



## Short Communication

# Pure agraphia in Romaji after left inferior frontal gyrus infarction: A case of selective deficit in syllable-to-grapheme conversion in Japanese



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## ABSTRACT

The Japanese writing system employs two distinct categories of characters: Kana and Kanji. The difference between Kana and Kanji writing corresponds roughly to that between phonological and lexical systems in Western languages. When typing, most Japanese use alphabetical characters based purely on phonological rules. In particular, the Romaji system is used, in which a syllable consists of a single vowel, a consonant–vowel compound (e.g., ka, ki), or a sequence of consonant–y(semivowel)–vowel (e.g., kya, kyu). We describe a right-handed Japanese patient who developed pure agraphia that affected Romaji writing but preserved Kana and Kanji writing and who had a lesion in the left pars opercularis of the inferior frontal gyrus that extended to the anterior precentral gyrus. The patient demonstrated literal paraphasia in spelling Romaji across modalities. Our findings suggested that the patient's agraphia in Romaji after a confined left frontal infarction was manifested by a selective impairment in syllable-to-grapheme conversion.

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## 1. Introduction

Cultural differences influence cognitive processes and thus the organization of neural networks in humans, and one such domain of influence is the writing system (Paulesu et al., 2000; Siok, Perfetti, Jin, & Tan, 2004). Current models for the writing system in Western languages postulate that there are two parallel routes: the phonological route and the lexical route (Ellis, 1988; Margolin 1984; Roeltgen, 1993). The former relies on a phoneme-to-grapheme conversion process, which is important for writing regularly spelled words (or non-words), whereas the latter relies on a whole-word retrieval process, which enables one to write familiar words regardless of their spelling regularity. Focal brain damage can affect spelling in different ways: dysfunctioning of the phonological and lexical routes gives rise to phonological agraphia and lexical agraphia, respectively (Beauvois & Dérouesné, 1981; Hillis, Chang, Breese, & Heidler, 2004; Marien, Pickut, Engelborghs, Martin, & De Deyn, 2001; Rapcsak, Arthur, & Rubens, 1988; Roeltgen, Gonzalez Rothi, & Heilman, 1986; Roeltgen, Sevush, & Heilman, 1983; Shallice, 1981).

The Japanese writing system employs two distinct categories of characters: Kanji (morphogram) and Kana (syllabogram). The relative difference comprising the regular-irregular dichotomy in

Western languages can appear to be a qualitative difference between Kana and Kanji. For instance, some Japanese patients with left frontal agraphia have shown particular difficulty in writing Kana with literal paraphasia (Abe, Yokoyama, Torifuji, & Yanagihara, 1993; Sakurai, Matsumura, Iwatsubo, & Momose, 1997; Tohgi et al., 1995), and these patients are considered to be showing phonological agraphia. However, in terms of linguistics, the phonological level of sound-to-letter conversion differs between Western regular words (phoneme-to-alphabet conversion) and Kana words (syllable-to-syllabogram conversion) (Iwata, 1984).

Most Japanese use another writing system for typing, an alphabetic writing system called Romaji, which is based on phonological rules learnt in elementary school. A syllable in Romaji consists of a single vowel, a consonant–vowel compound (e.g., ka, ki), or a sequence of consonant–y(semivowel)–vowel (e.g., kya, kyu) (Hattori, 1979). When typing with a software program for Romaji input, the moment an alphabetical letter string makes up a syllable unit, it automatically changes into a Kana character. Therefore, a whole word written in Romaji does not appear on the monitor. Japanese rarely use Romaji in handwriting, and Romaji words are seldom seen in Japanese printed materials. Consequently, the syllable-to-grapheme conversion can develop in Romaji writing without the development of whole-word imagery. Selective damage of the syllable-to-grapheme conversion system may result in impaired Romaji writing with preserved Kana and Kanji writing. Neither the mechanisms nor the neural substrates of the Romaji writing system have been investigated as of yet. We report here on a patient with isolated agraphia in Romaji after a left frontal

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infarct, indicating that there are two distinct sound-to-letter conversion systems and lexical routes developed in Japanese culture and that the left frontal region engages purely in sound-to-letter conversion without engaging in the associated whole-word imagery.

## 2. Case report

### 2.1. Case description

The patient was a 51-year-old, right-handed Japanese clerical worker with a 12th-grade education. He had been hypertensive for 12 years, for which he had received treatment. A year previously, he had suffered from the Tolosa-Hunt syndrome, a painful ophthalmoplegia caused by nonspecific inflammation of the cavernous sinus, and he had been treated with steroids. It is not known whether this condition had been associated with the onset of his infarction. He had been skilled at typing using Romaji input, having had 20 years' experience, but he suddenly realized that he was unable to type despite being able to operate a keyboard and to read alphabetical letters, and he was admitted to a nearby hospital. General physical and neurological examinations were unremarkable, but brain magnetic resonance imaging (MRI) demonstrated a small infarct confined to the left frontal lobe. After conservative therapy, he was referred to our hospital for further assessment and rehabilitation. On admission, the patient was alert and well-oriented. He showed no cranial nerve deficit and had no hemiparesis or ataxia. The muscle tone of his arms and legs was normal, as were his muscle stretch reflexes, and no pathological reflexes were elicited. Sensitivity to light touch, pain, temperature, vibration, and joint motion was also normal. A brain MRI performed 28 days after the onset of symptoms demonstrated a small lesion confined to the pars opercularis of the inferior frontal gyrus and its subcortical area, extending to the foot of the anterior precentral gyrus in the left hemisphere (Fig. 1). No other lesion was detected.

### 2.2. Neuropsychological examination

Neuropsychological evaluations were performed between one and two months after onset. Detailed data for the standard neuropsychological tests are shown in Table 1. The patient's intelligence and memory were normal. His performance of executive function was assessed by the Wisconsin Card Sorting Test, the Trail Making Test, and verbal fluency. The ability was impaired, which may not affect language performance as evidenced by the results of the Western Aphasia Battery (WAB) (Kertesz, 1982) Japanese version

(Sugishita, 1986), the Wechsler Adult Intelligence Scale-Revised, and Wechsler Memory Scale-Revised. The WAB is a standardized instrument for evaluating the language and praxis function of Japanese adults, discerning the presence, degree, and type of aphasia, and assessing writing ability in both Kana and Kanji. He showed no apraxia on the WAB. He showed no signs of unilateral spatial neglect or constructional disability. His linguistic ability was assessed by the WAB. He retained his ability to read words and sentences written in Kana or Kanji. He could read and write Arabic numerals. Writing of one- to four-digit numbers was performed without error during dictation (100% correct on 40 numbers). When writing, he used his right hand and produced orthographically well-formed Kana and Kanji, with occasional difficulty writing Kana. He was also able to read Romaji. In contrast, literal paraphasia was evident in his writing and typing of words in Romaji, even though he could name, write, and type each of the alphabetic characters. When typing, he used the fingers of both hands, which is typical for a person skilled in typing. He showed no other signs of aphasia.

### 2.3. Special assessments for agraphia

#### 2.3.1. The ability to write a single syllable in Romaji and Kana

To evaluate further the patient's ability to spell in Romaji as well as to write in Kana, we first gave him a monosyllable spelling test. An articulatory test in Japanese includes 101 single syllables. An examiner dictated the 101 syllables one at a time in a random order. At 23 days after onset, the patient's handwriting ability was experimentally examined. Three days later, his writing, typing (with no conversion to Kana), and orally spelling of these syllables in Romaji were simultaneously assessed. On the same day, he was also asked to transcribe into Romaji 101 Kana characters corresponding to the 101 syllables used in the monosyllable spelling test. He was also asked to write 101 syllables in Kana from dictation. He committed many errors in all tasks involving Romaji (Table 2), which were confined to literal paraphasia; the distribution of error types was remarkably similar across the tasks. In almost all cases, consonants were substituted for consonants and vowels for vowels, and the consonant substitutions were not determined by either phonological or orthographical similarity. The patient was not aware of all his spelling errors. He was able to write 94 of the 101 Kana characters correctly and could repeat all of the 101 syllables and read all of them when written in Romaji or Kana.

#### 2.3.2. The ability to write words and non-words in Romaji and Kana

Next, we assessed the patient's ability to write dictated words and non-words in Romaji and Kana. We presented him with eight

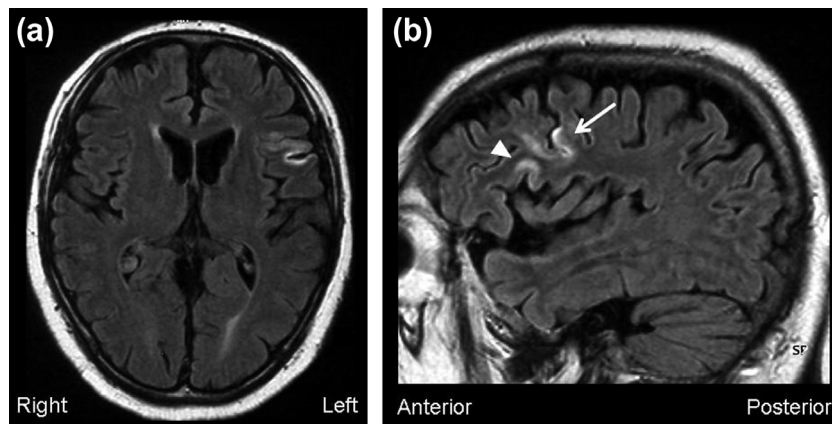


Fig. 1. MRI fluid-attenuated inversion recovery images (a, axial; b, sagittal) performed 28 days after onset, showing an infarct affecting the pars opercularis of the inferior frontal gyrus and its subcortical area (arrow head), extending to the foot of the anterior precentral gyrus (arrow) in the left hemisphere.

**Table 1**  
Performance on standard neuropsychological tests.

Tests	Score	Normative data; mean (SD)
<i>Language</i>		
WAB	Fluency	10
	Auditory word recognition	60
	Repetition	100
	Object naming	60
	Reading (Kanji and Kana)	9.8
	Writing (Kanji and Kana)	9.7
<i>Intelligence</i>		
WAIS-R	Full IQ	100
	Verbal IQ	92
	Performance IQ	110
J-RCPM		34
<i>Memory</i>		
Digit span	Forward, backward	5, 3
Spatial span	Forward, backward	5, 3
WMS-R	General index	92
	Verbal index	85
	Visual index	112
	Attention/concentration	101
BVRT	Immediate recall	9
	Delayed recall	9
<i>Executive function</i>		
WCST	Categories achieved	1
	Perseverative errors	6
	Difficulty maintaining set	0
TMT	Part A	39
	Part B	90
Verbal fluency	Animal	14
Praxis		
WAB	Praxis	60

WAB: Western Aphasia Battery, WAIS-R: Wechsler Adult Intelligence Scale-Revised, J-RCPM: Japanese Raven's Colored Progressive Matrices, WMS-R: Wechsler Memory Scale-Revised, BVRT: Benton's Visual Retention Test, WCST: Wisconsin Card Sorting Test, TMT: Trail Making Test.

**Table 2**  
Error distribution (number of errors and percentage error).

Error type	Examples	Hand-writing		Typing	Oral spelling	Transcription	
		Day-23	Day-26	Day-26	Day-26	Day-26	
		N (%)	N (%)	N (%)	N (%)	N (%)	
Substitutions	C → C	pe → me	23 (62.2)	25 (44.6)	19 (63.3)	8 (28.6)	22 (57.9)
	V → V	si → su	12 (32.4)	15 (26.8)	5 (16.7)	13 (46.4)	13 (34.2)
	C → C + V → V	re → mo	2 (5.4)	8 (14.3)	0 (0)	6 (21.4)	3 (7.9)
	C → V	kya → eya	0 (0)	1 (1.8)	0 (0)	0 (0)	0 (0)
	y → C	ryo → kto	0 (0)	0 (0)	1 (3.3)	1 (3.6)	0 (0)
Deletions		tya → ta	0 (0)	4 (7.1)	3 (10)	0 (0)	0 (0)
Insertions		e → ke	0 (0)	0 (0)	1 (3.3)	0 (0)	0 (0)
Substitution + deletion		syo → su	0 (0)	3 (5.4)	1 (3.3)	0 (0)	0 (0)
Total errors			37 (100)	56 (100)	30 (100)	28 (100)	38 (100)

N: number of errors, C: consonant, V: vowel, y: semivowel.

lists of ten stimuli. The first to the fourth lists consisted of one- to four-syllable words, and the fifth to eighth lists consisted of one- to four-syllable non-words. He wrote 17 out of 40 (42.5%) words and 23 out of 40 (57.5%) non-words incorrectly in Romaji. We obtained the following error probabilities for syllable units according to the number of characters in the words or non-words: 10% (1/10), 40% (8/20), 30% (10/30), and 18% (7/40) for words, and 30% (3/10), 35% (7/20), 30% (9/30), and 33% (13/40) for non-words. There was no significant difference in the percentage of errors for words and non-words ( $\chi^2 = 0.87$ ,  $p = 0.35$ ). The distributions of error types for both words and non-words were essentially identical to those in the monosyllable spelling tasks. In writing Kana from dictation, the patient committed only a few errors (2 out of 40 words, 1 out of 40 non-words, and 3 out of 200 kana characters; 1.5%) and could read all of the 40 words and 40 non-words when written in Kana or Romaji.

#### 2.4. Recovery

The patient's ability to write and type using Romaji improved gradually. Seven months after onset, his spelling accuracy in Romaji was 89% (90/101) when writing by hand and 97% (98/101) when typing on the monosyllable task.

### 3. Discussion

We have described a patient who manifested an impairment in spelling Romaji while his reading of Romaji was preserved after a confined infarction in the left inferior frontal gyrus. He could write and read Kanji, Kana, and Arabic numerals, and he showed no aphasia, agnosia, apraxia, limb clumsiness, constructional disability, or dementia. Therefore, the patient's symptom can be

interpreted as a focal sign after the onset of infarction, and it was consistent with the criteria for pure agraphia (Roeltgen, 1993).

Neither the mechanisms nor the neural substrates of the Romaji writing system have been investigated as of yet. The dissociation of writing abilities in Kana and Romaji is an important finding. Both writing systems rely on phonological-level associations (Hattori, 1979; Iwata, 1984). Kana characters are syllabic symbols, and a spoken syllable can be directly mapped to a Kana character (Kindaichi, 1988). Consequently, the term syllabograms should be used for Kana characters, although they are conventionally called phonograms (Iwata, 1984). When spelling in Romaji, in contrast, it is necessary to decompose a syllable into a grapheme unit (Hattori, 1979), which is the smallest orthographic unit representing a sound (Wydel & Butterworth, 1999). In addition to this difference at the phonological level of sound-to-letter conversion, Kana and Romaji writing systems have another important difference: Kana writing not only relies on syllable-to-syllabogram conversion but also on whole-word imagery. For instance, in a patient with Kana agraphia, writing of Kana non-words was more impaired than that of Kana words (Kokubo, Suzuki, Yamadori, & Satou, 2001). Therefore, the preservation of Kana writing ability may be explained by the compensatory use of the lexical route. However, preserved Kana writing in our patient cannot be explained by the use of the lexical route because the writing of Kana non-words, as well as of Kana words, was preserved. Therefore, our findings substantiate the presence of two independent sound-letter conversion systems in addition to the lexical route in Japanese: the syllable-to-syllabogram conversion system (Kana) and the syllable-to-grapheme conversion system (Romaji).

The patient did not show lexical errors, which most likely reflects a compensatory use of whole-word imagery that is also observed in Western patients with phonological agraphia. It has been suggested that the visual word images of the lexical system are developed from visual exposure (Roeltgen et al., 1986). As mentioned earlier, the Japanese rarely use Romaji in hand-writing, and Romaji words are seldom seen in Japanese printed materials. Furthermore, even when typing, a whole word written in Romaji does not appear on the monitor. Consequently, sound-to-letter conversion can develop in Romaji writing without whole-word imagery. The comparable error rates for words and non-words in writing Romaji in our patient support the notion that the Romaji writing system has no stored representations of the spellings of words. There might be an objection that the patient did not develop the lexical route in his writing process. However, he retained his ability to write Kanji, which heavily relies on the lexical route (Iwata, 1984). These findings suggest that the lexical route in his writing process, at least in Kanji and Kana, has been developed and preserved after an infarction.

Roeltgen's model of writing proposes two general categories of functions: linguistic components and motor components (Roeltgen, 1993). The former contains two parallel routes (phonological and lexical) and a graphemic buffer, whereas the latter includes graphic output programming and motor programming. Our patient could write all single alphabetic characters. The comparable error rates for written spelling, oral spelling, and typing indicate that his deficit cannot be explained by peripheral motor components, including graphic output programming and motor programming; his impairment existed in the phonological route or the graphemic buffer. There are several neuropsychological studies reporting on patients with selective damage to the graphemic buffer (Caramazza & Miceli, 1990; Caramazza, Miceli, Villa, & Romani, 1987; Schiller, Greenhall, Shelton, & Caramazza, 2001). These patients could spell dictated meaningless syllables consisting of a consonant and vowel flawlessly. Long words were affected more than short words, and their errors in spelling most often occurred in the final part of words. The absence of increasing errors for longer stimuli

and towards the end of a stimulus in our patient suggests that the graphemic buffer for Romaji itself functions well. Therefore, the patient's impairment existed in the phonological route.

Roeltgen et al. (1983) suggests that the phonological route is mediated by two sequential processes: (1) a segmentation process, in which syllable sequences of words or non-words are broken down into phonemes; and (2) phoneme-to-grapheme conversion, in which the segmented phonemes are translated into graphemes. In two sequential processes in the phonological route, the patient's segmentation process functioned well, making no phonological errors in spontaneous speech, repetition, oral naming, or reading (Caramazza, Miceli, & Villa, 1986). Furthermore, his paragraphic errors in Romaji writing occurred within the framework of each syllable, i.e., within a single vowel, a consonant-vowel compound, or a sequence of consonant-y(semivowel)-vowel. These findings suggest that our patient had a deficit in phoneme-to-grapheme conversion. Now, we will take a close look at phoneme-to-grapheme conversion in Japanese. Syllabic units play a major role in phonological processing in the Japanese language, but phonemic representations also develop (e.g., Mann, 1986). Instruction in Romaji writing helps Japanese people to become aware of segmental information at the phoneme level (Hattori, 1979). These previous studies allow us to hypothesize the presence of the segmentation processes in which syllables are broken down into phonemes in Japanese. However, this does not mean that phoneme-to-grapheme conversion indeed participates in the process of spelling alphabet letters as a mandatory cognitive component in Japanese. Hence, we should treat the phonological route without dividing into the two sequential processes, and we embrace the concept of syllable-to-grapheme conversion instead of phoneme-to-grapheme conversion.

There have been reports of both Western and Japanese patients with pure agraphia following damage to the left frontal lobe. Hillis, Wityk, Barker, and Caramazza (2003), Hillis et al. (2004) reported on five patients who presented with pure agraphia at the onset of a stroke, all of whom had lesions and/or hypoperfusion in Brodmann's areas (BA) 44, 45, and 6. The impaired phoneme-to-grapheme conversion mechanisms in these cases indicate that parts of BA 44 and 45 may be important for the phonological analysis needed for the phonological route, whereas a graphic output program may depend on BA 6. Marien et al. (2001) reported on a case of impaired use of the phoneme-to-grapheme conversion mechanisms for spelling following a lesion involving the left operculum, precentral gyrus, and insula. Rapp and Caramazza (1997) presented an analysis of the impaired writing performance of two dysgraphic subjects. One of them had lesions of the posterior left temporal and anterior and middle aspects of the peripheral left occipital lobe and the posterior left frontal region. He made well-formed letter substitution errors in written spelling with relatively intact oral spelling performance. The patient described by Anderson, Damasio, and Damasio (1990) showed impaired production of specific letter shapes following lesions in the precentral gyrus. Rapcsak et al. (1988) reported on a patient with lexical agraphia from a focal lesion of the left precentral gyrus. These reports seem to suggest that the inferior posterior frontal region, in particular BA 44 and 45 rather than BA 6, engages with the phoneme-to-grapheme conversion mechanism. Most Japanese patients with agraphia due to a left frontal lesion have been reported to have a Kana-dominant or Kana-specific writing impairment (Abe et al., 1993; Sakurai et al., 1997; Tohgi et al., 1995). They commonly make Kana errors such as paraphasia and rarely write badly formed Kana characters. Therefore, the frontal regions are essential for the sound-letter conversion (syllable-to-syllabogram conversion) mechanism in Japanese as well. However, most of the patients cited above had more extensive lesions and deficits in broader aspects of the language function. The present patient had no

speech or reading disorders from disease onset. The findings were obtained despite the fact that his brain damage was recent, and thus there had been little time for the functional reorganization of the neural networks. The patient's good recovery in terms of Romaji writing after seven months indicates that his impairment at stroke onset was an acquired rather than a premorbid deficit. Taken together, our findings suggest that agraphia in Romaji after a confined left frontal infarction is a manifestation of a selective impairment in syllable-to-grapheme conversion.

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