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Tremor and Other Hyperkinetic Movements

Case Reports

Restless "Lower Back" in a Patient with Parkinson's Disease

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Abstract

Background: In restless legs syndrome (RLS), the isolated involvement of other body parts in the absence of leg involvement is rare.

Case report: We report an 82-year-old male with a 1-year history of Parkinson's disease (PD) who developed an abnormal sensation limited to his "lower back." He fulfilled the four essential RLS criteria, with the major caveat that the criteria were applied in a modified manner to his lower back rather than his legs. The administration of a dopamine agonist completely eliminated his symptoms.

Discussion: Our patient's "restless lower back" may be a variant of RLS. Clinicians should pay attention to restlessness in other body parts in addition to the legs.

Keywords: Restless lower back, Parkinson's disease, restless legs syndrome

Citation: Suzuki K, Miyamoto M, Miyamoto T, et al. Restless "lower back" in a patient with Parkinson's Disease. Tremor Other Hyperkinet Mov 2013; 3: http://tremorjournal.org/article/view/195

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Editor: Elan D. Louis, Columbia University, United States of America

Received: July 11, 2013 **Accepted:** August 12, 2013 **Published:** October 7, 2013

Conflict of interest: The authors report no conflict of interest.

Financial disclosure: None.

Introduction

Restless legs syndrome (RLS), first fully described by Ekbom, is characterized by an urge to move the legs accompanied by abnormal leg sensations.[1] There are currently no reliable biological markers or tests for RLS, and the diagnosis of RLS should be made when the following four features are simultaneously present, as confirmed by a medical interview: 1) an urge to move the legs, 2) beginning or worsening of symptoms at rest, 3) partial or total relief by movement, 4) worsening or occurrence of symptoms only in the evening or night.[2] The diagnosis is often difficult because it mainly relies on the subject's complaints and descriptions of an unpleasant sensation in the legs, which vary among individuals.[3] The pathophysiology of RLS remains unclear, but it may be associated with central dopaminergic dysfunction, iron insufficiency, and/or an altered endogenous opioid system.[4]

The parts of the leg involved vary considerably in RLS patients. Other parts of the body, such as the arms, hips, trunk, or face, may also be involved in RLS, particularly in severe cases.[2] Although isolated involvement of body parts other than the legs is rare, sensory disturbances resembling RLS initially confined to the arms, abdomen, and perineum have been described.[5–8] Umehara et al[9] reported a patient who initially had abnormal sensations in the chest and back, which subsequently extended to the arms and legs.

We report a patient with Parkinson's disease (PD) with abnormal sensations and restlessness confined to the lower back, thought to be a variant of RLS.

Case report

An 82-year-old male with a 1-year history of PD complained of difficulty falling asleep because of abnormal itching sensations in his lower back. The patient noted an irresistible urge to move his lower back. These sensations only occurred during night and at rest, 4–5 days per week for 1 hour per night and lasted for 3 months. He could not resist rubbing his lower back on the bed sheet or with his fingers, and this resulted in relief of his symptoms, during which he had no abnormal sensations or restlessness in other parts of the body. Rocking his lower back was also effective. There were no skin lesions. The patient did not smoke or drink alcohol and occasionally drank caffeine-containing beverages. He had predominantly left-sided bradykinesia and rigidity and showed a resting tremor in the left hand while walking. He had been treated with 100 mg L-dopa with peripheral decarboxylase inhibitor twice daily at 6 am and 6 pm with a favorable response for 6 months. The Hoehn and Yahr scale score was 2, and the Unified Parkinson's Disease Rating Scale part III score was 14. The patient had no motor complications. His blood test results showed mild renal dysfunction: urea nitrogen was 29.6 mg/dL (reference value

Table 1. Patients with Restlessness Initially Occurring in Body Parts Other than the Legs

Author	Year	No./Age (Y)/Sex	Initially Involved Body Parts	Family History of RLS	Subsequent Emergence of RLS/* Follow-up Period of Restlessness	Periodic Limb Movement During Sleep	Laboratory Test Results	Comorbid Diseases	Treatment
Freedom et al[8]	2003	1/78/M	Arms	Negative	Yes/2 years	Yes	Unremarkable	Coronary artery disease, coronary bypass surgery, benign prostatic hypertrophy, hiatal hernia, and pulmonary tuberculosis	Partial improvement with clonazepam and gabapentin. Marked improvement with ropinirole
Horvath et al[5]	2008	2/39/M	Arms	Not described	Yes/several years	Not examined	Unremarkable except for an increased IgE level	Hay fever, asthma, coronary bypass surgery	Pramipexole 0.5–1.5 mg
Umehara et al[9]	2010	3/55/M	Chest and back	Negative	Yes/2 or 3 days	Not examined	Not described	Unremarkable	Clonazepam 0.5 mg
Pérez-Díaz et al[6]	2011	4/62/M	Abdomen	Negative	No/19 years	Yes	Unremarkable	Unremarkable	Pramipexole 0.18–0.65 mg
		5/62/M	Abdomen	Negative	No/6 years	Yes	Low ferritin level	Iron deficiency anemia due to Barrett esophagus	Pramipexole 0.18 – 0.70 mg, oral iron, pregabalin 150 mg
		6/62/F	Abdomen	Negative	Yes/1.5 years	Yes	Unremarkable	Anemia related to interferon and ribavirin therapy for hepatitis C infection	Pramipexole 0.18–0.36 mg
Present case	2013	7/82/M	Lower back	Negative	No/1 year and 3 months	Not examined	Unremarkable except for mild renal dysfunction	PD	Ropinirole 0.25–0.5 mg in conjunction with levodopa 200 mg

Abbreviations: IgE, Immunoglobulin E; PD, Parkinson's Disease; RLS, Restless Legs Syndrome.
*Follow-up period was between symptom onset in body parts other than the legs and development of RLS in the case of patients who subsequently developed RLS.

8.0–20.0), creatinine was 1.21 mg/dL (reference value 0.65–1.09) and the estimated glomerular filtration rate was 44.5 mL/min/1.73 m^2 (reference value >60.0). The remainder of the laboratory data was within the normal range, including liver and thyroid function and serum iron, ferritin, folic acid, vitamin B1 and B12, and hemoglobin levels. The abnormal sensation and restlessness in his lower back were not associated with prolonged standing, sitting, or lying in the same position, and changing position did not improve his symptoms, suggesting that positional discomfort was unlikely. Other medical conditions, such as peripheral neuropathy, radiculopathy, vascular problems, muscle cramps, and arthritis, were unlikely in our patient. Also, pain and motor complications, such as the wearing-off phenomenon, dystonia, akathisia, and tardive dyskinesia, were not observed. The patient fulfilled the four essential diagnostic criteria for RLS, with the major caveat that the criteria were applied in a modified manner to his lower back rather than his legs. A variant of RLS called "restless lower back" was considered. The administration of 0.25 mg ropinirole at 8 pm (1 hour before bedtime) completely eliminated his symptoms within a few days. After the 6-month follow up period, restlessness limited to the lower back recurred once, but increasing the dose of ropinirole to 0.5 mg resulted in immediate relief. He has not developed restlessness in other body parts, including his legs, over an 18-month period of dopaminergic therapy.

Discussion

Ondo et al reported that 20.8% of PD patients had symptoms of RLS. Compared with patients with isolated RLS, patients with PD with RLS had an older age at onset and were much less likely to report a family history of RLS. There is no evidence that RLS may predispose to subsequent development of PD.[10] Our patient had abnormal sensations and restlessness confined to the lower back, which was thought to be a variant of RLS, similar to restless abdomen and arms,[5,6] given that his symptoms showed an excellent response to dopamine agonist treatment and that he fulfilled the four essential RLS criteria with the major caveat that the criteria were applied in a modified manner to his lower back rather than his legs. Our patient did not show motor complications, such as dystonia, akathisia, wearing-off phenomenon, or tardive dyskinesia over an 18-month period of dopaminergic therapy. However, dopaminergic treatment may either mask or augment coexisting RLS symptoms in PD,[11] and an association between long-term dopaminergic treatment and RLS development in PD patients has been reported.[12] In addition, clinical overlap between RLS, wearing-off-related lower-limb discomfort, restlessness, and akathisia has been suggested.[11,13]

RLS may involve other body parts, such as the arms and trunk.[2] One-third of RLS patients have restlessness in the arms.[14] Approximately 50% of 230 RLS patients had symptoms in the arms, and in RLS patients with arm restlessness, RLS severity was greater than in those without arm restlessness.[15] Worsening of RLS symptoms and augmentation can include other parts of the body, including the face, arms, and trunk in addition to involvement of the legs.[2] The involvement of other body parts in the absence of the leg involvement is rare. Table 1 shows previously reported patients with restlessness initially confined to body parts other than the legs.[5–9] Dopaminergic therapy resulted in marked improvement of the restlessness in all patients, supporting the hypothesis that their isolated symptoms in other parts of the body are variants of RLS. Umehara et al[9] described a patient with restlessness initially confined to the chest and back, but the patient's symptoms subsequently extended to the legs. In contrast, our patient exhibited "restless lower back" as the initial and confined symptom over a follow-up period of 15 months.

Although the exact pathogenesis of RLS remains unclear, dysfunction of the dopaminergic A11 nucleus of the hypothalamus[16] and brain iron insufficiency[17] have been suggested. In idiopathic RLS, positron emission tomography/single positron emission computed tomography (PET/SPECT) studies have not produced consistent findings of striatonigral dopaminergic dysfunction.[18,19] In contrast, the typical motor symptoms in PD result from degeneration of the striatonigral dopaminergic neurons as confirmed by reduced striatal uptake observed in neuroimaging studies. In our patient, we suggest that striatonigral and/or hypothalamic dopaminergic involvement might have involved a projection to the T10-L2 spinal segments, resulting in abnormal sensations in the lower back. This hypothesis is in agreement with the suggestion that the T7–12 spinal segments may account for the restless abdomen.[6] However, why selective involvement of spinal segments corresponding to the lower back occurred in our patient is unclear. In PD, pain of central origin may involve restlessness, and unusual pain syndromes involving many parts of the body have been reported.[20] Parkinsonian akathisia is thought to result from a dopaminergic deficiency involving the mesocortical pathway, which originates in the ventral tegmental area.[20] Ondo recently described two patients showing semi-rhythmic leg movements that mimicked myoclonus.[21] The patients denied having any urge to move or abnormal sensations in the legs; however, the leg movements partially improved while standing and dramatically improved with pramipexole treatment, suggesting that these patients may represent the isolated motor component of RLS in the setting of spinal cord pathology. Moreover, restlessness in the legs without fulfilling RLS criteria, referred to as "leg motor restlessness," has been reported to be more frequent in untreated PD patients than in controls. It remains an open question whether patients with leg motor restlessness (but not fulfilling RLS diagnostic criteria) develop full-blown RLS.[22]

In conclusion, physicians should be aware that RLS-related restlessness can occur in other parts of the body in the absence of episodes of worsening or augmentation of restlessness. The diagnosis of a variant of RLS would be of great importance in view of the significant response to dopaminergic treatment, which considerably ameliorates patients' sleep problems and improves their quality of life.

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