A Case of Painful Legs and Moving Toes Syndrome in a Young Woman

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INTRODUCTION

Painful legs and moving toes syndrome (PLMT) is a rare syndrome characterized by pain in the lower extremities and involuntary movements of single or multiple toes.1 The largest case series on PLMT reported that the age of onset was 24–86 years.2 However, there are no previous reports of PLMT in young adults in Korea. Herein, we report the first case of PLMT in a 29-year-old Korean adult with a lumbosacral intervertebral disc herniation.

CASE REPORT

A 29-year-old woman presented to our clinic with complaints of bilateral foot pain and involuntary toe movements that she has been experiencing for the past three months. The symptoms began insidiously on the left side, but were recently noted intermittently on the right side as well. The patient described the pain as an intense pressure or a feeling of her toes being pulled. The pain was undeniably severe in the second toe. The pain and movements worsened when the patient lay down, and were relieved when she wore shoes or socks. She denied having an urge to move her feet or toes.

This patient had no history of trauma, neuroleptic drug use, or family history of this condition. Three years prior, she had developed left leg paresthesia with back pain and was diagnosed with intervertebral disc herniation at the L4/5 and L5/S1 levels. The symptoms decreased for one year after transforaminal lumbar epidural steroid injection. Before visiting our clinic, she had been prescribed pregabalin at another hospital. However, she discontinued the medication because it had no effect on the movements or pain in her toes. She was not taking any medications at the time of her visit. Physical examination revealed decreased extension power of the left great toe and involuntary repetitive abduction, adduction, and exten-
sion of the left toes approximately once every 5–6 seconds (Supplementary Video 1 in the online-only Data Supplement). Her serum ferritin level was 5.2 ng/mL (range: 9–89 ng/mL) and hemoglobin was 11.5 g/dL (range: 12–16 g/dL). Blood electrolyte, liver, kidney, and thyroid function levels were unremarkable. Nerve conduction study (NCS) and needle electromyography (EMG) were conducted at another hospital. The patient was informed that no abnormalities were present.

Intervertebral disc herniation in the left L4/5 and L5/S1 segments was detected upon lumbosacral spine magnetic resonance imaging performed 2 years ago (Fig. 1).

Oxcarbazepine (150 mg per day) and haloperidol (1.5 mg per day) were prescribed. However, the patient’s symptoms persisted. Intravenous iron supplementation (ferric carboxymaltose, 1,000 mg) was also ineffective.

This study was approved by the Institutional Review Board of Seoul National University Hospital (IRB No. H-2108-153-1248).

DISCUSSION

PLMT is a rare syndrome characterized by foot or leg pain and repetitive, stereotypical, and non-rhythmic toe movements.1,2 The age of onset is typically higher than the age of our patient at the time the symptoms appeared, and the mean reported age at onset in 76 patients with PLMT was 58 years.2 In Korea, PLMT has been reported in patients aged 713 and 164 years, but not in young adults. To the best of our knowledge, this is the first case of this condition in a young Korean adult patient.

However, the cause of PLMT remains unclear. Cryptogenic PLMT accounts for the majority of cases, but secondary causes such as peripheral neuropathy, radiculopathy, spinal cord lesions, mono-neuropathy, posttraumatic injury, and the use of neuroleptics have also been implicated.2,5

Several mechanism have been proposed for PLMT. One proposed mechanism is that peripheral nerve injury leads to changes in the afferent neuronal connections, which triggers involuntary movements of the digits.3 However, central neuromodulation has also been suggested as the mechanism of PLMT.2,3,6 In our case, pre-existing lumbosacral intervertebral disc herniation could be considered as a secondary cause. Initially, peripheral injury due to disc herniation might have evoked a peripheral pathophysiological process that progressed to central reorganization. This process may have led to the symptomatic progression (from unilateral to bilateral involvement). PLMT with unilateral lesions presenting with bilateral involvement has been reported previously.1,5,6 A pathophysiologic mechanism, similar to that noted in our case, has been reported in a Korean case of painless moving toes syndrome. In this condition, radiculopathy is thought to cause maladaptive changes in the peripheral connection and central reorganization, resulting in bilateral movements.7 In our case, because the patient no longer visited our hospital, NCS and EMG follow-up tests for the evaluation of radiculopathy could not be performed.

The nature of the pain and involuntary movements in this patient were typical of those described previously. These symptoms include tormenting pain, toe pulling, intense pressure, abduction, adduction, and extension of the phalanges.1 Up to 58% of patients can have exacerbating factors such as position-dependent symptoms, diurnal variations, cold temperature, external pressure, and the Valsalva maneuver.7 This patient’s symptoms were position-dependent and worsened when she lay down.

Differential diagnoses such as neuroleptic-induced dyskinesia, akathisia, restless legs syndrome, Wilson’s disease, hyperthyroidism, multiple sclerosis, and Huntington’s disease could be considered in young patients.3 Our patient had no history of psychological disorders or neuroleptic drug use, and had no remarkable family history. She did not complain of an urge to move, a key symptom of restless legs syndrome. Laboratory tests, including thyroid function tests, were negative, and physical examination did not indicate the possibility of other conditions.

In PLMT, the pain is more distressing than the movements.2 Our patient reported severe and unbearable pain. Therefore, pain mitigation is the primary management goal. There are no consistent guidelines for the treatment of this condition; however, gabapentin, pregabalin, carbamazepine, and antidepres-
sants can be prescribed for pain relief.\textsuperscript{2} In previous case reports of PLMT in Korea, gabapentin was generally effective,\textsuperscript{3,4} even in intractable cases.\textsuperscript{3} However, as our patient experienced no relief with pregabalin, other medications, including oxcarbazepine, haloperidol, and even high-dose intravenous iron therapy, were prescribed. However, these treatments had no effect.

To the best of our knowledge, this is the first reported case of PLMT associated with a lumbosacral intervertebral disc herniation in a young Korean woman. These observations suggest that PLMT should be considered for patients with pain and involuntary movements of the lower extremities.

**Supplementary Video Legend**

Video 1. The patient shows involuntary, repetitive abduction, adduction, and extension of the left toes.

**Supplementary Materials**

The online-only Data Supplement is available with this article at https://doi.org/10.13078/jsm.210026.

**Conflicts of Interest**

The authors have no potential conflicts of interest to disclose.

**ORCID iDs**

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**Author Contributions**


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**REFERENCES**