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Recovery of language function in Korean-Japanese crossed bilingual aphasia following right basal ganglia hemorrhage

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ABSTRACT

Few studies have investigated language recovery patterns and the mechanisms of crossed bilingual aphasia following a subcortical stroke. In particular, Korean-Japanese crossed bilingual aphasia has not been reported. A 47-year-old, right-handed man was diagnosed with an extensive right basal ganglia hemorrhage. He was bilingual, fluent in both Korean and Japanese. After his stroke, the patient presented with crossed aphasia. We investigated changes in the Korean (L1) and Japanese (L2) language recovery patterns. Both Korean and Japanese versions of the Western Aphasia Battery (WAB) were completed one month after the stroke, and functional magnetic resonance imaging (fMRI) was performed using picture-naming tasks. The WAB showed a paradoxical pattern of bilingual aphasia, with an aphasia quotient (AQ) of 32 for Korean and 50.6 for Japanese, with Broca's aphasia. The patient scored better in the Japanese version of all domains of the tests. The fMRI study showed left lateralized activation in both language tasks, especially in the inferior frontal gyrus. After six months of language therapy targeting L1, the Korean-WAB score improved significantly, while the Japanese-WAB score showed slight improvement. In this case, the subcortical lesion contributed to crossed bilingual aphasia more highly affecting L1 due to loss of the cortico-subcortical control mechanism in the dominant hemisphere. The paradoxical pattern of bilingual aphasia disappeared after lengthy language therapy targeting L1, and the therapy effect did not transfer to L2. Language recovery in L1 might have been accomplished by reintegrating language networks, including the contralesional language homologue area in the left hemisphere.

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Introduction

As the number of bilingual speakers increases worldwide, there is also a growing understanding of language presentation in these populations. In cases of bilingual aphasia, language recovery patterns may be variable. Parallel recovery, or concurrent improvement in both languages, is the most common type of language recovery in bilingual aphasia (Paradis, 2001). This is followed by differential recovery, which is better improvement in one language over the other. Unusual forms of recovery have also been reported. For example, paradoxical recovery is when one's first language becomes impaired, while the second language is relatively preserved and better recovered. This type of recovery has mostly been reported in standard or crossed subcortical bilingual aphasia (Aglioti, Beltramello, Girardi, & Fabbro, 1996; Aglioti & Fabbro, 1993a; Garcia-Caballero et al., 2007).

Bilingual aphasia typically accompanies cortical lesions. However, there is a growing body of literature addressing bilingual aphasia subsequent to subcortical lesions (Aglioti et al., 1996; Garcia-Caballero et al., 2007; Moretti et al., 2001; Paradis, 1995). Traditionally, the basal ganglia (BG) are associated with motor learning and speech motor control (Abutalebi & Green, 2007; Pyun, Jang, Lim, Ha, & Cho, 2013). Many previous reports have suggested that subcortical structures, especially the BG, govern implicit memory and native language, while the cortex plays a role

in explicit memory and acquired language (Aglioti & Fabbro, 1993a; Fabbro, 2001a, 2001b; Ullman, 2004a). Thus, the BG system seems to be essential to subcortical nodes in the neural network for correct output in the native tongue or any automatized language (Aglioti et al., 1996; Aglioti & Fabbro, 1993a). Recently another critical role of the BG in bilingual language processing was suggested. The BG system may resolve lexical competition in bilingual people, using inhibition through a left frontal-BG circuit. Language selection, set switching, planning, and lexical selection occur through a cortico-subcortical network (Abutalebi & Green, 2007). Therefore, damage to the BG may disrupt late integrative language processes such as suppression of competing alternatives in bilingual patients (Abutalebi & Green, 2007; Friederici, Kotz, Werheid, Hein, & Von Cramon, 2003).

Less than 3% of all reported patients with aphasia have crossed aphasia in dextrals (CAD), which is a right hemispheric brain injury in a right-handed person (Bramwell, 1899; Coppens, Hungerford, Yamaguchi, & Yamadori, 2002; Della Rosa et al., 2014). In patients with CAD, non-fluent aphasia and subcortical stroke are frequent. The aphasic symptoms are rarely transient, and full recovery is uncommon (Della Rosa et al., 2014; Ha, Pyun, Hwang, & Sim, 2012; Kim, Yang, & Paik, 2013; Marien, Paghera, De Deyn, & Vignolo, 2004). With regard to the recovery of patients with CAD and standard subcortical aphasia,

increasing evidence points to the importance of the BG system in establishing new language networks, including the contralateral homologue area for language (Della Rosa et al., 2014).

Cases of bilingual aphasia with coinciding crossed aphasia are extremely rare. Descriptions of its presentation may add to the understanding of language processes in the brain. In this study, we report a rare case of Korean-Japanese crossed bilingual aphasia subsequent to right BG hemorrhage. We also report the patient's recovery pattern of the two languages and functional magnetic resonance imaging (fMRI) findings.

Case report

A 47-year-old right-handed man went to the emergency room presenting with sudden headache and left-side motor weakness. The brain CT scan showed an intracranial hemorrhage in the right BG and thalamus (Figure 1). The patient subsequently underwent craniotomy and hematoma removal. However, the patient continued to show severe left-side motor weakness with severe left-side neglect and aphasia. The patient was transferred to the department of rehabilitation medicine one month after symptom onset.

At the time of transfer, a neurologic examination showed left hemiplegia. The score of manual muscle testing was 1/5 in left upper and lower extremities. And severe left hemineglect was present in the star cancellation test, on which the patient scored 13 of 54, with all targets deviated to the right side. The patient was fully dependent in all activities of daily living and was unable to walk or stand.

Language history and evaluation

The patient had 16 years of education and was extremely right-handed, as assessed by the Edinburgh Handedness Inventory. He was born in Korea and his native language was Korean. He learned Japanese as his second language after graduating from high school, making him a late bilingual. He went to college in Japan and lived there for five years. He was a high-proficiency bilingual, very fluent in Japanese, and

worked as a Japanese teacher at a language institute in Korea until the time of the accident. Although the lesion was in the right hemisphere of his brain, the patient presented with severe crossed aphasia. He was able to speak some Japanese, and he was more fluent in Japanese than his native Korean. While he was only able to produce meaningless syllables in Korean, he could produce appropriate phrases in Japanese. The patient himself confessed that speaking Japanese was easier than Korean.

To further analyze the discrepancy between the two languages, linguistic evaluations were completed in both Korean and Japanese. The Korean version of the Western Aphasia Battery (K-WAB) was completed one month after stroke onset and the result was an aphasia quotient (AQ) of 32 with Broca's aphasia. The Japanese version of the WAB (J-WAB) showed an AQ of 50.6 with Broca's aphasia (Table 1). The patient scored better in the Japanese version of all test domains, especially in the aspects of language production. He was not only significantly more fluent in Japanese with spontaneous speech, but Japanese comprehension, repetition, naming, reading, and writing skills were also better executed and were easier for the patient. While speaking, the patient frequently switched from Korean (L1) to Japanese (L2), while mixing of the two languages was observed less frequently. The patient was also able to translate from Korean to Japanese at the word level but unable to translate from Japanese to Korean at that level.

Cognitive assessment

To evaluate the patient's cognitive impairment, he underwent comprehensive cognitive assessment using the Computerized Neurocognitive Function Test (CNT[®], Maxmedica, Seoul, Korea). The patient showed poor performance in general, but his attention and visuoconstructive abilities were especially impaired. Digit span tests, visual span tests, and auditory and visual continuous performance tests showed scores in the 0.5 percentile. Rey-O complex figure test scores of copy and recall were in the 0.01 percentile, along with the score of Raven's colored

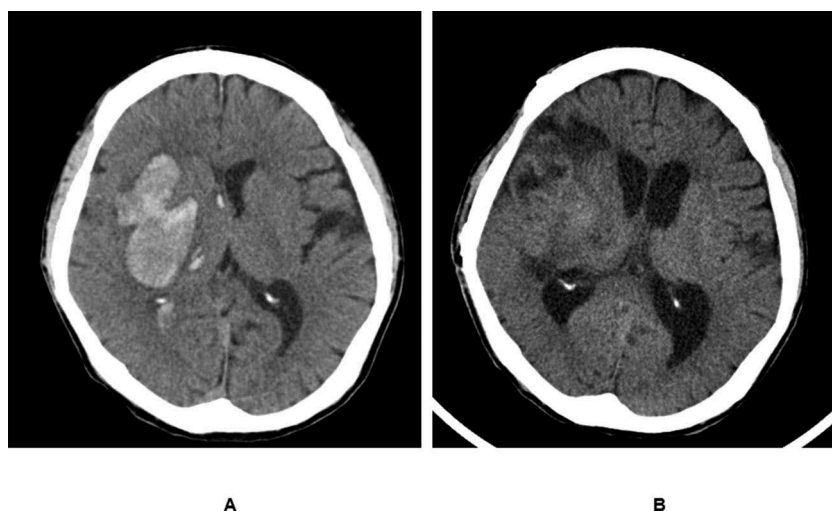


Figure 1. (A) Brain CT of case at stroke onset, showing a massive acute intracranial hemorrhage (ICH) in the right basal ganglia that extends to the frontal subcortical white matter. (B) Brain CT one month after onset, showing resolution of the right ICH.

Table 1. Results of initial and follow-up evaluation of the Korean and Japanese Western Aphasia Battery (WAB).

	Initial evaluation		Follow-up	
	K-WAB	J-WAB	K-WAB	J-WAB
Spontaneous speech (20)	3	6	13.5	12
Conversational question (10)	1	3	8.5	8
Picture description (10)	2	3	5	4
Auditory verbal comprehension (200)	125	151	167	193
Yes/No question (60)	48	51	51	60
Auditory word recognition (60)	47	48	54	57
Sequential commands (80)	30	52	62	76
Repetition (100)	17	50	72	67
Naming (100)	50	67	86	57
Object naming (60)	39	46	57	40
Word fluency (20)	3	5	13	3
Sentence completion (10)	4	8	8	4
Responsive speech (10)	4	8	8	10
AQ (100)	32	50.6	75.4	68.2
Aphasia type	Broca	Broca	Anomic	Broca

Table 2. Results of neuropsychological tests.

Cognitive test and domain	Raw score	Percentile	Interpretation
Attention			
Digit span forward	3	0.5	Abnormal
Visual span forward	3	0.5	Abnormal
Auditory CPT (135)	35	0.5	Abnormal
Visual CPT (135)	89	0.5	Abnormal
Memory			
Digit span backward	2	0.5	Abnormal
Visual span backward	2	0.5	Abnormal
Verbal learning (15)			
Trial 1	4	12.5	Abnormal
Trial 5	5	0.5	Abnormal
Delayed recall	3	0.5	Abnormal
Rey-O-CFT (36)			
Immediate recall	0	<0.01	Abnormal
Delayed recall	0	<0.01	Abnormal
Executive function			
Trail making test			
Type A	Failed		Abnormal
Type B	Failed		Abnormal
Wisconsin Card Sorting Test	Failed		Abnormal
Visuoconstructive ability			
Rey-O CFT (copy) (36)	0.5	0.01	Abnormal
Nonverbal intelligence			
RCPM (36)	3	0	Abnormal

Numbers in parentheses are the maximal score.

K-MMSE, Korean version of the mini-mental status exam; CPT, Continuous performance test; Rey-O CFT, Rey-O complex figure test; RCPM, Raven's colored progressive matrices.

progressive matrices (Table 2). Severe perseveration and poor inhibition control were observed during the cognitive test, suggesting frontal lobe dysfunction.

Functional MRI study

An initial fMRI study was performed two months after the stroke occurred, but follow-up fMRI study was not performed due to patient refusal. Two sessions of a picture-naming task were performed, once in Korean and once in Japanese. Each session involved subvocal naming of 40 black-and-white pictures depicting concrete nouns. Picture items were presented one at a time for 4 s each in 16 s blocks. This was followed by an image of a checker board for 16 s. Pictures were balanced in terms of frequency and number of syllables in the nouns depicted across the two sessions. The stimuli were presented using E-Prime software (Psychology Software Tools, Inc.,

Sharpsburg, MD, USA). The patient was familiarized with the procedures and tasks during a practice session conducted prior to the study.

Functional images sensitive to blood oxygenation level-dependent (BOLD) contrast were acquired on a Magnetom Tim Trio 3T system (Siemens Medical Solutions, Erlangen, Germany) using the following parameters: repetition time (TR) = 2000 ms, echo time (TE) = 30 ms, field of view (FOV) = 24 cm, flip angle = 9°, and matrix = 80 × 80. From each run, 166 functional volumes were obtained. The first six volumes were discarded to allow for signal equilibrium. Each volume was comprised of 36 axial slices of 4 mm thickness, covering the entire cerebrum. T1-weighted anatomical images were acquired with the following parameters: TR = 1900 ms, TE = 2.52 ms, inversion time = 900 ms, flip angle = 9°, slice thickness = 1 mm, FOV = 256 mm, voxel size = 1 × 1 × 1 mm. Data preprocessing and statistical analysis were performed with SPM8 software (Statistical Parametric Mapping; Wellcome Department of Cognitive Neurology, London, UK, <http://www.fil.ion.ucl.ac.uk/spm>). Functional scans were corrected for slice acquisition timing, realigned to the mean functional volume, and then co-registered with the anatomical T1 image, which was spatially normalized to the MNI T1 template of SPM8. Functional volumes were then normalized using the transformation parameters obtained from normalization of the anatomical image with 3 × 3 × 3 mm voxel size. Normalized images were smoothed with an 8-mm FWHM (full width at half maximum) isotropic Gaussian kernel. A voxel-wise threshold of $p < 0.001$ uncorrected and cluster size of 20 voxels were used. The patient provided informed consent before all procedures. The Health Service Human Research Ethics Committee and Committee on Experimental Procedures Involving Human Subjects of Korea University Anam Hospital approved this study.

Although initial evaluations and object naming scores were higher in J-WAB, the fMRI results showed similar cortical activation patterns during Korean and Japanese picture-naming tasks, with more lateralization in the left hemisphere. Both tasks activated the left inferior frontal gyrus, left superior parietal cortex, left pre-central and post-central gyri, and bilateral occipito-temporal cortex, except for supplementary motor area activity observed during the Japanese picture-naming task. No significant differences were shown between Korean and Japanese tasks, suggesting that a similar homologue language network is involved in both the L1 and L2 lexical retrieval processes during picture naming (Figure 2, Table 3).

Language therapy and follow-up evaluation

A speech-language pathologist conducted language therapy in Korean, five times per week for 30 minutes/session, for six months starting one month after stroke. During the early period at our rehabilitation department, therapy mainly focused on improving the patient's naming skills. Naming treatment included both semantic and phonologic treatment. Directions, cues, and feedback were given in Korean. Upon initiating language therapy, the patient's correct response rate for naming fruits and everyday objects during a therapy session was 25% (5 of 20) and 30% (6 of 20) of target words in the

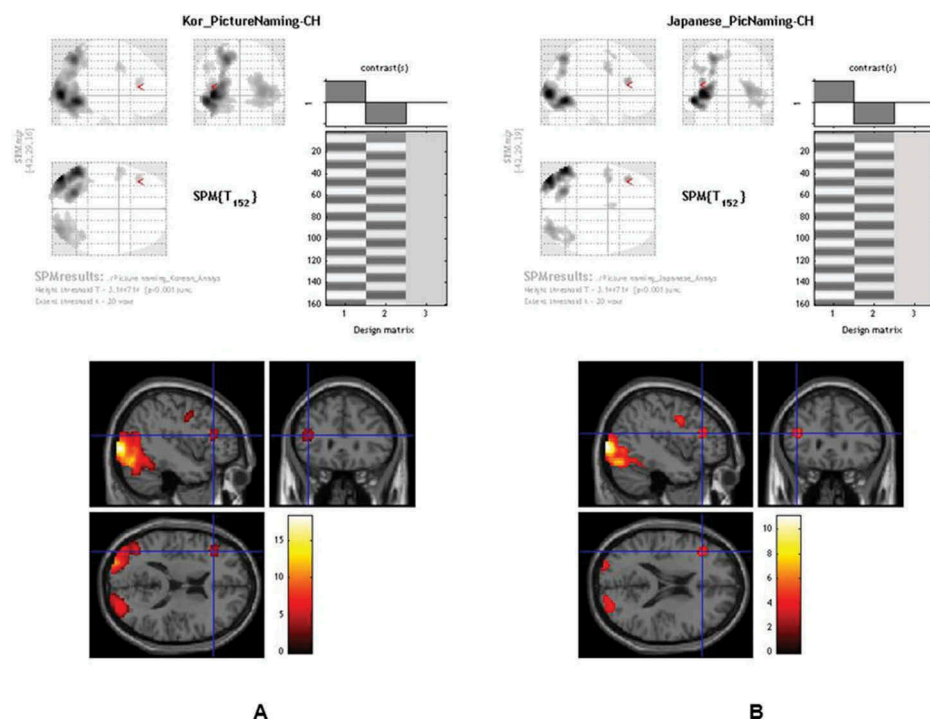


Figure 2. Results of the functional MRI study during the Korean (A) and Japanese (B) picture-naming task. The results showed more left lateralized cortical activation in both languages. Although the Korean picture-naming task seemed to activate more and larger areas than the Japanese, the difference between the two languages was not significant.

Table 3. Significant clusters of activation for overall Korean and Japanese picture-naming task.

Brain region	No. of voxels	MNI coordinates	z-Score
<i>Korean picture naming</i>			
L middle occipital gyrus	2374	−42 −88 −2	Inf
L inferior occipital gyrus		−51 −64 −11	Inf
L superior parietal lobe		−21 −70 49	Inf
R middle occipital gyrus	1051	33 −88 4	7.53
R middle occipital gyrus		42 −85 4	6.53
R superior occipital gyrus		24 −88 19	6.25
L postcentral gyrus	86	−54 −1 40	5.26
L precentral gyrus	60	−42 −4 37	3.84
L inferior frontal gyrus		−42 29 16	4.94
<i>Japanese picture naming</i>			
L middle occipital gyrus	816	−39 −91 1	Inf
L inferior temporal gyrus		−54 −67 −8	Inf
L fusiform gyrus		−42 −76 −17	7.10
L superior parietal lobe	247	−24 −67 52	6.70
L middle occipital gyrus		−27 −70 31	5.35
R middle occipital gyrus		30 −88 7	5.44
R inferior temporal gyrus	470	45 −55 −14	4.61
R inferior temporal gyrus		48 −67 −11	4.29
L inferior frontal gyrus	43	−42 29 19	5.32
L precentral gyrus	95	−42 2 34	4.65
L precentral gyrus	32	−48 −4 37	4.29
L precentral gyrus		−54 −1 43	4.22
L supplementary motor area		−3 8 55	4.00

category, respectively. There was a 20% correct response rate for writing the names of items. Phonemic paraphasia and perseveration were frequently observed during therapy. No remarkable interval change was seen after two weeks of language therapy. The patient showed significant progress at one month after treatment, with a correct response rate of 80% (24 of 30) of target words and shortened response time. He was transferred to another rehabilitation hospital and continuously received language therapy.

After six months of therapy, Korean became the stronger language. Japanese was only the more fluent language in the acute stage, with AQ score changes of 32 to 75.4 in K-WAB and 50.6 to 68.2 in J-WAB (Table 1). The patient's speech included repetitions in syllables and phrases, hesitations, and self-corrections. However, he was able to speak in up to three-word sentences, which was an improvement. The aphasia type changed to anomie as the patient made progress in language production. Similar speech patterns were found in the patient's Japanese, but his Japanese was not as strong as his Korean. While fluency as a whole did not significantly change, there was some improvement in his Japanese speaking ability.

Discussion

This case presented with crossed bilingual aphasia following right BG hemorrhage. Although subcortical aphasia cases tend to show non-fluent aphasia (Démonet, Puel, Celsis, & Cardebat, 1991; Fabbro, Clarici, & Bava, 1996; Mariën, Abutaleb, Engelborghs, & De Deyn, 2005), our case initially presented with Broca's aphasia in both languages with greater impairment of L1 and relative sparing of L2. This paradoxical impairment disappeared after six months of language therapy targeting L1.

To explain this kind of impairment, it is necessary to consider the neural systems involved in the language processing of a bilingual person. Paradis (1994) and Fabbro (2001a) posited that L1 and L2 are processed and represented by distinct neural systems. L1 processing is considered to be mediated by implicit memory, represented in the subcortical structures, while L2 is considered to be learned by explicit memory, represented in the cortical areas. Following these hypotheses,

Ullman described a Declarative/Procedural (DP) model (Ullman, 2004a). According to the model, procedural and declarative knowledge are mediated by distinct neural systems. Procedural knowledge involves the frontostriatal network, while declarative knowledge involves the temporal lobe. However, recent studies have found that procedural and declarative knowledge likely share a single neural network for L1 and L2 processing (Abutalebi, 2008; Abutalebi & Green, 2007). Functional neuroimaging studies have shown that bilingual people engage the same neural structures for L2 that are responsible for grammatical processing, regardless of their proficiency in L2. The manner in which this network operates depends on a person's proficiency in L2.

An inhibitory mechanism of the BG has been also suggested for the control of language selection in bilingual speech (Abutalebi & Green, 2007; Kroll, Bobb, Misra, & Guo, 2008). Based on the findings of neuroimaging studies, the prefrontal cortex, anterior cingulate cortex, inferior parietal cortex, and BG are involved in the control and selection of language used by a bilingual person (Abutalebi & Green, 2007). Selection of a target language may be controlled by the anterior cortical-subcortical circuit, while the left BG may supervise language output. Studies have reported that a lesion in the language-dominant subcortical structures leads to pathological switching and mixing among languages (Abutalebi, Miozzo, & Cappa, 2000; Mariën et al., 2005), as well as pathological fixation on a language (Aglioti et al., 1996; Aglioti & Fabbro, 1993a). In our case, the patient's paradoxical language selection toward L2 can also be described as a dysfunction of language control and selection. The patient's brain lesion extended from the BG to the prefrontal area, including Broca's area, which coincides well with the BG-prefrontal circuit. If we agree with the inhibitory control mechanism of the BG circuit, a lesion in the BG of the dominant hemisphere caused dysfunctional activation of L1, leading to spontaneous switching from L1 to L2.

To our knowledge, there is only one other officially reported case of crossed bilingual aphasia after a subcortical lesion (Garcia-Caballero et al., 2007). There have been no reported cases of Korean-Japanese crossed bilingual aphasia. Our findings share some similarities with that in Garcia-Caballero et al., including that the patient's first language (Galician) was more impaired than his second language (Spanish) after a right BG lesion. However, the recovery pattern of bilingual aphasia was different. The former case did not receive language therapy and showed paradoxical selective recovery of L2, even 16 months post-stroke. Our case received lengthy language therapy targeting the more impaired L1, and the paradoxical pattern of bilingual impairment disappeared at the final follow-up evaluation. Thus, our case was different from the previously reported cases of bilingual subcortical aphasia, which showed limited recovery of spontaneous speech and stable paradoxical selective recovery in L2 even after long-term follow-up (Aglioti et al., 1996; Aglioti & Fabbro, 1993a; Garcia-Caballero et al., 2007).

In our case, therapy in L1 benefits L1 but does not seem to transfer to L2. One explanation of greater improvement in L1 is Pitre's and Ribot's rule on bilingual recovery, in which the

most familiar and native language is better recovered (Paradis, 2004). However, a more plausible explanation is the effect of the language therapy that is used to enhance language recovery and associated functional changes of the brain. Recent studies have claimed that subcortical bilingual aphasia improved treated language and was associated with increased activation in language areas or homologue areas in the hemisphere contralateral to the lesion causing aphasia (Abutalebi, Della Rosa, Tettamanti, Green, & Cappa, 2009; Della Rosa et al., 2014). In this case, an fMRI study using Korean and Japanese picture-naming tasks revealed that the contralesional homologue language areas, including the left inferior frontal gyrus, were recruited for language production after a right BG lesion. The activation patterns in L1 and L2 were similar. This observation suggests that intensive language treatment induces behavioral change through reintegration of the remaining language network and control pathways for L1 and L2 (Abutalebi et al., 2009).

Bilingual aphasic patients may show diverse patterns of language impairment and recovery. Study of these patterns may increase the understanding of the cerebral control of different languages in a bilingual person's brain. This rare case of crossed bilingual aphasia contributes information about the BG in controlling language in bilinguals with right hemisphere dominance.

Disclosure statement

No potential conflict of interest was reported by the authors.

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