

In Response to:

Koo BB. The restless legs syndrome: Would you like that with movements or without? *Tremor Other Hyperkinet Mov.* 2015; 5. doi: 10.7916/D80P0Z0H

Editorials

Further thoughts on “The Restless Legs Syndrome: Would You Like that with Movements or Without?”: Summary of Evidence that the Presence of Periodic Limb Movements in Sleep Does Not Significantly Aid in the Diagnosis of Restless Legs Syndrome/Willis–Ekbom Disease

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Keywords: Restless Legs Syndrome, Periodic Limb Movements in Sleep

Citation: Walters AS. Further thoughts on “The restless legs syndrome: Would you like that with movements or without?”: Summary of evidence that the presence of periodic limb movements in sleep does not significantly aid in the diagnosis of restless legs syndrome/Willis–Ekbom Disease. *Tremor Other Hyperkinet Mov* 2015; 5. doi: 10.7916/D80R9NJ2

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Editor: Elan D. Louis, Yale University, USA

Received: May 30, 2015 **Accepted:** June 1, 2015 **Published:** July 1, 2015

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Funding: None.

Financial Disclosures: None.

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

I read with interest the article by Brian Koo “The Restless Legs Syndrome: Would you like that with movements or without?” published in *Tremor and Other Hyperkinetic Movements*.¹ In this contribution Dr. Koo does a superb job in marshaling evidence that restless legs syndrome/Willis–Ekbom disease (RLS/WED) and periodic limb movements in sleep (PLMS) share a common pathophysiology, pharmacology, genetics, and epidemiology, and that they may also share a similar clinical relevance particularly in relation to cardiovascular disease. The argument is then made that measurement of PLMS may add to the diagnostic accuracy of RLS/WED diagnosis.

Concerns over the accuracy of the RLS diagnosis first came about in epidemiology studies where only the basic four diagnostic criteria for RLS were administered to the general public in phone surveys in the absence of questions about differential diagnosis. It became apparent that under these circumstances false-positive diagnoses were being made because some disorders could meet all four diagnostic criteria for RLS and not be RLS.² These so called “mimics” have always been in the differential diagnosis of clinicians expert in the diagnosis of RLS. However, it became obvious that other diagnostic instruments would

have to be developed so that interviewers naïve to the differential diagnosis of RLS could administer questionnaires that exclude false-positive diagnoses. In one such instrument, the Cambridge–Hopkins questionnaire developed by Allen et al.,³ adding questions about two of these “mimics”, leg cramps and positional discomfort, increases the positive predictive value for the diagnosis of RLS from 39.2% to 87.2% without the need for polysomnography and the determination of PLMS.

Another approach by Benes et al.⁴ similar to the one recommended by Dr. Koo is to take the non-essential but supportive and associated criteria for RLS established by the international RLS Study and formally study to what degree they improve the diagnosis of idiopathic RLS/WED when added to the four essential criteria. The instrument that accomplishes this, the Diagnostic Index, includes the four essential criteria plus varying combinations of the supportive and associated criteria: sleep disturbance, family history of RLS, responsiveness to dopaminergic therapy, periodic leg movements, and a normal neurological examination. In a logistic regression analysis where essential and non-essential criteria for the diagnosis of RLS were

compared with an expert clinical diagnosis of RLS, the amount of variance attributed to the four essential criteria combined was 69.4% whereas this increased to 90.4% when there was a response to dopaminergic therapy and a normal neurological examination in addition to the four essential criteria. The further addition of periodic limb movements to the aforementioned items only increased the amount of attributed variance to 90.8% or a gain of only 0.4%. In other words, the presence of PLMS did little to improve the diagnostic accuracy of RLS.

In summary, the two different approaches by Allen et al. and Benes et al. both suggest that the diagnostic accuracy of RLS can be considerably improved by a good history and neurological examination without the need for polysomnography and the determination of PLMS.^{3,4}

An additional key factor is that polysomnography and its surrogate, actigraphy, are expensive, time consuming, and not available throughout much of the underdeveloped world. A recent survey of available sleep facilities in Saudi Arabia showed that the number of overnight sleep studies/year/100,000 people was only 18 compared with 427 in the United States, 370.4 in Canada, and 282 in Australia. Reasons cited for the failure of the development of sleep medicine in a large survey of hospitals in Saudi Arabia were no funds (31.8%), no space (50%), no sleep tech (81.80%), cannot interpret sleep study (40.90%), and no sleep specialist (81.80%).⁵ Readers can only suspect that things are undoubtedly worse in rural Africa and rural Latin America. It is simply not practical to recommend polysomnography as an important adjunct to the diagnosis of RLS to impoverished parts of the world, particularly since the diagnosis can be readily made with a proper history and examination as documented above. Unfortunately, this is even true for economically advanced countries like the United States. Dr. Koo says that 80–90% of patients with RLS have PLMS > 5/hour. What is not mentioned is that with one night of polysomnography the figure is 80.2% but it takes two nights of polysomnography to reach a figure of 87.8%.⁶ Most insurance companies in the United States will not pay for two nights of polysomnography in order that we might improve the accuracy of an RLS diagnosis.

An additional issue is that PLMS is a common incidental finding on polysomnography in subjects who do not have RLS. In one study, PLMS > 5/hour were found in 45% of community-dwelling elderly people, precisely the age group at which RLS is also at a maximum.⁷ PLMS can lend themselves to a false-positive diagnosis of RLS too.

Dr. Koo and I both agree that there may come a day when polysomnography will become necessary to document PLMS even in the absence of RLS if ongoing investigations into the connection

between PLMS and cardiovascular disease prove to be true in the long run. However, this day has not yet come. Dr. Koo and I are both actively involved in this area of investigation.^{8,9}

In conclusion, RLS/WED can be accurately diagnosed with a good clinical history that takes in to account a proper differential diagnosis in combination with a neurological examination. There is little need to use expensive and sometimes inaccessible technology such as polysomnography or actigraphy in order to document PLMS in most cases of RLS/WED. PLMS often occur as an incidental finding on polysomnography when no symptoms of RLS are present, which further confounds matters.

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