Background: According to recent functional studies, the medial frontal lobe of the dominant hemisphere plays a role in both word generation and speech initiation. To our knowledge, speech arrest with intact facial expression secondary to the right anterior cingulate cortex (ACC) lesion has not been previously reported.

Objective: To report 2 cases of speech initiation difficulties associated with a stroke of the right anterior cingulate gyrus with magnetic resonance imaging with tractography results.

Design: Two case reports.

Setting: Inpatient neurology clinic at a university medical center.

Patients: Two women who had acute and transient speech initiation problems.

Results: Speech evaluation revealed pure speech initiation difficulties with intact facial praxis and expression. In the patient who could be tested, writing ability was preserved. In addition to acute right ACC infarction, the magnetic resonance imaging also revealed anterior corpus callosum and/or posterior corpus callosum involvement. Tractography in patient 2 revealed fibers from the right ACC that would cross to the contralateral side. Reduced fiber numbers connecting the right supplementary motor area with the ACC were also observed, which differed from the left ACC tractography.

Conclusions: To our knowledge, this is the first case series of right ACC stroke with transient speech initiation problems. Because of the rare findings of the tractography, we suggest that in some patients, speech initiation required the participation of the right ACC in addition to the language network of the left hemisphere.

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IT HAS BEEN ESTABLISHED THAT THE medial frontal cortex plays a role in primate vocalization. Lesion studies have implicated the role of the anterior cingulate cortex (ACC) in spontaneous vocalization in monkeys, which involves either the region anterior and superior to the genu of the corpus callosum or the area above the corpus callosum near the cingulate sulcus. Sutton et al. also demonstrated the effect of lateralization of the hemisphere in primate vocalization and the damage of the ACC with impairment of vocal initiation with stimuli.

Speech initiation difficulties can be attributed to cortical or peripheral abnormalities (eg, neuropathy, myopathy, or vocal cord injuries). Akinetic mutism, a syndrome characterized by little or no spontaneous verbal initiation, has been related to lesions in the left or bilateral medial frontal areas. Reduced spontaneous speech has also been documented in reports after medial frontal surgery, suggesting that resection of the supplementary motor area (SMA) of the dominant hemisphere is critical for transient speech disorders. These findings and recent positron emission tomography studies point to the left ACC and/or SMA as important areas of language generation. However, to our knowledge, there has never been any report of speech initiation problems with lesions in the nondominant ACC. In this article, we will describe 2 cases of transient verbal output loss with a lesion in the right ACC resulting from right anterior cerebral artery territory infarction. We will offer speech evaluation and magnetic resonance imaging and tractography results to support our diagnosis and hypothesis.

REPORT OF CASES

CASE 1

An 82-year-old right-handed woman suffered from acute-onset speech loss and transient left-side weakness. Two weeks
prior to presentation she noticed 2 similar attacks. She had hypertension for 20 years and experienced acute myocardial infarction 2 years earlier. No stroke history was obtained. Upon admission, she was extremely anxious about her inability to speak, though she was capable of coughing and grunting. She was also able to follow all commands during the neurologic examination.

She scored 25 on the Mini-Mental State Examination by writing, because no verbal output could be obtained. She scored 29 of 30 in the Boston Naming Test. The Token test, an auditory comprehension test, revealed 100% word discrimination and 80% complex ideational comprehension. The patient could read the written paragraph and write the summary, but was not able to read them aloud.

The patient's cranial nerves were normal. She could open her mouth, eat food without choking, and suck liquid through a straw. She could perform facial praxis tests, such as forced eye closure, whistling, puffing the cheeks, and mimicking teeth brushing. Her gag reflex was normal. A strong grasp response was noted in her left hand. Laboratory test results showed that her blood cell count, biochemistry, homocysteine level, antinuclear antibody level, Venereal Disease Research Laboratory test, coagulation factor level, and tumor markers (carcinoembryonic antigen, cancer antigen 125, cancer antigen 19-9, and alpha-fetoprotein) were all within normal ranges. The patient underwent laryngoscopy and had normal vocal cord movement. No Pancoast tumor was noted, and thyroid and esophagus survey results were both normal. Combined, these tests ruled out the possibility of peripheral lesions. An electroencephalogram showed some theta activity over the right frontal area without epileptic discharge. Results from magnetic resonance imaging showed increased signal intensities in the right ACC and the anterior and posterior corpus callosum on diffusion-weighted images and T2-weighted images (Figure 1).

Left limb apraxia and grasp response were noted, because admission and muscle power improvement was faster than apraxia and speech ability. Patient 1 was able to walk 35 days after her stroke, but was only able to speak some words or short sentences 3 months postictus. When she attempted to speak in complete sentences, her first few words were clear, but they would trail off and fade. Although her sentences were short in length, the grammar was correct. Paraphasia and the use of jargon were never observed during 2-year follow-up.

CASE 2

A 74-year-old right-handed woman developed acute lower left leg weakness during toileting. Significant history included hypertension for 5 years without regular control and left internal capsule infarction 14 years earlier. She talked and responded to physicians the night she visited the emergency department. A remarkable decrease in word amount was observed the next morning. Her family noticed that she would only stare at people and not re-
spond using words. She could still obey all verbal commands during the neurologic examination, but she tended to respond by nodding or using hand gestures. Only during repeated questioning did she respond in phrases. In contrast to the first patient, she was not anxious about her deficits and would smile when she saw our embarrassed faces. During neurologic examination, she had no apraxia in her face or left limb. She ate and drank as usual. The brain diffusion-weighted images and T2-weighted images performed on day 3 showed an increased signal in the right ACC and anterior corpus callosum (Figure 2). Diffusion tensor images were obtained by using single-shot echo-planar imaging with gradients applied in 25 directions. Axial images were acquired using the following parameters: repetition time, 7000 milliseconds; time to echo, 72 milliseconds; field of view, 24 × 24 cm; matrix, 128 × 128 cm; thickness, 5 mm. Twenty-eight contiguous sections were obtained without an interslice gap. The size of the voxel was 1.8 × 1.8 × 3.0 mm. A B-value of 1000 s/mm² was used for all 25 directions. All selected images were transferred to our offline workstation equipped with Func- tool DTI (GE Medical System, Milwaukee, Wisc) for tractography plotting. Bilateral ACCs were each chosen as the main region of interest. Fibers of the right ACC and SMA were disrupted at the level of the stroke while most of the fibers still crossed the corpus callosum to the left hemisphere and went upward or downward (Figure 3A). In contrast, fibers from the left ACC only ran upward or downward in the ipsilateral hemisphere without connecting any regions in the right hemisphere (Figure 3B).

Patient 2 had no formal education and could not write her name or any Arabic numerals. It took her 1 hour to complete the Mini-Mental State Examination because of slow speech initiation. She scored 21 of 30 questions correctly. The Token test revealed 100% word discrimination and complex ideational comprehension. Her semantic fluency was low, only naming 1 animal in 1 minute and 2 pieces of fruit in 1 minute; however, she scored 27 of 30 items on the Boston Naming Test. It took her 56 seconds to start counting from 1 to 10, but she completed this task in 10 seconds. She could repeat sentences of up to 9 words with no paraphasia. Nine days after the stroke, her response latency shortened. Two weeks after the stroke, she could speak as usual and walk with minimal assistance. In repeated evaluation of semantic fluency, she was able to name 16 animals in 1 minute.

**COMMENT**

The speech disorder in these 2 patients was attributed to difficulties initiating speech without oral-buccal-facial apraxia after acute right ACC stroke. The ACC comprises the limbic cortex in Brodmann areas 24, 25, and 33, and the paralimbic cortex in Brodmann area 32. From sagittal cuts of magnetic resonance imaging, we
found that both of our patients had lesions on the right ACC rostral and superior to the corpus callosum (Brodmann areas 24a and b) that extended caudally to the cingulate sulcus while sparing the SMA area. The first patient’s ability to comprehend sentences, read, write, and name objects by writing was retained while difficulties initiating a sentence were prominent. In the second patient, diminished verbal output was found while retaining grammar ability and sentence comprehension. From our examinations, we had excluded the possibility of peripheral nervous system lesions, and the existence of seizure, Broca aphasia, transcortical motor aphasia, and bulbar and pseudobulbar palsy. We also determined that their speech problems were not caused by buccofacial apraxia, because they were both capable of performing tasks of facial praxis and responded by yelling and laughing to stimuli of pain and jokes, respectively. Both of our patients fit the definition of aphemia, which emphasized the “isolated loss of the ability to articulate words without loss of the ability to write and to comprehend spoken language.”

Figure 3. Tractography from the right anterior cingulate cortex (ACC) as the main region of interest revealed fiber disruption in medial frontal cortex. A, Most of the fibers from the right ACC crossed to the left hemisphere. B, Fibers from the left ACC ran upward or downward, or discontinued at the level of the corpus callosum without connecting to any region in the right hemisphere. (Alignment of figure: right is left, bottom is anterior.)

The ACC of the dominant hemisphere is associated with spontaneous vocalizations, expressing internal states, assessment of motivational content, task error monitoring, and verb generation. One patient reported by Carota et al developed verbal aspontaneity, hypergraphia, and depression after right posterior cingulate (Brodmann area 24c) and corpus callosum infarction. Their patient demonstrated that a lesion of the right posterior cingulate cortex caused impairments in behavioral expression of internal states, prosody, and initiation of spoken language. The involved areas in our patients were located more anteriorly in the right cingulate cortex. In both patients, the frustrated facial expression, sarcastic smile, and exaggerated perioral movement in the absence of spoken language reflected that their ability to express their internal emotional state was still intact. It also revealed the dissociation of verbal and emotional spontaneity in coordinating the same muscle groups of the face and vocal pathway. In patient 2, repeated inquiry helped to provoke a verbal response, which suggested that stronger stimuli help to integrate the remaining environmental monitoring–response network. Other medial frontal structures, such as the SMA, paracingulate sulcus, and dorsolateral prefrontal cortex, are also related to spontaneous speech production. Mark and Ulmer reported a speech-relevant tract in the medial subcallosal fasciculus, which connects the SMA and cingulate gyrus to the striatum and facilitates the initiation, preparation, and limbic aspects of spontaneous speech.
Supportive evidence that the SMA is critical for speech initiation included a recent functional magnetic resonance imaging study that describes how transient speech disorders developed in patients after tumor resection surgery. Another functional magnetic resonance imaging study showed that in healthy right-handed participants, prominent activity was centered in the paracingulate sulcus in a free-word generation task. This rarely extends to the corpus callosum or ACC if paracingulate sulcus activity is present, suggesting the importance of the paracingulate sulcus in word initiation. Extensive activation of the left dorsolateral prefrontal cortex, the ACC, and the SMA were also observed during word generation. It has been less clear what exact role these medial frontal structures played. However, all these structures were lateralized to the left hemisphere and are different from our patients.

Reports regarding right anterior cerebral artery infarction and ACC stroke are not uncommon. However, pure speech arrest has not been mentioned before. Because the ACC has premotor functions and contains neurons projecting into the spinal cord, and given the rarity of reports regarding this area and speech initiation, and the fact that the left hemisphere is the dominant hemisphere for language, we speculated that in some patients, speech initiation required the cooperation of the right hemisphere, which in this study, was the right ACC. It is difficult to conclude that the right ACC is the speech initiation center or one of the speech initiation networks. However, the lesion of the right ACC impaired speech initiation that could be related to disruption of a signal transmission through the right ACC to the left language network. The transient involvement in these 2 patients not only suggested the undergoing neuronal plasticity and recovering of the left homologous regions from diaschisis, but also hinted that the intact left hemisphere was critical for symptom recovery.

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