Spinal Cord (2011) 49, 754–756

www.nature.com/sc

# CASE REPORT

# Treatment-resistant sensory motor symptoms in persons with SCI may be signs of restless legs syndrome

S Nilsson[1], R Levi[1] and A Nordström[1,2]

[1]*Department of Community Medicine and Rehabilitation, Rehabilitation Medicine, Umeå University, Umeå, Sweden and* [2]*Sports Medicine, Department of Surgical and Perioperative Science, Umeå University, Umeå, Sweden*

**Study design:** Case report on the successful treatment with pramipexole in four men with chronic spinal cord injury (SCI) suffering from refractory symptoms that were previously considered to be manifestations of a post-traumatic spastic syndrome or neuropathic pain.
**Objective:** To raise awareness among health professionals regarding the diagnostic and therapeutic possibility of restless legs syndrome (RLS) and periodic limb movements (PLMs) in some patients with SCI responding poorly to conventional treatment for spasticity or neuropathic pain.
**Setting:** Neurorehabilitation department of the Rehabilitation Medicine Center of Northern University Hospital, Umeå, Sweden.
**Methods:** Medical records and clinical data were retrospectively reviewed.
**Results:** All cases obtained treatment with pramipexole, initially 0.09–0.72 mg day$^{-1}$. Two of the cases had RLS and PLMs, one RLS only and one PLMs only. All four reported symptoms in the lower extremities and one also in the upper extremities. Three patients with residual gait function reported RLS score with/without treatment as follows: 32/11, 37/12 and 33/12. One patient with complete paraplegia (with incomplete RLS score) reported 22/10. After a follow-up period of 16, 20, 43 and 49 months, respectively, all four still reported excellent outcomes. Two remained on initial dosage; one had increased dosage from 0.09 to 0.18 mg day$^{-1}$ and one from 0.27 to 0.80 mg day$^{-1}$ during the follow-up period.
**Conclusions:** In persons with SCI suffering from infralesional involuntary movements and/or dysesthesia and with poor response to conventional antispastic or analgesic treatment, the possibility of RLS or PLMs should be considered, as these conditions seem eminently treatable.
*Spinal Cord* (2011) **49,** 754–756; doi:10.1038/sc.2010.164; published online 30 November 2010

**Keywords:** spinal cord injury; restless legs syndrome; periodic limb movements

## Introduction

Spinal cord injury (SCI) typically causes an infralesional spastic syndrome. In addition, neuropathic pain at or below the neurological lesion level is common.[1]

Spasticity is preferably treated with baclofen, dantrolene sodium and other antispastic drugs,[2] whereas neuropathic pain usually responds to some degree to tricyclic antidepressants and/or antiepileptic drugs such as gabapentin and pregabalin.[3] However, some patients respond poorly to such treatment, raising the possibility of misdiagnosis. One additional neurological condition shown to occur after SCI is restless legs syndrome (RLS).[4] RLS can either be idiopathic or be comorbid with other disorders. RLS is characterized by unpleasant sensations associated with an urge to move the legs. Symptoms usually appear during evenings or during sleep. In about 80% of cases with RLS, there is the presence of repetitive stereotypical, involuntary movements of the legs, across the hip, knee and ankle joints. This symptom is known as periodic limb movements (PLMs).[4]

In our clinical practice, we have observed chronic SCI cases with troublesome and treatment-refractory symptoms, originally attributed to spasticity and/or central neuropathic pain. On closer scrutiny, they fulfilled the criteria for RLS/PLMs and subsequently responded excellently to the currently recommended treatment for this syndrome, that is, dopamine agonists. Four such cases are described in this case report.

## Patients and methods

Between 2005 and 2008, one of the authors (SN) detected symptoms of suspected RLS/PLMs among four SCI patients enrolled at the regional neuro-rehabilitation outpatient

Correspondence: Dr S Nilsson, Department of Community Medicine and Rehabilitation, Rehabilitation Medicine, Umeå University, Umeå 901 85, Sweden.
E-mail: Stefan.nilsson@vll.se
Received 31 May 2010; revised 26 September 2010; accepted 25 October 2010; published online 30 November 2010

center. What drew the attention to RLS/PLMs was that all four reported symptoms mainly during evenings and nights. Those patients with remaining motor function also reported relief when moving their extremities. All four cases were then prescribed oral pramipexole.

Treatment effect and dose adjustments were performed at subsequent outpatient visits, according to RLS scoring[5] (Table 1), clinical interview and examination.

No other attempts to treat cases of RLS/PLM were done.

## Results

Basic patient data regarding age, gender, time since SCI, time since debut of RLS, presence of PLM, neurological and ambulatory status, blood chemistry and treatment information are presented in Table 2. In order to determine whether the diagnosis of RLS was secondary to other conditions, B12, folate, creatinine, serum iron, ferritin and transferrin were measured and were found to be within normal ranges.

The initial dose of pramipexole varied between 0.09 and 0.72 mg day$^{-1}$. One patient fulfilled the clinical criteria of RLS, two for both RLS and PLMs, and one for PLMs. All four reported symptoms in the lower extremities and one also in the upper extremities. RLS score was used. Three patients with remaining gait function reported RLS score without treatment between 32/40 and 37/40. One patient with complete paraplegia (with incomplete RLS score) reported 22/32 (Table 3).

After a follow-up period of 16, 20, 43 and 49 months, respectively, all four still reported excellent results. Two

**Table 1** The scoring system for RLS symptom severity, developed by the International RLS Study Group[5]

| *Questions referring to the situation 'in the past week'* | *4* | *3* | *2* | *1* | *0* |
|---|---|---|---|---|---|
| Overall, how would you rate the RLS discomfort in your legs or arms? | Very severe | Severe | Moderate | Mild | None |
| Overall, how would you rate the need to move around because of your RLS symptoms? | Very severe | Severe | Moderate | Mild | None |
| Overall, how much relief of your RLS arm or leg discomfort did you get from moving around? | No relief | Mild relief | Moderate relief | Either complete or almost complete relief | No RLS symptoms to be relieved |
| How severe was your sleep disturbance due to your RLS symptoms? | Very severe | Severe | Moderate | Mild | None |
| How severe was your tiredness or sleepiness during the day due to your RLS symptoms? | Very severe | Severe | Moderate | Mild | None |
| How severe was your RLS as a whole? | Very severe | Severe | Moderate | Mild | None |
| How often did you get RLS symptoms? | Very often (6–7 days in 1 week) | Often (4–5 days in 1 week) | Sometimes (2–3 days in 1 week) | Occasionally (1 day in 1 week) | Never |
| When you had RLS symptoms, how severe were they on average? | Very severe (8 h or more per 24 h) | Severe (3–8 h per 24 h) | Moderate (1–3 h per 24 h) | Mild (less than 1 h per 24 h) | None |
| Overall, how severe was the impact of your RLS symptoms on your ability to carry out your daily affairs, for example, carrying out a satisfactory family, home, social, school or work | Very severe | Severe | Moderate | Mild | None |
| How severe was your mood disturbance due to your RLS symptoms—for example angry, depressed, sad, anxious or irritable? | Very severe | Severe | Moderate | Mild | None |

Abbreviation: RLS, restless legs syndrome.

**Table 2** Clinical data of the four SCI patients

| *Patient* | *Age at injury (years)* | *Type of injury* | *Age at onset of symptoms (years)* | *RLS* | *PLMs* | *Ambulatory capability* | *B12 pmol/l (145–637)* | *Folate nmol/l (7–40)* | *Creatinine μmol/ (60–100)* | *Serum iron μmol/l (9–34)* | *Transferrin g/l (1.87–3.19)* | *Ferritin μg/l (30–400)* |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | 21 | Incomplete paraparesis | 39 | Yes/lower extremities | Yes/lower extremities | Yes | 286 | 12 | — | — | — | 88 |
| 2 | 61 | Complete paraparesis | 61 | No | Yes/lower extremities | No | 456 | 18 | 64 | 18.3 | 2.33 | — |
| 3 | 25 | Central cord syndrome | 25 | Yes/upper and lower extremities | Yes/lower extremities | Yes | 338 | 15 | 54 | 23.8 | 2.0 | 142 |
| 4 | 23 | Incomplete paraparesis | 23 | Yes/lower extremities | No | Yes | 160 | 10.8 | 70 | — | — | 254 |

Abbreviations: PLMs, periodic limb movements; RLS, restless legs syndrome; SCI, spinal cord injury.

**Table 3** Treatment and outcome of treatment of the four patients

| *Patient* | *RLS score before treatment* | *Age when commencing treatment with pramipexole (years)* | *Initial treatment with pramipexole in mg per day* | *Length of treatment at follow-up (months)* | *Treatment with pramipexole in mg per day at follow-up* | *RLS score at follow-up* |
|---|---|---|---|---|---|---|
| 1 | 32/40 | 49 | 0.54–0.72 | 49 | 0.54–0.72 | 11/40 |
| 2 | 22/32 | 65 | 0.09 | 16 | 0.18 | 10/32 |
| 3 | 37/40 | 26 | 0.27 | 43 | 0.81 | 12/40 |
| 4 | 33/40 | 25 | 0.09–0.18 | 20 | 0.09–0.18 | 12/40 |

Abbreviation: RLS, restless legs syndrome.

reported no increase in drug dosage, one had increased drug dosage from 0.09 to 0.18 mg/day and one from 0.27 to 0.80 mg/day during follow-up.

No side effects of pramipexole were reported.

## Discussion

The limitations of a case study are obvious and restrict those conclusions that can justifiably be drawn. However, an increased awareness of the possibility of RLS/PLMs in persons with SCI is of importance, as adequate treatment is indeed available and the risk of confusing the patients' symptoms with those of post-traumatic spasticity and/or neuropathic pain is high. Conventional antispastic and analgesic treatment are unlikely to alleviate the symptoms of RLS/PLMs. In doubtful cases, a trial ex juvantibus with a dopamine agonist such as pramipexole might be warranted.

In conclusion, when treating patients with SCI suffering from infralesional involuntary movements or dysesthesia, especially if conventional antispastic and/or analgesic treatment fails, the possibility of RLS or PLMs should be considered.

## Conflict of interest

The authors declare no conflict of interest.

## References

1 Sjolund BH. Pain and rehabilitation after spinal cord injury: the case of sensory spasticity? *Brain Res Brain Res Rev* 2002; **40**: 250–256.
2 Adams MM, Hicks AL. Spasticity after spinal cord injury. *Spinal Cord* 2005; **43**: 577–586.
3 Siddall PJ, Taylor DA, Cousins MJ. Classification of pain following spinal cord injury. *Spinal Cord* 1997; **35**: 69–75.
4 Stiasny K, Oertel WH, Trenkwalder C. Clinical symptomatology and treatment of restless legs syndrome and periodic limb movement disorder. *Sleep Med Rev* 2002; **6**: 253–265.
5 Allen RP, Picchietti D, Hening WA, Trenkwalder C, Walters AS, Montplaisi J. Restless legs syndrome: diagnostic criteria, special considerations, and epidemiology. A report from the restless legs syndrome diagnosis and epidemiology workshop at the National Institutes of Health. *Sleep Med* 2003; **4**: 101–119.