Case Reports in Neurology

Case Rep Neurol 2015;7:59–62

DOI: 10.1159/000380942
Published online: March 21, 2015

1662–680X/15/0071–0059$39.50/0
www.karger.com/crn

Low Dose of Clonazepam Is Effective in the Treatment of Painless Legs and Moving Toes Syndrome: A Case Report

Sumihiro Kawajiri[a] Kazuyuki Noda[a] Aya Ikeda[b] Takahiro Koinuma[a]
Yuji Tomizawa[a] Nobutaka Hattori[b] Yasuyuki Okuma[a]

Department of Neurology at [a]Juntendo Shizuoka Hospital, Izunokuni, and
[b]Juntendo University School of Medicine, Tokyo, Japan

Key Words
Clonazepam · Painless legs · Painful legs · Moving toes

Abstract
Introduction: Painless legs and moving toes syndrome (PoLMT) is a rare movement disorder characterized by flexion, extension, abduction, adduction, and torsion of toes in the absence of pain. It is considered a variant of painful legs and moving toes syndrome, which is characterized by similar movements but is accompanied by pain. Although neuropathy, spinal cord compression, brain tumor, cerebral infarction, and Wilson's disease have been reported to be associated with PoLMT, the actual cause, trigger, and mechanism remain unclear. Therefore, a standardized treatment for PoLMT is not established yet. ***Case Presentation:*** We describe a 64-year-old Japanese woman with no past medical history who presented with nonrhythmic repetitive involuntary toe movement of the left foot in the absence of pain. She was diagnosed with idiopathic PoLMT and treated with a low dose of clonazepam (0.5 mg/day). The involuntary movement disappeared completely several days after treatment. ***Conclusion:*** A low dose of clonazepam is effective in the treatment of PoLMT.

Introduction

Painless legs and moving toes syndrome (PoLMT) is a rare movement disorder characterized by flexion, extension, abduction, adduction, and torsion of toes without pain. It is considered a variant of painful legs and moving toes syndrome (PLMT), which is character-

Sumihiro Kawajiri
Juntendo Shizuoka Hospital, 1129 Nagaoka
Izunokuni-shi, Shizuoka 410-2295 (Japan)
E-Mail skawaji@juntendo.ac.jp

ized by similar movements but accompanied by pain. Although neuropathy, spinal cord compression, brain tumor, cerebral infarction, and Wilson's disease have been reported to be associated with PoLMT, the actual cause, trigger, and mechanism remain unclear [1–4]. Therefore, a standardized treatment for PoLMT is not established. Here, we describe a patient with idiopathic PoLMT who demonstrated a good response to a low dose of clonazepam. We also present a corresponding video (online suppl. video 1, www.karger.com/doi/10.1159/000380942).

Case Presentation

We report a 64-year-old Japanese woman with no past medical history, no family history of PoLMT or PLMT, and no obesity who did not smoke or drink alcohol. She felt uncomfortable but did not have pain in her left foot about 3 months before onset. Although she did not hit her foot, did not have joint deformities, and did not have morning stiffness, she developed a slight swelling and a hot feeling in the dorsolateral part of the left foot that resolved spontaneously over several days about 2 weeks before onset. Upon neurological examination, she presented with repetitive involuntary toe movement of the left foot in the absence of pain. The movement comprised nonrhythmic extension, flexion, abduction, adduction, and torsion, and was most prominent in the 4th and 5th toes (video). She was able to consciously suppress the movement to some extent, but not completely. The movement was exacerbated when she was distracted. This occurred throughout the day at rest and disappeared during sleep. She had no sensory disturbance, reduced tendon reflexes, weakness, or muscle atrophy, which indicated that she had no other neurological deficits. CT and MRI did not show tumors, infarction, hemorrhage, or any other lesions. Whole spinal cord MRI did not reveal compression or any other lesions. Nerve conduction studies and somatosensory evoked potentials were within normal limits. Cerebrospinal fluids and blood examination, including vitamin B_1 and B_{12}, ceruloplasmin, iron, thyroid function, and autoantibodies indicating collagen diseases and rheumatic arthritis, were also all within normal limits. The patient's movement did not fulfill either periodic limb movement or restless legs syndrome because of the absence during sleep and the lack of urge to move her legs and toes, respectively. We excluded psychogenic movement disorder because she had no secondary gain and spontaneous remissions, her onset was not sudden, and the movement had no variability over time. Considering the form of the movement without pain and other clinical points, we diagnosed her with idiopathic PoLMT. The patient was treated with a low dose of clonazepam (0.5 mg/day). Thereafter, the involuntary movement improved and disappeared completely several days after treatment. She discontinued clonazepam because of the complete resolution. PoLMT recurred several days after the withdrawal, although to a lesser extent. Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal.

Discussion

PoLMT is assumed to be a variant of PLMT. The movements of both syndromes are basically identical to each other. PLMT is an uncommon illness, described by Spillane et al. [5] in 1971. A previous case report describes 76 cases of PLMT [6]. According to this report, symptom onset was more often on the left side and eventually became bilateral in 58% of the

patients. Various movements were described, but they tended to be constant and to wax and wane in severity. Peripheral neuropathy was the most common cause of pain (28%). No central nervous system involvement was described. The cause was not identified in 42% of the patients. Ropinirole, pramipexole, clonazepam, and botulinum toxin injection were applied for the treatment of the digit movements. Clonazepam was most commonly given to patients with PLMT for the movement, presumably because it had fewer side effects than other treatments and was noninvasive unlike botulinum toxin. However, the percentage of responders was about 25%, and the effective dose range was 2.0–5.0 mg/day [6], which was higher than that administered to our patient.

PoLMT is a rare illness and was first reported by Walters et al. [7] in 1993. To the best of our knowledge, only 10 cases have been reported so far. Not only peripheral nerve involvement but also central nervous system involvement, such as Wilson's disease, cerebral infarction, brain tumor, and spinal cord compression, have been reported [1–4]. Although the detailed mechanism remains unclear, some mechanisms are speculated, e.g., alterations in the afferent sensory information due to peripheral nerve damage can cause subsequent reorganization of efferent motor activity [8]. In the present case, we speculated that a peripheral involvement due to the swollen foot before onset might have triggered the above mechanism. In contrast, there are some reports suggesting that a dystonic mechanism contributes to this movement because repetitive transcranial magnetic stimulation was effective [9] and 1 patient also showed dystonic posturing [4]. If some etiology was identified, treatment for the primary diseases of patients with PoLMT was preferred, such as zinc acetate or resection for Wilson's disease or meningioma, respectively [2, 3]. In the absence of primary diseases, some reports found that quetiapine or gabapentin were effective [1, 10]. Interestingly, dystonic posture was also improved in a case treated with quetiapine, and another report indicated that gabapentin could improve dystonia of a patient with Wilson's disease [11]. Our case showed a good response to a low dose of clonazepam, which is often prescribed for patients with dystonia and dystonic tremor [12]. Based on these factors, we also speculated that the low dose of clonazepam in our patient might have affected dystonia to some extent.

To the best of our knowledge, this is the first case that showed a significant improvement of idiopathic PoLMT after a low-dose clonazepam treatment. Another case with meningioma had no response to clonazepam [2], indicating that clonazepam might be more effective in idiopathic PoLMT. Further investigations are needed to reach verifiable conclusions.

Disclosure Statement

The authors have no conflicts of interest to declare.

References

1 Bermejo PE, Zabala JA: 'Painless legs and moving toes' syndrome due to spinal cord compression. Eur Spine J 2008;17(suppl 2):S294–S295.
2 Yabe I, Kano T, Sakushima K, Terasaka S, Sasaki H: Painless legs and moving toes from parasagittal meningioma. Mov Disord 2012;27:586–587.
3 Papapetropoulos S, Singer C: Painless legs moving toes in a patient with Wilson's disease. Mov Disord 2006;21:579–580.
4 Boulos MI, Corrigan BW, Jewell DR, Murray BJ: 'Painless legs moving toes' triggered by ischemic stroke. Parkinsonism Relat Disord 2013;19:842–843.
5 Spillane JD, Nathan PW, Kelly RE, Marsden CD: Painful legs and moving toes. Brain 1971;94:541–546.
6 Hassan A, Mateen FJ, Coon EA, Ahlskog JE: Painful legs and moving toes syndrome: a 76-patient case series. Arch Neurol 2012;69:1032–1038.

7 Walters AS, Hening WA, Shah SK, Chokroverty S: Painless legs and moving toes: a syndrome related to painful legs and moving toes? Mov Disord 1993;8:377–379.
8 Dressler D, Thompson PD, Gledhill RF, Marsden CD: The syndrome of painful legs and moving toes. Mov Disord 1994;9:13–21.
9 Kim JY, Kim HJ, Park KI: Ameliorating effect of low frequency repetitive transcranial magnetic stimulation over the premotor cortex in a case of possible painless legs and moving toes syndrome. Parkinsonism Relat Disord 2012;18:702–703.
10 Di Fabio R, Casali C, Pierelli F: Quetiapine: an alternative treatment in painless legs and moving toes. Mov Disord 2008;23:1326–1328.
11 Lerose M, Di Fabio R, Serrao M, Loudianos G, Pierelli F, Casali C: Gabapentin can improve dystonia in confirmed Wilson disease. Can J Neurol Sci 2014;41:284–285.
12 Fasano A, Bove F, Lang AE: The treatment of dystonic tremor: a systematic review. J Neurol Neurosurg Psychiatry 2014;85:759–769.