INTRODUCTION

In patients with stroke, aphasia and language impairments negatively affect long-term social participation.1,2) In many cases, language dysfunction caused by aphasia recovers rapidly in the acute stage.3) Moreover, as shown in studies using functional magnetic resonance imaging (MRI), language functions change dynamically even in the chronic stage (i.e., 6 months after stroke onset) because of compensation of the right hemisphere and reactivation of the remaining region of the left hemisphere. This reorganization of language function in patients with aphasia has been proven in a study using functional MRI.4) From this theoretical background, it has been reported that intensive training improves language function and quality of life in aphasia patients in the chronic stage.5)

CASE REPORT

Constraint-induced Aphasia Therapy Improves the Use of Spoken Language and Word-finding Ability in Chronic Subcortical Aphasia: A Case Report

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Background: Constraint-induced aphasia therapy (CIAT) has been reported as a short-term, intensive language training program for improving language function in patients with chronic aphasia. We report the recovery of language function in a patient with chronic aphasia who was evaluated in the baseline assessment as having reached a plateau. Case: The patient with subcortical aphasia was a 62-year-old, right-handed man. At 192 days after left putamen hemorrhage, he visited our hospital to begin CIAT. The patient’s language and speech abilities were evaluated 1 month before and immediately before the start of CIAT. To evaluate the training effect, language function was assessed immediately after, 1 month after, 3 months after, and 6 months after the end of CIAT. The Western Aphasia Battery (WAB), the single-word-naming task in the Test of Lexical Processing in Aphasia (TLPA), and the Verbal Activity Log (VAL) were used to assess his language function and the amount of spoken language. From 1 month before CIAT to 6 months after CIAT, the WAB Aphasia Quotient increased by 6.1 points. Compared with before therapy, the errors of apraxia of speech in the TLPA disappeared from immediately after to 6 months after CIAT. Although the VAL score at 3 months after CIAT was higher than that before the start of CIAT, the score decreased after 6 months because of reduced opportunities for communication with friends. Discussion: CIAT improved the word-naming ability and amount of spontaneous, real-world spoken language in a patient with chronic aphasia.
In recent years, constraint-induced aphasia therapy (CIAT) has been reported as a short-term, intensive language training program for improving language function in patients with chronic aphasia.\textsuperscript{6,7} CIAT applies the theory of constraint-induced movement therapy (CIMT),\textsuperscript{8} and the effectiveness of this therapy has been previously reported.\textsuperscript{9} CIMT focuses on overcoming the learned nonuse resulting from damage to the central nervous system,\textsuperscript{8} and CIAT also encourages the use of speech by restraining the use of nonverbal strategies, such as gestures and writing,\textsuperscript{10} and providing situations in which patients must speak.\textsuperscript{6}

In Japan, it has been reported that the language function of patients with chronic aphasia improved after CIAT, which consisted of intensive training for 3 hours per day for 15 consecutive weekdays.\textsuperscript{11,12} Participants in these studies were patients who experienced aphasia for 6 months or longer after stroke onset, but because baseline assessments were not performed, the effects of spontaneous recovery could not be ruled out. Intensive language training improved the results of the aphasia test even in patients with chronic aphasia who had no change in language function in the pretraining baseline evaluation.\textsuperscript{13} In addition, it is necessary to investigate the training effect of the Japanese version of CIAT in cases where spontaneous recovery occurred. No studies have scrutinized the effect of the Japanese version of CIAT, which promotes the use of speech, on the improvement of word-naming ability. Here, we report the recovery of language function in a patient with chronic aphasia who was evaluated in the baseline evaluation as having reached a plateau. We show that the Japanese version of CIAT improved the patient’s word-naming ability and his level of spontaneous, real-world spoken language.

**CASE**

The patient was a 62-year-old, right-handed man who had worked as a chef. He had received 12 years of education. He was urgently admitted to the hospital because of paralysis of the right upper and lower limbs. The patient was diagnosed with left putamen hemorrhage (size: 3 × 2 cm) (Fig. 1) and underwent nonsurgical treatment. In the next hospital, he underwent a physical and language rehabilitation program for about 3.5 months and was discharged home 93 days after onset. At 192 days after onset, the patient visited our hospital to begin language rehabilitation on an outpatient basis. The patient’s Functional Independence Measure (FIM) score was 114/126 (motor FIM: 84, cognitive FIM: 30). Neurological findings included lucidity and right hemiplegia, and neuropsychological findings consisted of no nonverbal intellectual disability [Raven’s colored progressive matrices (RCPM): 31/36] and subcortical aphasia. Written informed consent was obtained from the patient to publish information in a case report.

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**Fig. 1.** Computed tomography scan showing left putamen hemorrhage.
Assessment

In the baseline evaluation, the patient’s language and speech abilities were evaluated 1 month before and immediately before the start of CIAT. To evaluate the training effect, language function was assessed immediately after the end of CIAT and at intervals of 1 month, 3 months, and 6 months after CIAT ended. Language function, in which word frequency and imageability were controlled, was assessed by the Japanese version of the Western Aphasia Battery (WAB) \(^{16}\) and the single-word-naming task in the Test of Lexical Processing in Aphasia (TLPA). \(^{15}\) The Verbal Activity Log (VAL) was used to assess the patient’s level of spontaneous real-world spoken language. \(^{16,17}\) We confirmed that the patient showed VAL scores of 3.0 points or less at 6 months or more after stroke onset and had moderate aphasia. We also confirmed the absence of the following factors: untreated serious illness, severe hearing loss, traumatic brain injury, and neurodegenerative disease.

Assessments before CIAT

The WAB Aphasia Quotient (AQ) was 75.6 at 1 month before CIAT and 77.3 immediately before CIAT. The patient’s WAB score was 65/80 at 1 month before CIAT and 70/80 immediately before CIAT. The patient’s spontaneous speech score declined, and we observed that he mainly uttered only words or two phrases when describing a picture. During word naming, once word finding became difficult for the patient, he was unable to resume speaking or answer the next question. Inconsistent articulation errors in the TLPA confirmed apraxia of speech. Given that some oral gestures, such as mouth opening, tongue protrusion, whistling gesture, blowing up cheeks, throat clearing, and tongue tapping, could be achieved upon verbal instruction, we judged that there was no oral apraxia. The patient showed good performance in yes/no questions and auditory word recognition for auditory verbal comprehension of the WAB, but the number of errors increased as the number of clauses increased in sequential commands. A previous study reported that the average change in AQ was 5.3 in patients with chronic aphasia. \(^{18}\)

Because the difference in AQ between 1 month before and immediately before CIAT was 1.7, we judged that the spontaneous recovery of language function had reached a plateau. The number of correct answers for the TLPA single-word naming was 138/200 (high frequency, 77; low frequency, 61) 1 month before and 129/200 (high frequency, 72; low frequency, 56) just before CIAT; therefore, we judged there was no recovery.

The learned nonuse of verbal language was confirmed by a reduction in spontaneous speech in daily life and scores below 2.5 on the two VAL scales. \(^{6}\)

CIAT

A Japanese version of CIAT, \(^{11,12}\) based on CIAT II, \(^{6}\) was used in this case. The therapy sessions involved the following activities:

1. Confirmation of homework and review of spontaneous real-world spoken language from the previous day (30 minutes)
2. Word repetition drills (20 minutes)
3. Phrase repetition drills (20 minutes)
4. Language card game (40 minutes)
5. Picture description (20 minutes)
6. Role-play activity (30 minutes)
7. Home skill assignment (10 minutes)

During the word and phrase repetition drills, the patient repeated as much as possible the words and phrases that were familiar to him, such as food and drinks provided at his restaurant and the tools he used at his work. These words and phrases were also used in the later role-play task. When the patient was unable to repeat or name words correctly, we encouraged him to speak the word by: (1) talking about his episodes related to the word, and (2) repeating the word and short sentences using the word many times. The difficulties of the tasks were adjusted by changing the number, type, and position of the syllables. In the language card game, the patient and the therapist were each given about ten picture cards. In an effort to accumulate pairs, the patient and therapist asked each other for a matching card by using the question: “Do you have XX (name)?” This activity required the patient (and therapist) to name the object on the card. If the opponent had the specified card, they provided the card to the questioner and then aimed to make the same pair as soon as possible. In the picture description, the patient explained a landscape painting of daily life drawn in black and white. The therapist urged the patient to explain the details of the picture following his initial overview. If the patient was unable to recall the target word, then the therapist presented the first syllable of the word and the episode associated with the word as a hint for verbal output. In the role-play activity, verbal conversation was practiced while assuming the daily situations of patients who require communication.

A “transfer package” (TP) \(^{9}\) was also introduced to transfer the therapeutic gain of CIAT to daily communication. As the first step of TP, we clarified what the patient wanted to achieve by improving speech before the start of CIAT. The patient and his wife agreed to communicate verbally.
as a daily task during the CIAT program instead of using nonverbal communication. When confirming homework, the therapist checked the patient’s schedule from the previous day and his use of speech in his daily activities. The therapist provided feedback each time and encouraged the patient to communicate by verbal language, as much as possible, using demonstrative pronouns and circumlocutions. The therapist also instructed the patient’s wife not to speak on the patient’s behalf.

The CIAT tasks were carried out for 3 hours per day, 5 days per week, for 3 weeks (45 hours). During the training, the patient was instructed not to use compensatory strategies, such as writing and nonverbal gestures, and he was encouraged to use only verbal language.

**Assessment after CIAT**

Immediately after CIAT, the WAB AQ had increased by 4.5 points from 1 month before therapy and by 2.8 points from immediately before therapy (Table 1). Immediately after CIAT and 3 months after therapy, the improvement from 1 month prior to treatment did not reach 5.3 points, which is the average change in WAB AQ in the chronic stage of aphasia.18 However, at 6 months after therapy, the WAB AQ score had increased by 6.1 points from 1 month before CIAT. Improvements were noted in auditory word recognition, sequential commands, and word-naming scores.

The number of correct answers for the TLPA immediately after CIAT increased by 10 points from 1 month before treatment and by 19 points from immediately before therapy. In particular, the number of correct answers for high-frequency words tended to increase after treatment. However, these improvements for high-frequency words had decreased at 3 and 6 months after treatment. In addition, the error type of paraphasia and the number of occurrences of apraxia of speech in the TLPA were evaluated. The criteria used to classify the paraphasia error types and apraxia of speech were based on previous reports19 as follows: 1) No response: no verbal response; 2) Phonological fragments: phonologically related to the target word; 3) Other phonological fragments: phonologically unrelated to the target word; 4) Formal paraphasia: not semantically similar but phonologically similar to the target word; 5) Mixed paraphasia: phonologically and semantically related to the target word; 6) Semantic paraphasia: semantically related to the target word; 7) Phonemic paraphasia: nonwords that are phonologically incorrect with the target word; 8) Monemic paraphasia: paraphasia consisting of two or more morphemes; 9) Irrelevant paraphasia: paraphasia that is semantically unrelated to the target word; 10) Perseverative paraphasia: paraphasia with verbal perseveration. Apraxia of speech was evaluated by nine characteristics, namely, sound substitutions, imprecise consonants, revisions, repetitions, prolongations, abnormal stress, slow rate, restricted pitch variation, and inconsistent errors.20

After CIAT, there were fewer errors in phonological fragments, semantic paraphasia, monemic paraphasia, irrelevant paraphasia, and apraxia of speech when compared with assessments 1 month before and immediately before therapy (Table 2). However, the number of errors in phonological fragments and semantic paraphasia after CIAT, monemic paraphasia after 3 and 6 months, and irrelevant paraphasia immediately after therapy only showed a one-point difference from that before CIAT; thus, there was no clear difference. Monemic paraphasia was not observed immediately after therapy but was observed again at 3 months after therapy. There was no significant change in errors of no response between before and after CIAT. However, instances of no response tended to become more frequent at 3 months after therapy when compared to immediately after CIAT. Errors of apraxia of speech were not observed immediately after CIAT.

| Table 1. Results of assessments before and after CIAT | 1 month before | Immediately before | Immediately after | 1 month after | 3 months after | 6 months after |
|------------------------------------------------------|---------------|-------------------|------------------|--------------|---------------|---------------|
| RCPM                                                 | 31/36         | -                 | -                | -            | -             | -             |
| WAB (AQ)                                             | 75.6          | 77.3              | 80.1             | -            | 80.7          | 81.7          |
| TLPA                                                 | 138/200       | 129/200           | 148/200          | -            | 141/200       | 142/200       |
| High frequency                                       | 77/100        | 72/100            | 87/100           | -            | 80/100        | 82/100        |
| Low frequency                                        | 61/100        | 56/100            | 61/100           | -            | 61/100        | 60/100        |
| VAL (patient)                                        |               |                   |                  |              |               |               |
| AS                                                   | 1.0           | 1.0               | 3.2              | 3.8          | 3.2           | 3.5           |
| HW                                                   | 1.0           | 1.0               | 3.8              | 3.8          | 3.5           | 2.1           |

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The VAL score at 3 months after CIAT was 2.5 points or higher, which is the criterion for learned nonuse of verbal language. However, the VAL at 6 months after CIAT was obviously below this level. In an interview, the patient’s wife revealed that the spread of COVID-19 had reduced the number of opportunities to go out or eat out. She also indicated that they used to spend time at home with friends before the outbreak but that their reduced number of visits had reduced opportunities for conversation.

### DISCUSSION

Although the recovery of language function in our patient had reached a plateau, CIAT contributed to improvements in word naming and the level of spontaneous real-world spoken language. To overcome learned nonuse of speech, CIAT restricts the use of nonverbal communication strategies and strongly encourages patients with aphasia to speak in therapy and daily life. Previous studies have indicated that CIAT permits participants to achieve many small, but discriminable, successes and may also increase the participant’s motivation to improve. The patient in this study, who had learned nonuse of speech, also demonstrated benefits from CIAT and may have overcome learned nonuse. This step may have improved his language ability and amount of speech.

A recent study reported that intensive language rehabilitation programs restore language function, even in patients with chronic aphasia. In Japan, the effect of CIAT on patients with chronic aphasia has been reported. Previous researchers have clarified that the language function of patients with aphasia recovers spontaneously even at 6 months after stroke onset. However, studies of the Japanese version of CIAT did not consider the effects of the spontaneous recovery of language function. Therefore, in this case, in addition to the reported evaluation protocol, we evaluated the patient’s baseline language ability to exclude the impact of spontaneous recovery on the effectiveness of CIAT. As a result, there was no obvious improvement in WAB or TLPA score in the baseline evaluation, indicating that the recovery of the patient’s language function had reached a plateau. In this case, the baseline evaluation was considered to be important in verifying the effect of the intervention.

The CIAT protocol restricts nonverbal strategies and strongly encourages a verbal strategy. This approach aims to improve the amount of speech, but no study has quantitatively clarified the functional improvement of word-naming ability in the Japanese version of CIAT. As an index of the improvement of word-naming ability, we compared the number of word-naming errors in the TLPA, which includes 200 words. The patient in this case had aphasia because of a subcortical lesion. It has been reported that various symptoms, such as apraxia of speech, comprehension impairment, repetition impairment, naming impairment, and paraphasia, are observed with subcortical lesions. There were several errors of no response, semantic paraphasia, and apraxia of speech in the TLPA, and language and speech impairments were observed in this case. Previous studies have reported that CIAT improves the naming ability in aphasia patients and the training effect is also effective in patients with apraxia.

| Type of error                   | 1 month before | Immediately before | Immediately after | 3 months after | 6 months after |
|--------------------------------|----------------|--------------------|-------------------|---------------|---------------|
| No response                    | 26 (13%)       | 42 (21%)           | 29 (14.5%)        | 37 (18.5%)    | 35 (17.5%)    |
| Phonological fragments         | 2 (1%)         | 1 (0.5%)           | 0 (0%)            | 0 (0%)        | 0 (0%)        |
| Other phonological fragments   | 0 (0%)         | 1 (0.5%)           | 0 (0%)            | 0 (0%)        | 0 (0%)        |
| Formal paraphasia              | 0 (0%)         | 1 (0.5%)           | 0 (0%)            | 0 (0%)        | 0 (0%)        |
| Mixed paraphasia               | 0 (0%)         | 0 (0%)             | 1 (0.5%)          | 0 (0%)        | 0 (0%)        |
| Semantic paraphasia            | 20 (10%)       | 23 (11.5%)         | 22 (11%)          | 19 (9.5%)     | 21 (10.5%)    |
| Phonemic paraphasia            | 0 (0%)         | 0 (0%)             | 0 (0%)            | 1 (0.5%)      | 0 (0%)        |
| Monemic paraphasia             | 4 (2%)         | 2 (1%)             | 0 (0%)            | 1 (0.5%)      | 1 (0.5%)      |
| Irrelevant paraphasia          | 3 (1.5%)       | 1 (0.5%)           | 0 (0%)            | 1 (0.5%)      | 1 (0.5%)      |
| Perseverative paraphasia       | 0 (0%)         | 0 (0%)             | 0 (0%)            | 0 (0%)        | 0 (0%)        |
| Apraxia of speecha             | 7 (3.5%)       | 8 (4%)             | 0 (0%)            | 0 (0%)        | 0 (0%)        |

Underlines show data where the number of errors is less than 1 month before CIAT and immediately before CIAT. Occurrence of errors including correct answers.
of speech. The TLPA score for high-frequency words was also improved in this case, suggesting that CIAT improves the naming ability. The number of phonological fragments, semantic paraphasia, monemic paraphasia, and irrelevant paraphasia were slightly reduced after CIAT, and their overall effects improved the TLPA score. No remarkable training effect was found for no response or semantic paraphasia after CIAT, but errors caused by apraxia of speech disappeared immediately after CIAT, and the effect was maintained until 6 months later. The effects observed in this case indicate that CIAT was an effective treatment for apraxia of speech.

Previous studies have reported that social participation is restricted in patients with aphasia in the chronic stage because of decreased communication ability. In addition, the reduction in communication opportunities by restrictions on social participation can lead patients with aphasia to learned nonuse of speech. In a previous study verifying the validity and reliability of the Japanese version of the VAL, the authors stated that the VAL score of patients with Broca’s aphasia in the chronic stage was an average of 2.47 points on the Amount scale (AS) and an average of 2.61 points on the How Well scale (HW). It is clear that many patients suffer from restricted verbal communication in daily life because of aphasia. CIAT encourages the use of speech in patients with chronic aphasia, and TP helps to generalize this progress into daily life. TP was designed to facilitate the transfer of therapeutic gains from the treatment setting to life situations. The introduction of TP into CIAT would have kept the patient’s attention on the use of speech in his daily life. These approaches contribute not only to improving language function but also to improving the amount of spontaneous real-world spoken language. In the present case, the VAL score at 3 months after CIAT was higher than before the start of CIAT, and the improvement in the VAL after CIAT was maintained. This result indicates that learned nonuse of speech would have been overcome by task selection and difficulty adjustment based on the patient’s life experiences and successes in verbal communication.

Previous studies have reported the effects of CIAT on language function and naming ability. Johnson et al. reported that CIAT did not significantly improve WAB in aphasic patients, but both the AS and HW of VAL improved, and the AS score was maintained until 6 months following CIAT. In contrast, the Japanese version of CIAT significantly improved the WAB and VAL scores after therapy. Regarding the improvement of naming ability, it has been reported that the scores of the Boston naming test improved more significantly in the group that took conventional training after CIAT than in the group that took CIAT after conventional training. In summary, CIAT provided improvements in VAL and naming ability, and the Japanese version of CIAT provided improvements in WAB and VAL. However, no previous report has analyzed the naming-error patterns and followed them up after CIAT. This report, which describes the effects of CIAT, is the first to demonstrate the relationship between improvement in WAB and VAL and improvement in word-naming errors caused by speech apraxia and elements of paraphasia.

In the current study, the VAL of the patient 6 months after CIAT was significantly lower than at 3 months after therapy. Although the previous study also reported that the VAL score gradually decreased after CIAT, the factors that contributed to this result are yet to be clarified. Improvement of apraxia of speech was maintained in this case, but instances of no response tended to increase after training. Increased frequency of no response may contribute to lower VAL scores. This result might have been affected by the spread of COVID-19, which restricted activities outside the home and reduced opportunities for communication with friends. Loss of communication opportunities because of COVID-19 restrictions could affect the language ability of patients with aphasia. An increase in the instances of no response after 3 months compared to immediately after CIAT may be attributed to the decrease in the communication opportunities as a result of TLPA. Thus, the language ability of this patient might have been affected from 3 to 6 months after therapy. In previous studies, the AS of the VAL at 6 months after CIAT was maintained. Therefore, the VAL score of this patient might have been maintained if his opportunities for speaking had not diminished. Taking into consideration the communication opportunities during the spread of COVID-19 infection, we found it necessary to provide guidance on communication activities after CIAT.

We report the case of a patient with subcortical aphasia and whose spontaneous recovery of language function had reached a plateau before his participation in CIAT. As a result, we noted improvement in word naming and the amount of speech. Future studies should investigate the effects of CIAT and maintenance of the training effects in a larger number of patients.
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CONFLICTS OF INTEREST

The authors have no conflicts of interest to declare.

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