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Rhythmic movements during sleep: a physiological and pathological profile

Abstract Rhythmic movement disorder (RMD) consists of rhythmic movements (RMs) that occur on falling asleep or during sleep, can involve any part of the body and have a reported frequency ranging from 0.5 to 2 Hz. RMs have been reported to occur in a high proportion of normal children as a self-limiting phenomenon starting and remitting within early infancy. However, there have also been descriptions of forms of RMD occurring against a background of mental retardation or persisting beyond childhood, or having onset in adulthood. So, the occurrence of RMs can be regarded as both a physiological and a pathological phenomenon. The few polysomnographic studies conducted in this field have shown that, in some forms of RMD, RMs are highly linked to arousal fluctuations. However, the mechanisms that underlie the genesis of RMs and are capable of leading to both physiological and pathological patterns of RMs are not fully understood. Here we emphasise the possibility that the central motor pattern generator, recently hypothesised to play a role in the genesis of motor phenomena during sleep in the cases of parasomnia and epileptic seizures, might account for the occurrence of RMs in both physiological and pathological conditions.

Key words Rhythmic movement disorder • Sleep • Central motor pattern generator

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Introduction

The heterogeneous phenomena grouped by the International Classification of Sleep Disorders [1] under the heading "Rhythmic Movement Disorder (RMD)", range from "auto-erotic", self-stimulatory behaviours occurring during quiet wakefulness or at sleep onset to more complex patterns of movements arising from stable, night-time sleep. RMD has a benign course and is not generally associated with a severe clinical picture [2].

However, despite being of relatively minor clinical importance, RMD appears to be of interest from a physiopathological point of view. The study of its physiopathogenesis is likely to give interesting insights into the complex mechanisms of motor control during sleep.

Our chances of understanding this disorder are limited by a lack of experimental and iatrogenic models of RMD and a lack of functional neuroimaging and genetic studies of human RMD. However, a few polysomnographic (PSG) investigations [3, 4] detailing the relationship between sleep structure and rhythmic movements (RMs) in RMD have shed some light on the physiopathogenesis of this motor disorder.

This paper is aimed at reviewing the clinical and PSG aspects of RMD on the basis of findings reported in the literature and in published [5–7] and unpublished personal data. A hypothesis regarding the physiopathogenesis of the disorder is put forward.

Clinical patterns of rhythmic movements during sleep

RMs during sleep can involve any part of the body. However, head banging/head rolling (so-called *iactatio capitis nocturna*), body rocking and body rolling are the ones most frequently encountered. Leg banging and hand banging have also been reported.

The movements have a reported frequency ranging from 0.5 to 2 Hz. Episodes of RMs, which can arise from any body position, show a variable duration, but they rarely exceed 15 min. Typically, RMs occur at sleep onset but they can persist throughout the night. RMs occurring at approximately 2-h intervals within the 24-h sleep/wake cycle (seemingly paced by the basic rest activity cycle, or BRAC [8]) have also been reported [9].

A subject can present different types of RMs, which may co-exist, or occur at different stages in the clinical course of the RMD. Generally, the clinical picture of RMD is mild and consists of sporadic, single episodes occurring at night. But severe forms also exist in which patients have been reported to present, on consecutive nights, several RM episodes occurring in clusters.

The clinical background against which RMs can occur is variable, ranging from absolute normality to pictures of cryptogenic or symptomatic mental retardation. There are no reports of RMD in association with other parasomnia, with the exception of restless legs syndrome (RLS) [10, 11].

RMD may be misdiagnosed as nocturnal epilepsy. As a matter of fact the relationships between RMD and epilepsy have been reported to be intricate in some cases [2, 12, 13].

RMs typically occur in early infancy and remit after the fourth year of life. However there have been reports both of RMs persisting into or relapsing in adulthood and of late-onset RMs [2, 11, 14]. In males the prevalence of some movements, such as head banging, is two to three times higher than in females.

In most of the cases pharmacological treatment is unnecessary. However in severe forms of RMD, both infantile and adult RMD episodes have been reported to show good response to clonazepam at low doses [5–7, 15].

Polysomnographic patterns of RMD

Full-night PSG investigations in RMD revealed that RMs can occur during both NREM and REM sleep. According to the most recent, updated review, RMs occur at falling asleep or during NREM sleep in 46% of PSG-documented RMD, during both NREM and REM sleep in 30%, and only during REM sleep in 24% [4]. The exclusively REM-related RMs have been reported to occur more frequently in adult forms of RMD.

As regards the occurrence of RMs in relation to the microstructural phasic events of NREM sleep, K-complexes have been found to have a close temporal relationship with the onset of RMs [3]. Recently we analysed the relationship between sleep and RMs in an infantile form of NREM-related RMD, basing our scoring of sleep on the cyclic alternating pattern (CAP) [5, 16].

The term CAP refers to spontaneous, periodic electroencephalographic (EEG) activity occurring during NREM sleep. CAP is thought to be an expression of sleep instability, and the rules and criteria for its visual scoring have now been standardised [16]. A CAP cycle is made up of two components: phase A and phase B, the latter being an interval of EEG background activity that separates two consecutive A phases. Phase A can be subdivided into: A1 phase, characterised by sequences of K-complexes and delta bursts, A2 and A3 phases, characterised by transient low-voltage fast EEG activities preceded by longer (A2) or shorter (A3) bursts of EEG synchronisation. Each CAP sequence is made up of at least two consecutive CAP cycles.

Phase A of this EEG biphasic pattern, characterised by repetitive activating and arousal events, is thought to be an expression of neural synchronisation and it has been reported to be associated with several motor phenomena occurring during sleep and having different aetiologies (e.g., periodic movements of the limbs (PMLs), bruxisms, sleepwalking and epileptic motor seizures) [17].

In the above-mentioned infantile form of RMD we observed, the RMs consisted of nocturnal multiple episodes of body rocking and head banging occurring during NREM sleep (Fig. 1). In our CAP analysis RMs showed a close relationship with CAP sequences regardless of the NREM stages. The RM episodes always were found to occur shortly after phase A.

Thus, the literature [3, 4] and our own [5] PSG findings have documented the existence of a close relationship between arousal fluctuations and RMD and have shown that RMs, like other motor phenomena occurring during sleep, are likely to be modulated by these fluctuations and by their underlying neurophysiological mechanisms.

The existence of RMD occurring exclusively during REM sleep raised the question of the possible relationship between RMD and REM behaviour disorder (RBD) [18]. However, PSG findings in REM-related RMD clearly document that RMs occur against a background of otherwise normal REM sleep, with preserved muscle atonia and normal occurrence of twitches. Furthermore subjects with RBD have never been reported to have a clinical history of RMD. So the existence of a relationship between the two disorders appears to be very unlikely.

Rhythmic movements during sleep: a normal phenomenon?

Some lines of evidence support the hypothesis that the occurrence of RMs during sleep is a physiological phenomenon and have suggested that RMs represent a positive stimulus for motor development in the early stages of life. The view that RMs during sleep are a physiological occurrence is supported by the fact that these movements are encountered in a high proportion of otherwise normal children [19] and that, in most cases, RMs constitute a self-limiting and

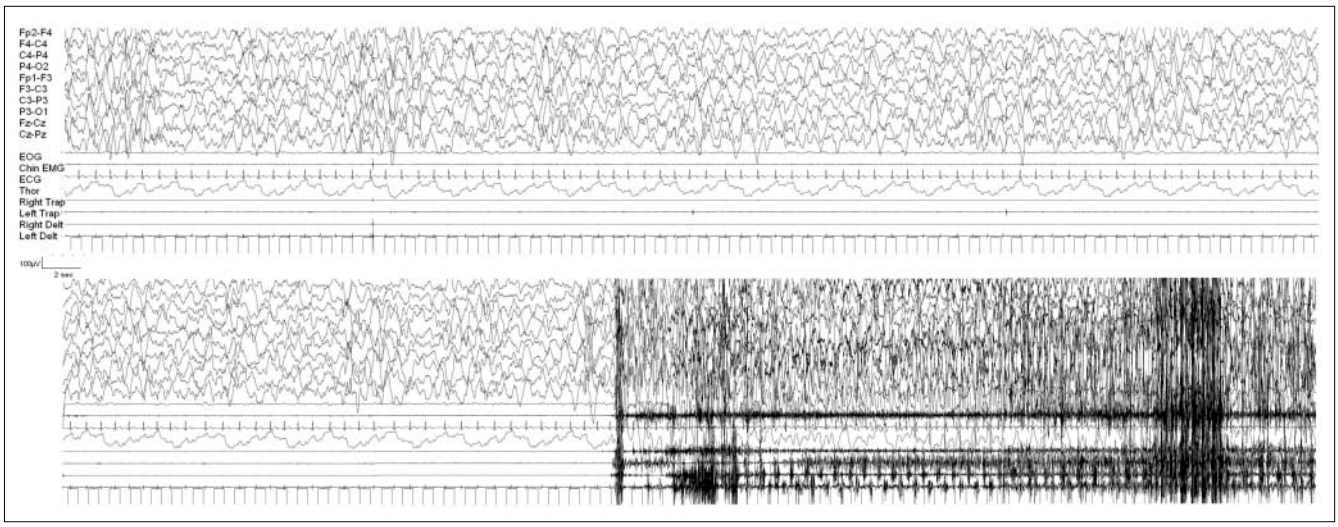

Fig. 1 Episode of body rocking and head banging arising from slow-wave sleep during phase A of a CAP sequence

temporary phenomenon that has onset early in life, and disappears spontaneously after the fourth year of life, as shown by Klakenberg's epidemiological survey [20].

Furthermore, their occurrence coincides with the milestones of psychomotor development in the first year of life. The temporal coincidence of body rocking and head banging with milestones of psychomotor development has been viewed as an indication that RMs should be regarded as normal manifestations. Some authors have even claimed that some RMs, namely body rocking and head banging, are associated with developmental precocity. The theory is that RMs may positively affect the motor system through vestibular stimulation triggered by head or whole body shaking. Vestibular stimulation has been hypothesised to exert an important positive influence on motor development in infants [21].

However, studies indicating that RMs are potentially able to exert a positive influence on motor development present several limitations due to their subjective and retrospective character. No prospective, longitudinal evaluation of motor skills in children with RMs during sleep has yet been conducted.

Rhythmic movements during sleep: a pathological phenomenon?

RMs can, but rarely, persist into, relapse in, or even have onset in later childhood and adulthood (the latter scenario corresponding to a kind of "late-onset" form of RMD). Forms of RMD have been reported to occur in combination with RLS [10, 11], but it is still not clear whether or not such associations share a common physiopathogenetic substrate.

Furthermore, it is interesting to note that RMD has certain features in common with RLS and PMLs. All show a male prevalence and are highly modulated by arousal level, appearing to be a "vigilance state-dependent" phenomena. Finally, they show a good response to clonazepam.

Personal observations

Our personal observations of RMD include both published [5–7] and unpublished cases.

The published cases (here referred to as cases 1, 2 and 3) are males, aged six, seven and 18 years respectively. The unpublished cases (here referred to as cases 4, 5 and 6) are also males, aged 15, 16 and 19 years, respectively.

In-lab, infrared video-audio PSG monitoring of RMs was carried out in all but case 6 (in whom RMs were captured only during at-home video-monitoring). Before coming to our notice, cases 1 and 3 had been misdiagnosed as having epileptic nocturnal seizures.

In cases 4 and 5 (15 and 16 years old at the time of their referral to our Sleep Centre), RMD consisted of head banging, which dated back to early infancy and had persisted into childhood. These movements used to occur only during quiet wakefulness/drowsiness prior to falling asleep at night and were recalled as semi-purposeful movements. Psychological investigation revealed emotional distress and a psychotherapeutic approach was advised. The efficacy of this approach is not known since the subjects were lost at follow-up early on.

In cases 1 and 2, the RMD consisted of head banging, which had begun at six months of age and had been followed by the appearance of body rocking from the age of about 12 months. In subject 1, the RMD had to be consid-

ered against a background of mental retardation due to fragile X syndrome. In both these cases, the clinical picture of RMD was one of rather violent, multiple RMs recurring almost nightly after sleep onset. Treatment with clonazepam at low doses proved to be successful in the long term (two years of follow-up).

Nocturnal video-PSG documented highly stereotyped, NREM-related RMs, which consisted of head banging alternating with body rocking, characterised by a constant frequency of 1 Hz in case 1, and of 2 Hz in case 2. When body rocking occurred while the subject was sleeping on his back, the subject, before starting rock, used to go on his hands and knees and to assume a position which reminds of that of a Muslim in praying. We think that the global movement by which the patients assume this "Muslim in praying" position before starting to rock is an integral feature of their RMD.

The RMD consisted of multiple episodes of head banging after sleep onset at a frequency of 2.5 Hz in case 3 and at a frequency of 3.5 Hz in case 6. In both cases the head banging had started within the two first years of life, had remitted after the age of four and then relapsed, against a background of neuropsychiatric normality, at the respective ages of 18 and 19 years. At this point, treatment with clonazepam was initiated and it proved to be successful in the long term (3 years of follow-up).

We did not observe EEG epileptiform abnormalities, either during wakefulness or during sleep EEG recordings, in any of the cases with the exception of case 1, affected by fragile X syndrome. In this subject, we found a right centro-temporal spike focus that proved to be independent of RMD manifestations and was likely an expression of the fragile X syndrome. Following clonazepam-induced remission of his RMD, this patient, at the age of nine years, developed partial motor seizures during sleep. These were successfully treated with valproate.

Conclusions

The polymorphism and heterogeneity of RMs, along with the lack of in-depth electrophysiological, neuroimaging and genetic studies, makes it difficult to understand their physiopathogenesis. For a long time all forms of RMD were regarded as expressions of emotional distress and psychological approaches to their treatment were encouraged.

However, growing evidence indicates that RMs that clearly derive from a dysfunction of motor control during sleep and constitute true sleep-related motor disorders also exist.

Some data indicate that RMs, like other motor disorders during sleep, are closely linked to arousal fluctuations, which would suggest that the brain-stem and thalamo-cortical reticular system are involved in their modulation. However, the mechanisms which underlie the genesis of RMs and which are capable of leading to both physiological (self-limiting, benign-course RMs of early infancy) and pathological patterns of RMs (forms persisting into or having onset in adulthood, in which the picture is generally one of several, multiple episodes per night) are not fully understood.

Recently, a system called the central motor pattern generator (CMPG) has been hypothesised to play a role in the genesis of motor phenomena during sleep in pictures of parasomnia and epileptic seizures, namely nocturnal frontal lobe epilepsy [22, 23]. The CMPG is a brain-stem neuronal network involved in the control of early locomotor function [24, 25] and is highly integrated with the reticular system of the brain stem. The CMPG is thought to be under the inhibitory control of the cortex.

Immaturity of the inhibitory cortical system in early infancy might account for RMs occurring during sleep in a few children in the first year of life, coinciding with their acquisition of the motor milestones. Accordingly, loss of inhibitory cortex function due to pathological conditions might account for the persistence, relapse or onset of RMs in childhood or adulthood.

The cortex-CMPG system, together with the reticular system, might be the common substrate of the spectrum of RMD manifestations.

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