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Left Leg Apraxia after Anterior Cerebral Artery Territory Infarction: Functional Analysis Using Single-Photon Emission Computed Tomography

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Key Words
Corpus callosum · Ideomotor apraxia · Single-photon emission computed tomography · SPECT · Subtraction ictal SPECT co-registered with MRI · SISCOM · Cerebral infarction

Abstract
Left hand apraxia is known as a unique symptom of callosal apraxia, but lower limb symptoms are rarely mentioned. We report a patient who experienced left ideomotor apraxia affecting both the upper and lower limbs after a stroke in the territory of the right anterior cerebral artery. His spontaneous gait was normal, but he was unable to move his left leg intentionally either by verbal command or by imitation. His leg symptoms gradually improved over time. We evaluated the change in cerebral blood flow in this patient using single-photon emission computed tomography. The results showed an increase in blood flow in the posterior corpus callosum; therefore, we suggested that the callosal pathway might contribute to left leg as well as left hand volitional movement.

Introduction

Previous reports have indicated that the movement of the nondominant side of the body involves bi-hemispherical events that require callosal participation [1]. Thus, corpus callosum damage is known to possibly cause left upper limb apraxia [2]. On the other hand, apraxia of the left lower limb has rarely been reported. Unlike the hands, leg functions are usually limited to walking and standing, which must be accomplished by the two legs working together.

Here, we describe a stroke patient who exhibited a left lower limb behavioral abnormality, despite having no sensory or motor deficits. We followed the clinical recovery of the patient and evaluated his cerebral blood flow using single-photon emission computed tomography (SPECT). To evaluate the cerebral blood flow, we applied a modification of novel analytic software known as subtraction ictal SPECT coregistered with MRI (SISCOM) [3].

Case Report

A 64-year-old, right-handed man was hospitalized and diagnosed as having had a cerebral infarction arising from the dissection of the anterior cerebral artery. He had a history of mild diabetes mellitus but no prior neurological problems. After the initial treatment, on the 30th day after onset, he was admitted to our hospital for rehabilitation. An MRI study showed that the responsible lesion involved the right frontal lobe, the anterior cingulate gyrus, and the corpus callosum extending from the genu to the truncus (fig. 1a, b). ^{123}I-iodoamphetamine (^{123}I-IMP) SPECT demonstrated decreased cerebral blood flow in bilateral frontal lobes, the cingulated gyrus, and the corpus callosum, with remarkable hypoperfusion especially notable in the right frontal lobes

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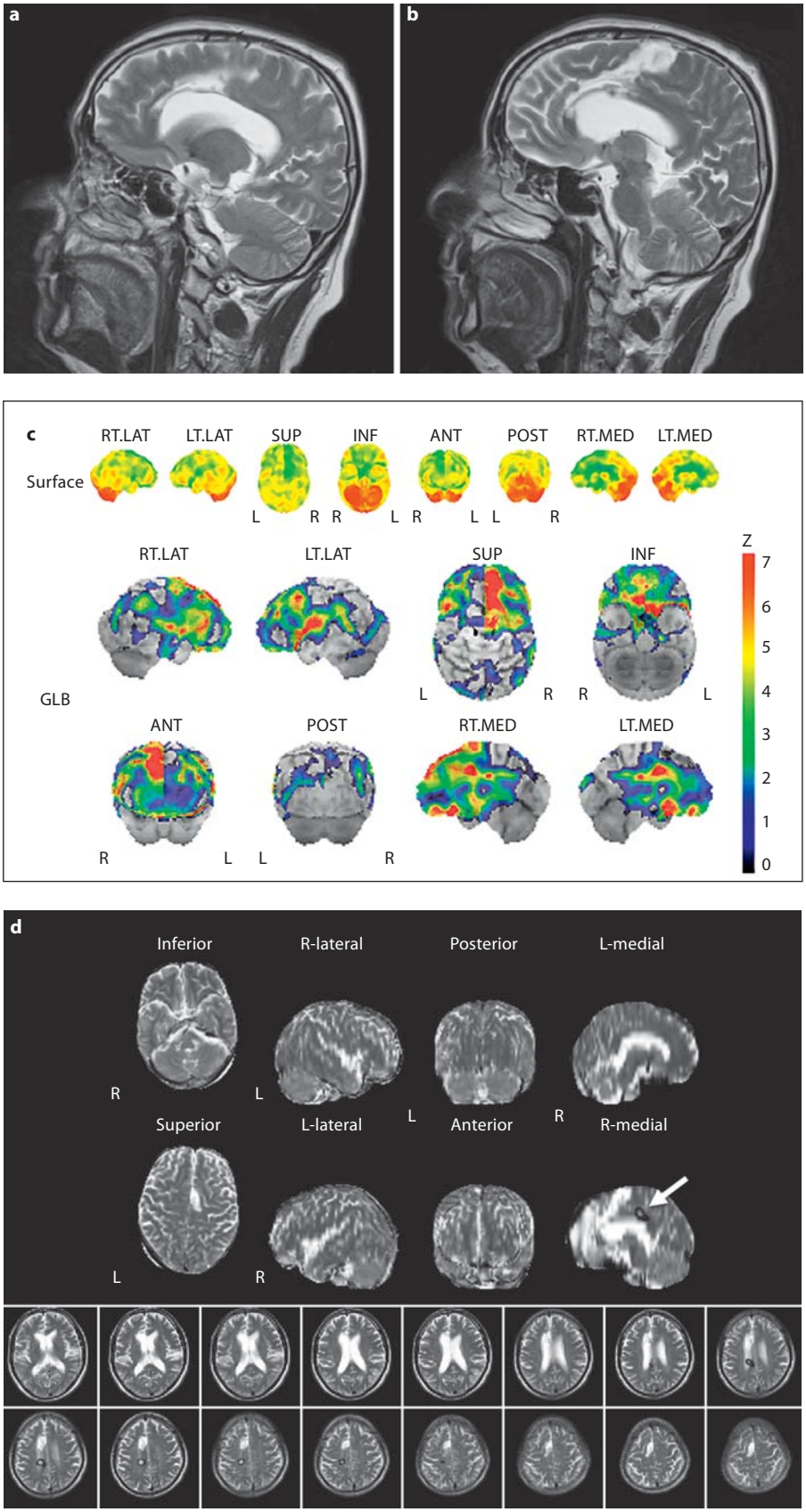

Fig. 1. a, **b** Sagittal T2-weighted magnetic resonance images show increased signals in the right frontal lobe, anterior cingulate gyrus, and corpus callosum extending from the genu to the anterior part of the truncus. **c** Brain ^{123}I-IMP SPECT at 2 months after stroke (3-dimensional stereotactic surface projection). The SPECT findings show decreased cerebral blood flow in bilateral frontal lobes, the cingulated gyrus, and the corpus callosum, with remarkable hypoperfusion in the right frontal lobes. **d** Subtraction SPECT findings coregistered with MRI results. The SPECT findings obtained 2 months after stroke onset were subtracted from the SPECT findings obtained 20 months after stroke onset. The SSCOM images demonstrated a significant increase in cerebral blood flow in the posterior truncus of the right corpus callosum.

(fig. 1c). The SPECT findings were explained by collateral vascular supply redistribution after anterior cerebral artery occlusion, as the distribution of the arterial branches was typical and no other lesions were present.

No evidence of motor or sensory disturbance in the patient's left leg was seen, and he was able to walk spontaneously. Good dorsiflexion of the left foot while climbing stairs was observed. He scored 28/30, 36/36 and 10/18 on the Mini-Mental State Examination, the Raven Colored Progressive Matrices, and the Frontal Assessment Battery, respectively. A mild grasp reflex in his left upper limb was seen. He showed clumsiness on the fist-edge-palm test in his left hand, although he could move his right hand smoothly. His clumsiness was remarkable when imitating nonsymbolic movements, but not symbolic ones. When he was asked to pantomime common objects, he did not make any errors, consistent with the classic concept of ideational apraxia, although his left arm movements were clumsy. We used the multiple object tests, such as (1) lighting a candle and (2) making a cup of Japanese tea, as these tasks require a movement sequence. All the sequences were correctly executed. In addition, his left arm never interrupted his right arm movement, and the movements of both arms were well coordinated. Behavioral Inattention Test scores (Conventional subset scores and Behavioral subset scores) were 146/146 and 79/81, respectively. Buccofacial apraxia and stuttering were absent. He had no signs of classical corpus callosum disconnection syndrome. The shapes drawn with his left hand were less precise, although he could write Japanese characters and could draw pictures using both hands. He was able to name objects placed in both his right and left visual fields. He also could identify objects, such as a comb or toothbrush, placed in either hand by palpation alone.

He was able to walk slowly, although he was unable to control the speed and direction of his movements. When he was asked to walk fast or to run, he was unable to do so. When we asked him to walk backward, he started walking forward. When he was asked to move his feet up and down, his left lower limb did not move, while his right lower limb moved correctly. When we instructed him to kick a ball with his left foot, he understood what he was supposed to do, but he hesitated and was unable to do so. Even when a response was observed, there was a considerable delay and his movements were clumsy. When he tried to put on his shoes, he was unable to lift his left foot, so he had to pick up his left shoe and put it on using his upper limbs. He tried to use a cyclic ergometer, but his left leg was unable to push the pedal. He was unable to imitate gestures, such as to extinguish a cigarette or to trace a circle on the floor with his left foot. Any efforts to concentrate attention to the left side failed to move the left lower limb. His left leg never interrupted his right leg movements during the neuropsychological evaluation or during daily living.

Conventional physical therapy and occupational therapy were provided after admission. Two months after onset, his upper limb grasp reflex disappeared. Three months after onset, he could run, walk fast and pedal a bicycle. Prior to being discharged, he was able to kick a ball without delay, although he still showed some clumsiness. Four months after onset, he started to work as a sales manager. His job required walking long distances, but he did not experience any problems. Six months after onset, his upper limb ideomotor apraxia had disappeared. After 1 year, he could put on his shoes without using his upper limbs and could kick a ball correctly and smoothly. However, some series of voluntary acts remained difficult. When he was asked to lift his left foot, he was unable to do so. He also continued to demonstrate clumsiness when imitating gestures using his left foot. A brain ^{123}I-IMP SPECT examination performed at 20 months after the stroke showed an improvement in the blood flow in the right frontal lobes, the cingulated gyrus, and the corpus callosum, although the hypoperfusion in the right frontal lobes had persisted.

Subtracted SPECT Coregistered with MRI

The SISCOM procedure is performed as follows. Ictal and interictal SPECT and T1-weighted 3D MR images are transferred to the same computer. Each patient's ictal and interictal SPECT scans are then registered to the same patient's MRI scan, using an automated image registration program (AIR). The AIR software is used to align the SPECT to the MRI scans of each subject using a 6-parameter rigid-body transformation. Before the coregistration of the SPECT and MRI, the outer scalp is removed from the MRI by applying a binary mask for the whole brain to the MRI in the manner described previously. The coregistered ictal and interictal perfusion SPECT images are then normalized according to the global mean voxel counts. Normalized ictal and interictal SPECT images are subtracted to obtain the ictal-interictal difference images. A binary mask is generated at a threshold of 35% of the maximum value for each SPECT image. The final mask is then generated from filling inner holes of implicit intersection of the two binary mask images. Mean and standard deviations of subtracted image of normalized ictal and interictal SPECT images are calculated. In the routine examinations, 2 standard deviations are selected as the Z score in the subtraction areas during the seizure and superimposed on the tomographic and surface rendering images of the patient's MRI.

In the present case, we performed the SISCOM procedure as described above. We used T2-weighted MR images instead of T1-weighted MR images to better visualize the lesion. In addition, the term SISCOM is not actually an appropriate description because this case report did not involve epilepsy. Therefore, we used the term subtracted SPECT coregistered with MRI (SSCOM), rather than SISCOM.

We performed a brain ^{123}I-IMP SPECT examination at 2 and 20 months after stroke onset and subtracted the previous SPECT findings from the latter ones. The result was then coregistered with MRI findings. The resulting SSCOM image showed a significant increase in cerebral blood flow in the posterior truncus of the corpus callosum (fig. 1d).

In the present case, a standard deviation of 2.7 on the subtraction image was regarded as indicating an area with a remarkable increase in perfusion.

Discussion

We report a patient with left inferior limb asymmetric apraxia. Unilateral motor neglect was excluded because his movements were not improved by directing the patient's attention. Since 'diagonistic apraxia' is defined as abnormal motor behavior of the left limb activated by voluntary movement of the right limb [6], his left leg motor disturbances were not consistent with di-

agonistic apraxia. His left leg symptoms featured a difficulty with movement initiation and clumsiness that seemed to arise from spatial and temporal errors, rather than a disturbance in the sequencing of a series of acts. Therefore, we think that his left leg symptoms may correspond to ideomotor apraxia, rather than ideational apraxia.

In 1907, Liepmann and Maas [4] first described left-handed apraxia in right-handed individuals as a diagnostic indicator of corpus callosum lesions. Since then, numerous reports and research on left upper limb apraxia and abnormal neural behavior have been published [5–7]. On the other hand, left lower limb symptoms have rarely been reported. We found a few case reports of lower limb manifestations in patients with corpus callosal strokes [8, 9]. In these cases, however, the infarction extended not only to the corpus callosum, but also to the cingulated gyrus, the supplementary motor area, and the medial prefrontal cortex. In addition, these reports presented only MRI findings and did not include the results of functional neuroimaging techniques. Thus, which of these brain lesions was responsible for the series of leg symptoms remained a clinical question. In the present case, the affected frontal lobe was considered to be a supplementary motor area for the left leg. Furthermore, the patient's performance on the Frontal Assessment Battery as well as his left hand forced grasping results probably indicated concomitant right frontal damage. Therefore, we investigated the recovery of apraxia over time and correlated the recovery with cerebral blood flow changes.

To compare the cerebral blood flow at different times in this patient, we used SISCOM [4]. SISCOM is a novel analytic software that is used clinically to detect epileptogenic foci, and previous research has demonstrated a high concordance. Recently, a few case reports have described the use of SISCOM for the evaluation of movement disorders [10, 11]. The correlation between SISCOM findings and stroke recovery has not been investigated. However, we think that using an analytic technique is a more objective and reliable protocol than side-by-side visual comparisons. Therefore, in this present study, we attempted to apply SISCOM for the evaluation of a stroke patient. We slightly changed the SISCOM procedure and used the term SSCOM, instead of SISCOM.

Interestingly, the SSCOM results demonstrated a significant increase in blood flow in the posterior corpus callosum. The patient also showed an improvement in his ability to kick a ball, put on his shoes, control his gait speed, and pedal a stationary bike. We assumed that the increased blood flow in the posterior corpus callosum contributed to the recovery of these movements.

Nevertheless, the patient continues to have difficulties with some kinds of volitional movements. The SSCOM findings showed no significant changes in blood flow in the anterior corpus callosum or the right frontal lobe. These findings suggest that the right frontal lobe and anterior corpus callosum might have been severely damaged. We speculated that these lesions might still be influencing the uncured left limb volitional movements.

We assumed that the left hemisphere in our patient was dominant for the volitional control of movements. Thus, the disturbances of the left limbs appear to have been produced by a failure in the transfer of motor control information from the left hemisphere. In general, there is one plausible recovery mechanism of apraxia that the right hemisphere may gradually acquire the ability to control its motor system volitionally and independently of the left. Hence, the most important result of this study is that SSCOM showed a significant increase in blood flow in the posterior corpus callosum. We therefore suggested that the improvement of some volitional motor activities resulted from the reestablishment of callosal connectivity between the major and minor hemispheres.

We hypothesized that the transmission of sensory and tactile information via the posterior corpus callosum might play an important role in volitional motor control not only for the left upper limb, but also for the left lower limb.

This present case study has some limitations. First, SISCOM was developed as an analytical software for epilepsy, not for stroke. The neural correlations between stroke and SSCOM findings are unclear. Secondly, the SSCOM results were only obtained in one patient. To confirm the relation between neurological symptoms and responsible lesions, further SSCOM studies are needed.

In conclusion, we report a patient who demonstrated unique left leg symptoms. An evaluation of the cerebral blood flow in this patient using SSCOM suggested that the recovery of apraxia in some patients may be associated with a functional improvement in the posterior corpus callosum.

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