

NOTE

PURE ANARTHRIA WITH PREDOMINANTLY SEQUENCING ERRORS IN PHONEME ARTICULATION: A CASE REPORT

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ABSTRACT

A 77-year-old left-handed man presented with pure anarthria following cerebral infarction. The lesion was restricted to the right precentral gyrus extending to the immediately underlying subcortical white matter and the frontal part of the insular cortex. Qualitative analysis of anarthria revealed that half of the phonemic-articulatory errors in spontaneous speech were sequencing ones. Sequential errors were detected at the phoneme level in both consonants and vowels, and at the syllable level. Most of the sequential errors were pre-positioning. Sequential errors were observed both within and across words. In clear contrast with anarthria, writing and comprehension was preserved, which suggested the problem was limited to oral expression. Our findings provide further support that the precentral gyrus and/or the insular cortex of the language dominant hemisphere is responsible for the temporal sequencing of the articulatory programming.

Key words: pure anarthria, precentral gyrus, insular cortex, articulatory sequencing

INTRODUCTION

Pure anarthria (Marie, 1906) or aphemia (Albert et al., 1981) is a phonemic-articulatory disorder (De Renzi et al., 1966) in which patients cannot properly articulate words or sentences. It differs from dysarthria because of the irregularity of the phonemic-articulatory deviations, which one cannot anticipate when and in what manner they occur. Dysarthria implies a distortion in sound production causing stable and predictable articulatory deviations, due to disordered muscular action of the human buccophonatory apparatus (Yamadori, 1985). Anarthria is most often seen as a partial symptom of Broca's aphasia. Programming of articulation was once regarded as a function of Broca's area, but several reports on pure anarthria have emphasized the importance of the precentral gyrus for articulation. There are only four reported cases of pure anarthria with lesions localized in the precentral gyrus, three of which had accompanying insular lesions (Sugishita et al., 1987; Lecours and Lhermitte, 1976; Tonkonogy and Goodglass, 1981; Saito et al., 1997). Lecours and Lhermitte (1976) argued that articulatory disorders were caused by a lesion in the Rolandic operculum causing disconnection between the foot of the inferior frontal gyrus and Broca's area through damage of the U-fibers. Tonkonogy and Goodglass (1981), however, reported a patient with lesions limited to the bilateral inferior frontal gyrus who did not show articulatory problems. In contrast, another case of their series developed articulatory impairment following a small infarction in the lower part of the left precentral gyrus. These reports seem to indicate that the lower part of the dominant precentral gyrus or the Rolandic operculum, but not Broca's area, is responsible for anarthria.

Other studies reported the importance of insular lesion for the impairment of articulation. Dronkers (1996) examined patients with and without apraxia of speech and

showed the double dissociation: the anterior insula was involved in patients with apraxia of speech but it was spared in patients without apraxia of speech. It was proposed that the left anterior insula is specialized in motor planning of speech. A study using positron emission tomography (PET) showed that the left anterior insula and the left premotor cortex were both activated in articulation (Wise et al., 1999).

Importance of the insula for articulation, however, remains to be settled. In clinical studies, lesions confined to the insula are very rare, because the insula is surrounded by many cortical and subcortical structures including the precentral gyrus, which share common vascular supply. Penfield and Faulk (1955) reported that total excision of the insular cortex did not result in phonemic-articulatory disorder.

Patients with pure anarthria are rare, and few case reports included sufficient description of both clinical findings and lesion localization. Thus, the precise role of each cortical structure in articulation is still unclear. We performed a systematic examination of a patient with pure anarthria in order to pin down the qualitative feature of the symptoms and the precise localization of the lesion.

CASE REPORT

A 77-year-old, left-handed, retired male executive with 16 years of education was referred to the Rehabilitation Department of Tohoku University Hospital, Japan, for evaluation and treatment of anarthria. His previous medical history included atrial fibrillation, glucose intolerance, and bronchial asthma. After retirement, he often volunteered for community services. He also enjoyed publishing a monthly private magazine for stamp collectors. He was a decent writer who kept a diary every day and frequently corresponded with his friends. Two days before admission, he suddenly became mute just after measuring peakflow of expiration to monitor his lung condition. He showed no disturbance in auditory and reading comprehension and could write sentences with some errors in kana (syllabogram). When eating, he felt food stuck in the left side of the mouth. He noticed no other disturbance in motor or sensory functions. On that day, he visited an outpatient clinic where a diagnosis of cerebral infarction was made. Six days later, he was referred to our service. Detailed neuropsychological examination was performed over the next two months. During this period, he showed some improvement, but the characteristic phonemic-articulatory disorder remained unchanged. Neurological examination revealed an alert, oriented and cooperative man with anarthria. Cranial nerves, motor, sensory and cerebellar functions were normal. Deep tendon reflexes were symmetrical without pathological reflexes.

Brain magnetic resonance imaging (MRI) performed 4 days after the onset revealed an area of high signal intensity in the right precentral gyrus extending to its subcortical region and the frontal part of the insula on fluid-attenuated inversion recovery (FLAIR) images and T2-weighted images (Figure 1 and Figure 2). Another lesion in the left parietal lobe showed hyperintensity on FLAIR and T2-weighted images and a cortical laminar necrosis on T1-weighted images, which suggested an older infarction compared to the frontal one. Measurement of the blood flow with 99mTc-ECD single photon emission computed tomography showed hypoperfusion in the left frontal area near the central sulcus.

NEUROPSYCHOLOGICAL ASSESSMENT

The patient was alert, cooperative and oriented in time and space. His digit span forward was 5. He obtained a full-scale IQ of 119, verbal IQ of 131, and performance IQ of 103 on the Wechsler Adult Intelligence Scale-Revised. He scored 31.5 out of 36 on the abbreviated Token Test. He was left-handed with L.Q. of -70 by the Edinburgh handedness inventory (Oldfield, 1971). His speech was marked with phonemic-articulatory and prosodic difficulties that could not be attributed to dysarthria, especially when he had to produce long sentences. No apparent abnormalities were observed in auditory and reading comprehension. On the Japanese edition of the Western Aphasia Battery 1 month after the onset, he scored nearly perfect except slight impairment in fluency and repetition tasks. He

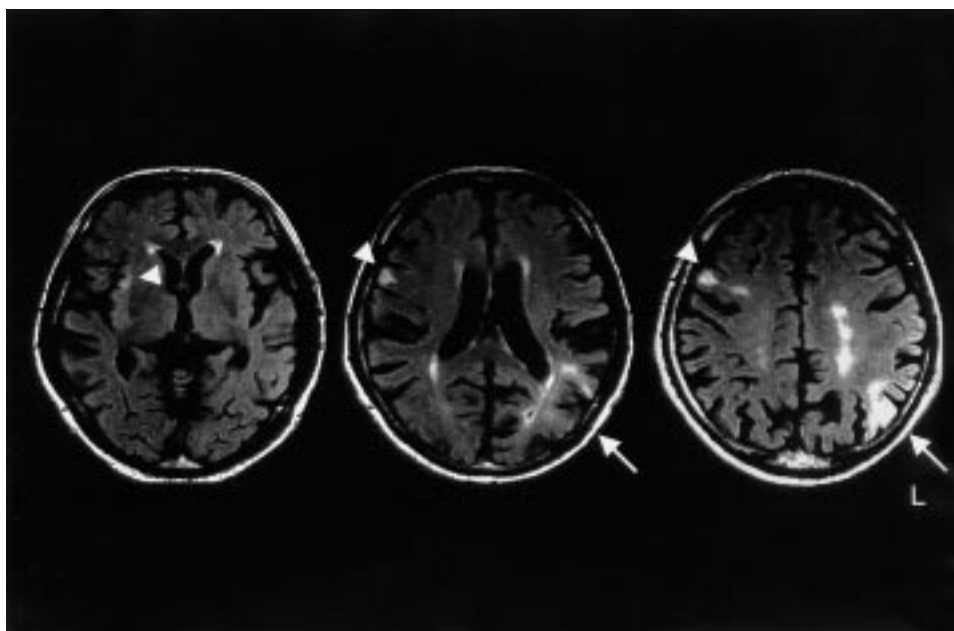


Fig. 1 – Axial fluid-attenuated inversion recovery (FLAIR) MR images obtained on the day of admission showing hyperintensity areas in the right precentral gyrus extending to the underlying subcortical white matter and the insular cortex (arrow heads). An additional hyperintensity area is observed in the left parietal cortex (arrows).

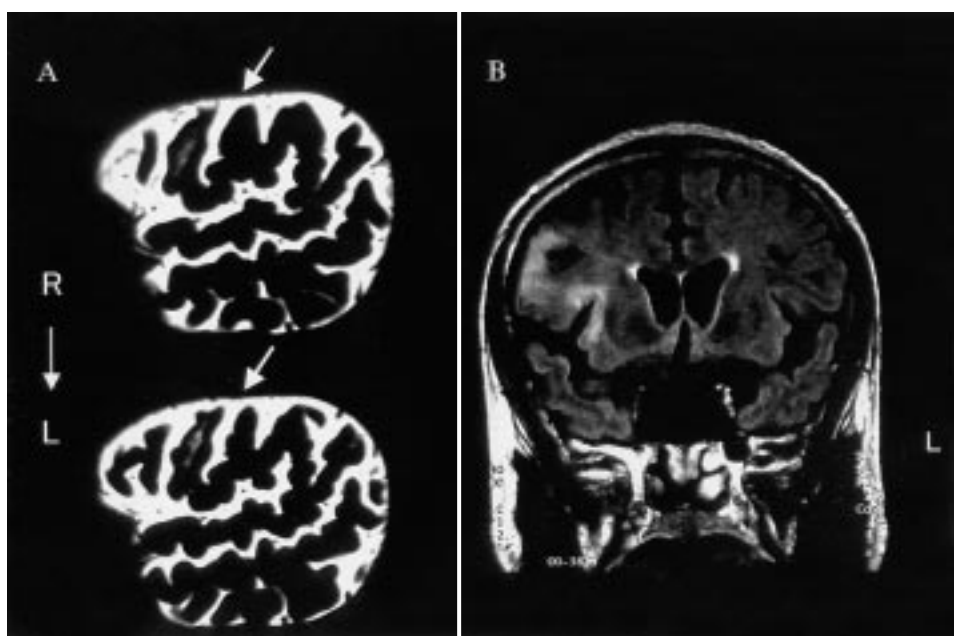


Fig. 2 – A: Sagittal T2-weighted MR images obtained one month later. Hyperintensity area in the right precentral gyrus is noted. The lesion is located mainly on area 6. Arrows indicate the central sulcus. B: A coronal FLAIR MR image showing hyperintensity area in the right precentral gyrus and the insula.

showed no evidence of acalculia, visual agnosia, buccofacial apraxia, ideomotor apraxia, ideational apraxia, or disturbance in constructional ability.

EXPERIMENTAL INVESTIGATIONS

Oral Expression

His speech was anarthric according to Marie's terminology, namely, effortful, nonfluent and dysprosodic though mostly comprehensible. Grammatical structure was well preserved. He was aware of his own disturbances and repeatedly tried to correct them with some improvement. Distortions in articulation were irregular and varied from time to time and from word to word. Anarthria was most prominent in spontaneous speech and reading aloud sentences. His articulation was relatively good at the syllable and word level. He could articulate 46 basic Japanese syllables fairly clearly when he pronounced them separately. Repetition of separate words and object naming were fair.

We further tried to examine the qualitative features of his anarthria using his spontaneous speech recorded on audiotape. We classified phonemic-articulatory error types into sequential and non-sequential. Sequential errors were defined as those caused by inadequate order of phonemes or syllables, in which the distance between the original position of a target and its actual position was within 2 syllables. They were subdivided into pre-positioning, post-positioning, and metathesis according to La Pointe and Johns (1975). Two experienced neurologists and a speech pathologist transcribed the speech sample. First, one examiner orthographically transcribed the 5-minute speech sample. The resulting transcript was independently verified or modified by two other examiners, yielding a total of three transcripts. The three transcripts were then compared and consolidated in order to produce a composite transcript that reflected agreement of at least two of the three examiners. No further analysis was performed on the utterances for which no agreement was reached (approximately 10% of the error classifications were not agreed and were excluded from the analysis). In case the patient discontinued articulating a word halfway, errors in the discontinued word were evaluated as far as it was intelligible from the context of the sentence, or by his corrected utterance.

Table I shows the results of this qualitative error analysis. About half of the phonemic-articulatory errors were sequential. Sequential errors were detected at the phoneme level both on consonants and vowels, and at the syllable level (Figure 3A, 3B). Most of the sequential errors were pre-positioning. Sequential errors were observed across words (as in Figure 3A), and were also observed within words (as in Figure 3B). A few pre-positioning errors were seen in compound words characterized by 'rendaku'. 'Rendaku' is a well-documented phenomenon in Japanese phonology. An initial obstruent consonant, which is voiceless when the word is used as an independent unit, becomes voiced when the same word is used as the second member of a compound word (e.g., /ori/ + /kami/ → /origami/ 'paper folding') (Fukuda and Fukuda, 1999). For example, /sanbjakurokudzju:/ '360' is a compound word, which is composed of /san/ 'three', /haku/ 'hundred', /roku/ 'six', and /dzju:/ 'ten'. /haku/ is usually voiced to be /bjaku/ when it comes next to /san/ in a compound form. Interestingly, pre-positioning occurred in its original (voiceless) form to replace /san/ with /hjan/ instead of /bjan/.

TABLE I
Distribution of Phonemic-articulatory Error Types

Error category	Percentage of total errors
Non-sequential	47.3
Sequential (total)	52.7
Sequential (pre-positioning)	44.6
Sequential (post-positioning)	6.8
Sequential (metathesis)	1.3

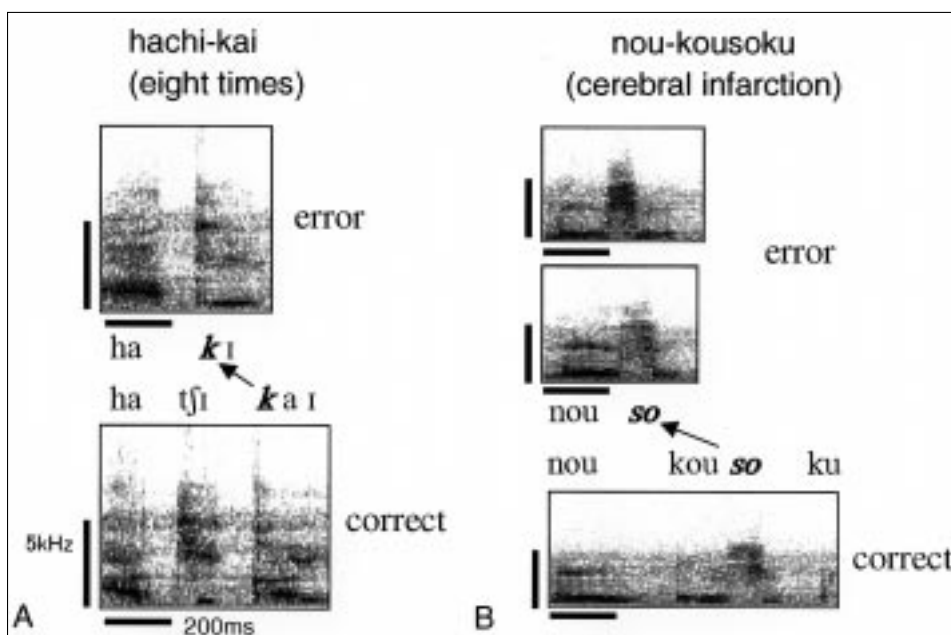


Fig. 3 – Examples of sequential errors (pre-positioning). A: Consonant 'k' of the syllable 'ka' was pre-positioned and merged with vowel 'i' of the preceding syllable 'chi'. In the next trial the patient could articulate the target word. Pre-positioning was seen across words. B: Syllable 'so' was pre-positioned twice, and then he succeeded in articulating the target word correctly. Pre-positioning occurred within a word. Hyphens indicate boundary of words.

Written Expression

Although he could write from the very beginning of the disease, he noticed that he tended to substitute or omit kana characters. He could detect and correct all of the mistakes by himself. The following translated example of his letter written on the next day after the onset showed that the contents were richly expressed and grammatical structures were perfect.

“Dear friend,

Flowers in my garden are starting to bloom and I heard cherry-blossoms have just come out in Sendai. While spring is really here, something strange happened to me. Yesterday afternoon I suddenly became unable to speak. I ran into a clinic nearby and was further examined in another clinic. This morning I was told that I had an attack of cerebral infarction. Although I have lost my voice, other parts of my brain function well. But when I write sentences, I often make mistakes in hiragana characters and have to correct them.”

Difficulty in writing kana improved quickly and no mistakes were observed during our neuropsychological examinations.

DISCUSSION

The present case was characterized by pure anarthria, which could be clearly confirmed through intactness of writing and comprehension. On the MRI, a well-demarcated, restricted lesion was found in the right precentral gyrus extending to the insular cortex.

Qualitative analysis of anarthria revealed severe sequential errors. La Pointe and Johns (1975) reported that sequential errors existed in patients with apraxia of speech but their

proportion was relatively small; between 1 and 16% of all the errors. Only one case in the literature is reported (Sasanuma, 1971) in which more than 70% of total phonemic-articulatory errors were classified as sequential. MR images performed later revealed a cortical lesion in the left precentral gyrus and the insula extending to the underlying subcortical region and the caudate nucleus (Kawachi, 1987). Lesions common both to Sasanuma's (1971) and our case were the precentral gyrus and the insula. These regions may be responsible for articulatory sequencing, which codes the proper timing for each phoneme and syllable.

Monoi et al (1983) reported that sequential errors (transposition) were frequently seen in patients with conduction aphasia. Although sequential errors in patients with conduction aphasia are superficially similar to those in our case, they are mainly paraphasic errors. They may be caused in the phonological system, as writing also includes sequential errors which usually parallel those observed in spoken output (Luria, 1966). In contrast, anarthria is likely caused at the level of articulatory control, when letter-grapheme transcriptions are already completed. Although our case showed slight difficulty in writing at the acute stage, it resolved very quickly. Thus, it seems that the sequential errors seen in our case are distinctly different from those seen in conduction aphasia.

Studies of non-speech movements in primates and humans demonstrated that the supplementary motor area (SMA) is important in executing sequential movements (Tanji and Shima, 1994; Tyzka et al., 1994). However, speech disturbances related to lesions in the SMA include poor initiation of speech and echolalia, but not sequential errors (Ziegler et al., 1997). Reporting a case with a lesion in the SMA, Ziegler et al. (1997) concluded that the speech impairment was not a problem of linguistic processing proper, but rather, a problem of initiating speech. These findings suggest that SMA is not directly involved in articulatory sequencing.

There is no behavior comparable to human articulation in other primates, which makes it difficult to study how articulation is processed in the brain. Primates can make innate calls but these are profoundly different from speech articulation. Articulation is a temporospatially controlled, learned, finely tuned vocal movement. In that respect, although phylogenically separated, birdsong has much to share with human articulation. Birdsong carries very limited amounts of meaning, and there are points that cannot permit simple comparison (Doupe and Kuhl, 1999). Yet it is possible that the basic principle underlying their mechanical aspects has something in common with humans. Vu et al. (1994) produced sequential errors in birdsong essentially the same as those seen in the present case, by stimulating a structure called HVC in zebra finches. They also showed that, when stimulating a structure called RA, syllables were distorted but the overall temporal pattern was not affected. HVC and RA are considered to correspond roughly to the human motor association areas and the primary motor area, respectively. Based on this analogy, we presume that the precentral gyrus and/or the insula perform similar functions as those of songbirds' HVC. If this is the case, rostral lesions in the precentral gyrus (involving area 6) are more likely to cause sequential errors, and caudal lesions (involving area 4) to cause distortion of phonemes. This could be a good model to explain why there are cases of pure anarthria with mainly sequential or nonsequential errors.

Whether the articulatory programming, which may take place in the precentral gyrus and/or the insula, involves purely motor processes or linguistic processes in addition remains unclear. We observed that the first syllable in a second member of a compound word which usually should be voiced ('rendaku'), was prepositioned without being voiced. 'Rendaku' is a productive morpho-phonological operation of Japanese (Fukuda and Fukuda, 1999). Our observation suggests that the sequencing of phonemes is determined before phoneme pronunciations are programmed. Thus symptoms in our case involved disturbances not only in the motor component of articulation but also in the linguistic processing. The precentral gyrus and/or the insula of the language dominant side seem to play roles in linguistic processing. This is why we prefer to use the term "pure anarthria" rather than "apraxia of speech".

Our case, a left-handed man, demonstrated pure anarthria with a right-sided lesion. It is possible that anomalous distribution of language function in this case affected his symptoms. Mori et al. (1989) proposed that the cortical localization of language functions tended to be

anomalous in cases with pure anarthria. There are two possible mechanisms for causing pure anarthria. First, the region responsible for articulation is lateralized to the opposite side with respect to areas subserving other language functions. In that case, large lesions can affect only articulation and spare other linguistic processes. Secondly, a small region responsible for articulation is selectively damaged in the language areas located in one hemisphere. Various language functions in the present case seem to be localized in the right hemisphere, since the previous infarction in his left parietal lobe did not cause any language disturbances. Furthermore, the lesion in the right frontal lobe was limited to a small area that was a mirror image of the lesion observed in a right-handed case with similar symptoms (Sasanuma, 1971). Considering the small lesion in the strategically important area for articulation, the second hypothesis seems to be adequate to explain pure anarthria in this case.

In conclusion, the present case suggests that the lower part of the precentral gyrus and/or the insula of the language dominant side plays an essential role in coding the overall sequential structure of articulation. Articulation of each phoneme presumably is coded in the primary motor cortex, thus forming a hierarchical programming process for human articulation.

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