


## Restless limbs and bladder: an atypical variant of restless legs syndrome case report

### *Huzursuz uzuvlar ve mesane: atipik huzursuz bacak sendromu varyantı vaka sunumu*

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### ABSTRACT

Restless Legs Syndrome (RLS) primarily affects the legs, but atypical variants involving other body parts have been described. We present a 42-year-old patient presented with 10-year history of urinary urgency along with pain, numbness, and itchiness in the right hand and foot, which worsened and started affecting the left side over the past six months. The symptoms occurred predominantly at night, improved with urination and movement, and affected daily activities. Symptoms completely resolved with pramipexole treatment. The diagnostic criteria for RLS acknowledge atypical variants involving various body parts. Reported cases include genital, bladder, abdominal, bowel, head, oral, and phantom limb variants. Clinicians should consider therapeutic testing, such as with low-dose dopamine agonists, when there is a high clinical suspicion.

**Keywords:** Restless legs syndrome, pramipexole, dopamine agonists.

### ÖZ

*Huzursuz Bacaklar Sendromu (HBS) öncelikle bacakları etkiler, ancak diğer vücut bölgelerini içeren atipik varyantlar da tanımlanmıştır. Bu yazıda 10 yıldır olan, son altı aydır sol tarafını da etkilemeye başlayan, idrar hissi ile birlikte sağ el ve ayaklarda ağrı, uyuşma ve kaşıntı şikayeti olan 42 yaşında bir hasta sunulmuştur. Semptomlar ağırlıklı olarak geceleri ortaya çıkmakta, idrar yapma ve hareketle düzelmekte ve günlük aktiviteleri etkilemekteydi, pramipeksol tedavisi ile tamamen düzeldi. HBS için mevcut tanı kriterleri, çeşitli vücut bölgelerini içeren atipik varyantları kabul etmektedir. Bildirilen vakalar arasında genital, mesane, abdominal, bağırsak, baş, oral ve fantom uzuv varyantları bulunmaktadır. Klinisyenler, yüksek klinik şüphe halinde düşük doz dopamin agonistleri gibi terapötik testleri düşünmelidir.*

**Anahtar Sözcükler:** Huzursuz bacak sendromu, pramipeksol, dopamin agonistleri.

### INTRODUCTION

Restless Legs Syndrome (RLS) is a sensorimotor sleep disorder primarily affecting the legs (1). While severe RLS can sometimes involve other body parts, atypical cases with little or no leg involvement have been described, representing likely variants of RLS (2). These variants can

affect the head (3), oral cavity (4), arms (1), abdomen (5), bladder (6), genital area (7) or even phantom limbs (8). Similar to classical RLS, variant cases typically respond to dopaminergic treatment, such as low dose dopamine agonist pramipexole.

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## Case Presentation

A 42-year-old female visited our clinic with a 10-year history of right hand and foot pain, numbness and itchiness in tips of toes and fingers, urging her to move them. Initially occurring once a month, especially when her menstruation was close, the symptoms worsened over the past six months, becoming a nightly occurrence. The symptoms would start abruptly in the right leg when the patient lay down to sleep and would improve with urination and movement of the right limbs. The patient experienced these episodes five to twenty times per night, lasting three to four hours with episodes occurring approximately every ten minutes. In severe occasions, the symptoms would extend to the left extremities. This affected the patient's daily activities and even prevented long-distance travel due to the frequent urge to urinate. The patient had previously been evaluated and followed by Department of Urology, cystoscopy revealed no abnormalities, treated by anticholinergics and mirtazapine which did not improve the symptoms. Neurological examination showed slightly brisk deep tendon reflexes in all extremities, most prominent in the right upper extremity. Blood work revealed low folate levels, while ferritin, iron, thyroid, and vitamin D were within normal range. Electromyography did not show polyneuropathy. Cranial and spinal MRI scans showed no abnormalities.

We initiated treatment with pramipexole at 0.25mg and increased to 0.5mg per day, resulting in the complete resolution of symptoms. The patient reported that they had forgotten to take pramipexole for one night, and as a result, the pain and urges persisted until the next morning.

The patient mentioned an improvement in their ability to travel and work more efficiently, and they were relieved from their pain and other troublesome symptoms.

## DISCUSSION

The International Restless Legs Syndrome Study Group (IRLSSG) diagnostic criteria updated in 2014 acknowledges the presence of atypical presentations of RLS in various body parts (1). Instead of considering these conditions as separate restless syndromes, we support the perspective of categorizing them as RLS variants. The exact mechanism of RLS and its

variants is not fully understood, but it may involve hyperexcitation of the spinal cord and alterations in the dopaminergic system in the brain and spinal cord (6). As most RLS patients initially benefit from dopamine agonists, insufficient response to dopamine agonists should raise concerns about the diagnosis of RLS (1). Recently, several genetic variants have been associated with the risk of RLS such as PTPRD, BTBD9, and MEIS1 (10).

Clinicians reported variants of RLS with a wide range of symptoms, all responding to dopamine agonists. The genital variant is characterized by persistent and excessive arousal sensations and may be associated with small fiber sensory neuropathy of the dorsal nerve of the clitoris (7). The bladder variant manifests as an urge to void (6). The abdominal variant presents with abdominal twitching, tickling, and epigastric sparks (5). The bowel variant is indicated by an urge to defecate, often accompanying irritable bowel syndrome (9). Chronic tension type headaches and treatment response with dopamine agonists may be a signal to head variant (3). A dopa-responsive abnormal oral sensation resembling burning mouth syndrome can also be classified as a RLS variant (4). Furthermore, RLS can occur in phantom limbs (8).

Following the exclusion of other diagnoses through MRI, blood work, cystoscopy, and electromyography, and considering the pramipexole response without a concurrent response to anticholinergics and mirtazapine, we diagnosed our patient with a variant of RLS. Our report is noteworthy due to the presence of one-sided arm and leg (hemi-) symptoms, accompanied by bladder urgency. This presentation is distinct from previously reported cases, which predominantly featured either isolated bladder symptoms or a combination of bladder symptoms along with leg involvement (2, 6).

Increased awareness of atypical RLS variants is crucial for accurate diagnosis and appropriate management. Clinicians should be aware of the atypical variants of RLS and consider therapeutic testing, such as with low dose dopamine agonists, when there is a high clinical suspicion.

**Conflicts of interest:** The authors have no conflicts of interest to declare.

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