time after onset (days) | 81           | 50             |
| Lesion area  | Right putamen  | Right putamen  |
| FMA-UE motor function (/66) wrist (/10) | 31/3 | 14/0 |
| MAS for the affected forearm flexor muscles | 1 | 3 |
| MMSE         | 29             | 24             |
| Side of USN  | Left           | Left           |
| In the peripersonal space | | |
| Line bisection | +            | +              |
| Star cancellation | –           | +(left side:2/right side:1) |
| Copying of a flower | +            | –              |
| In the extrapersonal space | | |
| Line bisection | +            | –              |
| Reading      | –              | –              |

+ = Neglect, – = Normal. FMA-UE = Fugl-Meyer Assessment Upper Extremity (Fugl-Meyer et al., 1975). MAS = Modified Ashworth Scale (Bohannon & Smith, 1987). USN = Unilateral spatial neglect. In the peripersonal space = on the table, In the extrapersonal space = on the white board (150 cm from the participants). Left side/right side = the number of omissions in left side/right side.
condition. In addition, participants rested for 5 minutes between the two different conditions.

If the participants went the wrong way, they were corrected. If any side of the wheelchair collided with a chair, this was counted as a collision. If the chair was moved by the collision, the assessor returned the chair to the original position after the participants passed by.

Procedures

A single system design/A baseline-intervention-baseline (ABA’) design was applied (Fig. 2). Each phase was carried out consecutively for 2 weeks, except on weekends. Phase A and phase A’ consisted of the manual wheelchair driving task only. Phase B consisted of the wheelchair driving task with limb activation using FES. When participants drove the wheelchair using sound limbs, the affected forearm extensor muscles were moved passively using FES simultaneously. During passive limb activation, the affected limb was kept on the arm rest.

Outcome measures

To examine the effect of passive limb activation by FES on wheelchair driving, the assessor followed participants and recorded the distance that participants drove the wheelchair during the 8 minutes (hereinafter referred to as the distance), and the number of collisions with obstacles and the wall (hereinafter referred to as collisions). The distance travelled was measured from the start point to the arrival point as a straight-line distance along the centre of the passage using a measuring tape.

As a reference, the following neglect outcome measure using the Behavioural Inattention Test (BIT) (Ishiai, 1999; Wilson et al., 1987) were also carried out in two different spaces to examine the participant’s characteristics of USN at the beginning and the end of each phase because USN in both the peripersonal space and the extrapersonal space affects wheelchair driving.

The line bisection task, the star cancellation task and the copying of a flower onto an A4 sheet from subtests of BIT were conducted to assess USN in the peripersonal space on the table.

The line bisection task from the BIT and the reading task were to assess USN in the extrapersonal space displayed on a white board, 150 cm from the participants. The reading task involved reading all of the letters (9 x 4 lines, 36 letters) written on a sheet and was checked for omissions. The size of the materials was modified to match for the distance from the participants.

Analysis

The data of outcome measures were plotted on a graph. The levels and slopes of the celeration line were used for

Figure 1 Wheelchair driving course and setting of the obstacles

Figure 2 The experimental procedure
visual analyses to assess the transition of outcome measures (Carter, Lubinsky, & Domholdt, 2011). Furthermore, binomial tests were conducted to evaluate significant differences in the outcome measures between phases.

Results

Distance

The results were plotted in Fig. 3. For participant 1, in Phase A, the slope of the celeration line was 2.4 in the CW condition and −2.2 in the CCW condition. In Phase B, the level increased compared with that of Phase A for both conditions. The slope also increased to 2.8 in the CW condition and 4.8 in the CCW condition. In Phase A', the level decreased compared with that of Phase B for both conditions. The slope increased to 5.2 in the CW condition and 8.6 in the CCW condition. In addition, the results of the binomial test indicated that it increased significantly for Phase B compared with Phase A (p = .002, p < .05.) and compared with Phase A' (p = .002) in both conditions.

For participant 2, in Phase A, the slope of the celeration line was 0.7 in the CW condition and 2.0 in the CCW condition. In Phase B, the level slightly increased in the CW condition, and decreased in the CCW condition compared with that of Phase A. The slope decreased to 12.5 in the CW condition and 9.3 in the CCW condition. In Phase A', the level increased compared with that of Phase B for both conditions. The slope decreased to 0.1 in the CW condition and 1.8 in the CCW condition. There was no significant difference between Phase B and the other phases.

Collisions

The results were plotted in Fig. 4. For participant 1, in Phase A, the slope of the celeration line was −1.9 in the CW condition and −1.6 in the CCW condition. In Phase B, the level decreased in the CW condition, and increased in the CCW condition compared with that of Phase A. The slope increased to 0.6 in the CW condition and −0.8 in the CCW condition. In Phase A', the level decreased compared to that of Phase B for both conditions. The slope decreased to −0.6 in the CW condition, and increased to −0.2 in the CCW condition. There was no significant difference between Phase B and the other phases.

For participant 2, in Phase A, the slope of the celeration line was 0.4 in the CW condition and −0.1 in the CCW condition. In Phase B, the level increased in the CW condition, and decreased in the CCW condition compared with that of Phase A. The slope increased to 1.2 in the CW condition and 0.0 in the CCW condition. In Phase A', the level increased compared with that of Phase B for both conditions. The slope decreased to 0.9 in the CW condition and −0.1 in the CCW condition. There was no significant difference between Phase B and the other Phases in the CW condition. On the other hand, in the CCW condition, the results of the binomial test indicated a significant decrease for Phase B compared with Phase A' (p = .02., p < .05).

The results showed that the trend of collisions particularly decreased in Phase A and stabilised in the later stage in Phase B and Phase A' for participant 1. For participant 2, collisions did not decrease during the intervention phase in
both conditions but increased for Phase A’ in the CCW condition.

**Neglect outcome measure using BIT**

The results of the neglect outcome measure are presented in Table 2. For participant 1, USN in the peripersonal space remained unchanged until the end of the Phase A’. In contrast, USN in the extrapersonal space had a trend of improvement. For participant 2, USN in the peripersonal space had a tendency to improve as time passed. However, the omissions on the star cancellation task remained unchanged in both the affected side and the sound side. This result indicated that participant 2 had neglect symptoms and general attention deficit.

**Limb activation by FES**

FES evoked contraction of wrist extensor muscles and sufficient wrist extension in participant 1, however, the evoked contraction was minimal and only slight wrist extension was noted in participant 2.

| Table 2 | The Results of the Neglect Outcome Measure Using Behavioural Inattention Test. |
|---------|---------------------------------------------------------------------------------|
| Neglect outcome measure | Participant 1 | | | Participant 2 | | | |
| | Begin A | End A | End B | End A’ | Begin A | End A | End B | End A’ |
| In the peripersonal space | | | | | | | | |
| Line bisection (%) | | | | | | | | |
| Upper | 19 | 14 | 18 | -11 | 19 | 23 | 4 | 7 |
| Middle | 34 | 40 | 10 | 23 | 7 | 23 | 1 | 10 |
| Lower | 43 | 52 | 55 | 73 | 23 | 21 | 17 | 9 |
| Copying of a flower | + | - | + | + | - | - | - | + |
| Star cancellation (Left/Right) | 0 (0/0) | 0 (0/0) | 1 (0/1) | 1 (1/0) | 3 (2/1) | 2 (1/1) | 5 (4/1) | 3 (2/1) |
| In the extrapersonal space | | | | | | | | |
| Line bisection (%) | | | | | | | | |
| Upper | -12 | 9 | 5 | 8 | 0 | 4 | 12 | 1 |
| Middle | 16 | 24 | 19 | 4 | 2 | 11 | 6 | 1 |
| Lower | 19 | 38 | 13 | 11 | 10 | 16 | 2 | 6 |
| Reading | 0 | 0 | 0 | 0 | 0 | 0 | 0 | 0 |

The results of the line bisection show a percentage of rightward displacement, copying of a flower shows the correctness of copying, and the star cancellation and reading task show the number of omissions. Begin A = Beginning of Phase A, End A/B/A’ = End of Phase A/B/A’ (Left/Right)= (the number of omissions in the left/right side).
Discussion

The purpose of this study was to examine the effect of passive limb activation by FES on wheelchair driving for patients with USN and to compare the differences in the effect of passive limb activation on various features of USN. The results suggest that passive limb activation might improve wheelchair driving in some patients with USN. Two participants had different responses, therefore, the findings should be considered individually with cautions.

For participant 1, the distance travelled significantly improved after passive limb activation was applied. This improvement was different from the learning effect because the celeration line on the intervention phase significantly increased compared with that on the baseline phase. It is inferred that passive limb activation ameliorated the asymmetric spatial representation and reduce USN, especially in the extrapersonal space related to wheelchair driving. Participant 1 might aware the obstacles and the corner of the passage in far positions and perform the wheelchair driving task more quickly. The results of participant 1 indicated that USN was found in both peripersonal and extrapersonal spaces on the neglect outcome measure, and USN in the extrapersonal space had a tendency to improve after the intervention phase. Previous research also reported that passive limb activation reduced USN on cognitive tasks in the extrapersonal space (Frassinetti, Rossi, & Ladava, 2001). Passive limb activation combined with the tasks which demand attention in the extrapersonal space might improve USN in the extrapersonal space, not only on the cognitive tasks but also on ADL such as wheelchair driving. From visual analysis, the ameliorative trend of the distance was more obvious in the CCW than the CW conditions. However, Jacquin-Courtois et al. (2008) reported that the learning effect of practicing wheelchair driving for patients with left USN is commonly more apparent in the CW (turning right) than the CCW conditions (turning left) due to the behavioural orientation deficit in the affected side. Thus, passive limb activation might also reduce the deviation of the spatial perception and response better in the CCW condition. On the other hand, collisions only improved during Phase A, regardless of passive limb activation. This result corresponds to previous research that patients with USN adjust to a route by locating the obstacles on the sound side as they go through the obstacles (Punt, Kitadono, Hulleman, Humphreys, & Riddoch, 2011). Because of this, participant 1 could reduce the number of collisions during Phase A, and an effect of passive limb activation on collisions could not be revealed in this study. Moreover, the number of collisions remained consistent during different phases while distance travelled improved during intervention phase. According to the speed-accuracy trade-off laws (Fitts, 1954), the increase in distance (i.e., the increase in speed) during the wheelchair driving task should increase the number of collisions. Hence, the increase in speed but similar frequency of collisions confirms that the performance of the wheelchair driving task was improved by passive limb activation. We also think that passive limb activation by FES might be useful to improve wheelchair driving for patients with USN in the extrapersonal space which is essential for wheelchair driving.

For participant 2, the distance travelled did not increase and the number of collisions did not decrease during the intervention phase. In other words, the performance during the wheelchair driving task was not improved by passive limb activation. Participant 2 had omissions over both affected and sound sides during the star cancellation task, probably due to a general attention deficit. According to this result, general attention deficits might prevent the improvement of performance during wheelchair driving. Previous research also indicated that patient with USN and general attention deficits wheelchair driving by limb activation (Punt et al., 2011). Moreover, Sugahara, Kamakura, and Maeda (2010) reported the importance of general attention in wheelchair driving for patients with USN. Accordingly, limb activation might not work in patients with hypergeneral attention deficits, and participant 2 did not indicate improvement during the wheelchair driving task due to general attention deficit. Furthermore, participant 2 indicated hyper tonus in flexor muscles of the affected wrist and fingers and FES only evoked slight wrist extension. In this study, limb activation was used in accordance with the hypothesis that the affected limb movements in the affected side activate the somatosensory area in the damaged hemisphere and integrate somatosensory with spatial perception in the affected side (Robertson & North, 1992). Hence, in participant 2 the slight range of passive movement by FES might not be able to activate the somatosensory and association areas in the damaged hemisphere to integrate spatial perception, and the effects of limb activation on wheelchair driving were not demonstrated in Participant 2.

As mentioned above, the effect of passive limb activation on ADL might be influenced by unique feature of USN in each individual and the range of passive limb movement in the neglect space. Additionally, the effect of it might also depend on the behavioural tasks and the spaces carrying out. Therefore, passive limb activation should be applied considering the feature of USN and the range of passive movement on the affected limb in the neglect space, the occupations and its spaces practiced. Passive limb activation might enhance the effects of the intervention on ADL, for example, if patients with USN need to go to the lavatory or the dining room, passive limb activation by FES are applied with the intervention on wheelchair driving.

This study adopted an ABA design and revealed the effects of limb activation on ADL for some patient with USN, however, the effects were not maintained until Phase A’. In previous research, limb activation was undertaken as three 45-minute sessions per day, 4 days a week for 4 weeks, and the effect of limb activation persisted for 1 month (Samuel et al., 2000). The results of this study are different from the previous study in terms of frequency and regime of intervention. Further study is necessary to examine the effect of passive limb activation by FES on various occupations client need to do in the peripersonal space, such as eating, and in the personal space, such as shaving, dressing, and getting up, etc.

We conducted this case study and discussed the effects of limb activation by FES on manual wheelchair driving for 2 participants with USN based on hypotheses in previous studies (Ladavas et al., 1997; Polanowska et al., 2009;...
Robertson & North, 1992), however, we did not compare the brain images before, after, and during the intervention, in order to highlight the individual variation.

Conclusion

This study examined the effect of passive limb activation by FES on ADL, especially for a wheelchair driving task among patients with USN. The results suggest that passive limb activation by FES could enhance the effects of the intervention on ADL.

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