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Painful Leg Moving Toes Syndrome: A Report of Two Cases and Update of Clinical Manifestation, Pathophysiology and Management

Samih Badarny[1*,2], Yazid Badarny[3], Kamal Hassan[2,4] and Adnan Zaina[5]

[1]*Department of Neurology, Galilee Medical Center Nahariya, Israel*
[2]*Faculty of Medicine, Bar Ilan University, Safed, Israel*
[3]*Department of Neurosurgery, Rambam Medical Center, Haifa, Israel*
[4]*Department of Nephrology and Hypertension, Galilee Medical Center Nahariya, Israel*
[5]*Department of Endocrinology, Zvulun Medical Clinic, Kyriat Bialik, Israel*

Abstract

Introduction: Painful Leg and Moving Toes (PLMT) is a rare disorder of unknown etiology. It is characterized by pain and abnormal movement of toes. These symptoms are troublesome to patients but not life-threatening. There is no curative treatment; however, supportive therapy and symptomatic therapy somewhat ameliorate the conditions. Thus, also to avoid extensive or invasive examinations, physicians should be aware this condition.

Case presentation: The first patient had sensory symptoms that began in both legs, and gradually involuntary movements appeared in the toes of one leg, which spread to the other leg. The second had no leg pain, but involuntary movement of the toes was the sole clinical presentation. Diagnosing these conditions as PLMT, we prescribed pregabalin, clonazepam, baclofen, local corticosteroid injections, epidural blocks and warm-water soaks, which led to partial improvement of these symptoms.

Conclusion: PLMT presents in different forms, symptoms usually do not worsen over time and not life-threatening, and thus, in this sense, benign condition. We, presenting these two cases, wish to update the clinical presentation, electrophysiological findings, pathophysiology, associated diseases, treatment and management of PLMT.

Keywords: Painful leg • Moving toes • Involuntary movement • Central or peripheral origin • Differential diagnosis

Abbreviation: PLMT: Painful Leg and Moving Toes; PNS: Peripheral Nervous System; CNS: Central Nervous System; CRPS: Complex Regional Pain Syndrome; TENS: Transcutaneous Electrical Nerve Stimulation

Introduction

Painful Leg and Moving Toes (PLMT) is of unknown etiology, no definite diagnostic criteria first described in 1971 by Spillane [1]. Thus, diagnoses are made clinically and by ruling out other conditions. PLMT is not-life-threatening and its treatment should be symptomatic. The largest study published by Hassan et al. the others reported as case reports [2]. The syndrome is not uniform, and can present differently from one case to the next and this heterogeneity is not clear [3]. We are describing two atypical cases of PLMT, one with abnormal movements involved both legs and the second had no leg pain at all.

Case Report

We present 43-year-old female who, three years prior, began to feel as if something was moving inside her left foot as well as a burning sensation. Gradually this sensation became a visibly involuntary movement of the toes of her left foot, which would vary during the day but noticeably worsen when she thought about it. Similar movements appeared in her right foot several months after their appearance in the left foot. The burning sensation slowly intensified, together with onset of pain with intermittent tingling sensation in both feet and calves, which became more problematic than the involuntary toe movements. The patient known with diabetes mellitus and hypertension 10 years earlier, and has had no trauma or back pain in her neither history nor previous exposure to neuroleptics. Her physical examination showed involuntary movements of non-rhythmic, adduction/abduction, fanning, of the toes more on the left foot than the right. Tendon reflexes in legs were missing, with a decrease in pain and touch sensation in the form of short socks. EMG revealed mixed demyelinated, axonal sensory motor polyneuropathy, CT of lumbar spine was normal, vitamin B12, TSH, and blood tests of rheumatic functions were all normal. She was prescribed with pregabalin 75 mg twice daily that was increased slowly to 300mg twice daily, resulting in some improvement in pain but no changes in involuntary movements.

Clonazepam and baclofen resulted in mild benefit for involuntary toe movements. Epidural block L5-S1 was used later with short transient reduction of the movements. She is still on pregabalin, mainly for some pain relief. The second patient is a 28-year-old woman complaining of five years of involuntary movements of the toes of her right foot when at rest, standing or walking, which is especially prominent when she is wearing shoes, regardless of whether it is a closed or open shoe. She had no pain in her foot or leg. According to her medical file, a number of neurologists and orthopedists have examined her in the recent past, and foot X-ray and lumbar spine CT, EMG and EEG have all been normal. She had no history of back pain, trauma or exposure to neuroleptics in the past. On examination, in plantar flexion/extension, rhythmic movements of the right toes could be seen when the foot was resting on the floor or in the air while sitting with crossed legs. Local corticosteroid injections

***Address for Correspondence:** Badarny S, Department of Neurology, Galilee Medical Center Nahariya, Israel, E-mail: samihb@gmc.gov.il*

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did not improve her condition, and warm-water soaks with clonazepam and baclofen were recommended. Drugs were stopped after a short time due to side effects but the soaks continued, with some improvement. Later the patient did not agree to further suggested treatments when it was explained to her that there is no definitive treatment to completely cure this phenomenon.

Discussion

PLMT usually occurs in middle-age females in one limb, or rarely both legs are involved. The syndrome is characterized by sustained pain from thigh to foot described as deep throbbing, ache, burning, cramp, dullness and numbness, sometimes with allodynia, and accentuated with walking, though resting does not alleviate the discomfort. Rarely, the syndrome is painless, as described in our second patient. The second part of this syndrome is the involuntary movements of all toes or a single toe, with varying frequency, stereotyped, constant but with rises and falls in severity, suppressed while standing and with several variant forms such as flexion/extension, abduction/adduction, dystonic, myoclonic, circular with continual wriggling and writhing movements [4]. Both pain and involuntary movements can appear at the same time, however usually the pain precedes the involuntary movements. Movements typically do not change from one moment to the next, cease during sleep, are controlled voluntarily for only few seconds, and cannot be mimicked by a healthy person [5] (Figure 1). Verhagen et al. described a variant of PLMT presenting symptoms in arm and fingers, which is now known as painful hand and moving fingers [6]. PLMT should be distinguished from spinal myoclonus which, although it is painless, has rhythmic movements that are faster and jerkier [7]. Restless leg syndrome is also a possible diagnosis, but usually is painless, which is not the case with PLMT; furthermore, the former involves an urge to move legs, and is sleep-related [8]. Chorea has a random nature, is not rhythmic and each movement differs from the other, not stereotyped and repetitive such as in PLMT. Some patients have dystonic toe posture that can be suppressed consciously or by pressure to the foot, similar to dystonia [2]. There are some similarities with CRPS that manifest with pain and movement symptoms such as tremor, dystonia and myoclonus which are related to local trauma, however the vascular abnormalities, trophic changes and edema which characteristic of CRPS are not signs of PLMT [3,9].

It is debatable whether PLMT is an organic or psychogenic disorder, but the precise pathogenesis underlying the development of the syndrome remains unclear [10]. Although toe movements cannot imitate voluntarily, which is against psychogenic cause, however this does not mean with certainty that the phenomenon is organic. Reduction of movements when distracted, sudden onset and remission and other nonorganic signs will help in reaching diagnosis [11] (Figure 2). In electrophysiological studies, there are wide ranges of EMG findings that can see in patients with PLMT, from normal to interesting findings. Nerve conduction velocity and needle EMG with evidence of peripheral neuropathy or chronic radiculopathy, cramps and fasciculations have found in some patients with PLMT [12]. Surface EMG can demonstrate a pattern of short, erratic, low amplitude discharges or a pattern of complex, long duration, high amplitude bursts, or random, irregular or semi continuous bursts [13]. The motor unit discharges fire in time with toes movements as irregular or semi-rhythmic phasic bursts with co-contraction and bursts lasting 10 milliseconds up to 2 seconds with rates of 1.5 to around 200 Hz [2-13]. Hassan et al. reported that PLMT associated with other diseases such neuropathy in one third, and 11% with limb trauma [2]. Tibial nerve lesions and various neuropathies have been documented [14]. Back pains with cauda equine, spinal stenosis or disc degeneration have also been reported, but most cases show very mild findings of the vertebral column on CT and MRI [5-14]. Neuroleptic drugs were suspected to cause PMLT as part of tardive dyskinesia, others suggested that can be allied to minor local trauma without evidence of nerve injury [13,15]. Herpes zoster radiculitis or myelitis have also been described as possible sources in several reports [6,16]. The sympathetic system has also been suggested, although sympathectomies and blocks were used in PLMT treatment with only transient relief [5,13]. Recently Nishioka et al. reported the results of neurological, electrophysiological and brain MRI investigations of ten patients with PLMT syndrome [10]. Eight showed hypo perfusion of prefrontal, occipital, cerebellum and thalamus, and hyper-perfusion of the anterior cingulate gyrus, parietal lobes bilaterally on brain SPECT. These areas relate to the pain matrix. Since other electrophysiological tests did not indicate specific abnormalities, the researchers concluded that PLMT is a centralized pain disorder (Figure 3).

Treatment of this phenomenon is only symptomatic as long as we do not know the definite cause. In their extensive work of 76 cases, Hassan et al. reported some improvement in leg pain or in toes movement, or both by blocking tibial nerve and epidural space with steroids, local massage,

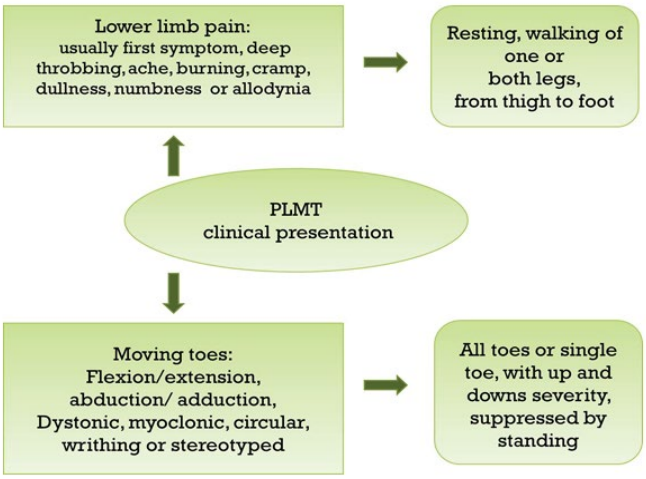

Figure 1. Clinical presentation of painful leg and moving toes.

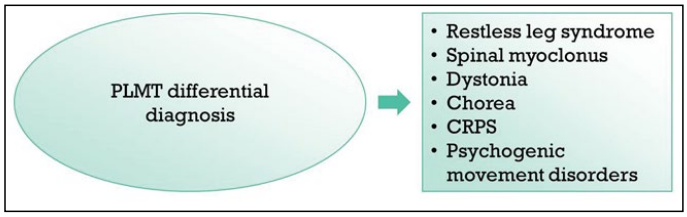

Figure 2. Differential diagnosis of painful leg and moving toes.

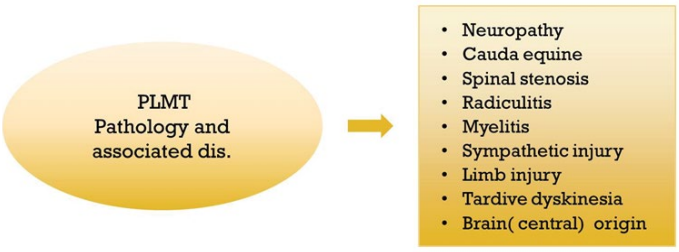

Figure 3. Pathology and associated diseases of painful leg and moving toes.

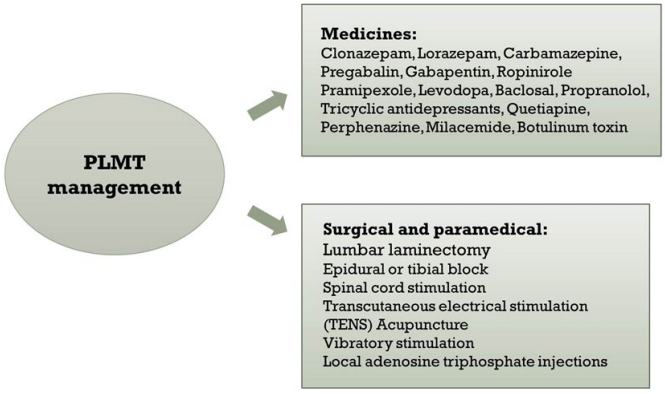

Figure 4. Treatment and management of painful leg and moving toes.

transcutaneous electrical stimulation, or acupuncture, or with medications such as levodopa, pramipexole, ropinirole lorazepam, clonazepam, carbamazepine, pregabalin, gabapentin and/or botulinum toxin, individually or in various combinations. Surgical procedures such as multiple spinal laminectomies did not help in several patients. An average of one third of PLMT patients responded to treatment, with no differences in demographic data such as age, sex, and time of onset. In fact, there was no significant difference in any of the variables investigated between those who responded positively or negatively to treatment. Another group of drugs used in management of PLMT are tricyclic antidepressants, such as baclosal, propranolol, quetiapine, perphenazine and milacemide [16,17]. Transcutaneous Electrical Nerve Stimulation (TENS) and vibratory stimulation used with some improvement in both pain and toes movements [18]. Epidural anesthesia with spinal cord stimulation has been used in three patients with partial improvement [19]. Sympathetic block was already used since 1971 by Spillane et al in four patients with transient improvement in only one patient, but there was no significant improvement following sympathectomy. Guieu treated two patients by injection of adenosine triphosphate, and reported pain relief but did not mention any effect on toes movement [20]. Although some have theorized prevalence of autoimmune disease with this phenomenon, there have been no immunosuppressive therapies except in one failed case (Figure 4).

Conclusion

In summary, this is a very uncommon phenomenon that can be missed by physicians in various fields including neurologists and pain doctors who are not familiar with the syndrome. We should be aware of the syndrome, reach diagnosis as soon as possible, diminish the patient's concern, and save unnecessary investigations.

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