Hatakeyama *et al. BMC Neurology* (2024) 24:374
https://doi.org/10.1186/s12883-024-03892-w

BMC Neurology

CASE REPORT

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A case of isolated dystextia due to subcortical infarction: a novel condition of digital device era

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Abstract

Background In recent years, cases of dystextia (texting disabilities) and dystypia (typing disabilities) have been reported. However, reports describing isolated dystextia without aphasia or other cognitive impairments are rare, and the detailed pathophysiology is not fully understood. Most Japanese people use the alphabetical spelling system (Romaji) for texting and typing. Herein, we report the case of a man with isolated dystextia and dystypia resulting from Romaji conversion difficulties.

Case presentation A 48-year-old, right-handed Japanese man developed texting and typing difficulties. The standard neuropsychological tests showed no signs of aphasia or other cognitive impairments, except for slight executive dysfunction. Thus, isolated dystextia and dystypia were diagnosed. Furthermore, the patient experienced Romaji conversion difficulties. Magnetic resonance imaging revealed a subcortical infarction in the left cerebral hemisphere. Single photon emission tomography revealed hypoperfusion, including in the left dorsolateral frontal cortex.

Conclusions The left dorsolateral frontal cortex may be related to Romaji conversion in Japanese individuals. Therefore, diaschisis of the left dorsolateral frontal cortex due to subcortical lesions may have impaired Romaji conversion, leading to dystextia and dystypia, in this patient.

Keywords Dystypia, Dystextia, Romaji, Subcortical lesion, Diaschisis, Cerebral infarction, Hypoperfusion

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Background

As communication with digital devices becomes increasingly ubiquitous, deficits related to the usage of these new technologies will likely become more widespread as a presenting symptom of acute or chronic neurological dysfunction. In particular, texting messages on smartphones and keyboard typing play further significant roles in people's lives. In recent years, cases of dystextia (texting disabilities) [1, 2] and dystypia (typing disabilities) [3] have been reported. Since reports describing isolated dystextia without other cognitive impairments are very rare, the detailed pathophysiology is not fully understood [4].

Most Japanese people use the alphabetical spelling system (Romaji) for texting and typing, wherein a syllable consists of a consonant-vowel compound. In Japanese texting, the consonants are first selected and then vowels are determined using the number of taps (toggle input) or the direction of the slide (flick input). In Japanese typing, a person types the key of the alphabet corresponding to a consonant or vowel, and an alphabetical letter string that forms a syllable unit is automatically converted into a syllabogram (Kana). Herein, we report the case of a Japanese patient who developed isolated dystextia and dystypia after cerebral infarction, where difficulties in Romaji conversion may have contributed to the disability.

Case presentation

The patient was a 48-year-old, right-handed Japanese man educated up to the 12th grade and with a history of type 2 diabetes mellitus, hypertension, and dyslipidemia. Additionally, the patient had nasopharyngeal cancer and had received chemoradiation therapy, with no problems with activities of daily living. The patient was skilled at texting messages using a smartphone with toggle input. The patient was also skilled at typing using Romaji input, and the patient's typing speed was considerably faster than that of writing.

The patient suddenly realised the clumsiness of the right hand, difficulty in articulation, and word-finding difficulty. Furthermore, the patient unconsciously tapped the key, either more or less than correctly, while texting a message on a smartphone. Additionally, the patient was unable to correctly type with the keyboard and required more than 1 min to input one word.

Twenty-nine days after onset, the patient was admitted to our hospital with a diagnosis of cerebral infarction. A neurological examination revealed that the patient was fully alert, cooperative, and mentally stable. Furthermore, slight dysarthria, clumsiness of the right hand, and abnormal sensations on the right side of the body were observed. After admission, dysarthria and clumsiness of the right hand improved; however, difficulties in texting and typing did not completely recover.

On admission, brain magnetic resonance imaging (MRI) revealed a cerebral infarction extending from the lateral side of the left thalamus to the left corona radiata (Fig. 1a). Brain magnetic resonance angiography (MRA) on admission showed no signs of stenosis of the intracranial arteries (Fig. 1b). Neck MRA performed 35 days after onset of cerebral infarction revealed stenosis of the left internal carotid artery (Fig. 1c). Electrocardiogram-monitoring on admission and transthoracic echocardiogram performed 39 days after the onset of cerebral infarction revealed no signs of a cardiac source of embolism. Single photon emission tomography (SPECT) with Technetium-ethyl cysteinate dimer (^{99}Tc-ECD) 41 days after onset revealed decreased cerebral blood flow (CBF) in the left dorsolateral frontal lobe, subcortical area of the right cerebral hemisphere, and the right cerebellar hemisphere, in addition to the infarct area (Fig. 1d).

Neuropsychological evaluations were performed 30–40 days after onset. Table 1 shows the detailed data of the standard neuropsychological tests. Although the patient experienced word-finding difficulty at disease onset, the Japanese version of the Western Aphasia Battery revealed no signs of aphasia, alexia, or agraphia. The patient showed no signs of spatial neglect, constructional disabilities, short-term memory impairment, or general intelligence deficit. During Part B of the Trail Making Test, Japanese edition (TMT-J), he took slightly more time than the average and had one connection error, suggesting slight executive dysfunction.

Furthermore, we investigated texting and typing difficulties by presenting 100 auditory words 39–40 days after onset [5]. The patient was required to write in Kana, text, and type the same material. Errors were not detected in writing. However, the number of texting errors per 100 words was 11. We classified texting errors into three categories: consonant errors, vowel errors, and errors in both consonants and vowels. All consonant errors were tapped to keys adjacent to the target, possibly caused by the clumsiness of the right hand, whereas vowel errors are not thought to be caused by clumsiness. The number of typing errors per 100 words was 12. We also classified typing errors into four categories: spatial error (mistyping the key adjacent to the target), phonological error (mistyping the key not adjacent to the target), omission, and order inversion. Phonological errors and omissions are not thought to be present in healthy individuals skilled at typing (Table 2).

Next, we assessed the patient's Romaji conversion ability. We randomly presented 60 auditory syllables one at a time; subsequently, the patient was required to write the same material in Kana and Romaji. The number of errors and the time required to write every five syllables were recorded. The number of errors was zero (writing in Kana), and one (written in Romaji). However, the average

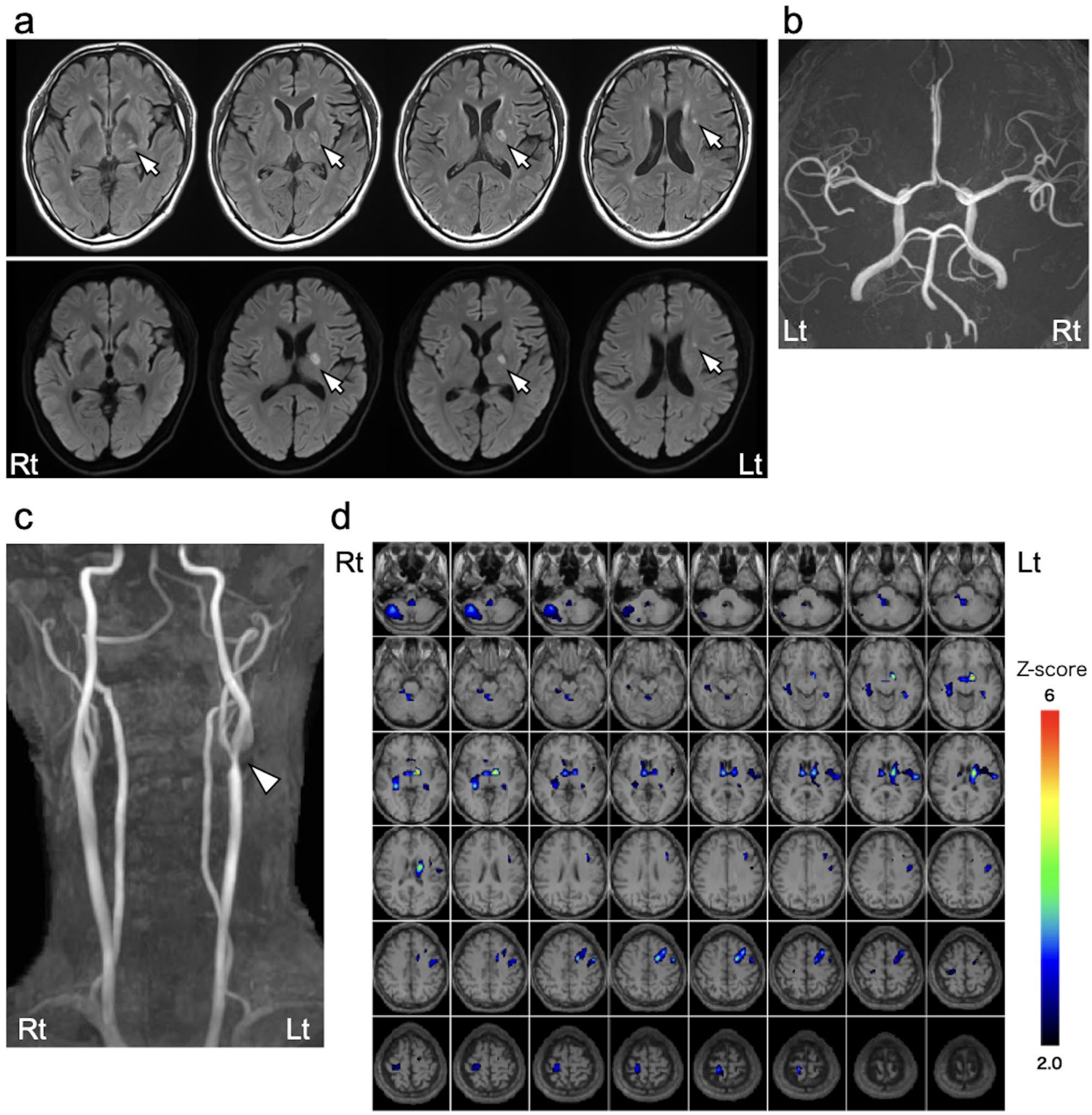

Fig. 1 Imaging evaluations: **a**. Magnetic resonance imaging (MRI) fluid-attenuated inversion recovery images (upper row) and diffusion-weighted images (bottom row) on admission showing cerebral infarction extending from the lateral side of the left thalamus to the left corona radiata (arrow). **b**. Brain magnetic resonance angiography (MRA) on admission showed no signs of stenosis of the intracranial arteries. **c**. Neck MRA performed 35 days after onset of cerebral infarction revealed stenosis of the left internal carotid artery (arrowhead). **d**. Single photon emission tomography with ^{99}Tc-ECD at 41 days after the onset revealed decreased cerebral blood flow in the left dorsolateral frontal lobe, the subcortical area of the right cerebral hemisphere, and the right cerebellar hemisphere in addition to the infarct area. Data are analysed with the Easy Z-score imaging system. Hypoperfusion with a Z-score > 2 is coloured. ^{99}Tc-ECD: Technetium-ethyl cysteinate dimer

time required for writing in Kana was 15.7±4.5 s, and that for writing in Romaji was 21.0±6.8 s. An unpaired *t*-test revealed a significant difference between the two conditions ($p=0.035$).

Discussion and conclusions

The patient exhibited isolated texting and typing impairments without aphasia, alexia, agraphia, apraxia, visuospatial impairments, or general intelligence impairments. Additionally, texting and typing errors could not be

Table 1 Performance on standard neuropsychological tests

Tests		Score
General intelligence		
J-RCPM		35/36
HDS-R		30/30
Attention and Executive function		
Digit span	Forward, Backward	6, 4
TMT-J	Part A	29 s
	Part B	65 s
Language		
WAB	Spontaneous speech	
	Fluency	10/10
	Comprehension	
	Auditory word recognition	60/60
	Sequential commands	80/80
	Repetition	100/100
	Object naming	60/60
	Written word stimulus-picture choice matching	6/6
	Writing	
	Writing of dictated words (Kanji, Kana)	6/6, 6/6
	Copying a sentence	10/10
Praxis		
WAB	Praxis (right hand, left hand)	60/60, 60/60
Visual perception		
BIT	Line crossing	36/36
	Letter cancellation	38/40
	Star cancellation	54/54
	Line bisection	9/9
Figure copy	Crossed pentagon, Necker cube	Normal, Normal

J-RCPM: Japanese Raven's Coloured Progressive Matrices, HDS-R: Revised Hasegawa Dementia Scale, TMT-J: Trail Making Test, Japanese edition, WAB: Western Aphasia Battery, BIT: Behavioural Inattention Test

Table 2 Error distribution of the word dictation task

Tests	Error patterns	Example	Number of errors
Writing in Kana			0
Texting	Consonant	wa→ya	1
	Vowel	su→si	10
	Consonant and vowel	ko→na	1
Typing	Spatial	ya→ys	4
	Phonological	ha→ka	2
	Omission	ta→t	5
	Order inversion	so→os	1

explained solely by hand clumsiness. Therefore, isolated dystextia and dystypia were diagnosed. Our patient also showed slight executive dysfunction on the TMT-J. The relevance of slight executive dysfunction to texting and typing cannot be denied; however, not all patients with slight executive dysfunction show texting and typing impairments. Thus, we speculated that slight executive

Table 3 Summary of published cases of isolated dystextia and dystypia due to stroke

Author / year [Ref no.]	Dystextia or dystypia	Age (y), Sex	Pathophysiology	Lesion site
Otsuki, et al. 2002 [3]	Dystypia	60, M	Conversion disturbance of phonological information to an appropriate corresponding performance	Left frontal lobe
Ryu, et al. 2012 [10]	Dystypia	64, M	Visuospatial memory impairment	Bilateral frontal subcortical area (predominantly in the left side)
Ogura, et al. 2013 [6]	Dystypia	51, M	Impairment in syllable-to-grapheme conversion	Left frontal lobe
Cook, et al. 2013 [11]	Dystypia	68, M	-	Left temporo-parietal lobe
Suzuki, et al. 2015 [7]	Dystypia	77, F	Impairment of writing and reading abilities for Romaji	Left middle cerebral artery territory
Maeda, et al. 2021 [8]	Dystypia	53, M	Difficulty in converting syllabogram to Romaji	Left parietal lobe
Chen, et al. 2023 [4]	Dystextia	54, M	-	Left frontal lobe
Yamamoto, et al. 2024 [9]	Dystypia	58, M	Difficulty with recalling romaji spelling	Left internal capsule
This case	Both	48, M	Difficulties in syllable-to-Romaji conversion	Left thalamus to left corona radiata

dysfunction did not play a major role in texting and typing impairments in our patient.

We reviewed case reports of isolated dystextia and dystypia due to stroke in the English literature on Medline (Table 3). Several hypotheses have been proposed to explain the underlying mechanisms of dystypia. Otsuki et al. [3] hypothesised that dystypia is caused by a disturbance in the conversion of phonological information to an appropriate corresponding performance in a Japanese patient. Additionally, Ogura et al. [6] reported that a Japanese patient exhibited disabilities in Romaji writing but not in Kana or Kanji writing; additionally, the patient exhibited typing disabilities. In addition, several cases of Japanese patients with dystypia who showed impairment in the Romaji spelling system have been reported [7–9]. In contrast, Ryu et al. [10] reported a case of a Korean patient with dystypia associated with visuospatial memory impairment. In our patient, we did not evaluate visuospatial memory in detail; however, our patient required a longer time for dictation in Romaji than in Kana, suggesting that this patient had difficulties in syllable-to-Romaji conversion, leading to dystypia. The detailed underlying mechanisms of dystextia have not been reported; however, texting with a smartphone also requires syllable-to-Romaji conversion. Thus, dystextia may be caused by the same difficulties as dystypia.

The lesions responsible for dystypia can be roughly divided into two categories. One is the left frontal lobe [3, 6, 10], and the other is the left temporoparietal lobe [8, 11]. With respect to Romaji conversion impairment, Ogura et al. [6] reported an infarct affecting the pars opercularis of the inferior frontal gyrus, extending to the foot of the anterior precentral gyrus in the left hemisphere. Although the patient described by Maeda et al. [8] exhibited a left angular infarction, the patient also exhibited decreased CBF in the bilateral frontal lobes. Therefore, the left frontal lobe may play a role in Romaji conversion in Japanese individuals. In our patient, the infarct lesion on MRI was restricted to the left thalamus and the left corona radiata. However, SPECT revealed decreased CBF in the left dorsolateral frontal lobe, subcortical area of the right cerebral hemisphere, and the right cerebellar hemisphere. Among these hypoperfusion areas, the left frontal lobe was thought to have caused Romaji conversion impairment in our patient.

A patient with dystypia and dystextia described by Adachi et al. [12] exhibited hypoperfusion in the left inferior precentral and posterior inferior frontal gyri on SPECT, although a cerebral infarction restricted to the left corona radiata and the putamen was observed on MRI. Therefore, it was hypothesised that the left subcortical infarction caused diaschisis of the left frontal lobe in their case. In our patient, hypoperfusion of the left frontal lobe was primarily considered to be associated with diaschisis due to subcortical infarction, although stenosis of the left internal carotid artery may have partly contributed to the hypoperfusion of the left cerebral hemisphere. Additionally, the thalamus has been reported to correlate with both the contralateral cerebral and contralateral cerebellar hemispheres [13, 14]. Thus, hypoperfusion of the right cerebral and cerebellar hemispheres was thought to be caused by diaschisis in our patient.

Abbreviations

CBF	Cerebral Blood Flow
MRA	Magnetic Resonance Angiography
MRI	Magnetic Resonance Imaging
SPECT	Single Photon Emission Tomography
^{99}Tc-ECD	Technetium-Ethyl Cysteinate Dimer
TMT-J	Trail Making Test, Japanese edition

Acknowledgements
None.

Author contributions
MH: Conception, Acquisition, Drafted the work, TK: Analysis, ST: Acquisition, TK: Acquisition, ST: Acquisition, MK: Revised the work, OO: Revised the work.

Funding
The authors have no funding sources to declare.

Data availability
The data that support the findings of this study are available from the corresponding author, MH, upon reasonable request.

Declarations

Consent for publication
Written consent was provided by the patient prior to the publication of this case report.

Competing interests
The authors declare no competing interests.

Received: 19 March 2024 / Accepted: 30 September 2024
Published online: 05 October 2024

References

1. Cawood TJ, King T, Sreenan S. Dystextia – a sign of the times? Ir Med J. 2006;99:157.
2. Nakamura K, Kuroha Y, Hatakeyama M, Kimura AM, Nakamura Y, Murakami Y, et al. Corticobasal syndrome mimicking Foix-Chavany-Marie syndrome with suggested 4-repeat tauopathy by tau PET. BMC Geriatr. 2023;23:838.
3. Otsuki M, Soma Y, Arihiro S, Watanabe Y, Moriwaki H, Naritomi H. Dystypia: isolated typing impairment without aphasia, apraxia or visuospatial impairment. Eur Neurol. 2002;47:136–40.
4. Chen WC, Huang P. Selective dystextia secondary to a left frontal hemorrhagic infarct. Neurol Sci. 2023;44:749–51.
5. Sakurai Y, Uchiyama Y, Takeda A, Terao Y. On-Reading (chinese-style pronunciation) predominance over Kun-Reading (native Japanese pronunciation) in Japanese semantic dementia. Front Hum Neurosci. 2021;15:700181.
6. Ogura K, Fujii T, Suzuki K, Mori E. Pure agraphia in Romaji after left inferior frontal gyrus infarction: a case of selective deficit in syllable-to-grapheme conversion in Japanese. Brain Lang. 2013;127:1–5.
7. Suzuki Y, Inatomi Y, Yonehara T, Hirano T. Dystypia after ischemic stroke: a disturbance of linguistic processing for Romaji (Romanized Japanese)? Rinsho Shinkeigaku. 2015;55:8–12.
8. Maeda K, Yamamoto Y, Shiraishi T, Fujioka E. Dystypia associated with diaschisis of the middle frontal gyri after left angular infarction. J Stroke Cerebrovasc Dis. 2021;30:105803.
9. Yamamoto W, Inatomi Y, Matsuda M. Dystypia in a patient with subcortical ischemic stroke. Rinsho Shinkeigaku. 2024;64:163–70.
10. Ryu DW, Kim JS, Yang DW, Kim YI, Lee KS. Dystypia without aphasia associated with visuospatial memory impairment in a patient with acute stroke. Alzheimer Dis Assoc Disord. 2012;26:285–8.
11. Cook FA, Makin SD, Wardlaw J, Dennis MS. Dystypia in acute stroke not attributable to aphasia or neglect. BMJ Case Rep. 2013;2013.
12. Adachi H, Numata J, Nishida K, Yamasaki H, Futamura N. Dystextia and dystypia due to phonological errors after ischemic stroke: a case report in a Japanese patient. Neurocase. 2024;29:50–7.
13. Zhang D, Snyder AZ, Fox MD, Sansbury MW, Shimony JS, Raichle ME. Intrinsic functional relations between human cerebral cortex and thalamus. J Neurophysiol. 2008;100:140–8.
14. Förster A, Kerl HU, Goerlitz J, Wenz H, Groden C. Crossed cerebellar diaschisis in acute isolated thalamic infarction detected by dynamic susceptibility contrast perfusion MRI. PLoS ONE. 2014;9: e88044.

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