

## Early-Onset Dystonia after Supplementary Motor Area Infarction

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A 63-year-old patient with the right supplementary motor area infarct developed early-onset dystonia in the left upper extremity. The mechanisms involved in the genesis of focal dystonia are discussed with emphasis on cortical basal ganglia circuit and efferent projections from the supplementary motor area. **Key Words:** Movement disorders—dystonia—infarction—supplementary motor cortex—acute onset.

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### Case Report

A 63-year-old man presenting with sudden-onset stiffness of the left limbs was admitted to our hospital. He had a history of right putaminal hemorrhage 10 years earlier but no family history of either involuntary movement or illicit drug use.

On admission, neurologic examination showed disorientation, mild dysarthria, and left central facial palsy. In addition, posture of the left limbs was characterized by flexion at the elbow, pronation of the forearm, flexion at the wrist with grasping, and extension of the foot.

Blood testing including serum copper, ceruloplasmin, and magnesium were normal. Acute infarctions in the right supplementary motor area (SMA) and left precentral gyrus were identified on diffusion-weighted imaging (Fig 1, A). T2\*-weighted imaging revealed old hemor-

rhage in the right putamen and left thalamus (Fig 1, B). Occlusion of the left internal cerebral artery at its origin was observed on magnetic resonance angiography.

Dystonic posture of the left upper extremity appeared on the second day after admission. On day 6, his dystonic posture was characterized by abduction of the shoulder, flexion at the elbow, rotation of the forearm, and extension of the fingers (Fig 1, C). This posture was enhanced during walking. Focal dystonia gradually improved spontaneously, with no medication. At 1 month after onset, only mild dystonic posture with flexion at the elbow persisted.

In a review of the literature involving 56 cases of post-stroke movement disorders, dystonia was the second most common after chorea.<sup>1</sup> In addition, the onset of post-stroke dystonia is frequently delayed, with a latent period ranging from 2 weeks to 4 years.<sup>1,2</sup>

Dystonia is thought to result from increased thalamocortical excitation as a consequence of decreased inhibitory output from the globus pallidus internus and subthalamic nucleus to the thalamus. The fact that poststroke movement disorders are relatively rare, even with marked basal ganglia damage, is explained by the marked plasticity of the brain within the basal ganglia anatomical networks, partly as a result of parallel processing, and compensatory mechanisms that provide certain resilience and protection against clinically evident loss of motor control.<sup>3,4</sup>

Efferent projections from the SMA to the putamen, caudate nucleus, and subthalamus have been found subcortically.<sup>5</sup> Dystonia in the present patient could be considered likely to be because of the damage of efferent projections from SMA to subthalamus in addition to old

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Received August 23, 2013; revision received September 25, 2013; accepted September 27, 2013.

This work was supported by JSPS (Japan Society for the Promotion of Science) KAKENHI (grant number 24591309).

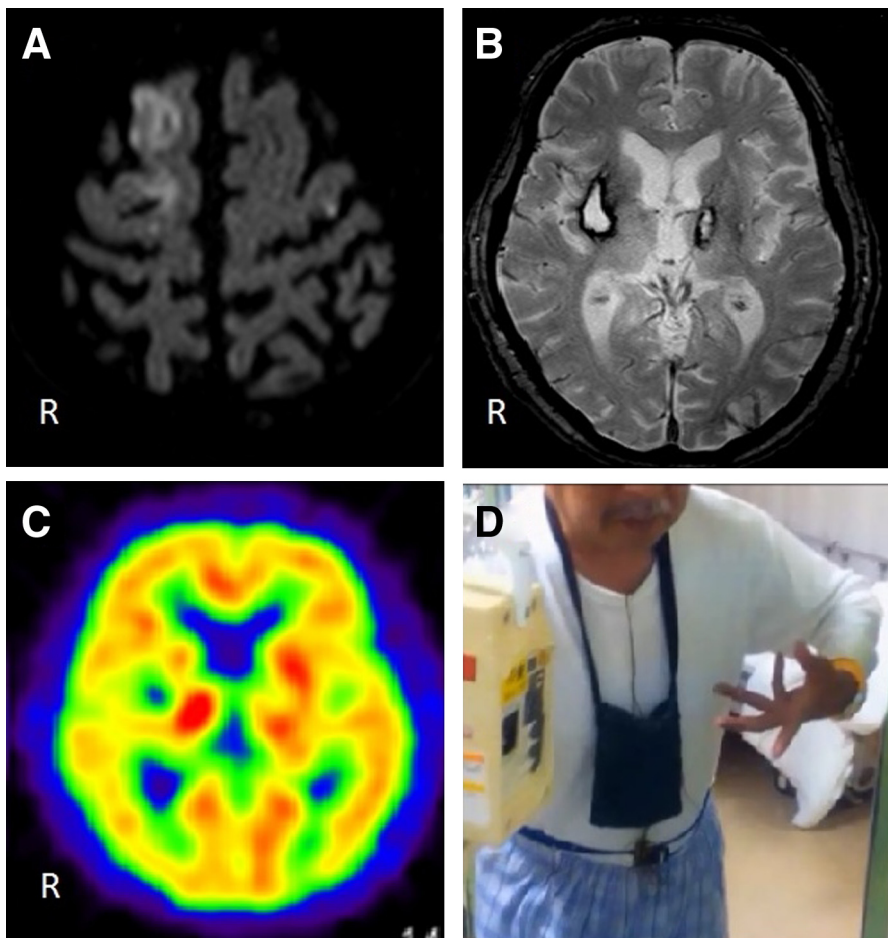
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1052-3057/\$ - see front matter

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<http://dx.doi.org/10.1016/j.jstrokecerebrovasdis.2013.09.028>



**Figure 1.** (A) Diffusion-weighted imaging showing hyperintense signals in the right SMA and left precentral gyrus. (B) T2\*-weighted imaging showing old hemorrhage in the right putamen and left thalamus. (C) Dystonia in left upper extremity. (Color version of figure is available online.)

putaminal hemorrhage. This case seems to be useful for clarifying the pathogenesis of dystonia.

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