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## Videographic aspect of 3 cases of Painful Legs and Moving Toes Syndrome

Kelly Guichard, Theo Michaud, Imad Ghorayeb

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2 **Videographic aspect of 3 cases of Painful Legs and Moving Toes**  
3 **Syndrome**

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6 Kelly Guichard<sup>1,2</sup>, Theo Michaud<sup>3</sup>, Imad Ghorayeb<sup>3,4,5\*</sup>

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8 <sup>1</sup>Clinique du Sommeil, Bordeaux University Hospital (CHU), Bordeaux, France

9 <sup>2</sup>Université de Bordeaux, SANPSY, USR 3413, F-30000 Bordeaux, France

10 <sup>3</sup>Département de Neurophysiologie Clinique, Pôle Neurosciences Cliniques, CHU de  
11 Bordeaux, F-33076 Bordeaux, France

12 <sup>4</sup>Université de Bordeaux, Institut de Neurosciences Cognitives et Intégratives d'Aquitaine,  
13 UMR 5287, F-33076 Bordeaux, France

14 <sup>5</sup>CNRS, Institut de Neurosciences Cognitives et Intégratives d'Aquitaine, UMR 5287, F-  
15 33076 Bordeaux, France

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22 \*Corresponding author: Imad Ghorayeb, Department of Clinical Neurophysiology, CHU de  
23 Bordeaux, Place Amélie Raba Léon, 33076 Bordeaux cedex, France; Tel: +33556795653;  
24 Fax: +33556796109  
25 E-mail address: imad.ghorayeb@u-bordeaux.fr

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## 34 **Introduction**

35           The Painful Legs and Moving Toes Syndrome (PLMTS) is a rare chronic neurological  
36 ailment. It is characterized by painful sensations in the feet or the calf with involuntary  
37 movements of the toes during wakefulness, and is often confused with Restless Legs  
38 Syndrome (RLS) [1]. Most descriptions of PLMTS are limited to case reports/series, and only  
39 two cases showed a videographic vignette [2,3]. In order to enrich the scarce literature on the  
40 topic and help sleep specialists and neurologists for a better recognition of this condition, we  
41 here describe three videographic confirmed cases of PLMTS with unique clinical diversity.

42

## 43 **Cases description**

44           Medical history, complementary investigations, previous treatment attempts and  
45 ongoing treatment are listed in table 1.

46           Case 1 was a 63-year-old woman who was referred for a pharmaco-resistant RLS that  
47 has developed since 2016. She complained from paresthesia involving the toes and the sole of  
48 both feet with right side predominance, along with involuntary movements of the toes while  
49 she was at rest and particularly at night. She was on fluoxetine for depression but  
50 antidepressant withdrawal did not improve symptoms. Examination revealed abnormal,  
51 asynchronous and irregular athetotic-like movement of the toes with a subtle right  
52 predominance consistent with PLMTS. Carbamazepine at 200 mg a day was of temporary and  
53 moderate efficacy.

54           Case 2 was a 47-year-old woman who was referred for a painful complaint affecting  
55 the legs, mostly at rest, with tingling and burning sensations of the soles along with cramps, a  
56 left predominance and nocturnal worsening, that has developed since 2015. Because of a  
57 medical history of dysuria, extensive neurological investigation was performed and was  
58 negative for neuropathy, amyotrophic lateral sclerosis and parkinsonism. Examination  
59 showed dystonic posture of the left toes with myoclonic jerks on both sides along with  
60 frequent episodes of athetotic-like crawling movements of the right toes. She admitted better  
61 improvement with clonazepam than with pregabalin with however a short duration efficacy  
62 and declined botulinum toxin.

63           Case 3 was a 73-year-old man who was referred for suspicion of RLS because of a 30-  
64 year history of painful complaint affecting the toes and the calf with night predominance. Pain  
65 was mostly perceived at rest and was partially relieved by movement. Examination showed  
66 moderate but almost permanent athetotic-like movements of the left toes along with sporadic  
67 myoclonic jerks of the right. He reported partial efficacy with paracetamol.

**68 Video analysis**

69 All patients were examined and video-recorded while awake between 10 and 12 am.

70 Video file of case 1 showing dyskinetic movements involving both feet.

71 Video file of case 2 showing three different sequences of dystonic posture of the left toes and  
72 probably of the right big toe.

73 Video file of case 3 showing moderate dyskinetic movements with left toes predominance.

74

**75 Discussion**

76 This article reports video-documented cases series of three patients with PLMTS.  
77 Compared to previous video-cases [2,3], where only rhythmic/arhythmic flexion-extension  
78 digit movements can be seen, our patients displayed a variety of irregular and asymmetrical  
79 dyskinetic involuntary movements ranging from myoclonic to athetotic-like, thereby  
80 expanding the motor clinical repertoire of PLMTS. Furthermore, no alternating pattern of  
81 movement between the two legs was observed in our patients but rather asynchronous  
82 movements with frequent side predominance. By contrast to the previous video-case [2],  
83 periodic limb movements (PLM) were seen during PSG that was performed only in case 1,  
84 but this association may be fortuitous as PLM syndrome is prevalent in the general population  
85 [4]. In any case, and as reported in the literature, pain was the major concern in our patients  
86 and none complained from insomnia.

87 Due to possible confusion with RLS, PLMTS may be underdiagnosed. Several clinical  
88 features are however unique to PLMTS as involuntary movements are strictly restricted to the  
89 feet and particularly the toes, had no clear circadian pattern although more noticeable when  
90 patients are at rest or not engaged in their daily activities (case 1, case 2 and case 3) thereby  
91 probably accounting for the perceived partial relief upon movement (case 3). In PLMTS,  
92 sensory symptoms are strictly reported as painful whereas, when present, they are described  
93 as disagreeable in a majority of patients with RLS.

94 The pathology of PLMTS is still unclear but it probably does not share similar RLS  
95 mechanism as, by contrast to the latter condition, no relief is obtained with dopamine agonists  
96 and no exacerbation of PLMTS symptoms is noted under selective serotonin release  
97 inhibitors, at least in the reported cases. Therefore, although of partial and limited efficacy  
98 duration, symptomatic therapeutic options so far failed to overcome debilitating symptoms of  
99 pain and to reduce involuntary movements of the toes, as in our cases. Better recognition of  
100 PLMTS will help refining its nosological framework and a better understanding of its  
101 underlying mechanism.

102 **Table 1:** Medical history, investigations and treatments of cases

Cases	Medical history	Investigations	Previous treatments	Ongoing treatment
Case 1 Woman 63 year-old	<ul style="list-style-type: none"> <li>Recurrent depressive disorder on Fluoxetine</li> <li>Breast adenocarcinoma in remission</li> </ul>	<ul style="list-style-type: none"> <li>Normal blood test (no iron deficiency)</li> <li>Normal cerebral and spinal MRI</li> <li>Normal electromyography</li> <li>Polysomnography revealed mild obstructive sleep apnea syndrome (IAH=16.9) and periodic legs movements (PLMi=38.7/h)</li> </ul>	Alternative therapy (acupuncture, reflexology, osteopathy) Pramipexole, Clonazepam, Pregabalin, Gabapentin, Paracetamol codeine, Botulinum toxin	Carbamazepine 200mg/day
Case 2 Woman 47 year-old	<ul style="list-style-type: none"> <li>Dysuria syndrome</li> <li>Raynaud syndrome</li> <li>Renal colic</li> <li>Surgical sterilization</li> </ul>	<ul style="list-style-type: none"> <li>Normal blood and cerebro-spinal fluid tests (no iron deficiency)</li> <li>Normal cerebral and spinal MRI and normal DATscan</li> <li>Normal electromyography</li> <li>Normal somatosensory evoked potential</li> </ul>	Pregabalin, Gabapentin, Duloxetine, Thiocolchicoside	Clonazepam (6 drops)/day
Case 3 Man 73 year-old	<ul style="list-style-type: none"> <li>Psoriatic arthritis on Methotrexate 15mg/week</li> </ul>	<ul style="list-style-type: none"> <li>Normal electromyography</li> </ul>	Paracetamol 1g/day	Paracetamol 1g/day

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105 **References:**

- 106 1. American Academy of Sleep medicine. *International Classification of Sleep Disorders,*  
107 *3rd Ed.* American Academy of Sleep medicine; 2014. Accessed September 4, 2017.  
108 <http://www.aasmnet.org/store/product.aspx?pid=849>
- 109 2. Zellini F, Ferri R, Abbafati M, et al. Video-polysomnographic aspects of painful legs  
110 and moving toes syndrome. *Sleep Med.* 2017;33:43-46.
- 111 3. Fan Y-Y, Xu Y, Gao X-G. Spontaneous aching pain and peculiar involuntary  
112 movements: a case report of painful legs and moving toes and review of the literature.  
113 *Case Rep Med.* 2014;581402.
- 114 4. Haba-Rubio J, Marti-Soler H, Marques-Vidal P, et al. Prevalence and determinants of  
115 periodic limb movements in the general population. *Ann Neurol.* 2016;79(3):464-474.

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