“Painless legs and moving toes” syndrome due to spinal cord compression

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Abstract Painful legs and moving toes is a rare syndrome characterized by spontaneous neuropathic pain in the lower limbs associated with peculiar involuntary movements of the lower extremities, especially the toes and feet. Although its origin is unknown, it is associated to some pathologies. With regard to the painless variant, very few patients have been reported and most of them are idiopathic. We report a patient with involuntary movements of the toes similar to those seen in painful legs and moving toes syndrome, but without any associated pain and due to a spinal compression. We conclude that spinal lesions may produce the painless variant as it has been reported with the painful form.

Keywords Gabapentin · Spinal compression · Moving toes

Text

“Painful legs and moving toes” syndrome is a rare condition, firstly described by Spillane in [6] about six patients who complained about pain in one or both feet and showed abnormal movements in the distal lower limbs. However, the “painless legs and moving toes” syndrome was discovered in 1993, in which the dystonic movements were not accompanied by the typical pain [8]. Although the pathophysiology remains unknown, both pictures have been proposed as the same entity, they may have an hereditary component [3] and have been related with spinal

cord lesions, lumbosacral radiculopathy, neuropathies, peripheral trauma, drugs, Wilson’s, and Hashimoto’s diseases [4, 5]. Despite that, most cases are idiopathic and most of these associations have been described in the painful form.

Concerning the painless variant, the known data are very short, since there are very few cases in the literature and most data come from the complete variant. We present a patient with a “painless legs and moving toes” syndrome due to a spinal compression. As far as we know, this is the only case in the bibliography with the same characteristics.

We report an 82-year-old woman who suffered from colonic diverticulosis, generalized arthrosis and multiple vertebral compressions. She described a 3 weeks picture of paresthesias and weakness in both lower limbs, that impeded the gait. On examination, a slight symmetric paraparesis that limited her walking (a score of M4 in manual muscle testing), global hyporeflexia and a left extensor plantar reflex were present. Additionally, dystonic movements in her toes stood out. They were low frequency (one Hz), flexion–extension and separation movements, more prominent in the left side, and they interfered with deambulation. They were increased by sensitive stimulation and persisted during sleep. Blood analyses, electrocardiogram, chest radiography and electromyography did not display any pathological finding. In the MRI (Fig. 1) a D11 vertebral compression fracture with displacement of the posterior part was appreciated. There was also a spinal cord hyperintensity from D10 to D12 that indicated a spinal contusion. Moreover, there were significant signs of arthrosis and osteoporosis. After the admission, the patient got progressively worse with a more marked paraparesis and dystonic movements. A treatment with corticoids (dexamethasone 16 mg/day) and gabapentin (900 mg/day) was started. Due to an initial

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Fig. 1 Sagittal T2-weighted magnetic resonance imaging of the thoracolumbar region of the spinal cord. A D11 osteopenic vertebral fracture and a secondary spinal contusion may be appreciated

improvement with these drugs and the patient's rejection to the surgical treatment, she was initially treated conservatively. After 2 weeks and a worsening of neurological symptoms, the patient was subjected to a decompressive laminectomy developing an epidural haematoma (displayed in a new MRI) with a marked paraparesis (a score of M1 in manual muscle testing) and the disappearance of the abnormal movements. The patient had to undergo a new surgery for this complication but she did not get better. After one year follow-up an important paraparesis (M2 in manual muscle testing) persisted.

Although the etiology of this syndrome remains unclear some theories have been proposed. Neural discharges formed in the lesion site may reorganize the central information processing, what could mean an anomalous information transmission from the dorsal horns to the ventral motor neurons producing the toes movement. Some secondary changes have been described in the central nervous system such as an imbalance between excitatory/inhibitory stimuli and formation of new synapses [2]. Different interruption degrees in the sensory afference and different central reorganization processes may take place in

a wide range of clinical possibilities, including isolated pain, abnormal movements and pain (painful variant) and isolated abnormal movements (painless variant) [1]. Within the known causes of the painful variant, the most common one is related with spinal cord lesions, so the development of the painless form as a consequence of this kind of lesion supports a common origin for these two entities. Concerning the disappearance of the toes movement after the surgery, there are two possible explanations. The first one is that the new lesion of the spinal cord resulted in a new information transmission to the ventral motor neurons, and the other one is that the disappearance of the abnormal movements of the lower limbs was just the consequence of the extreme muscle weakness (with a score of M1 in manual muscle testing).

Gabapentin has been proposed as an effective treatment for this syndrome in the last years. Its mechanism of action is mediated through voltage-dependent calcium channels blockade of the ventral horn neurons and it has been suggested that it may interrupt the events that produce the clinical picture [7]. The treatment response of our patient was partial and could be related with the inflammation decrease or with the beneficial effects of gabapentin.

Conflict of interest statement None of the authors has any potential conflict of interest.

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