THE ROLLING BODY: A CASE OF DRUG-RESISTANT SLEEP RELATED RHYTHMIC MOVEMENT DISORDER ASSOCIATED WITH PERIODIC LIMB MOVEMENTS IN SLEEP AND SLEEP RELATED BRUXISM

S. Novello, G. Macorig, S. de Biase, P. Dolso, G. L. Gigli, M. Valente
Neurology Unit, Department of Experimental and Clinical Medical Sciences, University of Udine Medical School, Italy

Introduction
Sleep related rhythmic movement disorder (RMD) is characterized by repetitive, stereotyped, rhythmic motor behavior that occur predominantly during drowsiness or sleep and involve large muscle groups. This disorder is typically seen in children, rarely can persist in adulthood provoking self-inflicted bodily injury, excessive daytime sleepiness, and insomnia (1). Only when RMD interferes with normal sleep, treatment with benzodiazepines can be considered (2). The aim of this case report is to describe clinical and polysomnographic data of an adult affected by several forms of RMD, associated to other sleep disorders.

Case history
A 44-year-old male consulted our service because of a long history of insomnia and body rolling, worsening in time. RMD started when he was 6-years-old, and it continued through adulthood occurring almost every night, usually in the second part during wake time. Family history was negative for epilepsy and sleep disorders. He didn’t have clinical conditions mimicking RMD and didn’t report a history of apneas. In the previous years he tried several drugs to treat insomnia (melatonin, zolpidem, trazodone, lormetazepam, alprazolam, and clonazepam) without significant improvement and sometimes with side effects. He suspended all therapies one week before admission. To confirm the presence of RMD, we performed a video-polysomnographic (V-PSG) recording. Once the RMD was diagnosed we tried to treat it, monitoring with V-PSG recordings for other three nights the effect of three drugs. V-PSG recordings documented a mean frequency of 33 episodes per night. In addition to body rolling, he presented episodes of head rolling, leg rolling, and leg banging every night. Moreover during N1 and N2 sleep the exams showed an increased periodic leg movements during sleep (PLMS) index and the presence of several episodes of sleep related bruxism. We tried to treat these disorders with pramipexole, gabapentin, and diazepam. All these drugs didn’t improve both sleep parameters and RMD. Only pramipexole treatment documented a positive and complete effect on PLMS (PLMS index: 70 to 0) and partial on bruxism.

Conclusions
Despite a wide experience regarding the benzodiazepines’ efficacy on RMD, an acceptable control on symptoms cannot be reached in some cases. Moreover, as previously suggested (3), we suppose that PLMS and bruxism probably don’t share common pathogenic substrate with RMD and this association could be explained by the occasional co-occurrence of RMD with other frequent sleep disorders. This hypothesis is supported by the different response of these disorders after pramipexole administration.

References