

Unusual presentation of more common disease/injury

Isolated astasia in acute infarction of the supplementary-motor area

Yuko Wada, Yo Nishimura

Department of Neurology, Nishi-Kobe Medical Center, Kobe, Japan

Correspondence to Yuko Wada, wada@nmc-kobe.org

Summary

Astasia, which is the inability to stand in the absence of motor weakness or marked sensory loss, is an uncommon clinical feature of stroke in the thalamic ventrolateral region. The authors describe a patient with a unilateral supplementary motor area (SMA) infarction presenting with contralateral astasia. On neurological examination, he would lean to the left side and would fall unless supported. He showed no muscle weakness, sensory deficits or cerebellar ataxia. Magnetic resonance imaging of the brain showed acute infarction only involving the right SMA. On the basis of the anatomy that the SMA is connected to the vestibulocerebellar system through the ventrolateral nucleus of the thalamus, the authors concluded that contralateral astasia probably resulted from disruption of this connection following infarction of the SMA.

BACKGROUND

Astasia, which is the inability to stand unsupported despite normal strength, resembles the marked balance impairment in patients with vestibulocerebellar disease.¹ Astasia is an uncommon manifestation of thalamic syndrome² and its duration is transient when it occurs.³ It can be caused by lesions of the thalamic ventrolateral region.³ Recently, there has been one report of astasia associated with a cingulate lesion.⁴ However, to the best of our knowledge, there have been no reports of astasia associated with a lesion in the supplementary motor area (SMA).

We describe the case of a patient presenting with transient contralateral astasia as the characteristic feature of an infarction of SMA and discuss the underlying neurological mechanisms of this phenomenon.

CASE PRESENTATION

A previously healthy 61-year-old right-handed man presented with a suddenly inability to stand; whenever he attempted to stand or walk, he would lean to the left. He noted no other symptoms. On the next day, he was admitted to our hospital. On admission, his blood pressure was 138/96 mm Hg, and his pulse rate was 96 beats/min and regular. He showed normal consciousness and orientation. There was no personality change or apparent intellectual deterioration. No deficit of language, visual cognition or praxis was observed.⁵ There was neither motor nor verbal inertia. There was no motor neglect. He was unable to stand unsupported because there was an inclination to veer his body toward the left, characterised by marked left-sided pulsion, and needed support on his left side to prevent him from falling. When asked to stand up from a sitting position, he was able to stand up by pulling himself up with both hands. However, his ability to sit straight was preserved. Cranial nerves were intact. His strength and the muscle tone of the extremities and trunk were normal. All the tendon reflexes were normal, with no pathological reflexes. Superficial sensation, and joint position, and vibration senses were normal. His finger-to-nose, heel-knee and shin-tapping test were normal.

INVESTIGATIONS

Laboratory test results, including those of rheumatological (anti-DNA, antinuclear, antiphospholipid and lupus anticardiolipin antibodies) and coagulative (antithrombin III, prothrombin time, S and C-protein and fibrinogen) tests, were normal. Transesophageal echocardiogram (TEE) with air contrast showed a patent of foramen ovale with a right-to-left shunting, accentuated with the Valsalva maneuver. Right atrial pressure was not elevated, and no involvement of the valves was observed. On the day of admission, diffusion-weighted and T2-weighted MRI of the brain revealed a high-intensity lesion in the right SMA (figure 1). There were no other abnormalities in the cerebrum, brainstem or cerebellum. MR angiography and carotid ultrasonography findings were normal, as was that of Holter monitoring.

OUTCOME AND FOLLOW-UP

Although we did not detect deep venous thrombosis of the patient, we suspected that he had a paradoxical embolism. Because TEE of the patient showed foramen ovale with a right-to-left shunting.

The patient was treated with an anticoagulant. The next day his symptom began to improve, and 2 days after admission he was able to walk independently.

DISCUSSION

Our patient presented with a transient inability to stand unsupported and marked truncal instability, characterised by contralateral pulsion due to an acute SMA infarction. He noted no other neurological symptoms or signs, such as motor weakness, sensory deficit, incoordination or parkinsonism. Thus, we consider this manifestation as an astasia. Astasia, which is the inability to stand unsupported despite normal strength, resembles the marked balance impairment observed in patients with vestibulocerebellar disease.¹ Masdeu and Gorelick² first described the cases of patients with thalamic astasia, and their patients fell toward the side contralateral to the side of lesions. They also described that thalamic astasia may mimic cerebellar disease. In other reports

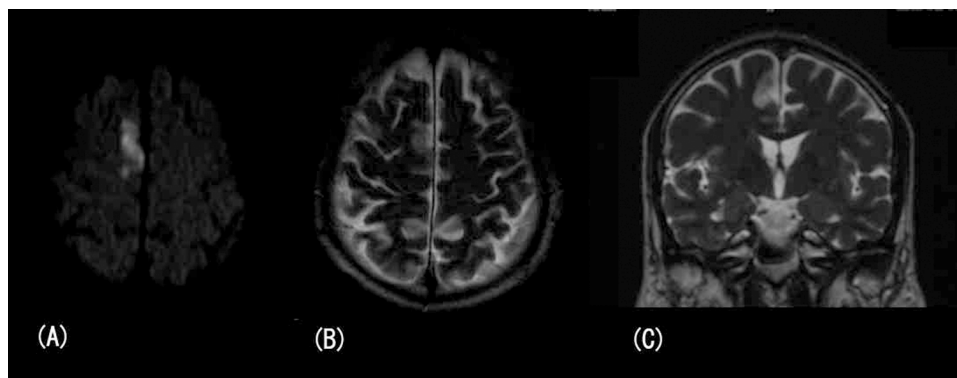


Figure 1 Diffusion-weighted (A) and T2-weighted (B) MR images show a high-intensity lesion in the medial frontal lobe (supplementary motor area). (C) T2-weighted MR image demonstrates a high-intensity lesion in only the supplementary motor area.

of astasia associated with other lesions, the direction of astasia was also contralateral to the site of lesions, such as the midbrain or posterior cingulate region.^{4 6}

Our patient has only one lesion located in the unilateral SMA, in the absence of other lesions in the primary motor cortex, thalamus, brainstem or cerebellum. It has been shown that SMA lesions cause various abnormalities of motor function, such as the planning or initiation, including speech function.^{7 8} Chung *et al*⁷ reported the case of a patient who developed severe gait disturbance characterised by markedly impaired postural reflexes after apparent SMA seizure. However, their patient did not show contralateral pulsion. The exact mechanism of contralateral pulsion associated with SMA lesion has not been confirmed in either humans or animals. To the best of our knowledge, no cases of unilateral SMA lesions that cause contralateral pulsion as the sole symptom have been reported.

Astasia as the main clinical manifestation of supratentorial involvement has been associated with the thalamic ventrolateral region,^{1 3} or midbrain tegmental lesions.⁶ According to Solomon *et al*² fastigial fibers of the vestibulo-cerebellar pathway project to the medial ventrolateral nucleus of the thalamus, and disruption of this pathway may be responsible for thalamic astasia. SMA also receives a thalamic input from the ventrolateral part of the thalamus, which receives inputs from cerebellar nuclei in animals.⁹ Disruption of this connection by SMA infarction may have been responsible for astasia in our patient.

Contralateral astasia associated with medial frontal lesions has rarely been described, and recently, only one case has been reported.⁴ Kataoka *et al*⁴ described a patient with posterior cingulate infarction who showed contralateral pulsion similar to that of our patient. They considered their patient's manifestation as astasia. Because the cingulated motor area receives a major thalamic input from the ventrolateral thalamus, disruption of this connection by a posterior cingulate infarction may be responsible for astasia in their patient. Moreover, SMA is also connected to the ventrolateral part of the thalamus, which receives inputs from the basal ganglia⁹ as well as the cingulated motor area. Disruption of this connection by SMA infarction also have contributed to the patient's astasia.

Astasia in our patient was transient, with resolution within 2 days. This finding was in accordance with those of previous reports.^{1 3} In Masdeu and Gorelick's report³ the

inability to stand unassisted lasted for an average of 3 days in six patients with infarction, and two of these patients presented with the inability to stand for only 1 day. They considered that the bilateral representation of fastigial vestibulocerebellar projection to the medial ventrolateral nucleus of the thalamus may be one of the factors for the transient nature of thalamic astasia. This finding may explain in part the transience of astasia in our patient.

In summary, the present finding raises the possibility that unilateral small SMA lesions can cause transient astasia following the disruption of the connection between SMA and the ventrolateral part of the thalamus.

Learning points

- ▶ Astasia is an uncommon clinical feature of stroke, such as thalamic infarction.
- ▶ The role of SMA has been demonstrated in animals, but remains unclear in humans. Clinicians should be aware of the possibility of a SMA stroke in patients with contralateral astasia.

Competing interests None.

Patient consent Obtained.

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