

Transient periodic limb movement secondary to acute bleeding in a patient with a spine meningioma

Dokyung Lee, Tae-hwan Roh, Tae-Beom Ahn

Department of Neurology, College of Medicine, Kyung Hee University, Seoul, Korea

Abstract

We report a 71 years old woman with transient periodic limb movement disorder and myoclonus that appeared after acute gastrointestinal bleeding. The abnormal movements dissolved as the blood loss was restored. The woman was later found to have an asymptomatic spine meningioma. Iron deficiency due to acute bleeding may be the trigger resulting in the abnormal movements in this patient with subclinical spinal meningioma.

INTRODUCTION

Periodic limb movement disorder (PLMD) is frequent in patients with restless leg syndrome (RLS). PLMD shares pathomechanisms with RLS such as iron deficiency, and disinhibited diencephalic pathway.¹ PLMD can be associated with spinal cord lesions, and sometimes coexist with myoclonus.² However, lack of a close temporal relationship between the myoclonus and an apparently longstanding spinal lesion make it difficult to establish a firm cause-and-effect relationship between the two.

CASE REPORT

A 71-year-old woman was admitted due to acute dyspnea and chest pain. She had no history of constipation, urinary dysfunction, and sensory and gait disturbance. Her hemoglobin concentration (Hb) was 6.0 g/dl, compared to 13.1 g/dl 6 month earlier. She had a massive hematochezia 5 days earlier and tarry stools until the admission. A colonoscopy showed ulcerative lesions at the ascending colon and rectum, and a capsule endoscopy revealed an additional ulceration with recent bleeding at the jejunum, which later found to be a gastrointestinal stromal tumor.

Sudden involuntary movements appeared in her legs during the preparation of transfusion. Neurologic examination was normal, except mildly decreased deep tendon reflexes in the lower extremities. The abnormal movements resembled periodic limb movement, and were more frequent in the left leg. Myoclonus infrequently appeared in the right leg. The abnormal movements of both legs were out of phase and repeated in

every 15 seconds during the night (Video 1). The abnormal movements were not accompanied by pain, other sensory symptoms or restless urge to moving legs. The transfusion started 6 hours after the onset of the involuntary movements, which raised the Hb up to 8.1 g/dl in the same night and 10.3 g/dl in the next morning. Her abnormal movements disappeared during sleep, without specific medication. Spine MRI showed an extra-axial meningioma compressing the thoracic spinal cord (Figure 1). Sensory and motor evoked potential showed a central conduction defect. She declined surgical treatment of the tumor because of the operative risk and lack of neurologic deficit. She did not suffer any neurologic symptom related to the meningioma subsequently, and neurologic examination could not find any significant additional abnormalities during the 2 year follow-up.

DISCUSSION

The close relationship between PLMD and iron status was suggested in the pediatric patients, but such association was not found in adults with obstructive sleep apnea.³ In our patient, the anemia developed over 5 days due to massive bleeding. As the patient had no neurologic symptoms and signs before the bleeding and after correction of anemia, the spinal meningioma by itself was not sufficient to result in the involuntary movements. The rapid rise of Hb (6.0 → 8.1mg/dl) seemed to be able to prevent further recurrence of PLMD. Thus, decreased iron, secondary to the acute blood loss, could have played a key role in the patient's transient PLMD.

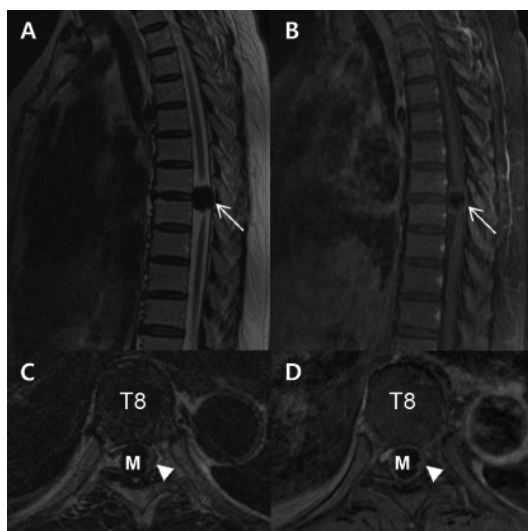


Fig.1 Magnetic resonance imaging of the thoracic spine (A and C, T2 weighted image; B and D, T1 weighted enhanced image) showed an intradural extramedullary mass in T7-8 level (white arrow, about 15x21 mm in size). In axial image, the spinal cord (arrow head) was being squashed by the mass (M=mass).

Transient abnormal movements were rarely related to an underlying structural spinal lesion.⁴ In our patient, the longstanding spinal tumor was by itself unable to cause the abnormal movement, until the development of the anemia. However, the evoked potential study showed central conduction defect, which implied a deranged intra-spinal neural conduction despite the absence of neurologic deficits. It was possible that the descending inhibitory control was marginal under the “normal” condition, restraining the generation of disinhibited abnormal movements. The unstable equilibrium might be vulnerable to some perturbations, such as the dysfunction of dopaminergic diencephalic pathway, which is regulated by the iron dependent tyrosine hydroxylase (TH).⁵ The patient’s anemia might have started with the massive bleeding and reach critical level after 5 day of tarry stool, resulting in the dysfunction of TH.

Polysomnography could not be done because of acute medical illness and rapid disappearance of her symptoms. After the episode, she and her family did not notice abnormal leg movement during sleep. Thus, her PLMD was more compatible with a symptomatic and transient PLMD rather than ongoing PLMD during daytime or sleep.

Legend to the Video

Video 1. [http://neurology-asia.org/content/18/2/neuroasia-2013-18\(2\)-225-v1.mp4](http://neurology-asia.org/content/18/2/neuroasia-2013-18(2)-225-v1.mp4). In this video, 7 episodes were recorded, with different phenotypes and duration. The first 2 episodes consisted of slow flexion of the ankle and the knee, and sequential jerky hip movements in the left leg (2 and 17 second). There were 3 episodes

only with slow ankle and knee flexion in the left leg (34, 51, and 61 second). In the right leg, jerky hip flexion followed the abnormal flexion of the left leg in the first 2 episodes (22 second). In the later segment, an episode of myoclonic jerk of the right hip preceded the involuntary flexion of the left leg (73 second). In all the episodes, the abnormal movements of both legs were out of phase.

ACKNOWLEDGEMENTS

This research was supported by the Kyung Hee University Research Fund in 2010. (KHU-20100650)

DISCLOSURE

Conflicts of Interest: None

REFERENCES

1. Stiasny K, Oertel WH, Trenkwalder C. Clinical symptomatology and treatment of restless legs syndrome and periodic limb movement disorder. *Sleep Med Rev* 2002; 6:253-65.
2. Lee MS, Choi YC, Lee SH, Lee SB. Sleep-related periodic leg movements associated with spinal cord lesions. *Mov Disord* 1996; 11:719-22.
3. O’Brien LM, Koo J, Fan L, Owusu JT, Chotinaiwattarakul W, Felt BT. Iron stores, periodic leg movements, and sleepiness in obstructive sleep apnea. *J Clin Sleep Med* 2009; 5:525-31.
4. Marrufo M, Politsky J, Mehta S, Morgan JC, Sethi KD. Paroxysmal kinesigenic segmental myoclonus due to a spinal cord glioma. *Mov Disord* 2007; 22:1801-3.
5. Qu S, Le W, Zhang X, Xie W, Zhang A, Ondo WG. Locomotion is increased in a11-lesioned mice with iron deprivation: A possible animal model for restless legs syndrome. *J Neuropathol Exp Neurol* 2007; 66:383-8.