

CASE REPORT

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An Idiopathic Case of Painless Legs and Moving Toes

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ABSTRACT Painful legs and moving toes is an uncommon movement disorder. A painless variant has been reported in few case reports. Painless legs and moving toes is mainly related to the spinal cord pathologies, or reported as secondary to the neurodegenerative diseases or as a tardive phenomenon. Here we report a 27-year-old female patient complaining of involuntary semi-continuous writhing movements in her toes for 9 years. She had no family history for any movement disorder. As a result of detailed clinical and laboratory investigations including cranial and spinal neuroimaging, biochemical tests, viral serology, autoimmune and paraneoplastic biomarkers, she was diagnosed as having idiopathic painless legs and moving toes.

Keywords: Painless legs and moving toes; painful legs and moving toes; pathophysiology

Painful legs and moving toes (PLMT) is an uncommon movement disorder described by Spillane et al. in 1971.¹ It is characterized by the involuntary continuous or semi-continuous twisting and writhing movements in the toes, which are associated with pain in one or both lower extremities. These movements may be in flexion/extension or abduction/adduction or sinusoidal in morphology. The pathophysiology of PLMT is unclear. The proposed mechanism is an injury to the peripheral nervous system between plexus to peripheral nerves, causing a change in the sensory and motor mechanisms at the spinal level.² Peripheral neuropathy, radiculopathies, trauma, Wilson's disease, infections like herpes zoster myelitis or human immunodeficiency virus (HIV), or drugs such as neuroleptics and chemotherapeutic agents were reported among the underlying etiologies of PLMT.³

Rare variants of PLMT were also reported, though the disease itself is a rare disorder. These include painless legs and moving toes, painful arms and mobile fingers, painless arms and mobile fingers, and

painful mouth and mobile tongue.⁴ Painless variant of the disease was described by Walters et al. in 1993.⁵ Similar to PLMT, irregular, involuntary movements in the toes affecting unilateral foot or bilateral feet are present, though the pain in feet and/or legs does not accompany these movements. Here we present a young female patient who was diagnosed as having idiopathic painless legs and moving toes.

CASE REPORT

A 27-year-old female patient was admitted complaining of abnormal movements in her toes. The patient reported the presence of involuntary small movements in her toes, which started 9 years ago in her right foot, and then occurred bilaterally in both feet. These movements emerged especially when she rested in supine position after getting tired of a busy day or exercise. They occurred for at least 4 to 5 days in a week, at any time of the day without circadian characteristics. A pain in the feet or legs did not accompany these movements, ever.

For the video/videos of the article:



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Past medical history of the patient was unremarkable. Her family history did not reveal any kind of movement disorders or chronic systemic diseases. She was a non-smoker and denied the use of alcohol and/or substance.

The neurological examination was all normal; the cranial nerves, and the muscle strength were intact, the deep tendon and plantar reflexes were normal, sensory and cerebellar system examinations were normal. The involuntary, semi-rhythmic and semi-continuous extension and flexion movements in her toes, being prominent on the right, were observed on the examination (Video 1). Distracting maneuvers failed to lead any change in the shape, duration, or the frequency of the movements.

A detailed work-up was performed including complete blood count, thyroid function tests, urea, creatinine, iron, ferritin, folic acid, vitamin B₁₂ levels, serum ceruloplasmin and serological tests (hepatitis markers, and HIV), all of which were resulted to be within normal limits. Detailed autoimmune panel (including NMDA-R, AMPA-R1, AMPA-R2, CASPR2, LGI1, GABA-R) and paraneoplastic panel (anti-Hu, Yo, Ri, Tr, PCA-2, Ma, CV2-1, ANNA-3, amphiphysin antibodies) were all negative. The electromyographic (EMG) investigations, conduction studies and needle EMG were normal. The cranial and lumbosacral spinal magnetic resonance imaging were unremarkable. Informed consent was obtained from the patient for the publication.

In the light of these data, the patient was diagnosed as having painless legs and moving toes. Because the detailed work-up failed to demonstrate any pathology, our patient was accepted to have idiopathic painless legs and moving toes. Despite of the semi-continuous movements for 9 years, the patient did not ask for the treatment due to the lack of pain or any disturbing sensation.

DISCUSSION

There are only case reports of painless legs and moving toes in the literature. The first case described by Walters et al. in 1993 was a 59-year-old man with an undetermined etiology.⁵ One year later, 3 more cases were reported by Dressler et al., the etiology was also

undetermined in 2 of these patients, while severe dysraphism was noted in one patient.⁶ Similar case reports with spinal cord pathologies were subsequently reported.^{7,8} Other cases of painless legs and moving toes reported in the literature as case reports have emerged in the settings of Wilson's disease, stroke, parasagittal meningioma, Parkinson's disease or multiple system atrophy.^{3,4,9-12} The painless legs and moving toes was also reported as a tardive syndrome related to the chronic neuroleptic usage.¹³ Dziewas et al. reported a 70-year-old mother (since the 50 years of age) and 38-year-old daughter (since her childhood) with painless legs and moving toes; no associated medical conditions were present, and a possible hereditary component was suggested in the pathophysiology of this disorder.¹⁴

The pathophysiology of painless legs and moving toes is still unknown. Because it was accepted as the same entity with the PLMT, a similar pathophysiology was suggested as a disturbance in the afferent sensory signaling secondary to an injury in the peripheral nervous system, causing alterations in the segmental and/or suprasegmental organization of the efferent motor systems.^{2,13} A detailed work-up is mandatory for the demonstration of the underlying etiology. However, idiopathic cases are also present, as in the reported patient. Whether a long-term follow-up is needed in these patients or not deserves further attention.

It is questionable whether the painless legs and moving toes should be treated or not. In contrast to severe and intractable pain in the PLMT, the surgical treatment of the underlying cause or the drugs like gabapentin, levodopa or quetiapine were reported to be effective in controlling the involuntary movements in this rare and painless variant of PLMT.^{7,8,11,12,15} However, because of the absence of the pain, some patients may even not seek for the treatment.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or mem-

bers of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: Gülçin Benbir Şenel; **Design:** Esra Koçhan Kızılkılıç, Başak Yılmaz Öz, Gülçin Benbir Şenel; **Control/Su-**

pervision: Gülçin Benbir Şenel; **Data Collection and/or Processing:** Esra Koçhan Kızılkılıç, Başak Yılmaz Öz, Merve Şimşek, **Analysis and/or Interpretation:** Gülçin Benbir Şenel, Nurten Uzun Adatepe; **Literature Review:** Esra Koçhan Kızılkılıç; **Writing the Article:** Esra Koçhan Kızılkılıç; **Critical Review:** Gülçin Benbir Şenel, Nurten Uzun Adatepe.

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