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HIGHLIGHTS

- Anarthria could be caused by a lesion not related to language lateralization.
- This case suggests that bilateral hemisphere work together in articulation network.
- This case will be considered on neural correlates of anarthria lesion.

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Transient Anarthria in a Patient With Non-Dominant Hemispheric Lesion: A Case Report

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ABSTRACT

Anarthria is a complete loss of speech. It usually results from lesions in bilateral neural substrates that control articulation. Recently, lateralized cortical control of speech articulation in the dominant hemisphere has been reported. However, anarthria by non-dominant hemispheric lesion has not been reported yet. Here we report a rare case of transient anarthria caused by right hemispheric infarction after brain surgery in a right-handed patient. This report suggests that anarthria could be caused by a lesion not related to language lateralization. This report is expected to contribute to studies on neural correlates of anarthria lesions.

Keywords: Anarthria; Articulation; Dysarthria; Corticobulbar Tract; Non-Dominant Hemisphere

INTRODUCTION

Speech production requires coordinated movement of phonatory and articulatory muscles for precise articulation [1]. Dysarthria is a motor speech disorder resulting from impaired neuromuscular control over speech production [2]. The most severe form of dysarthria is anarthria meaning a complete loss of speech. Patients with anarthria might have intact language without speech production or cognitive function. They have a desire to communicate in spoken language [2,3]. Because loss of verbal communication ability could negatively affect the quality of life [4], some researches have tried to make a neuroprosthesis decoding speech from the cerebral cortical activity for patients with anarthria based on the neural pathway of the human articulation system [5].

Motor act of speech is primarily mediated by the motor cortex. The Broca area only formulates the articulatory code for motor cortex execution [6]. Compared with aphasia, anarthria has been usually described as bilateral lesions such as bilateral anterior choroidal artery territory infarction involving corticobulbar tracts [7,8], ischemic bilateral opercular syndrome [9], and ischemic stroke of pyramidal decussation [10]. Anarthria can also be induced by unilateral dysfunction of the ventral premotor cortex in the language dominant hemisphere [11,12]. This finding is consistent with results of a neuroimaging study showing that anterior insula and premotor cortex of the language dominant hemisphere are

Conflict of Interest
The Corresponding author of this manuscript is an editor of *Brain & NeuroRehabilitation*. The Corresponding author did not engage in any part of the review and decision-making process for this manuscript. The other authors have no potential conflicts of interest to disclose.

specialized in articulatory planning [13]. However, there have been few reports on anarthria in patients with a unilateral language non-dominant hemispheric lesion. Herein, we report a rare case of anarthria caused by right hemispheric infarction after a brain tumor surgery in a right-handed patient.

CASE REPORT

A 53-year-old woman with a 3-day history of headache and mild dysarthria was admitted to the Department of Neurosurgery. She was right-handed. She had no previous history of a neurological disease. T2-weighted image of brain magnetic resonance imaging (MRI) showed 5.7-cm sized meningioma in the right frontoparietal lobe and peritumoral edema causing mass effect (**Fig. 1**). Consequently, she received craniotomy and gross total tumor removal. Intraoperatively, the distal branch of the right middle cerebral artery was placed at the posteromedial margin of the tumor. Adhesion of the distal branch to the arachnoid membrane of the tumor was noted. In addition, the brain cortex adjacent to the meningioma was friable due to edema caused by tumor compression. After surgery, she developed anarthria and left central-type facial palsy. There was no speech production except occasional sound which was not understandable. However, she was able to communicate in hearing and writing. Slight weakness of left peri-oral facial muscle was observed in spontaneous facial expression; however, facial asymmetry due to muscle weakness was absent at rest. She could protrude her tongue with slight left deviation. On postoperative day 3, video-fluoroscopic swallowing study (VFSS) was performed and there was no significant impairment of mastication and deglutition on VFSS.

At 2 days after the operation, follow-up brain MRI demonstrated acute ischemic lesion in the medial margin of the tumorectomy site with persistent cerebral edema. There was no other intracranial lesion (**Fig. 2**). This lesion was located in the right primary motor cortex responsible for motor control of the face area (**Fig. 3**). When compared with pre-operative MRI images (**Fig. 1A**), sulcal effacement including the sulci surrounding the Broca's area disappeared apparently in post-operative MRI (**Fig. 2I**) which implies decreased intracranial pressure after the surgery. However, cerebral edema involving white matter in the right fronto-parietal lobe persisted. To treat the cerebral edema in the right hemisphere,

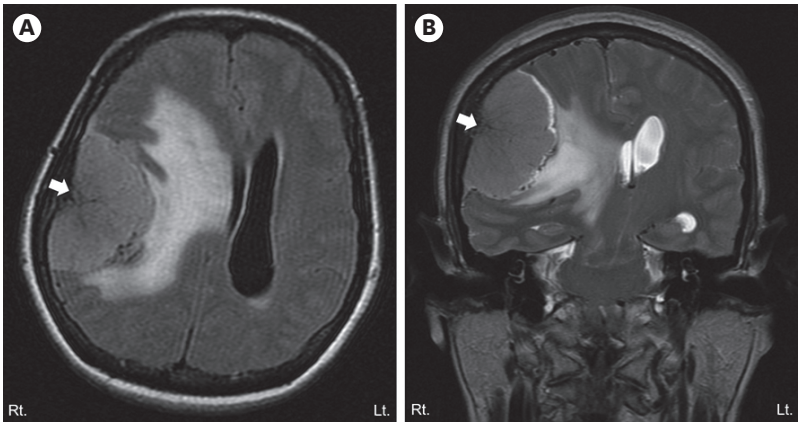

Fig. 1. Pre-operative brain magnetic resonance imaging findings. Axial T2 fluid attenuated inversion recovery image (A) showing a 5.7-cm sized meningioma in the right frontoparietal lobe (arrow) with a peritumoral cerebral edema causing midline shifting. Coronal T2 turbo spin echo image (B) of the meningioma (arrow) is shown. Rt., right; Lt., left.

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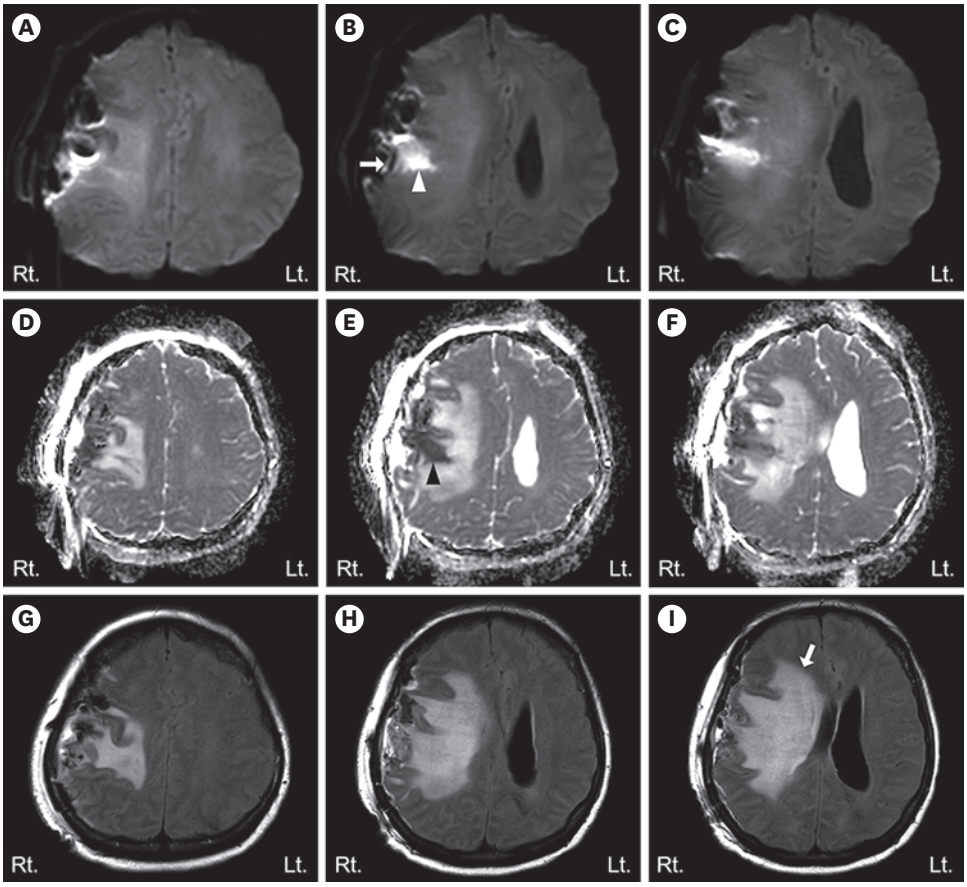

Fig. 2. Post-operative brain magnetic resonance imaging image showing an acute ischemic lesion in the medial margin of the tumorectomy site. (A-C) are axial DWI. (D-F) are axial ADC images. (G-I) are axial T2 FLAIR images. A high signal intensity lesion (white arrowhead) in the precentral gyrus (white arrow is central sulcus) on the DWI (B) and corresponding low values (black arrow) in ADC image (E) are shown. T2 FLAIR images (G-I) showing remnant peritumoral cerebral edema involving white matters adjacent to genu of corpus callosum in the right frontoparietal lobe with midline shifting (white arrow).
Rt., right; Lt., left; DWI, diffusion weighted images; ADC, apparent diffusion coefficient; FLAIR, fluid attenuated inversion recovery.

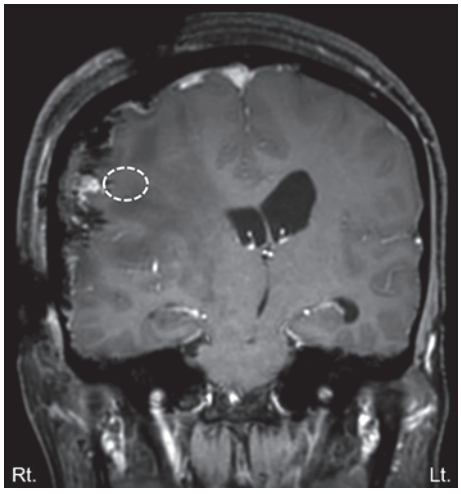

Fig. 3. Post-operative acute infarction lesion in the primary motor cortex of the face region (white dashed circle) and correlates with coronal T1 fat suppression gradient echo image of brain magnetic resonance imaging.
Rt., right; Lt., left.

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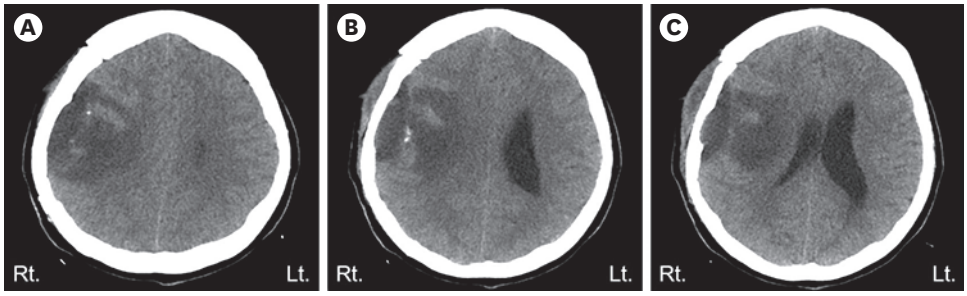

Fig. 4. Axial brain computed tomography scan images at 24 days after surgery. The extent of cerebral edema in white matters of the right hemisphere and the amount of midline shift are decreased.
Rt., right; Lt., left.

dexamethasone was administered by intravenous injection for 7 days and tapered off in oral administration for 11 days. She was able to speak one syllable within 5 days after the steroid therapy. One week after the surgery, she was transferred to the Department of Physical Medicine and Rehabilitation (PMR). Although anarthria was improved and she could speak in short sentences, moderate to severe dysarthria with slurred speech and imprecise articulation persisted on the day she was transferred to the Department of PMR. The feature of apraxia of speech such as self-correction behavior was not observed. On neurological examination, left central facial palsy and tongue deviation to the left side were observed. There was no motor or sensory deficit for any of the 4 extremities. Edinburgh handedness inventory revealed dominance of the right hand. Laterality quotient was 100. Language function was assessed with a Korean-Western Aphasia Battery. The aphasia quotient (AQ) was 86.8. Writing skill was spared. She received comprehensive rehabilitation including speech therapy. The frequency of speech therapy was twice per week and duration of each session was 30 minutes. Follow-up speech and language evaluation were performed after 2 weeks of speech therapy (total 4 sessions). Mild dysarthria was noted and production accuracy of Korean consonant category was 77%. Her speech intelligibility was improved that she was able to communicate with others by spoken language. AQ was increased to 92.6. Aphasia was not suspected. On discharge (at 4 weeks after brain surgery), she only had difficulty in speaking long sentences. Brain computed tomography scan showed decreased extent of cerebral edema in white matters of the right hemisphere (**Fig. 4**). When she was followed up in an outpatient clinic at 2 months after the surgery, mild degree of dysarthria remained.

DISCUSSION

This patient developed a transient anarthria following a right hemispheric infarction after brain meningioma surgery. Brain MRI demonstrated acute ischemic lesion in the primary motor cortex of the face area and large extent of peritumoral cerebral edema. Mild degree of left central-type facial palsy and left glossal palsy were accompanied. However, her language function was preserved. Anarthria showed good recovery. Complete loss of speech persisted for less than 5 days. Only mild dysarthria was noted at 3 weeks after the surgery.

The most common etiology of anarthria was amyotrophic lateral sclerosis (44%) and followed by other neurodegenerative diseases (15%) such as frontotemporal dementia and multiple systemic atrophy [2]. Although the prevalence of dysarthria after acute stroke has been reported to be 41.5% [14], the incidence of anarthria following unilateral hemispheric stroke is not well-known due to its rare occurrence. The primary motor cortex projects

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the corticobulbar tract. Fibers of the corticobulbar tract then go on to the synapse of the hypoglossal nerve, facial nerve, and trigeminal nerve motor nuclei bilaterally to control phono-articulatory muscles [15]. Therefore, anarthria is typically caused by bilateral lesions of the corticobulbar tract [7]. In case reports, anarthria could be developed by unilateral cerebral infarction of the language dominant hemisphere [16]. Meta-analysis of functional neuroimaging studies has demonstrated core components of human speech production network including the Broca area (Brodmann area 44), anterior insula, caudate nucleus, cerebellum, premotor cortex (Brodmann area 6), and face region of M1 (Brodmann area 4) and their connectivity [17]. Lesion-based analysis [18] and functional neuroimaging studies [13] have revealed lateralization in speech articulation, with left insula and left premotor cortex in a right handed person having a dominance in motor planning of articulation. These studies have elucidated the mechanisms by which anarthria may develop in a unilateral language dominant brain lesion.

However, the case in this report was unusual in that anarthria was caused by infarction in the face region of a language non-dominant primary motor cortex. The left hemisphere of her brain was supposed to be dominant for language considering her right-handedness and preserved language function despite of large extent of cerebral edema in the right hemisphere. The Broca's area of the dominant hemisphere might be compressed by the elevated intracranial pressure pre-operatively. However, dysarthria was aggravated immediately after the tumor removal and there was no significant clinical symptom or sign for increased intracranial pressure. This clinical course was not correlate with the possibility with the increased intracranial pressure. In addition, Foix-Chavany-Marie syndrome (FCMS) could be considered in the differential diagnosis of anarthria in this patient, which is characterized by bilateral severe weakness of orofacial muscles with automatic-voluntary dissociation [19]. However, this case showed mild weakness of unilateral oro-facial muscles and these clinical findings were different from the features of FCMS. Development of anarthria in this report could not be explained in the absence of interhemispheric connectivity in speech production network because her corticobulbar tract in the left hemisphere was intact. The primary motor cortex works in concert with feedback control system composed of somatosensory and auditory cortex in the bilateral hemisphere for tuned speech corrective motor commands [20]. Previous study has supported the hypothesis that combined function of bilateral hemisphere is crucial for the process in articulation network and emphasized the role of ventral premotor cortex [21]. Anatomically, there is homotopic connection between lateral frontal cortices including posterior third of middle frontal gyrus, inferior frontal gyrus and ventral premotor cortex which is mediated by ventro-lateral fibers of anterior corpus callosum [22]. One possible mechanism for this case could be that anarthria was induced by the unilateral corticobulbar tract lesion in the presence of a pre-existing partial interruption in interhemispheric connection of speech processing network due to extensive peritumoral cerebral edema adjacent to anterior corpus callosum in the right hemisphere (**Fig. 2I**). Unlike a previously reported case of bilateral corticobulbar tract infarction with anarthria remained at 3 months of follow-up [8], the present case showed a good prognosis in that the anarthria improved to mild dysarthria within 3 weeks. Anarthria in this case might have improved promptly because the interhemispheric connection of speech processing network was recovered fast as the amount of cerebral edema decreased. Diffusion tensor image was not performed in this case. Thus, the absence of tractography is the limitation of our study. Further neuroanatomy studies are needed to clarify the interconnectivity between bilateral hemispheres in the neural network of speech production.

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We described a patient who presented with transient anarthria following infarction involving language non-dominant hemisphere. This report suggests that anarthria could be caused by a lesion not related to language lateralization and that bilateral hemisphere might work together in speech articulation. This report is expected to contribute to studies on neural correlates of anarthria lesion to understand the articulation network.

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