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Familial RLS with Nocturnal Eating - Case Report and Review of Literature

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Abstract

Restless Leg Syndrome (RLS) is a common neurological disorder, which disrupts life and sleep considerably for those who have it. Its diagnosis is clinical, based on essential criteria of International RLS Study Group. It can be idiopathic or associated with various medical and other neurological disorders. Idiopathic RLS can be sporadic or may have a familial inheritance, with several genetic loci reported. RLS has a strong association with Periodic Limb Movements (PLM), both during sleep and wakefulness. Very few studies of familial RLS/PLM in sleep and their associations have been reported from India. Nocturnal eating is a non-motor manifestation of RLS with several clinical implications. We report an Indian family with RLS and PLMs as well as dysfunctional nocturnal eating.

Keywords: Restless leg syndrome; Familial; Nocturnal eating

Introduction

Ever since the Restless Legs Syndrome (RLS) was described by Ekbom in 1945 [1], it has been noted with a variable age of onset [2] and to occur in several members of the same family. Nocturnal Eating (NE) and RLS were initially described as separate sleep disorders [3]. However, recent research shows that these disorders may be more related than previously thought. Presently, no family studies of RLS/NE are available from the Indian subcontinent. Here we present a case with this association and review the available literature.

Case Presentation

Our proband, a 56-year-old married female, presented with complaints of paresthesia, restlessness, tingling and crawling sensations in both legs during the evening hours with an urge to move the legs or walking for relief, along with intermittent rhythmic movements of limbs on and off for the last 16 years. These movements usually woke her up after about 1 h to 2 h of sleep, requiring her to walk around the house, engage in activities such as emptying clothes out of the cupboard and rearranging the contents to get some relief. The unpleasant sensations were more common during periods of rest and improved by walking and other activities. This clinical history was suggestive of a diagnosis of RLS with PLMs and a nocturnal compulsive behavior. Along with this, for the last 4 years, she had been observed to wake up at night to eat excessive amounts of sweet and cookies in spite of having had dinner a few hours ago. On questioning, the patient explained that the urge to eat prevented her from falling asleep and once food was ingested, she could go back to sleep. She also reported loss of appetite on the following day and such symptoms occurred 2 to 3 times in a week. She did not have any psychiatric or medical comorbidities nor was she on any long-term medications. A neurological examination was unremarkable with no evidence of Parkinsonism or peripheral neuropathy. A complete blood count, biochemical profile, thyroid function tests, blood sugar, serum cortisol, vitamin B12 and iron studies were normal. Neuroimaging did not reveal any abnormalities of the brain or pituitary. Nerve conduction studies including Sympathetic Skin Response (SSR) were also normal. Vitamin D levels were found to be insufficient and appropriate supplementation was initiated along with dopamine agonist, ropinirole leading to relief in symptoms of both RLS and NE. A detailed family history revealed that the patient had 6 daughters and 1 son, the elder 5 daughters

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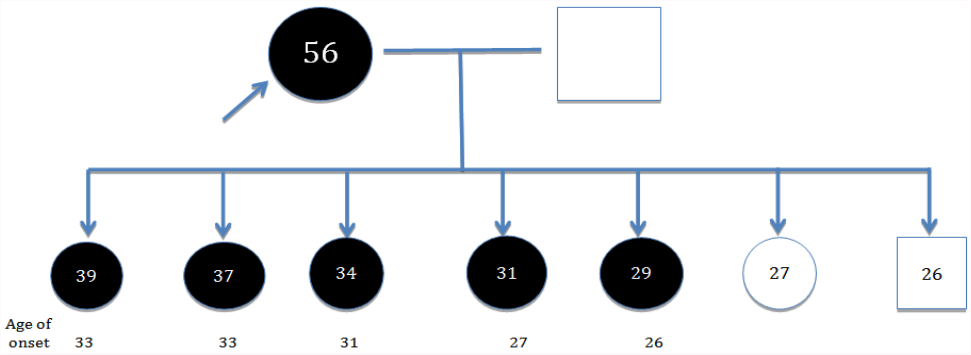

Figure 1: Family Tree with age of onset and respective IRLS scores.

Table 1: Definition and Diagnostic Criteria for Sleep-Related Eating Disorder.

A. Recurrent episodes of involuntary eating and drinking occur during the main sleep period.
B. One or more of the following must be present with the recurrent episodes of involuntary eating and drinking:
1. Consumption of peculiar forms or combinations of food or inedible or toxic substances.
2. Insomnia related to sleep disruption from repeated episodes of eating, with a complaint non restorative sleep, daytime fatigue, or somnolence.
3. Sleep-related injury.
4. Dangerous behaviors performed while in pursuit of food or while cooking food
5. Morning anorexia.
6. Adverse health consequences from recurrent binge eating of high caloric food.
C. The disturbance is not better explained by another sleep disorder, medical or neurologic disorder, mental disorder, medication use or substance use disorder (hypoglycemic states, peptic ulcer disease, reflux esophagitis, Kleine-Levin syndrome, Kluver-Bucy syndrome, and nighttime extension of daytime Anorexia Nervosa (binge/purge subtype), bulimia nervosa, and binge eating disorder).

Table 2: Provisional Criteria for Night-Eating Syndrome.

Morning anorexia, even if the subject eats breakfast
Evening hyperphagia, in which ≥ 50% of the daily energy intake is consumed after the last evening meal
Awakenings at least once a night
Consumption of snacks during the awakenings
Repetition of the provisional criteria for ≥ 3 months
Subjects do not meet criteria for bulimia nervosa or binge eating disorder

having symptoms suggestive of RLS with a variable age of onset in late twenties to early thirties. IRLS score for the index case was found to be 24. The age of onset of symptoms and the respective IRLS scores of the affected family members were as follows: 33 years & 14, 33 years & 14, 31 years & 10, 27 years & 9, and 26 years & 9 (Figure 1).

Discussion

- RLS is characterized by discomfort of lower limbs and sometimes upper limbs with an urge to move and is often associated with disturbed sleep. PLMS first described by Lugaresi et al. [4] in which one or both legs contract rhythmically and intermittently during sleep and can be noted in up to 88% of patients with RLS. The prevalence of RLS in the general population varies between 1.2% to 15% [5,6]. It is twice as common in females in comparison with males. Familial RLS was described in the initial series of Ekbom [1]. Since then, increasing number of families with RLS have been reported with significant heterogeneity both in terms of age of onset and clinical presentations even within the same family. The largest collection of complete family study data on RLS was reported from Canada [7]. This case series survey showed that out of 247 fully investigated probands, 192 had RLS aggregation in their respective families with a familial rate of 77%. They also noted a variable range of age of onset in familial RLS (28 ± 15 years) with most of the family members having early onset disease and mild to moderate symptoms to start with, similar to our observation. In another study of RLS patients from Germany, 42% showed a "definite" and 12% showed a "possible" positive family history [8]. They also demonstrated an earlier age of onset with a "definite" compared to "negative" family history (35.45 years compared to 47.17 years), with few patients noting symptoms as early as in second decade. The earlier recognition of RLS symptoms in families with hereditary RLS could be one reason for earlier age of onset reported in studies. This association of age of onset with familial and sporadic forms of RLS also supports the opinion that RLS should be divided into early-onset disease with a clear genetic component and late-onset disease with an unclear etiology. Studies have shown familial RLS to have a genetic etiology with linkage studies identifying several loci including chromosome 12q, 14q, 9p, 20p, 2q, 16p [9]. However, the data from Indian subcontinent is lacking on the same. RLS is associated with non-motor phenomena as well, such as mood and anxiety disorders as well as other nocturnal compulsions like nocturnal smoking that interfere with sleep [10,11]. However, as noted in our patient, there is some evidence supporting an association between eating disorders and RLS. "They often have to get up and walk, 'like a caged bear', to quote one of my patients, or they go into

the kitchen and get something to eat" – Karl-Axel Ekbom, Neurology, 1960. This interesting association of RLS and NE was noted by Ekbom himself in his original series [12] and later conclusively established by researchers who studied this aspect in detail. NE in RLS can be attributed to several reasonsDrug-related especially zolpidem which is usually co-prescribed for patients with RLS who have insomnia or fragmented sleep.

- A behavioral response to reclaim lost sleep and to tide over the disturbing interludes of fragmented sleep and unpleasant leg sensations.

- Sleep Related Eating Disorder (SRED) in which there is a certain compulsion to eat where patient is not fully aware of the act and further becomes amnesic of the event. The ICSD-2 criteria (Table 1) [13].

- NE syndrome in which there is a predilection to eat during the night more succinctly described as a circadian phase shift in eating behavior. Although the distinction from SRED is vague, the patient here is fully conscious during the episodes. Diagnostic criteria for SRED was proposed by Birketvedt et al. (Table 2) [14].

In a case control study, which examined 100 subjects with RLS with a mean age of 41.7 ± 16.2 years and IRLS score of 14.2 ± 10.2, 33% of them had documented SRED [15]. They also observed a high incidence of obsessive-compulsive behavior in RLS subjects as opposed to controls, which is the probable psychopathological substrate for the eating disorder. A subgroup analysis failed to demonstrate any significant clinical or demographic differences in the profiles of RLS patients with and without SRED. In our report the patient had an unequivocal and invariable urge to eat 2 to 3 times a week. These episodes were associated with other forms of compulsive behavior like arranging her wardrobe. Except for morning anorexia, other cardinal features of SRED were lacking. There was no prior consistent use of zolpidem or dopaminergics to implicate them. So, are these NE episodes merely a method of "killing time"? In a study that explored this question, 88 patients with RLS and 42 patients with insomnia were interviewed elaborately [16]. They found that 61% of patients with RLS had NE while only 12% of insomnia subjects reported such behavior. SRED was exclusively found in the RLS group. Patients with RLS who had NE had more pronounced nocturnal awakenings than those without NE, but the insomnia group despite having a higher incidence of nocturnal awakenings had lesser incidence of NE. Furthermore, the RLS patients reported that they felt an urge to eat, the realization of which enabled them to return to sleep. In conclusion, NE should be regarded as a specific type of compulsive behavior, which can be classified as a non-motor manifestation of RLS, and the diagnosis of SRED should be made only when the diagnostic criteria for the same are met.

Conclusion

We report a unique Indian family with RLS with variable age of onset in those affected, and PLMs and dysfunctional NE noted in the proband. A detailed history of these associations in family members of patients with RLS/PLMs should hence be carefully sought. Early recognition and treatment of these disorders and their correlates can markedly improve the quality of life in these patients.

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