

Case Reports

Possible Functional Moving Toes Syndrome

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Abstract

Background: Moving toes syndrome has been classically described as an organic movement disorder, on occasion related to peripheral nerve injuries. The association between nerve trauma and movement disorders has become a controversial topic, and the functional etiology of moving toes syndrome has recently been proposed.

Case Report: We describe two cases of moving toes syndrome with clinical features typically suggestive of a functional movement disorder.

Discussion: The presence of entrainability and distractibility in the described patients is an indication of attentional influences on their involuntary movements. However, it is possible that if there is a subcortical origin, the toe movements could be influenced by voluntary commands.

Keywords: Functional movement disorder, psychogenic movement disorder, moving toes syndrome

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Ethics Statement: All patients that appear on video have provided written informed consent; authorization for the videotaping and for publication of the videotape was provided.

Introduction

Painful legs moving toes (PLMT) syndrome is characterized by repetitive movements of the toes, typically in association with pain of the affected leg. Sometimes it occurs without pain, and can then be called “painless legs moving toes syndrome.” Both syndromes together can be referred to as “moving toes syndrome” (MTS). MTS is usually unilateral, occurs predominantly in the lower extremities, and no specific clinical or

electrophysiological markers have been established. Pain is present in most cases, predominantly described as tingling, numbness, aching, shooting, and/or cramps. Medical treatments including antiepileptics, analgesics, anesthetic nerve blocks, and botulinum toxin injections have not demonstrated a consistent benefit across patients.^{1,2}

The etiology of MTS is unclear but its association with radiculopathy, neuropathy, and spinal cord lesions has supported its organic

origin.³ Clinical features such as suppressibility with voluntary movements of the affected extremity and exacerbation with postural changes have been reported.³ We describe the presence of clinical features typically suggestive of functional movement disorders in two patients with diagnoses of MTS. Patients were evaluated at the National Institutes of Health and signed informed consent for videotaping and publication in a medical journal.

Case report

Case 1

A 52-year-old female, with a history of bipolar disorder and gastric bypass, presented with pain and involuntary movements of her left toes for 2 years. She denied a history of peripheral or spinal trauma or neuroleptic use. The movements were constant, asymmetric, and associated with throbbing pain of the extremity. She described improvement with ambulation and worsening with rest and fatigue. The symptoms had progressively worsened over time and several medication trials had failed to provide benefit, including trihexyphenidyl, levodopa, gabapentin, quetiapine, and botulinum toxin injections. No details regarding medication doses were available. Magnetic resonance imaging (MRI) of her brain was unremarkable, and a previously reported vitamin B12 deficit had already been corrected. On examination, there were no sensory or motor deficits. She demonstrated dystonic posture of her left foot and asynchronous movements of the left toes in different directions (abduction/adduction of the fifth toe, flexion/extension of the remaining toes). She displayed entrainment and distraction of the left toe movements with both hand and right toe tapping. This case is a typical presentation of PLMT with evidence of entrainability and distractibility on examination (Video 1, Segment 1).

Case 2

A 35-year-old female presented with continuous twitching movements of the right toes after open reduction and internal fixation of a right ankle fracture. She experienced mild postoperative pain of the extremity, which quickly subsided without the need for analgesic therapy. The movements disappeared during deep sleep and with motion of her toes and ankle. She improved during ambulation and while standing, and when applying pressure to the affected toes. She was able to consciously suppress the movements for brief periods of time. She denied pain, numbness, or weakness of the right foot. Neurological examination revealed repetitive involuntary movements of the right toes (flexion/extension and abduction/adduction) with the exception of the big toe. The movements were distractible and entrainable, with left foot and right hand tapping tasks at variable frequencies. The clinical picture of these movements is consistent with painless legs MTS (Video 1, Segment 2).

Discussion

We present two cases of MTS with the presence of functional characteristics on examination. In the first case, the patient demonstrated typical features of PLMT with poor response to medical treatment, and not associated with peripheral trauma or neuropathy. Our second case was preceded by a surgical procedure on the affected leg without residual pain upon evaluation; the movements were also typical of MTS. While the movements in patient 1 might be considered to be similar to tardive dyskinesia, there was no history of neuroleptic exposure before movement onset. Therefore, the presence of tardive dyskinesia in this case is unlikely. In addition, it would be unusual for tardive dyskinesias to present exclusively in one segment, and, in particular, only in the toes.²



Video 1. Two Cases of Moving Toes Syndrome with Clinical Characteristics of Functional Movement Disorders. Segment 1. Painful Leg Moving Toes. The segment demonstrates asynchronous movements of the left toes at rest. The movements occur in different directions (abduction/adduction of the fifth toe, flexion/extension of the remaining toes). The patient was subsequently asked to tap with the right hand; distraction and, on occasion, entrainment of the left toe movements were observed. In the final section of this segment, distraction with up and down movements of the opposite foot abolished the left toe movements and tapping with the opposite foot caused entrainment at low frequencies. **Segment 2. Painless Leg and Moving Toes.** The segment demonstrates repetitive involuntary movements of the right toes (flexion/extension and abduction/adduction) with the right foot at rest. Purposeful movements of the affected foot and pressing on the floor suppressed the involuntary movements. Subsequently, distraction with left foot motion (up-down) stopped right toe movements. In the final section of this clip, right hand tapping caused entrainment of the toe movements and distraction with left hand tapping abolished the toe movements.

The etiology of MTS remains unclear; however, several case series have associated this phenomenon with the presence of radiculopathy, peripheral nerve, or spinal cord lesions. It is believed that central plasticity (maladaptive) responses to peripheral nerve injuries could result in a variety of movement disorders, including tremor, dystonia, and MTS.⁴ However, the association between peripheral nerve injuries and movement disorders has become a controversial matter.⁵

MTS has typically been described as an organic disorder linked to various types of peripheral nerve injuries. Typically, no clear association of MTS with signs of functional movement disorders has been made. However, a recent report pointed out the presence of functional features in two cases of MTS where attention tasks showed distractibility and entrainability of the toe movements.⁶ These cases have questioned the organicity of this condition, setting the need for a redefinition of its clinical features.

Entrainability and distractibility are thought to be manifestations of a functional disorder, since they show that the origin of the involuntary movement is shared or can be co-opted by the voluntary movement or an element of more focused attention. This is clearly relevant for movements that have been shown to be produced by voluntary mechanisms even though they are experienced as involuntary.⁷ There is, however, another possibility to consider here: the origin of MTS is not known, but if it is subcortical, then the subcortical generator might be partially influenced by voluntary commands. An analogy might be the locomotor generator in the spinal cord that has not only intrinsic rhythmicity but can be controlled voluntarily. Some evidence for this was the occasional voluntary control observed in Case 2 and in

previous case series describing the presence of transient suppressibility with voluntary movements.³

We present further evidence of attentional influences on MTS by reporting two cases whose physical examination demonstrated entrainability and distractibility of the toe movements, both features typically found in functional movement disorders.

However, it is difficult to establish a definitive pathophysiological diagnosis and, at present, we would still consider this an organic condition.

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