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CASE REPORT

A Multigenerational Family with Persistent Sleep Related Rhythmic Movement Disorder (RMD) and Insomnia

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In the *International Classification of Sleep Disorders 2nd Edition* (ICSD -2), sleep related rhythmic movement disorder (RMD) is classified as a disorder characterized by rhythmic movements of large muscle groups in different parts of the body. These are repetitive, stereotyped, rhythmic motor behaviors that occur predominantly during drowsiness or sleep,and are typically seen in infants and children. Episodes often occur at sleep onset, at any time during the night, and during quiet wakeful activities at a frequency of 0.5–2 sec), lasting < 15 min. The prevalence is high in infants (59%), dropping to 5% at the age of 5 years. When persisting to older childhood or beyond, association with mental retardation, autism, or other significant pathology is reported.[1] Few cases in adults of normal intelligence have been reported in the literature.[2-5] There is a strong association with attention deficit hyperactivity disorder, suggesting a similar pathogenetic mechanism.[5] There is also one adult case report occurring during strictly REM sleep.[6] Mayer et al reported 24 subjects with RMD that persisted into adolescence and adulthood. Twenty of the subjects were adults, and 16 of them had the condition since childhood. Of these 20, 16 had no other sleep disorders (but 2 had a family history of RMD), and 4 had obstructive sleep apnea.[7] This was the first ever report of familial RMD.

Keywords: Sleep related rhythmic movement disorder, adults, insomnia, familial condition

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We have identified a multigenerational family in which 50% of members have either prominent insomnia or both insomnia and RMD simultaneously, and the other 50% do not have either condition. No family member has RMD only. The insomnia in the family was multifactorial: each subject had a different comorbid condition contributing to their inability to fall asleep and stay asleep. In subject A, insomnia is comorbid with bipolar disorder; in subject C with menopause; in subject E with developmental delay; subject F started in adulthood and is comorbid with his generalized anxiety disorder; subject G meets the diagnostic criteria of idiopathic or childhood onset insomnia; and symptoms in subject I started after a bout of inflammatory demyelinating condition (possibly early onset multiple sclerosis). Although comorbid conditions vary from subject to subject, insomnia is a prominent complaint with all of the above affected family members, and they all have sought medical attention for it. Despite a variety of treatments, both pharmacological and (in one case) cognitive behavioral therapy for insomnia, the subjects continue to experience prominent sleep disturbances.

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The subjects with RMD have had it since childhood, and it has persisted into adulthood. All 3 have the body rocking type of RMD, occurring only at the beginning of the night during drowsiness, for several minutes, with a frequency of 2-3 times a week. It did not interrupt their sleep, and their only complaint was that it was a source of embarrassment in certain situations. Two family members (F and G) who have both conditions underwent polysomnography after they failed to respond adequately to insomnia treatments. Polysomnography showed severely reduced sleep efficiency and prolonged sleep latency, despite zolpidem 10 mg in the case of G. No other abnormalities were identified. None of the subjects complained of any symptoms suggestive of sleep apnea, restless legs syndrome, or any other sleep disorder. See Figure 1.

DISCUSSION

Our three subjects with insomnia and RMD were of normal intelligence. Both mother and sons are gainfully employed and fully functional, and subject F has graduated college. In all 3, the RMD started in childhood and has persisted unabated into adulthood. Other family members also have insomnia, albeit secondary or comorbid with other conditions. We postulate that the familial tendency to have fragile sleep and therefore cause prevalent complaints of prominent insomnia is at the root of the 3 subjects' continued frequent RMD. This may have developed as a self-soothing mechanism but has persisted because of their tendency to have light sleep. This behavior can persist into

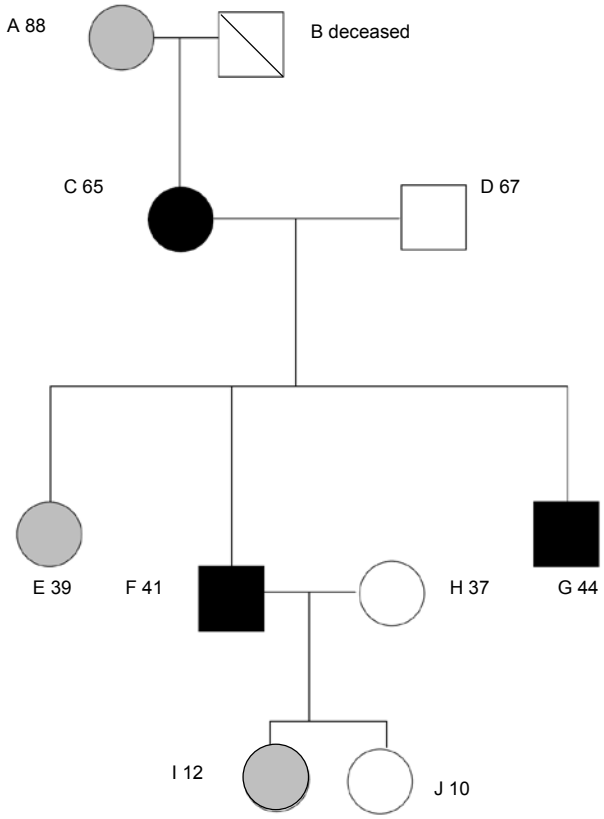

Figure 1—The black shapes represent the members of this family who have both conditions, the grey shapes have insomnia only, and the open shapes do not have either condition.

adulthood either by perceptual reinforcement or by entraining a central oscillator (similar to a tic).[6] Our subjects, however, did not have tics or obsessive compulsive behaviors otherwise.

The two subjects in the third generation may have learned this behavior from their mother. Although behavioral and other aspects are of greater importance than genetics in the familial occurrence of RMD,[6] the RMD and the tendency for fragile sleep may conceivably have a common genetic substrate, possibly affecting GABA receptors. Increased cortical and subcortical arousals have been demonstrated in RMD subjects,[4] similar to insomniacs.

In conclusion this is, to out knowledge, the first report of a family with both RMD persisting into adulthood and insomnia. It is very important to emphasise two important points; the RMD could have been a learned behavior, and most of our insomniac subjects had comorbid conditions that may have caused or complicated the symptoms of insomnia. Because of this and the small number of the cohort (e.g., subjects E and G are childless) we were unable to perform gene mapping; therefore, we cannot say with certainty that there is a common pathophysiological mechanism to RMD and insomnia, but we hope this will lead to larger studies into this rare but interesting sleep disorder.

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DISCLOSURE STATEMENT

This was not an industry supported study. The authors have indicated no financial conflicts of interest.

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