Brainstem Infarction Presenting with Neurogenic Stuttering

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Abstract

We reported a sixty-year-old man who developed acquired stuttering after a brainstem infarction. Infarctions were detected in the midbrain and upper pons. Neurogenic stuttering of this patients indicated that the midbrain and upper pons could be lesion sites responsible for acquired stuttering. We speculated that the reticular network extending from the brainstem to the frontal cortices, and the periaqueductal gray matter could be closely related regions generating neurogenic stuttering. (Internal Medicine 42: 884-887, 2003)

Key words: brainstem infarction, midbrain, neurogenic stuttering, periaqueductal gray matter, palilalia, acute multiple brain infarction

Introduction

Stuttering is classified into developmental and acquired stuttering. The major causes of acquired stuttering are neurological disorders such as stroke or neurodegenerative diseases. Neurogenic stuttering is reported to occur with damage to the cerebral hemisphere, extrapyramidal system, diffuse affection of the central nervous system (1, 2), basal ganglia (3, 4), corpus callosum (5), and thalamus (6). In most of the previously reported cases of neurogenic stuttering, the hemispheric lesions were either bilateral or left-sided, while right-sided lesions were rare (7). We report in this article that brainstem infarction can cause neurogenic stuttering.

Case Report

A 60-year-old hypertensive man experienced sudden onset of vertigo and was admitted to our hospital. This blood pressure was 133/75 mmHg, and pulse was 53/min and regular. A general physical examination did not show any abnormalities. On neurological examination, he was alert and fully orientated. He had eye movement disorder (paralytic pontine exotropia). He had truncal and limb ataxia, and could not walk. Muscle bulk, tone and strength were normal. Sensation and deep tendon reflexes were normal, while involuntary movements, pathological reflexes, and autonomic dysfunction were absent. He had slurred dysarthria and repetitive speech disorders with the following characteristics. In spontaneous speech, repetition was restricted to syllables mainly at the beginning, but sometimes at the end of a word. Repetition of words or phrases was not observed at that time. The speed of speech gradually increased, while the volume gradually decreased. These disorders were sometimes observed during repetition or reading. For example, he pronounced “so-so-so-soudesu (yes in English)” instead of “soudesu”, “jibundewa (for me) yukkuri (slowly) shabetteiru (speaking) tsu-ts-tsumori (intend to)” instead of “jibundewa yukkuri shabetteiru tsu-tsu-tsumori”, and “kowka (this place) Ezu desu-su-su-su (is)” instead of “kokowa Ezu desu”. Standard Language Test of Aphasia (SLTA) demonstrated that auditory comprehension, naming of objects, reading and writing were preserved. We diagnosed that there were no signs of aphasia. Brain magnetic resonance imaging (MRI) showed multiple high intensity lesions in the midbrain paramedian area and upper pons on T2 and diffusion weighted images (Figs. 1, 2). On MRI and 99mTc-ethylcysteine-dimer (ECD) single photon emission computed tomography (SPECT), there are no abnormal lesions in the thalamus, basal ganglia, nor cerebrum. Vertebral angiography showed severe stenosis at the proximal portion of the right vertebral artery and at the distal portion of the left vertebral artery (Fig. 3). The stroke pattern was classified as atherothrombotic infarction and he was treated with antiplatelet therapy. His stuttering gradually improved and disappeared after four months.
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Discussion

The speech disturbance of the present patient was characterized by involuntary repetition of mainly the first, and sometimes the last syllable of a word, gradual increasing the speed of speech, and gradual decreasing of the loudness of speech (decrement of volume). This disturbance was mainly observed in spontaneous speech and sometimes in repetition or reading. Stuttering or palilalia is a possible explanation of this repetitive speech disorder.

Developmental (congenital) stuttering is a common form of stutter, which is characterized by the involuntary repetition of the first syllable of a word. Initiation of the word is followed either by a machine gun-like repetition (stutter) or the presentation of the syllable followed by a prolonged silence (stammer). In newly acquired stuttering, which is mainly caused by neurological disorders such as stroke or neurodegenerative diseases, repetitions and prolongations are not restricted to the initial syllable (8). Palilalia is one of speech disorders characterized by involuntary repetition of words and phrases during verbal output (8). Gradual increasing the speed of speech, and gradual decreasing of the loudness of speech (decrement of volume) are characteristics of palilalia. We think the speech disturbance in the present patient can be classified as stuttering, because the repetition was limited to syllables, even though his speech disorder contained a component of palilalia.

There are two reports describing repetitive speech disorder due to paramedian thalamic and midbrain lesions. Yasuda et al (9) reported a patient with paramedian thalamic infarcts, who developed repetitive speech disorder, and they concluded that the disorder was palilalia. That patient gradually showed an increased rate of speech, reduced loudness, and repeated individual words or syllables 5 to 10 times. Abe et al (10) reported another patient with repetitive speech disorder resulting from infarcts in the paramedian thalami and midbrain. They described that repetitive speech disorder could be distinguished from stuttering by the points of compulsive repetitions, constant rate and monotonous tone, and from palilalia by the points of repetition restricted to the first syllable, and they concluded it to be “stuttering-like repetition”. The ischemic lesion in our patient exclusively involved the midbrain and upper pons, and the lesion was thought to be an incomplete form of paramedian thalamic and midbrain infarction. This means a lesion of the midbrain and upper pons can play a very important role in inducing repetitive speech disorder in a case of paramedian thalamic and midbrain infarction.

We note the important role of the brainstem in the speech mechanism relating stutter formation. First, the brainstem, basal ganglia, and cerebellum have connections with the frontal motor, premotor, and supplementary cortices, and generate skilled motor movements including speech (11, 12). Andy and Bhatnagar (13) reported that therapeutic mesothalamic stimulation to their four stuttering patients suffering from chronic pain, seizures, and somatosensory disorders also had an ameliorating effect on the stuttering. They indicated that the concurrent amelioration suggests that both
chronic pain and stuttering may be implicated by similar or related reticular electrophysiologic generators, couched in overlapping reticular networks extending from the brainstem to the thalamus, and that the acquired stuttering may be recruited as one component of a larger syndrome complex. Second, a small region of the brainstem, the midbrain periaqueductal gray matter (PAG) is recently proposed to be the crucial brain site for mammalian voice production (14). It can play a role not only in the production of emotional or involuntary sounds, but also as a generator of specific respiratory and laryngeal motor patterns essential for human speech and song, and the lesion of PAG could cause stuttering as improper voice production. In our patient, and in those of Yasuda et al (9), and Abe et al (10), brainstem infarction seemed to extend to the midbrain aqueductal area.

We can find some other case reports that describe brainstem lesions which seem to extend to the midbrain aqueductal area causing neurogenic stuttering or palilalia. Ciabarra et al (15) reported a patient with a small pontine infarct, who developed neurogenic stuttering. The infarct was a linear lesion existing in the upper medial pons which extended to the upper pontine tegmentum, very close to the midbrain tegmentum. Kimura et al (16) reported a patient with neuro-Behçet disease, who developed palilalia. That patient had a linear lesion of the left paramedian midbrain extending to the midbrain tegmentum. It is not clear why some patients present with stuttering and others present with palilalia, even though they have similar lesions. Further accumulation of patients along with further analysis is needed.

The stroke pattern in the present patient is thought to be classified as an atherothrombotic infarction, and it also showed features of acute multiple brain infarction (AMBI) on MRI. AMBI is reported to be often seen in the vertebrobasilar arterial territory and caused by several mechanisms including artery to artery emboli and hemodynamic hypoperfusion (17). The present patient is particularly interesting because AMBI was localized within the brainstem and included a lesion which was classified as branch athero-
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Figure 3. A vertebral angiography showed severe stenosis at the proximal portion of the right vertebral artery and at the distal portion of the left vertebral artery (arrows).

matous disease. The generating mechanism of our patient is thought to be caused by artery-to-artery emboli, because the lesions were not located in the watershed areas of the brainstem.

References