

THE SYNDROME OF PAINFUL LEGS AND MOVING TOES – A CASE REPORT

A K Y Tan, C B Tan

ABSTRACT

The syndrome of painful legs and moving toes is an uncommon and distressing condition with pain in the feet or legs and involuntary movements of the toes. It can follow spinal cord or cauda equina trauma, lumbar radiculopathy, injury to the feet, peripheral neuropathy or without any preceding causes. Ephaptic transmission in damaged nerve roots or peripheral nerves with central reorganisation may be the underlying mechanism of the syndrome. Treatment is difficult. We report a case of this syndrome following peripheral neuropathy, with a good early response to the GABA agonists baclofen and clonazepam. The role of different GABA agonists in the treatment of this condition needs to be better defined.

Keywords: painful legs, moving toes, ephaptic excitation, GABA agonists, peripheral neuropathy

SINGAPORE MED J 1996; Vol 37: 446-447

INTRODUCTION

The syndrome of 'painful legs and moving toes' was first described by Spillane et al in 1971⁽¹⁾. This condition is characterised by pain in the feet or legs and involuntary movements of the toes. This distressing syndrome has been known to follow spinal cord or cauda equina trauma⁽²⁾, lumbar root lesions⁽¹⁻⁵⁾, bony or soft tissue injuries of the feet⁽⁶⁾, peripheral neuropathy⁽⁷⁾ or without any antecedents. Treatment is usually unsatisfactory. We report a case of 'painful legs and moving toes' following peripheral neuropathy with a good initial therapeutic response to the combination of baclofen and clonazepam.

CASE REPORT

A fifty-seven year old housewife developed progressively painful cramps of both calves. The pain was described as burning and severe, unrelieved by oral analgesic medication and it disturbed her sleep. One month later, she noticed involuntary wriggling movements of her toes on both feet. These toe movements persisted throughout the day and appeared to be absent when she was asleep.

She has a history of diabetes mellitus for 17 years and she was initially treated with oral hypoglycaemic medication. Two years later, she required insulin injections because of poor blood sugar control. She has the diabetic complications of retinopathy, nephropathy and peripheral neuropathy for five years. There was no history of back pain, injury or sciatica.

On physical examination, involuntary wriggling movements of the toes of both feet could be seen. Semi-rhythmic flexion-extension and occasionally abduction movement of the phalanges, especially the big toes, were present almost continuously and they could not be voluntarily suppressed. The calves were not swollen and they were slightly tender when palpated.

She has bilateral diabetic proliferative retinopathy. The cranial nerves and limb power were normal. Tendon reflexes were all absent and position sense was lost over the big toes and

both ankles. A glove and stocking distribution of hypoesthesia was detected. There was no tenderness or step over the thoracic and lumbar spine and the straight-leg raising test was normal.

Nerve conduction velocities of the peroneal and tibial nerves bilaterally were impaired with absent F wave responses. Sensory nerve action potentials could not be obtained from both sural nerves. Electromyogram of the muscles of the lower limb demonstrated a reduced interference pattern. Electrophysiological studies of the upper limbs were within normal limits. This suggested a sensori-motor polyneuropathy affecting mainly the lower limbs.

The patient was prescribed baclofen 10 mg tid and clonazepam 2 mg tid and the pain in her legs together with the involuntary toe movements improved tremendously. After eight months of effective therapy, the pain and toe movements recurred. Increasing doses of baclofen and clonazepam had only a minor beneficial effect.

DISCUSSION

In the syndrome of 'painful legs and moving toes', pain usually precedes the toe movements. The pain affects the calves and feet, and does not follow a dermatomal or peripheral distribution. It has been described as constant, burning, crushing or throbbing with co-existent hyperpathia, hyperaesthesia and allodynia. The onset may be unilateral but often becomes bilateral.

The toe movements are spontaneous and involuntary with a wriggling and writhing pattern. They consist of flexion, extension, abduction and adduction of the toes and are intermittent or continuous. An act of voluntary effort can occasionally diminish the movements but only momentarily.

The patient had diabetic peripheral neuropathy. She developed painful legs and moving toes five years after the diagnosis of peripheral neuropathy. It has been postulated that ephaptic excitation in damaged peripheral nerves may cause abnormal impulse transmission in peripheral sympathetic and sensory nerves, leading to reorganisation of the central processing of sensory information. This reorganisation leads to pain over wide areas in a non-dermatomal distribution⁽⁸⁾. Several studies have reported on changes in the pattern of neuronal activation after peripheral nerve injury occurring in the dorsal horn, dorsal column nuclei, ventral thalamus, somatosensory cortex and they have also found alterations in basal ganglia neurotransmitters⁽⁹⁾. This may lead to imbalances between the excitatory and inhibitory signals.

Treatment of this syndrome is exceedingly difficult. The distressing nature of the pain has driven patients to suicide. Neurologists prescribe combinations of baclofen,

Department of Neurology
Tan Tock Seng Hospital
Moulmein Road
Singapore 308433

A K Y Tan, MBBS, MRCP (UK), FAMS
Senior Registrar

C B Tan, MBBS, M Med (Int Med), FAMS
Consultant

Correspondence to: Dr A K Y Tan

benzodiazepines, carbamazepine and antidepressants but the effects are unpredictable and short-lived. Lumbar sympathetic blockage with local anaesthetics, phenol and guanethidine has been tried, with about 50% of patients experiencing only transient relief of pain and movements. The patient in this case report obtained very good relief of both pain and toe movements with a combination of baclofen and clonazepam. The beneficial effect, unfortunately, lasted only eight months. Baclofen and clonazepam, are both GABA receptor agonists. Baclofen affects the GABA B-type receptor while clonazepam, the GABA A-type receptor. Chronic administration of these drugs could possibly cause a downregulation of the GABA receptors and result in a diminution of therapeutic effectiveness. It is hoped that the development and usage of more potent GABA receptor agonists would eventually help alleviate the tremendous suffering of patients with painful legs and moving toes.

REFERENCES

1. Spillane JD, Nathan PW, Kelly RE, Marsden CD. Painful legs and moving toes. *Brain* 1971; 94: 541-56.
2. Nathan PW. Painful legs and moving toes: evidence on the site of the lesion. *J Neurol Neurosurg Psychiatry* 1978; 41: 934-9.
3. Wulff CH. Painful legs and moving toes. A report of 3 cases with neurophysiological studies. *Acta Neurol Scand* 1982; 66: 283-7.
4. Barrett RE, Singh N, Fahn S. The syndrome of painful legs and moving toes. *Neurology* 1981; 31(suppl 1): 79.
5. Schoenen J, Gonce M, Delwaide PJ. Painful legs and moving toes: a syndrome with different pathophysiologic mechanisms. *Neurology* 1984; 34: 1108-12.
6. Schott GD. Painful legs and moving toes: the role of trauma. *J Neurol Neurosurg Psychiatry* 1981; 44: 344-6.
7. Montagna P, Cinignotta F, Sacquegna T, Martinelli P, Ambrussetto G, Lugaresi E. Painful legs and moving toes associated with polyneuropathy. *J Neurol Neurosurg Psychiatry* 1983; 46: 399-403.
8. Dressler D, Thompson PD, Gledhill RF, Marsden CD. The syndrome of painful legs and moving toes. *Mov Disord* 1994; 9: 13-21.
9. DeCaballos ML, Baker M, Rose J, Jenner P, Marsden CD. Do enkephalins in basal ganglia mediate a physiological motor rest mechanism? *Mov Disord* 1986; 1: 223-33.