

Restlessness in right upper limb as sole presentation of restless legs syndrome

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ABSTRACT

Restless legs syndrome (RLS) rarely affects the upper limb during the initial course of disease. We present a patient who complained of symptoms suggesting RLS in the right upper limb as the sole manifestation of illness. Bilateral cervical ribs and depression were co-incidental findings. Patient responded well to dopaminergic therapy.

Key words: Cervical ribs, depression, restless leg syndrome

Introduction

Restless legs syndrome (RLS) commonly involves the lower limbs and may affect the arms. Symptoms in arms often follow symptoms in the legs and appear late in the course of disease.^[1,2] To the best of our knowledge, there are only 2 case reports that describe RLS symptoms in the arm as the initial manifestation of RLS.^[3,4] We present a patient who complained of symptoms suggesting RLS in right upper limb as the sole manifestation of her illness.

Case Report

A 28-year-old female was presented with a 7-month history of tickling sensation, felt deep inside the muscles of right upper limb. The symptoms started slowly over a period of 1 month and progressed gradually. The arm and the forearm were affected in equal magnitude. The sensations appeared during night and were associated with an urge to move the affected extremity. Movement of the right arm and forearm brought relief but the symptoms recurred with the cessation of movements.

Typically, symptoms lasted for 4-6 hours/day for at least 4 nights a week. She also complained of daytime symptoms like lethargy and somnolence on almost all days. These symptoms were not following any dermatomal pattern; not associated with any motor weakness; never experienced during the day; and never interfered with the use of any of the upper limbs in daily chores. She did not notice any aggravating factor for the symptoms except the diurnal variation. The patient did not report any history to suggest medical, surgical or psychiatric illness before the onset of these symptoms. Similarly, onset of symptoms was not associated with any drug intake or any stressful life event. As reported by her, none of the family members had RLS or any psychiatric disorder.

She had been taking treatment (Diclofenac 50 mg orally twice a day and Vitamin B complex formulation orally once a day) for 15 days, without any improvement. Her X-ray neck AP view depicted bilateral cervical ribs [Figure 1]. Mental status examination showed decreased psychomotor activity and depressed anxious affect. Neurological assessment-motor power, deep tendon reflexes and sensory examination (touch, pain, pressure, temperature, graphesthesia) of all the limbs did not show any abnormality. Specifically, in both the upper limbs, power in abductors, adductors and rotators of shoulder; flexors and extensors of arm, forearm and hand; and hand grip was adequate (Grade 5) and bilaterally equal. Provocation tests for the disputed thoracic outlet syndrome were done.^[5] Elevated arm stress test, Wright's

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Figure 1: X-ray showing bilateral cervical ribs

test, Cyriax test and cervical rotation lateral flexion test were performed, which were negative.^[5] Her radial pulses were of equal volume on both the sides and she was not having any sensory or vascular symptoms on movement of either of the shoulders. Considering the clinical features and clinical examination, further examination through radiological investigation of cervical spine or level-1 polysomnography was not considered.

Diagnosis of the RLS was made. Severity of the symptoms was adjudged with the help of IRLS Hindi version^[6] and Insomnia Severity Index (ISI) Hindi version.^[7] IRLS score was 34 and ISI score was 14. Her total iron binding capacity (TIBC) was 526 µg/dl (range = 274-385 µg %) and serum ferritin was 4.02 ng/ml (range = 22-112 ng/dl). She was given tablet pramipexole 0.25 mg once a day in the evening along with the oral iron therapy (ferrous sulfate 150 mg twice a day).

She was followed up after 2 weeks. She reported a significant improvement in RLS (IRLS score 7). Despite improvement in RLS, she had difficulty in falling asleep and non-refreshing sleep persisted (ISI score = 10). Her clinical history was reviewed, which revealed that sleep problems and sadness of mood were present since past 6 months, i.e., after the onset of RLS symptoms. Her diagnosis was reviewed and major depressive disorder with co-morbid insomnia was added to the initial diagnosis of RLS. Hamilton rating scale for depression (HAM-D) was administered (Score = 23). Duloxetine 20 mg once a day orally along with clonazepam 0.125 mg twice a day orally was added to the pramipexole and oral iron therapy.

Follow-up after 3 weeks showed a substantial improvement in the mood (HDRS = 11) and the sleep (ISI = 4) with almost complete remission of RLS symptoms (IRLS = 2).

Discussion

In the present case, the symptoms of RLS were seen in one arm only, and that too, in the absence of leg symptoms. In addition, daytime symptoms did not improve till the psychiatric co-morbidity was treated.

RLS commonly affects the legs. In the cases described earlier, the patients were having sensory symptoms in both arms.^[3,4] Clinical picture was different in the present case as symptoms were seen in one arm only. An urge to move the affected extremity was reported with increment of problem during the night. In addition, arm movement brought relief and rest worsened the symptoms. Thus, all of the 4 essential criteria for RLS were met.^[1,2] Earlier, the arm restlessness and the periodic arm movements were reported in nearly 43% of idiopathic RLS patients, but in association with leg symptoms.^[8] A good response to pramipexole further substantiated the diagnosis.^[2]

Presence of bilateral cervical ribs and co-morbid psychiatric symptoms necessitated to consider other explanations of symptoms viz., thoracic-outlet-syndrome (TOS), cervical radiculopathy, depression and anxiety in this case. We are discussing each of these diagnostic entities here to have a clearer understanding. Cervical ribs may be seen in nearly 1% of population, however, these are symptomatic in just 10% of the patients.^[5] Neurogenic TOS is the most common presentation (90%).^[5] One interesting phenomenon seen in TOS is the release phenomenon when symptoms are predominantly seen during night.^[5] However, all the provocation tests for TOS, including Cyriax test was negative in this case., so as the history and clinical examination.^[5] In addition, movement of the affected limb relieves symptoms in RLS. Contrarily, elevation of arms, especially lifting a load aggravates symptoms of TOS.^[9] Hence, TOS was ruled out. Absence of dermatomal localization and relief with movement ruled out cervical radiculopathy. Anxious patients may present with restlessness, however, in such cases restlessness is primarily psychic. Somatic restlessness, even if present, involves whole of the body rather than a particular body part. Depression when co-morbid with anxiety may present with somatic restlessness. However, in this case, restless sensations were limited to one limb; responded well to dopamine agonists; well before the start of antidepressant and clonazepam; and lastly, depression appeared after onset of RLS. Hence, psychiatric illness or antidepressant as a cause for limb symptoms can be ruled out. She was suffering from iron deficiency anemia; however, details regarding the etiological diagnosis are beyond

the scope of this report. However, considering this, oral iron therapy was initiated along with pramipexole. Significant improvement in her symptoms of the arm was noticed at the end of 2 weeks, before initiating duloxetine. Pramipexole has never been reported to improve neuropathy and, oral iron therapy cannot replenish iron stores in such a short duration. One report is available that used oral iron therapy for the management of iron-deficient RLS and showed that RLS symptoms were present even after 12 weeks.^[9] On the other hand, pramipexole has been found to be effective within 2 weeks of treatment.^[10]

Insomnia is commonly associated with RLS as well as the major depressive disorder (MDD). Sleep deprivation induced by RLS may produce daytime symptoms e.g., loss of energy, inability to concentrate and psychomotor retardation, which may be mistaken for depression.^[11] Diagnosis of MDD was considered in the present case because patient complained of 2 cardinal features of depression—sad mood and anhedonia.^[11] Antidepressants are known to aggravate symptoms of RLS to variable extents.^[12] Antidepressant-associated RLS often appears early in the course of antidepressant therapy particularly with selective serotonin reuptake inhibitors and tricyclics, and are more common in men as compared to women.^[12,13] In general, norepinephrine reuptake inhibitors and bupropion have been found safe.^[12,14] Further, antidepressants may not be directly responsible for the appearance of RLS, as it is seen only in a small number of patients.^[12] Rather, hypothyroidism and estrogen therapy may be the intermediate substrates for development of RLS after antidepressants.^[15] Considering these facts, we chose to prescribe duloxetine, which brought relief to the patient.

In conclusion, RLS may affect only one arm in the early course of illness, albeit rarely. A high index of suspicion is required not to miss such cases.

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