

LETTER TO THE EDITOR

Multifocal Myoclonus as a Manifestation of Acute Cerebral Infarction Recovered by Carotid Arterial Stenting

Hyangkyoung Kim,¹ Jun Soo Byun,² Mark Hallett,³ Hae-Won Shin^{3,4}

Departments of ¹Surgery, ²Radiology, and ⁴Neurology, Chung-Ang University College of Medicine, Seoul, Korea

³Human Motor Control Section, National Institute of Neurological Disorders and Stroke, National Institutes of Health, Bethesda, MD, USA

Dear Editor,

Many types of hyperkinetic movement disorders, including tremor, myoclonus and dystonia, can occur both in the acute and chronic stages of stroke.¹ Among the various movement disorders, myoclonus is rare and occurs after recovery from the acute stage of stroke.¹ We describe a patient who presented with the abrupt onset of myoclonus as a manifestation of contralateral cortical infarction. This patient's symptoms resolved after carotid artery stenting.

A 71-year-old man was admitted due to the abrupt onset of twitching movement of his left hand and face for 7 days and paralysis of his right hand for 3 days. His previous medical history included coronary artery stenting due to acute myocardial infarction 10 years earlier, as well as hypertension and diabetes. The initial neurological examination showed brief, rhythmic and shock-like twitching movements in his left arm and face. The muscle power of his left hand and face was normal. The muscle power in his right hand had decreased to grade IV. Diffusion-weighted brain MRI showed high signal intensity in the left basal ganglia including the caudate nucleus and the putamen, bilateral centrum semiovale and frontal cortices (Figure 1A). Brain CT angiography showed severe stenosis of both proximal internal carotid arteries (Figure 1B). Valproic acid and levetiracetam were administered with antiplatelet medications for the relief of myoclonus. Despite marked improvements, involuntary movements persisted. Transfemoral percutaneous

arteriography showed 88% and 83% stenosis in his right and left internal carotid arteries (ICAs), respectively (Figure 1C). Carotid artery stenting was performed on the patient's left internal carotid artery the day after admission (Figure 1D). Stenting of the right ICA was conducted two days after the procedure on the left ICA (Figure 1D). Beginning the day after the procedure, myoclonus disappeared in the left arm and face. The weakness in the patient's right arm gradually improved over the next month.

The involuntary movements in our patient were continuous, rhythmic and jerky twitching confined to his left arm and face. This muscle twitching was not synchronous but occurred independently in the arm and face. It was neither focal nor generalized. Therefore, the movements might be defined as multifocal myoclonus, which could have both cortical and subcortical origins.² Acute infarction in the right frontal cortex and centrum semiovale in a diffusion-weighted brain MRI suggested that, in this patient, myoclonus might have been caused by cortical and subcortical lesions. The brain MRI also showed acute infarction in the left basal ganglia, centrum semiovale and frontal cortex, which resulted in right arm weakness in this patient. The signal intensity of the infarction in the left side was higher than that on the right side. The symptoms corresponded to the three days from the onset of myoclonus on the left face and arm to the onset of weakness on the right arm.

The time interval between acute stroke and the development

Received: September 14, 2016 Revised: October 26, 2016 Accepted: November 7, 2016

Corresponding author: Hae-Won Shin, MD, PhD, Department of Neurology, Chung-Ang University College of Medicine, 102 Heukseok-ro, Dongjak-gu, Seoul 06973, Korea / Tel: +82-2-6299-1503 / Fax: +82-2-6280-8734 / E-mail: shinhw@cau.ac.kr

© This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

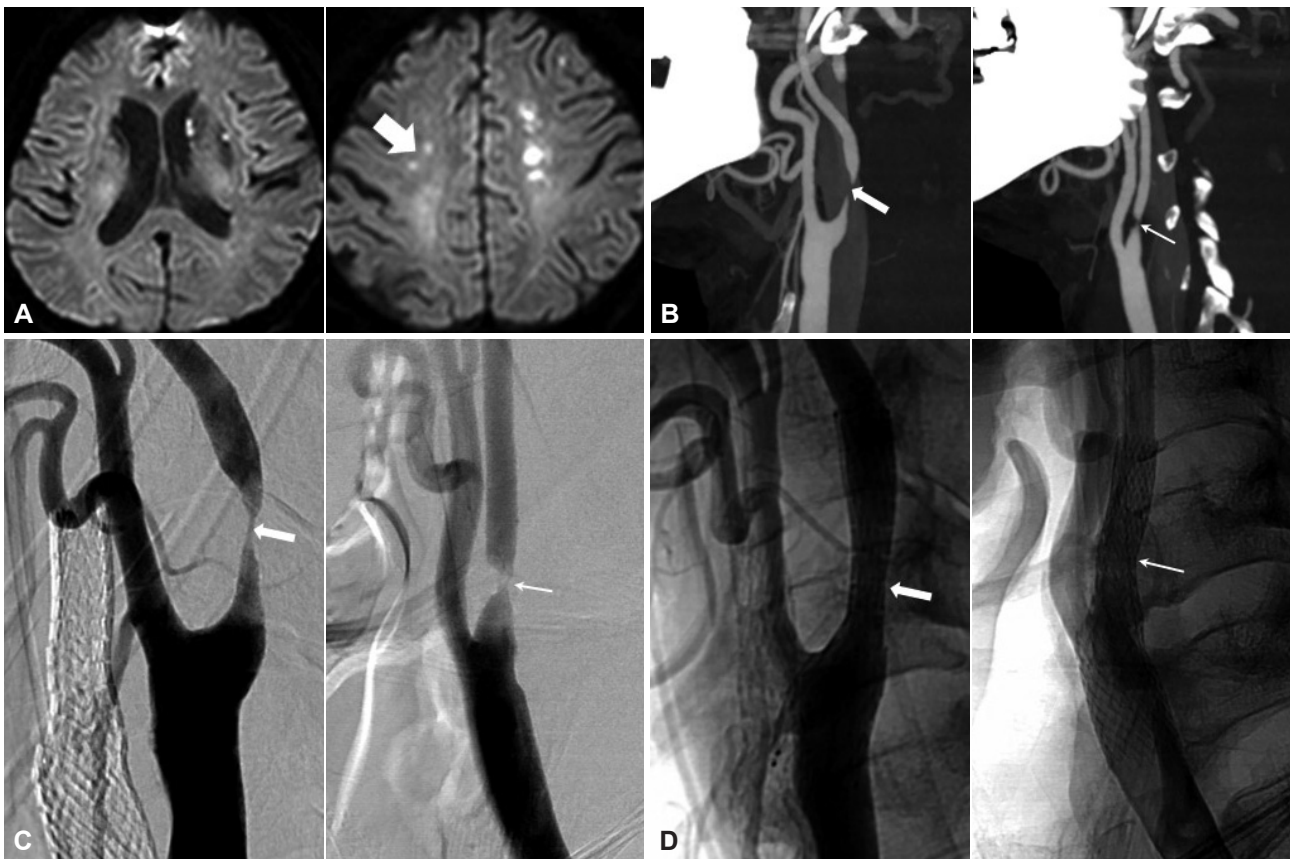


Figure 1. Brain MRI on diffusion-weighted image shows multifocal acute infarctions in the caudate nucleus, putamen of left basal ganglia and bilateral centrum semiovale (A, thick white arrow indicates right centrum semiovale). Brain CT angiography shows severe stenosis in the bilateral (B, thick arrow: right, thin arrow: left) proximal internal carotid arteries. Initial bilateral internal carotid artery (ICA) angiogram shows severe stenosis in the bilateral proximal ICA (C, thick arrow: right, thin arrow: left). After angioplasty and stent deployment, the bilateral ICA flows are nearly completely normalized (D, thick arrow: right, thin arrow: left).

of movement disorders varies depending on the type of movement disorders.³ Myoclonus along with tremor and dystonia occurs as a delayed complication of stroke. Myoclonus is a very rare type of delayed movement disorder after stroke compared with dystonia or tremor. Reorganization of the basal-ganglia motor circuits or generation of hyper-excitable areas after the acute stage of insult might cause delayed hyperkinetic movement disorders.^{1,3} In our patient, however, the abrupt onset of involuntary movement developed as a manifestation of the acute stroke. The exact mechanisms underlying acute myoclonus due to a destructive lesion of cortical regions are unclear. One possible mechanism is that destruction of subcortical structure connecting the motor cortex might have caused hyper-excitable or paroxysmal discharges to the post-synaptic receptors, resulting in spontaneous discharges of the corticospinal tract. Although there were no epileptic discharges on the EEG, the condition could be

symptomatically classified as ‘epilepsia partialis continua (EPC)’.⁴ EPC has been defined as ‘continuous muscle jerks of cortical origin.’ However, EPC from subcortical lesions with preserved cortical area and an absence of EEG abnormality have been observed.^{4,5} Therefore, the multifocal myoclonus in our patient could be classified as ‘subcortical EPC’ because there were no epileptic discharges on the EEG and the right motor cortex was preserved on the brain MRI.

Along with acute stroke lesions, these involuntary movements might also have been caused by hemodynamic failure due to severe carotid stenosis because the myoclonic episodes improved just hours after the revascularization procedures.⁶ Limb-shaking transient ischemic attack (TIA), another involuntary movement disorder related to cerebral ischemia, has been associated with high-grade stenosis or occlusion of the ICA.³ Although our patient showed myoclonus that was similar to limb-shaking TIA, there were several differences from the typical fea-

tures of that syndrome. First, in contrast to limb-shaking TIA, where abnormal movements are transient and occur from a period of several minutes to hours, the myoclonus jerks in this patient continued all day long. Second, myoclonus was extending to the face as well as the arm. Finally, myoclonus on the patient's face and arm was not affected by postural change or blood pressure change. These features are not compatible with the typical presentations of limb-shaking TIA.

A recent observation by Kim⁷ suggested that limb-shaking TIA could be categorized as a myoclonus, especially negative myoclonus. Kim⁷ insisted that "limb shaking," which is not strictly medical terminology, has been used to describe a type of symptom in patients with carotid stenosis due to a lack of knowledge on the clear pathophysiology of the development of limb-shaking TIA. He also insisted that limb shaking in carotid TIAs might be asterixis, which is a form of myoclonus that arises from a hypo-perfusion syndrome due to carotid stenosis. Impaired postural tone under the control of the cortico-subcortical pathway might cause an intermittent failure of postural control and then lead to negative myoclonus in patients with carotid stenosis. However, because perfusion MRI was not performed during the clinical course, we could not establish if the symptoms in our patient were from hypo-perfusion due to carotid stenosis.

Taken together, the positive and negative myoclo-

nus in this patient might have been caused by acute stroke lesions and carotid arterial stenosis with regard to positive and negative myoclonus, respectively. Despite the limited evaluation in this patient, this case might expand the concept of limb-shaking TIA as a type of etiology for myoclonus. This case is unique in that multifocal myoclonus occurred as a manifestation of acute stroke with carotid stenosis and was completely resolved by angioplasty with carotid arterial stent insertion.

Conflicts of Interest

The authors have no financial conflicts of interest.

Acknowledgments

Dr. Kim receives institutional research support from Chung-Ang University.

REFERENCES

1. Handley A, Medcalf P, Hellier K, Dutta D. Movement disorders after stroke. *Age Ageing* 2009;38:260-266.
2. Hallett M. Myoclonus: relation to epilepsy. *Epilepsia* 1985; 26 Suppl 1:S67-S77.
3. Béjot Y, Giroud M, Moreau T, Benatru I. Clinical spectrum of movement disorders after stroke in childhood and adulthood. *Eur Neurol* 2012;68:59-64.
4. Guerrini R. Physiology of epilepsy partialis continua and subcortical mechanisms of status epilepticus. *Epilepsia* 2009; 50 Suppl 12:7-9.
5. Juul-Jensen P, Denny-Brown D. Epilepsia partialis continua. *Arch Neurol* 1966;15:563-578.
6. Baquis GD, Pessin MS, Scott RM. Limb shaking--a carotid TIA. *Stroke* 1985;16:444-448.
7. Kim JS. Symptoms of transient ischemic attack. *Front Neurol Neurosci* 2014;33:82-102.