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Rhythmic movement disorder in childhood: an integrative review

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Conflicts of interest

The authors have no conflicts of interest to declare relevant to this publication

Abbreviations: Attention deficit hyperactivity disorder (ADHD), International Classification of Sleep Disorders (ICSD), Non-rapid eye movement (NREM), Obstructive sleep apnea (OSA), Rapid eye movement (REM), Rhythmic movement disorder (RMD), Rhythmic movements (RMs).

Keywords: Child, Rhythmic Movement Disorder, Sleep

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Summary

Rhythmic Movement Disorder consists of repetitive stereotypic movements, such as head banging or body rocking, that recur every second or so and may last from a few minutes to hours, usually prior to sleep onset. This review of childhood rhythmic movement disorder highlights the lack of systematic research into core aspects of the condition, relying heavily on small case series or case reports. Interpretation is further limited by almost universal failure to confirm the core the diagnostic criteria (C) of the International Classification of Sleep Disorders (III), namely that the rhythmic movements should have clinical consequences. Nonetheless, a number of themes emerge. Rhythmic movement disorder is likely to start in infancy and have a developmental course with spontaneous resolution in early childhood in many cases. Factors associated with persistence are, however, unclear. Associations with ADHD and neurodevelopmental disorders are intriguing, require further study and may shed light on the underlying cause of the condition. There is a pressing need for a systematic approach to classify rhythmic movement disorder, to allow standardization of the much needed research into the underlying aetiology and treatment of this relatively neglected sleep disorder.

Introduction

The International Classification of Sleep Disorders III (ICSD-III)(1) describes rhythmic movement disorder (RMD) as rhythmic, stereotyped, repetitive gross motor movements, occurring at approximately one second intervals, predominately at sleep onset and sometimes during sleep. Typically these involve violent whole body rocking or rolling and may involve striking a body part (often the head) against a surface(1). Episodes of movements may be brief or last up to around 20 minutes(1). Importantly, the diagnostic criteria stipulate that these movements must cause significant sleep interference, impaired daytime functioning or actual or potential injury in order to be classified as RMD. Full diagnostic criteria are illustrated in figure 1. While primarily occurring in childhood, RMD can persist into adolescence and adulthood(1-3). The prevalence of RMD has been estimated to be as high as 66% in infants(4), though the few published prevalence studies have significant methodological limitations. The intensity and amplitude of movements, as well as noise generated when the movements are associated with head-banging, or noisy vocalisations, can be distressing to parents(5). Identification and effective management of RMD in childhood is important for both the index child and the extended household. A further consideration, although rare, is the risk of injury(1, 3, 6-10) . There have been no systematic studies quantifying either the risk of injury nor the daytime consequences of RMD and no clinical trials evaluating the treatment of RMD. Current and, to date, management is guided by clinical experience and published case studies(11).

Study question and purpose

This review aims to provide a comprehensive summary of the current research on the classification, prevalence, pathophysiology, diagnosis and treatment of childhood RMD, where childhood is defined from birth to 18 years.

Methods

An integrative review allows for the inclusion of both theoretical and empirical literature in fields where high quality data is lacking(12). A systematic search was conducted from February to April 2016 using MEDLINE, PubMed and PsycINFO databases. The following synonyms for RMD (taken from the ICSD-III(1)) were used : "rhythmic movement disorder"; "RMD"; "jactatio capitis nocturna"; "headbanging"; "body rolling"; "body rocking"; "rhythmic du sommeil". The MeSH terms "sleep" and "sleep disorders" were used to exclude papers focusing on movement disorders unrelated to sleep. An additional search using the term "rhythmic movement disorder" alone was conducted to capture publications lacking the keyword "sleep". No time limit was used. In addition to searching computerised databases, ancestry searching and expert consultation networking were also used, as recommended by Conn et al(13). Inclusion criteria included age (0 and 18 years) and publications in English. Unpublished manuscripts (e.g. dissertations); comments or letters and review articles summarising data already included and lacking novelty were excluded from the review.

The quality of each paper was classified according to level of evidence as described by Melnyk and Fineout-Overholt(14) (see figure 2).

Results

The literature search identified 1047 abstracts. Of these, 924 failed to meet the inclusion criteria, most relating to adult non-sleep related movement disorder research. 196 papers related to sleep related rhythmic movements (RMs) were reviewed in their entirety and where the same data was published in different journals, duplicates were removed. 36 remaining papers fitted the inclusion criteria. An additional 11 relevant papers were identified via ancestral searching and two were identified via expert consultation. Thus a total of 49 papers were included (figure 3) and are summarised in Appendix 1.

The majority of studies presented low levels of evidence (figure 4): one of level II; two of level III; three of level IV; 10 of level V; 12 of level VI and 21 of level VII quality (of which 17 were case reports). The one level II and two level III studies were of experimental design investigating the effects of rocking on arousal levels, motor development and sleep disordered breathing in infancy. These are important studies as they uniquely offer evidence to explain the possible aetiology of RMD.

A minority (19) of the included papers explicitly addressed whether the participants met Criteria C of the current ICSD III, namely that the rhythmic movements had a clinical consequence. In a further two papers this was implied as the term 'RMD' was used, and in three papers this was not applicable. This important distinction is highlighted in appendix 1. This limitation means that for many papers the results can only be implied to be applicable to RMD since clinical consequences were not adequately documented in most instances. Where possible in the text this has been made clear but the use of the term RMD where it was reasonable to assume, or specified that Criteria C was met, or the term RMs(rhythmic movements) where this was not the case.

School-aged children (5 - 13 years) were the most commonly studied (18 studies), followed by teens (13 - 18 years, 13 studies), toddlers (1 - 3 years, 8 studies), pre-school children (3 years - 5 years, 7 studies) and infants (0-12 months, 5 studies). Of the review articles included, seven focused on children (no age limits were specified) and seven did not specify an age range.

Nine themes relating to RMD were identified (the number of publications relating to each theme are indicated in parenthesis): diagnosis (eight); demographic features (six); sleep stage distribution (13), consequences for the child or their family (14); classification (12); aetiology (23); neurophysiology (two); co-morbidities and associated factors (18); persistence to five years and older (30); and treatment (17). Each theme will be discussed with reference to the relevant literature.

Diagnosis

As traditionally conceptualized, Rhythmic Movement Disorder (RMD) consists of repetitive stereotypic movements that recur every second or so and may last from a few minutes to hours, usually prior to sleep onset. Amongst the common forms are head banging and body rocking. RMD usually occurs in early childhood and disappears in late childhood. To be diagnosed with RMD there must be a clinical consequence such as significant sleep disturbance, impaired daytime functioning or actual physical injury. If a clinical consequence is not present the rhythmic movements are simply noted but the term RMD is not employed. The ICSD-III(1) provides a clear description of the diagnostic features of RMD although criteria C, namely that RMs should have a clinical consequence for the child (figure 1), is frequently overlooked in the literature. When diagnosing a sleep-related movement disorder, we also suggest that the following elements are considered(5) (figure 5): the relationship of the movements to sleep or wakefulness; whether movements are simple or complex and whether epilepsy could be mimicking a movement disorder. In practice RMD is readily distinguished from seizures. However, the presence of tonic-clonic movements, tongue biting, incontinence, risk factors for epilepsy or an inability to cease movements on command should prompt further neurological assessment(11, 15).

Diagnosis in clinical practice most often relies on parental report(16). Parent report, however, may be unreliable, even in the presence of noise-generating head-banging. Strauss et al.(10) reported that parents underestimated episodes of head-banging when compared with objective overnight audio recording. In support of this, Thorpy and Glovinsky(16) suggest that polysomnography can identify short episodes which may be missed by an observer. In practice exhausted parents, even when co-sleeping, may not recall all episodes, even if they wake-up to attend to the child. If parental report is unreliable then objective assessments are needed. This is not only helpful to establish a diagnosis but also importantly for assessing severity of RMs (duration, frequency and temporal occurrence) and measuring treatment response. At present there is no 'gold standard' for measuring RMs in sleep. While the

American Academy of Sleep Medicine describe polysomnographic scoring criteria(17) (figure 6) polysomnography may have limitations as some children completely suppress RMs during a single night of laboratory study (personal communication CMH). This is supported by observations from Stepanova et al.(18) who reported that polysomnography underestimated the number of RMs compared to parental report. Home assessment offers ecologically valid assessment of RMs in the child's natural setting over multiple nights and detailed analysis of film can produce a hypnogram of movement events. Videosomnography (11) has potential advantages in diagnosis allowing direct visualisation whole body movements and also provides rich data on approaches to injury risk reduction, sleep hygiene, parenting practices etc.). At present there are no agreed standards for scoring of RMs using videosomnography technology (with or without additional sensors such as EMG and accelerometry) although this is a promising area for future research(19). Accelerometry alone can usefully quantify movement amplitude and sleep quality but cannot distinguish periods of RMs from normal movements when a child is awake(20). For example figure 7a illustrates actigraphy in a child where parents gave a convincing clinical history of RMD. On first inspection the periods of increased activity during the night could be interpreted as RMs, however, in this case simultaneous videosomnography illustrated that these episodes represented normal behaviors, as the toddler bounced at the cot-side. This is strikingly similar to the appearance of videosomnographically confirmed RMD in a similar aged child (Figure 7b). The explosion of technologies in sleep medicine(21), including remote telemedicine technologies, alongside new approaches using 3D video may open the doors to automated quantification of RMs in sleep and address the limitations of current approaches.

A final point to note is that parents may fail to seek help if they perceive the problem to have no cure, or if they consider the behaviour to be normal(22). Indeed, the risk of injury associated with head-banging could explain why head-banging is the commonest form of RM reported in case studies (Appendix 2); parents may seek specialist help to prevent injury, rather than out of concern for the effects of the behavior on the child's sleep. Hence,

education of primary health care workers and parents about this condition is an important agenda.

Demographic features

The limited studies reporting the population prevalence of RMD provide a wide range of estimates. Klackenberg(4) reported rocking, head-banging and head-turning in 66% of infants aged 9 months. Prevalence in this study decreased with age, with rocking, head-banging and head-turning reported in 45% of 18 month olds (n=194) but only 6% of 5 year olds (n=198)(4). Although these prevalence rates are frequently quoted in the literature they should be interpreted with caution as they were based entirely on parental report. Such high prevalence rates in the very young suggest a self-limiting physiological normal variant(4, 23). Whether these movement are precursors of RMD in vulnerable individuals, or a completely different phenomenon is unclear. Importantly, the proportion of these children who would have qualified for an RMD diagnosis (as defined by criteria C of the ICSD-III(1)) cannot be estimated as this information was not provided.

Petit et al.(24) prospectively studied 1058 children from 2.5 to 6 years of age. Parental report indicated an overall prevalence of RMs in 9.2% during the period studied, with the highest reported prevalence at 2.5 years of age(24). This study has a number of strengths: the sample was large and the use of randomised 3-level stratified sampling increased the likelihood of the participants representing the chosen population (Quebec, Canada). Furthermore, prospective data collection minimised recall bias(24). However, in common with Klackenberg's(4) data, it was unclear if criteria C of the ICSD-III(1) was fulfilled.

Nevés et al.(25) reported RMs occurring every night in 1.4% of 1407 children aged 6.2-10.9 year olds. Cases were ascertained by child and parent questionnaire response(25) and the inclusion of the child as active participant was a strength of this study. However, RMD was assessed using a question which exclusively focused on rocking or swaying before sleep onset(25), excluding other RMD subtypes and those children with RMs after sleep onset.

Furthermore, lack of reference to criteria C of the ICSD III means it is impossible to know how many of these children would qualify for a diagnosis of RMD.

Laberge et al(26) recruited 1353 10 year olds and collected retrospective data for the period between three and ten years of age. The sample was followed up annually to the age of 13 years(26). They reported a 17.2% prevalence of body-rocking across this 10 year age span (3 to 13 years)(26). Importantly, this study shed some light on the natural history of body-rocking. Onset between the age of 3-10 years was reported in 89% of affected children and spontaneous remission in 71%(26). Retrospective data could however, be subject to recall bias and no consideration is given as to whether the movements existed prior to the age of 3 years. Once, again, the failure to acknowledge clinical consequences of these movements, and the focus solely on body-rocking, prevents generalisation of this data.

Taken together data from these 4 prevalence studies(4, 24-26) suggest that RMs are common in young children and become less prevalent with increasing age(4, 24, 26). However, a crucial limitation is the failure to report criteria C of the ICSD-III(1)diagnostic criteria for RMD. This in part reflects the state of knowledge at the time of each study. In 1971, when Klackenberg(4) reported on a Swedish cohort, there was no ICSD. Furthermore, the diagnostic criteria for RMD have changed with successive editions of the ICSD. The revised 1st edition(27) in 2001 specified mild impairment of psychosocial functioning for a diagnosis of moderate RMD and a signs of physical injury or significant impairment of psychosocial functioning for a diagnosis of severe RMD (27). The requirement for RMs to significantly interfere with normal sleep, impair daytime function or cause injury in order to be considered a disorder was introduced with the 2nd edition of the ICSD in 2005(28). Importantly, no prevalence studies have been published that adhere to these ICSD-III (1) criteria. This is an important goal of future research.

There are mixed findings on the association between gender and the prevalence of RMD. Sallustro and Atwell(29) reported almost three fold male to female ratio of head-banging in

children aged 3 months to 6 years. This data, however, included both children with RMD and children who only exhibited RMs when awake, furthermore, data on gender split in the clinical RMD sub-group was not reported. Petit et al.(24) in contrast, found no association between head-banging and gender. No association has been reported between body-rocking and gender(24, 26).

There are also contradictory findings on the relationship between RMD and socioeconomic status, family stress and maternal depression. While persistent RMD (body-rocking or head-banging) was found to be related to lower socioeconomic status and maternal depression by Petit et al(24), Laberge et al.(26) found no association between body-rocking and socioeconomic status or family adversity, although ~~did not~~ cases that persisted were not studied. Other sub-types of RMs have not been investigated so it is unclear if socio-demographic associations are sub-type specific. Although isolated cases have been reported in children with stressful home/social environments(30-32), there are more reports in children where this is not the case (6, 7, 9, 15, 33-40). While case reports can yield helpful insights into a rare condition they are inevitably highly selected and findings cannot be generalised.

Sleep stage distribution

Polysomnographic features of RMD have been reported in a total of 43 children, either as case series or case reports. Historically, RMD was believed to be a predormitum phenomenon(41), predominately associated with sleep onset(4, 16, 29). Indeed, the first edition of the ICSD(27) (both original and revised) classified RMD as a sleep-wake transition disorder. However, Kohyama et al.(42) collated published polysomnographic findings in 33, mostly adult, patients with RMD and noted that 13 cases uniquely exhibited RMs during sleep and never during the sleep-wake transition. Moreover, Mayer et al.(3) observed that RMs occurred during sleep in all 24 patients, even when movements were reported by the individual or bed partner to be confined to sleep onset. Furthermore, RMs

have been observed in all sleep stages. Dyken et al.(43), reported RMs in every sleep stage across the seven children studied, with the majority in N2 sleep. Of the seven children, three had attention deficit disorder, two had mental retardation, one had suffered neglect and abuse and one had seizures(43). Nonetheless, it is likely that these cases were representative, as three larger case series(3, 18, 42) also reported RMs in all stages of sleep. While the majority of affected children exhibit RMs in some, but not all, sleep stages(3, 15, 18, 31, 33, 42, 43), a minority have been reported to demonstrate RMs pervasively across the sleep cycle in each sleep stage(9, 18, 34, 42). Of particular note, 15 children across seven publications(18, 31, 33, 34, 38, 42, 43) have been reported to have RMs arising from REM sleep (six children had reported neurodevelopmental problems(18, 31, 38, 43), ADHD being most common(18, 38, 43)). Caution should be exercised however, in moving away from a construct of RMD as a disorder of transition into sleep. As highlighted noted in the ~~previous~~ section on diagnosis, children frequently suppress their RMs completely in the laboratory environment, hence PSG studies that report a predominance of sleep related RMs may be biased towards reporting movements that the child is unable to suppress. What is clear, however, from these data is that RMs can exist throughout the sleep cycle.

Consequences of RMD

Although the presence of injury and/or daytime impairment is a fundamental criteria for the diagnosis of RMD, there is sparse data on these crucial aspects of the condition.

Injury is often a significant concern to parents. While local trauma such as callus, ecchymosis and hair loss(10) is commonly seen ~~can occur~~ as a result of RMs with head-banging, ~~While there are a handful of case reports of more serious injuries, they are~~ exceptionally rare(11). Jeannet et al.(36) reported Hirayama disease, a cervical myelopathy associated with neck flexions, in a 10 year old girl with RMD (violent head- and body-rocking). Treatment and cessation of RMs in this case prevented further progression of weakness(36). However, this appears to be a unique case and no reports of brain injury or

retinal damage in children who head-bang were found in this review. However, this absence of evidence is not necessarily evidence of absence of subtle trauma. Systematic investigation with retinal and brain imaging would confirm this.

A key consideration in childhood RMD is sleep quality. The literature once again provides clues, but little consistency, largely limited by lack of objective measures of sleep with polysomnography or actigraphy. Future research in a larger sample of children is needed to understand the implications of different movement sub-types for sleep quality and in turn daytime function.

Sleep quality could potentially be impacted through persistent movements at sleep onset restricting total sleep time, or, through movements during sleep disrupting sleep quality(11). Conversely, RMD could be a symptom of an underlying sleep disorder that directly disrupts the continuity of sleep, such as periodic limb movement disorder (PLMD) or sleep apnea(44). The possible association between ADHD and persistent RMD is of note in this context, as PLMD is more prevalent in children with ADHD(45, 46) and could be a mediating factor. Importantly, Nevéus et al(25) found that children with RMD were at the highest risk of daytime sleepiness compared with all other sleep problems investigated. Poor sleep quality has significant consequences for the child; poor concentration, difficult behavior and impaired memory and decision-making capabilities(22, 47). These downstream consequences have not been studied in childhood RMD. Understanding the impact of RMD on sleep quality is clearly an important research agenda.

Finally to consider the social consequences of RMD. Children report feeling embarrassed by their behavior, leading to avoidance of social situations(2, 11, 16, 37) such as sleep-overs and school camps. RMD associated with head-banging can disturb other family members and neighbours(33). Indeed, it is very often this aspect that brings children to the attention of physicians. Once again however, this aspect of the condition has not been systematically studied.

Classification of RMD

Movement sub-type

The ICSD-III(1) recognises several subtypes of RMD. Head-banging(3-7, 9-11, 16, 18, 19, 22-24, 29, 31, 32, 34-36, 38, 40-43, 46-55), body-rocking(2-7, 9, 11, 16, 18, 19, 23-26, 29, 30, 34, 37, 38, 41, 43, 46, 47, 49-51, 53) and head-rolling(3, 4, 11, 16, 18, 23, 24, 29, 31, 39, 41, 42, 47) predominate in the literature. Other RMD sub-types include body-rolling(3, 9, 11, 15, 23, 31, 39), leg-banging(11, 23, 43), leg-rolling(11, 34), hand-banging(23, 34) and arm-banging(34). Whilst head-banging, body-rocking and head-rolling are most commonly identified in population studies, multiple case reports describe a broader range of movements that do not fit standard categories. For example single cases reports of isolated leg-rolling(34) and head punching(8) (33). Furthermore, case reports have highlighted the fact that children may engage in multiple types of RMs(6, 7, 9, 31, 34, 38, 39, 48).

No study has reported the distribution of RM subtypes within a clinical population. However, general population based studies provide some indication of the likely prevalence of RM sub-types. Sallustro and Atwell(29) , in a questionnaire based study of 525 healthy Californian children, aged 3 months to 6 years reported body-rocking in 92 of 483 (19.1%), head-banging in 26 of 506 (5.1%) and head-rolling in 30 of 473 (6.3%)(29), suggesting that body-rocking was most common variant. The large sample size, balance of gender (51% male), socioeconomic status, range of ethnicity (71% white, 21% Hispanic, 6% black, 1% Asian) and use of both urban and rural locations(29) allow some generalisation of this data. However, a significant limitation of the study was case-ascertainment. Movements were not required to be sleep related; indeed 44.2% of body-rockers, 33.3% of head-bangers and 43.5% of head-rollers did not exhibit movements when tired or at bedtime(29), suggesting only a minority could be candidates for RMD. Klackenberg's(4) study of 212 Swedish children randomly selected from the prospective longitudinal Stockholm study reported that at 9 months (n=203), body-rocking was more common (43%) than head-banging (28%) or

head-rolling (24%). At 12 months (n=207), head-banging was most commonly reported (39%) whereas at 36 months (n=202), head-rolling was the most common (7%)(4). Once again, case ascertainment was potentially inaccurate, as data relied entirely on parent report and did not report impact of the movements on the child. Stepanova et al.(18) studied seven children and three young adults with RMD using a combination of video-polysomnography and parental report. Head-banging and body-rolling were the most frequently reported movements in this small clinical sample(18).

Generating a meaningful classification system for this disorder has been impeded by the nature of the evidence base, largely relying as it does on single case reports (Appendix 2) and/or population studies based on parental report. While this of itself is interesting, and likely reflects the rarity of the disorder in clinical practice, it does limit our ability to generalise about the nature of the condition.

Movement frequency

The ICSD-III(1) states that the typical frequency of RMs is between 0.5 to 2 seconds. This is supported by Dyken et al(43) who studied seven children with RMD aged between 1-12 years and reported that all 37 RM episodes observed using video-polysomnography, had a movement frequency of between 0.5-2 Hz. Stepanova et al.(18) also reported all RMs within the 0.5-2Hz frequency range, with 1Hz being the most common frequency. Of the five case reports(9, 15, 33, 34, 37) that used polysomnography and documented the frequency of the child's RMs, all reported a frequency value within the 0.5-2Hz range (Appendix 2).

Movement duration during sleep period

There is a wide variation in the reported duration of RMs during the sleep period.

Klackenberg(4) reported that the total time spent in RM episodes over one night ranged from a few minutes to a few hours, Stepanova et al.(18) reported a 6 second to 85 minute range while Dyken et al.(43) reported a range of 1 to 59 minutes. Individual episodes were reported to last between 4 seconds and 21 minutes.

The number of individual episodes per night across case series and case reports has varied from a minimum of one episode to a maximum of 116 episodes reported by Stepanova et al(18).

This inter-individual variation in both sub-type of movement and in time domain parameters, such as the duration of episodes and the number of episodes each night, is reflected in the broad description given in the ICSD-III(1) but there is limited data to explain such variation.

Research and case-reports which use video-polysomnography provide the most detailed assessment of RMs and allow assessment of sleep quality. However, the largest single polysomnographic study to date (Stepanova et al.(18)) included only 7 children. As previously noted the sleep laboratory environment may impact on RM semiology. At present there is no consensus on the most appropriate technology for the identification and classification of RMD and no standardised approach for scoring movement episodes. We have reported a scoring algorithm for home videosomnography which demonstrates a high degree of inter-rater reliability(56, 57). Such approaches in the future would allow standardisation of RMD research.

Pathophysiology of RMD

As mentioned previously, a high reported prevalence of infantile RMs, suggest a normal developmental variant(4, 23), indicating a potential advantage of RMs in early life(23). Sallustro and Atwell(29) compared the attainment of 12 developmental milestones in infants with RMs (body-rocking, head-banging or head-rolling) and infants with none. Children who body-rocked (n=92) reached five developmental milestones (sitting up without support, getting up onto hands and knees, crawling, cruising, walking without support) at a significantly earlier age; children who head-banged (n=26) reached two milestones (holding head erect, walking without support) significantly earlier(29). The researchers suggested that RMs could confer an advantage to motor development through increased vestibular stimulation(29). No differences were found for children who head-rolled (n=30)(29), possibly

due to the small amplitude of such movements. Golbin et al.(49) noted that many of the RMs observed in RMD occur in planes of movement that mirror the semi-circular canals, lending support to the vestibular stimulation theory. This hypothesis was tested by Clark et al.(58) in a controlled experimental study. Twenty-six infants were exposed to vestibular stimulation over a four week period, consisting of 16 sessions in a rotating chair(58). Handling effects were controlled for by the control group attending the laboratory(58). Reflexes and motor skills significantly improved in the intervention group compared to controls(58). The researchers concluded that vestibular stimulation may facilitate earlier maturation of the vestibulo-ocular reflex and faster attainment of motor skills(58). The vestibular stimulation hypothesis, while intriguing, does not explain why such movement would be preferentially associated with sleep. An alternative hypothesis was proposed by Groswasser et al(59) who studied the effects of a rocking mattress in 18 infants (aged 46-71 weeks) over two nights using polysomnography. Eight infants were born prematurely and had apneas whilst on a neonatal unit(59). The other ten were full-term infants referred for investigation of sleep disordered breathing(59). The rocking mattress was associated with a significant decrease in the frequency of obstructive breathing events in 16 of 18 infants, from a median of 2.5 to 1.8 episodes per hour in preterm infants and from 1.5 to 0.7 in full-term infants(59). The extent to which such induced rocking movements mimic RMs is unclear and no prior polysomnographic studies of RMD cases has been designed to study sleep disordered breathing. This warrants further study.

Another theory is that RMs are a learned behavior which sooth the child at sleep onset and following night awakenings(11). Vrugt et al.(60) investigated the effects of vertical rocking frequencies on the behavior of 64 infants. Whereas infants who were not rocked tended to increase in arousal over time, infants receiving 1.5Hz rocking decreased in arousal over the rocking period(60). Frequencies of 0, 0.5, 1 and 1.5Hz were investigated and it was observed that the proportion of infants who slept increased with increasing rocking frequency(60). It has also been suggested that rocking is experienced as soothing as it

mimics the mother's movements(4), heartbeat(16) or respiratory rhythm(16). While these findings support the self-soothing hypothesis they do not explain more violent behaviors such as head-banging, nor reports of RMs during sleep(18).

Etzioni et al(61) tested the learned behavior hypothesis by trialling a sleep restriction and hypnotic medication regime in six children with RMD, aiming to reinforce the child's belief in their ability to fall asleep. Both episodes of RMs and sleep onset latency decreased significantly after treatment; RMs were completely abolished in all children at two weeks(61). Only one child re-experienced occasional movements at four weeks and at one year follow up(61). Whilst this study provides support for the learned behavior hypothesis, the sample size was small and little detail was given on the manifestation of each child's RMD(61).

Learned behavior has also been suggested as potentially causative in several case studies(2, 7, 30, 39). Attarian et al.(2) reported familial RMD and insomnia in a mother and two sons. An additional two family members also had insomnia(2). Albeit that affected family members were adults at the time of study, all had childhood onset RMD and it is suggested that the RMs may either have been learned from the mother by the two sons(2) or the familial trait of fragile sleep creates a pre-condition for RMs to be used as a self-soothing mechanism(2). Similarly, Evans(7) proposed that a 17 year old boy who had insomnia and severe body-rocking and head-banging at night, developed RMD as a learned behavior to reduce anxiety at bedtime. A number of case reports have noted the link between RMs and sleep onset latency. Lindsay et al.(39) reported that a 13 year old boy with frequent head and body rolling was unable to fall asleep without using these repetitive movements, while Blunden et al.(30) reported a 6 year old boy who rhythmically rocked over a teddy bear for 30-75 minutes at bedtime and demonstrated an increase in sleep onset latency when rocking time was limited. Interestingly, these movements were not exhibited when initiating sleep after night wakings(30).

A failure to coordinate sensory and motor information at sleep-wake transition has been suggested as a possible cause of RMD by Vetrugno and Montagna(41), although they

acknowledge this theory is not supported by empirical evidence. Similarly, Yeh et al.(33) reported RMD in sleep stages N1 and 2 and REM in two boys aged 11 and 13 years and a 22 year old woman and hypothesised that rhythmic activation of motor pattern generators in the brainstem and spinal cord(33) could be involved. Detailed functional imaging during RMD episodes would be challenging and as yet no such data is reported.

Fifty years ago psychoanalytic theory gave credence to the interpretation of head-banging as an autoerotic behavior used in the absence of environmental stimulation(54). While this interpretation is no longer mainstream, case reports have identified family stress and difficulty as potentially contributing to RMD(30-32) supported by the fact that family stress is associated with child sleep problems more generally(53).

Finally, isolated cases of RMD have been reported following head injury(48) and otitis media(6).

In summary the aetiology of RMD remains at best conjecture and requires further study. It is likely to represent the final common pathway of multiple aetiologies(47) where a child becomes reliant on a behavior which was initially physiological, and perhaps beneficial in infancy to induce sleep, but becomes established as a debilitating and intrusive behavior. Further research is needed to establish the underlying psychological, behavioral and neurological features of this condition.

Neurophysiology

The neurophysiological basis of RMD is intriguing. Recent reports of movements during REM sleep are intriguing. Preserved REM skeletal muscle tone is rare in childhood and mostly associated with narcolepsy and brainstem tumours. A recent case series however, reported REM sleep behavior disorder in children with a spectrum of neurodevelopmental disorders including ADHD, autism, Smith-Magenis syndrome and Tourette's syndrome(62). The authors(62) note an association between autism and impaired GABA signalling(63) and

hypothesised that deficiency of GABA could in turn diminish spinal neuron hyperpolarisation during REM sleep. It is possible that similar mechanisms exist in RMD, particularly persistent RMD which has been reported to be associated with neurodevelopmental disorders(18, 42). However, against this theory is the fact that only a minority of RMs occur in REM sleep. Other potential neuronal pathways which have been hypothetically linked to RMD include the vestibulo-spinal, tecto-spinal and reticulo-spinal tracts(42). These tracts predominately innervate the axial and proximal muscles and are fully myelinated during early life when the majority of RMs tend to appear(42). Whether the presence of RMs in particular stages of sleep predict how severely a child's sleep is affected by RMD requires further study.

Co-morbidities and associated factors

Co-morbid sleep disorders

Individual case reports(2, 7, 38) and small case series(3, 18) have noted other sleep disorders to be comorbid with RMD including: bruxism(18), somniloquy(18, 38), obstructive sleep apnea (OSA) (3, 18), primary snoring(18, 44), somnambulism(25), insomnia(2, 7, 25) and restless legs syndrome(41). Findings are, however, inconsistent. Liukkonen et al.(44) reported a significant correlation between parent report of snoring frequency and the presence of RMs in a cross-sectional study of sleep disordered breathing in 1471 children aged 1-6 years old. Mayer et al.(3) reported the coexistence of OSA in five adult patients with RMD and observed that arousals occurring at the end of apneas frequently induced RMs. No similar polysomnographic evidence is available in children. Laberge et al.(26) found no associations between parental reports of body-rocking and somniloquy or somnambulism but did not comment on other sleep disorders. Klackenberg(4) reported no associations with bruxism in children with RMs up to the age of eight years old.

Other co-morbidities

Clinical experience suggests that many children with RMD are healthy and typically developing(11). One of the authors(46) reviewed the relationship between attention deficit hyperactivity disorder (ADHD) and sleep-related movement disorders and noted a probable bi-direction relationship between ADHD and RMD. ADHD does occur commonly in the literature in children with RMD(46), Stepanova et al.(18) reported a diagnosis of ADHD in six of their ten cases; Dyken et al.(43) reported attention deficit disorder in three of seven cases of RMD. Furthermore, RMD appears to be more prevalent in the attention deficit hyperactivity disorder (ADHD) population(46). Further experimental studies with strict case definition are required to determine the direction of the relationship between ADHD and RMD. Impaired attention could be the result of sleep disruption or RMD and ADHD may share a common predisposition to increased motor activity(46).

Laberge et al(26) reported that body-rockers had significantly higher anxiety levels compared with non-rockers of the same age, though the use of parental report could be have bias reporting. Isolated cases have reported RMD as an anxiety-reducing behavior(7, 30). Certainly, children experiencing extreme emotional deprivation engage in self-soothing rocking in the daytime(64) – whether this translates to a uniquely sleep related behavior however, is unclear.

Stepanova et al(18) reported that seven of their ten cases of RMD also had allergic symptoms and five had positive perinatal histories (high risk pregnancy and prematurity (n=3), hyperbilirubinaemia (n=2)). These associations are not mentioned elsewhere in the literature. Haywood et al.(11), in a personal practice article, noted an association with autism spectrum disorders and learning difficulties but provided no research data to support this.

(11)

In summary no single study has been designed to investigate co-morbidities in RMD. The possible link with ADHD and neurodevelopmental disorders is intriguing and requires further

study. Further data on co-morbidities could potentially inform our understanding of the aetiology of RMD.

Persistent RMD

There are no data that report the average age of onset and remission of RMD using the ICSD-III(1) diagnostic criteria. Contemporary knowledge is therefore bedevilled by the case ascertainment problems highlighted earlier. Sallustro and Atwell(29), reported the average age of onset of RMs (both sleep and non-sleep related) as 6.4 months for body-rocking, 9.4 months for head-banging and 9.7 months for head-rolling. It is often asserted that RMD resolves spontaneously in early childhood(11, 23) and by 5 years of age, the prevalence of RMD has been reported to drop to around 2.6-6%(4, 24). Although the few prevalence studies do have limitations, for the purpose of this review, we have defined 'persistent RMD' as failure to resolve spontaneously before the age of 5 years. While the ICSD-III(1) does not provide an age limit for diagnosis, it too focuses on the low rate of persistence at five years. Mayer et al.(3) studied 24 patients with persistent RMs, four aged between 11 and 18 years old and 20 aged over 18 years old. All were reported to have developed RMs during childhood: 16 had onset before 5 years of age and eight had onset between 5 and 17 years of age(3). They also observed that there was a male preponderance in their sample (n=20)(3), however, no description of the sampling method is given so selection bias is possible. Persistence is associated with, but not exclusively seen, in children with developmental or intellectual disorders(11). Kohyama et al.(42) identified seven cases of co-existing psychiatric disorders or intellectual disability in 27 cases of RMD over ten years old. Six cases of co-existing ADHD were identified by Stepanova et al.(18) in ten children with persistent RMD. Mayer et al.(3) reported psychopathology in only five patients of 24 patients. Thus the limited existing literature offers a mixed viewpoint on whether an association between persistent RMD and neurodevelopmental/psychiatric disorders exists. Thus there is insufficient evidence to fully understand the true natural history of the condition.

Treatment

There have been no therapeutic randomised controlled trials in RMD and the available evidence is limited to clinical perspectives, case studies or small sample observational studies. Parental reassurance and the avoidance of reinforcing RMs through parental attention have formed the mainstay of recommendations(11, 35, 55, 65), although no empirical support is given to demonstrate how useful these approaches are in practice. The presumed self-limiting nature of RMs in pre-school children has been emphasised(11).

Several authors(5, 11, 16, 41) recommend reducing injury risk for example by ensuring that the child's bed is stable(11), using bedrails(11), moving the bed away from the wall(11) and, where necessary, the use of padding(11, 16).

In 2003, Kuhn et al.(47) systematically reviewed psychological treatments for RMD using the Chambless criteria which categorise interventions as 'well established' if they produce benefits that exceed another treatment or placebo or 'promising' if initial empirical evidence from two or more studies evaluating the same treatment protocol exist but fail to meet the full Chambless criteria). No psychological treatments were identified that met either criteria(47). Behavioral approaches, including teaching replacement behaviors, reward systems and aversion therapy, have been reported in a small number of quasi-experimental studies and case reports, however, no two studies have replicated a consistent treatment protocol(47). A review of cognitive behavioral treatments for childhood sleep disorders(53) in 2005 also noted the scarcity of data on head-banging and body-rocking. Although more than a decade has passed since these reviews(47, 53), quality research into treatment of RMD remains elusive. Etzioni et al.(61) trialled a three week sleep restriction regime in a sample of six children with RMD. Sleep restriction by one hour per night for 2 weeks and an incremental return by 10 minutes per night to baseline sleep in week 3, with chloral hydrate 0.5ml/kg at bedtime in week one, aimed to re-establish the child's belief in their ability to fall asleep without RMs(61). A high success rate was reported(61), however, this has not been replicated in a larger sample nor in a randomised controlled trial. Other behavioral

techniques used with partial or complete success and reported in case studies include aversive stimulation(51) and improving sleep hygiene(6).

There are no randomised controlled trials of pharmacological treatments for RMD and current knowledge is based on limited case studies. Despite this, major texts and reviews continue to support these approaches(5, 16, 23, 41). The benzodiazepines clonazepam(31, 33, 34), flurazepam(52) and oxazepam(37) have been reported to have partial success in eliminating RMs although have no effect in some patients(8, 33, 52). Rapid development of tolerance(37) has also been reported. Drake(48) used the tricyclic antidepressant imipramine to eliminate RMs in a 16 year old girl. However, the pathophysiology of the RMs in this patient may differ from typical cases, as the RMs developed following a head injury. Lee(8) reported that use of the dopamine antagonists haloperidol and pimozide resulted in a reduction in the intensity and duration of head-punching in a 17 year old boy. No other studies have reported tricyclic antidepressants or dopamine antagonists in children with more typical presentations of RMD.

Currently the treatment of RMD relies heavily on clinical experience with few studies providing strong evidence in favour of a particular treatment approach. In addition to the lack of high quality evidence, reporting bias created by the tendency for case reports to only focus on successful treatment is likely.

Conclusion

Rhythmic movement disorder is arguably the Cinderella of sleep disorders, with a poverty of reliable research to guide clinical practice. Where it persists in childhood, clinical experience bears witness to a distressing condition with the potential to disrupt the sleep of the index child and extended family, with adverse consequences all too familiar to sleep physicians. There is a challenging agenda for clinical research to establish fundamental facts about this condition and its treatment.

Practice points

- 1) Rhythmic movements in infancy may promote motor development but beyond infancy rhythmic movement disorder may persist and children should be offered clinical review
- 2) Parental report may be unreliable and objective observation of movements is recommended. Importantly the movements must have a consequence to be considered a disorder
- 3) No single treatment approach has sufficient high quality evidence to be recommended leaving clinician discretion and family preference as the best current approach.

Research agenda

- 1) Population prevalence of rhythmic movement disorder with a clinical consequence (as opposed to simple sleep related rhythmic movements without a clinical consequence) must be established
- 2) Detailed phenotyping and follow up of a clinical population is required to understand the natural history and risk factors associated with this condition
- 3) Randomised controlled studies of quality treatment trials with behavioral, psychological and pharmacological approaches are needed in carefully defined clinical populations using agreed scoring criteria.

References

- *1. Medicine AAsS. The International Classification of Sleep Disorders. 3rd ed. Darien IL, editor: American Academy of Sleep Medicine; 2014.

2. Attarian H, Ward N, Schuman C. A multigenerational family with persistent sleep related rhythmic movement disorder (RMD) and insomnia. *J Clin Sleep Med*. 2009;5(6):571-2.
- *3. Mayer G, Wilde-Frenz J, Kurella B. Sleep related rhythmic movement disorder revisited. *Journal of Sleep Research*. 2007;16(1):110-6.
- *4. Klackenberg G. Rhythmic movements in infancy and early childhood. *Acta Paediatr Scand*. 1971;60(s224):74-83.
5. Walters AS. Clinical identification of the simple sleep-related movement disorders. *Chest*. 2007;131(4):1260-6.
6. Bramble D. Two cases of severe head-banging parasomnias in peripubertal males resulting from otitis media in toddlerhood. *Child Care Health Dev*. 1995;21(4):247-53.
7. Evans J. Rocking at night. *J Child Psychol Psychiatry*. 1961;2:71-85.
8. Lee SK. A Case with Dopamine-Antagonist Responsive Repetitive Head Punching as Rhythmic Movement Disorder during Sleep. *J Epilepsy Res*. 2013;3(2):74-5.
9. Manni R, Terzaghi M, Sartori I, Veggiotti P, Parrino L. Rhythmic movement disorder and cyclic alternating pattern during sleep: a video-polysomnographic study in a 9-year-old boy. *Mov Disord*. 2004;19(10):1186-90.

10. Strauss CC, Rubinoff A, Atkeson BM. Elimination of nocturnal headbanging in a normal seven-year-old girl using overcorrection plus rewards. *J Behav Ther Exp Psychiatry*. 1983;14(3):269-73.
11. Haywood PM, Hill CM. Rhythmic movement disorder: managing the child who head-bangs to get to sleep. *Paediatrics and Child Health*. 2012;22(5):207-10.
12. Whittemore R, Knafk K. The integrative review: updated methodology. *Journal of Advanced Nursing*. 2005;52(5):546-53.
13. Conn VS, Isaramalai SA, Rath S, Jantarakupt P, Wadhawan R, Dash Y. Beyond MEDLINE for literature searches. *J Nurs Scholarsh*. 2003;35(2):177-82.
14. Melnyk BM, Fineout-Overholt E. Evidence-based practice in nursing & healthcare : a guide to best practice. 3rd ed. Philadelphia: Lippincott Williams & Wilkins; 2014. xxvi, 625 p. p.
15. Kaneda R, Furuta H, Kazuto K, Arayama K, Sano J, Koshino Y. An unusual case of rhythmic movement disorder. *Psychiatry Clin Neurosci*. 2000;54(3):348-9.
16. Thorpy MJ, Glovinsky P, B. Parasomnias. *Psychiatric Clinics of North America*. 1987;10(4):629-32.
17. Berry RB, Brooks R, Gamaldo CE, Harding SM, Lloyd RM, Marcus CL, et al. The AASM Manual for the Scoring of Sleep and Associated Events:

Rules, Terminology and Technical Specifications. 2.2 ed. Darien, Illinois: American Academy of Sleep Medicine; 2015.

- *18. Stepanova I, Nevsimalova S, Hanusova J. Rhythmic movement disorder in sleep persisting into childhood and adulthood. *Sleep*. 2005;28(7):851-7.
19. Walters AS, Lavigne G, Hening W, Picchiatti DL, Allen RP, Chokroverty S, et al. The scoring of movements in sleep. *J Clin Sleep Med*. 2007;3(2):155-67.
20. Morgenthaler T, Alessi C, Friedman L, Owens J, Kapur V, Boehlecke B, et al. Practice parameters for the use of actigraphy in the assessment of sleep and sleep disorders: an update for 2007. *Sleep*. 2007;30(4):519-29.
21. Singh J, Badr MS, Diebert W, Epstein L, Hwang D, Karres V, et al. American Academy of Sleep Medicine (AASM) Position Paper for the Use of Telemedicine for the Diagnosis and Treatment of Sleep Disorders. *J Clin Sleep Med*. 2015;11(10):1187-98.
22. Stores G. Aspects of sleep disorders in children and adolescents. *Dialogues in Clinical Neuroscience*. 2009;11(1):81-90.
23. Manni R, Terzaghi M. Rhythmic movements during sleep: a physiological and pathological profile. *Neurological Sciences*. 2005;26 Suppl 3:s181-5.

24. Petit D, Touchette E, Tremblay RE, Boivin M, Montplaisir J. Dyssomnias and parasomnias in early childhood. *Pediatrics*. 2007;119(5):e1016-25.
25. Nevéus T, Cnattingius S, Olsson U, Hetta J. Sleep habits and sleep problems among a community sample of schoolchildren. *Acta Paediatr*. 2001;90(12):1450-5.
26. Laberge L, Tremblay RE, Vitaro F, Montplaisir J. Development of parasomnias from childhood to early adolescence. *Pediatrics*. 2000;106(1 Pt 1):67-74.
27. Medicine AaOs. The International Classification of Sleep Disorders, revised: Diagnostic and Coding Manual. Chicago, Illinois: American Academy of Sleep Medicine; 2001.
28. Medicine AaOs. The International Classification of Sleep Disorders: Diagnostic & Coding Manual. 2nd ed: American Academy of Sleep Medicine; 2005.
29. Sallustro F AC. Body rocking, head banging, and head rolling in normal children. *The Journal of Pediatrics*. 1978;93(4):704-8.
30. Blunden S, Nair D. An unusual clinical phenomenon: a case of bedtime ritual with apparent sexual overtones. *Clin Child Psychol Psychiatry*. 2010;15(1):55-64.

31. Hashizume Y, Yoshijima H, Uchimura N, Maeda H. Case of head banging that continued to adolescence. *Psychiatry Clin Neurosci.* 2002;56(3):255-6.
32. Ross RR. Treatment of nocturnal headbanging by behavior modification techniques: a case report. *Behav Res Ther.* 1971;9(2):151-4.
33. Yeh SB, Schenck CH. Atypical headbanging presentation of idiopathic sleep related rhythmic movement disorder: three cases with video-polysomnographic documentation. *J Clin Sleep Med.* 2012;8(4):403-11.
34. Su C, Miao J, Liu Y, Liu R, Lei G, Zhang W, et al. Multiple forms of rhythmic movements in an adolescent boy with rhythmic movement disorder. *Clin Neurol Neurosurg.* 2009;111(10):896-9.
35. Happe S, Ludemann P, Ringelstein EB. Persistence of rhythmic movement disorder beyond childhood: a videotape demonstration. *Mov Disord.* 2000;15(6):1296-8.
36. Jeannot PY, Kuntzer T, Deonna T, Roulet-Perez E. Hirayama disease associated with a severe rhythmic movement disorder involving neck flexions. *Neurology.* 2005;64(8):1478-9.
37. Walsh JK, Kramer M, Skinner JE. A case report of jactatio capitis nocturna. *Am J Psychiatry.* 1981;138(4):524-6.
38. Mattewal A, Casturi L, Subramanian S. A child with REM sleep disturbance. *J Clin Sleep Med.* 2010;6(1):97-100.

- *39. Lindsay SJ, Salkovskis PM, Stoll K. Rhythmical body movement in sleep: A brief review and treatment study. *Behaviour Research and Therapy*. 1982;20(5):523-6.
40. Balaschak BA, Mostofsky DI. Treatment of nocturnal headbanging by behavioral contracting. *Journal of Behavior Therapy and Experimental Psychiatry*. 1980;11(2):117-20.
41. Vetrugno R, Montagna P. Sleep-to-wake transition movement disorders. *Sleep Medicine*. 2011;12 Suppl 2:S11-6.
- *42. Kohyama J, Matsukura F, Kimura K, Tachibana N. Rhythmic movement disorder: polysomnographic study and summary of reported cases. *Brain & Development*. 2002;24(1):33-8.
- *43. Dyken ME, Lin-Dyken DC, Yamada T. Diagnosing rhythmic movement disorder with video-polysomnography. *Pediatric Neurology*. 1997;16(1):37-41.
44. Liukkonen K, Virkkula P, Aronen ET, Kirjavainen T, Pitkaranta A. All snoring is not adenoids in young children. *Int J Pediatr Otorhinolaryngol*. 2008;72(6):879-84.
45. Konofal E, Lecendreux M, Cortese S. Sleep and ADHD. *Sleep Med*. 2010;11(7):652-8.
46. Walters AS, Silvestri R, Zucconi M, Chandrashekariah R, Konofal E. Review of the possible relationship and hypothetical links between

attention deficit hyperactivity disorder (ADHD) and the simple sleep related movement disorders, parasomnias, hypersomnias, and circadian rhythm disorders. *J Clin Sleep Med*. 2008;4(6):591-600.

47. Kuhn BR, Elliott AJ. Treatment efficacy in behavioral pediatric sleep medicine. *Journal of Psychosomatic Research*. 2003;54(6):587-97.
48. Drake ME, Jr. Jactatio nocturna after head injury. *Neurology*. 1986;36(6):867-8.
49. Golbin AZ, Guseva VE, Shepovalnikov AN. Unusual behaviors in sleep as 'compensatory' reactions, aimed at normalizing the sleep-wake cycle. *Human Physiology*. 2013;39(6):635-41.
50. Manni R, Terzaghi M. Comorbidity between epilepsy and sleep disorders. *Epilepsy Res*. 2010;90(3):171-7.
51. Martin RD, Conway JB. Aversive stimulation to eliminate infant nocturnal rocking. *Journal of Behavior Therapy and Experimental Psychiatry*. 1976;7(2):200-1.
52. Reimao R, Lefevre AB. Evaluation of flurazepam and placebo on sleep disorders in childhood. *Arq Neuropsiquiatr*. 1982;40(1):1-13.
53. Sadeh A. Cognitive-behavioral treatment for childhood sleep disorders. *Clinical Psychology Review*. 2005;25(5):612-28.
54. Silberstein RM, Blackman S, Mandell W. Autoerotic Head Banging. *Journal of the American Academy of Child Psychiatry*. 1966;5(2):235-42.

55. Wiggs L. Behavioural aspects of children's sleep. *Archives of Disease in Childhood*. 2009;94(1):59-62.
56. Gwyther A. A study to determine the prevalence and phenotype of rhythmic movement disorder in children with Down Syndrome [Unpublished undergraduate study report]: University of Southampton; 2016.
57. Gwyther A, Wood I, Gogo E, Cheung T, Jones L, Hill C. Videosomnography assessment in childhood rhythmic movement disorder: validation of a novel scoring algorithm. 23rd Congress of the European Sleep Research Society; Bologna2016.
- *58. Clark DL, Kreutzberg JR, Chee FK. Vestibular stimulation influence on motor development in infants. *Science*. 1977;196(4295):1228-9.
59. Groswasser J, Sottiaux M, Rebuffat E, Simon T, Vandeweyer M, Kelmanson I, et al. Reduction in obstructive breathing events during body rocking: a controlled polygraphic study in preterm and full-term infants. *Pediatrics*. 1995;96(1 Pt 1):64-8.
- *60. Vrugt DT, Pederson DR. The Effects of Vertical Rocking Frequencies on the Arousal Level in Two-Month-Old Infants. *Child Development*. 1973;44(1):205-9.

- *61. Etzioni T, Katz N, Hering E, Ravid S, Pillar G. Controlled sleep restriction for rhythmic movement disorder. *Journal of Pediatrics*. 2005;147(3):393-5.
62. Lloyd R, Tippmann-Peikert M, Slocumb N, Kotagal S. Characteristics of REM sleep behavior disorder in childhood. *J Clin Sleep Med*. 2012;8(2):127-31.
63. Chao HT, Chen H, Samaco RC, Xue M, Chahrour M, Yoo J, et al. Dysfunction in GABA signalling mediates autism-like stereotypies and Rett syndrome phenotypes. *Nature*. 2010;468(7321):263-9.
64. MacLean K. The impact of institutionalization on child development. *Dev Psychopathol*. 2003;15(4):853-84.
65. Mindell JA, Owens JA. *A Clinical Guide to Pediatric Sleep: Diagnosis and management of sleep problems*. 2 ed: Lippincott Williams & Wilkins; 2009. 352 p.

Figure Legends

Figure 1: Diagnostic criteria for rhythmic movement disorder as described in the International Classification of Sleep Disorders, 3rd edition(1).

Figure 2: Levels of evidence. Level I indicates the highest level of evidence; Level VII indicates the poorest level of evidence. Adapted Melnyk and Fineout-Overholt (2014)(14).

Figure 3: Flow chart showing the literature selection process.

Figure 4: Publications included in the integrative review organised by evidence level and type of study.

Figure 5: Differential diagnosis of the sleep-related movement disorders in childhood. Adapted from Walters et al (2007)(19) using the International Classification of Sleep Disorders, 3rd edition(1).

Figure 6: American Academy of Sleep Medicine scoring criteria for polysomnographic assessment of rhythmic movement disorder(17).

Figure 7: Actigraphy in two toddlers, one without RMD (a) and one with RMD (b) illustrating potential for mis-interpretation in absence of video

Appendix 1: Summary of included publications.

Appendix 2: Summary of case reports and small case series

Appendices

Appendix 1: Summary of included publications

Author (date)	Aim of study	Type of study	Evidence level	Sample/ population	Themes	ICSD-III criteria C explicitly met?
Attarian et al. (2009)²	Case report on a multigenerational family with persistent RMD and insomnia	Case report	VII	3 family members with RMD	Aetiology Consequences of RMD Persistent RMD	Yes
Balaschak and Mostofsky (1980)⁴⁰	Case report on the treatment of head-banging by behavioural contracting in a 16 year old male	Case report	VII	16 year old male with RMD	Aetiology Persistent RMD Treatment	No
Blunden et al. (2009)³⁰	Case report on an unusual presentation of RMD consisting of rocking at sleep onset with apparent sexual overtones	Case report	VII	6 year old male with RMD	Aetiology Classification Consequences of RMD Persistent RMD	Yes
Bramble (1995)⁶	Case series on two cases of head-banging following otitis media in toddlerhood	Case series	VI	2 cases of RMD (aged 12 and 11 years old)	Aetiology Persistent RMD Treatment	Yes
Clark et al. (1977)⁵⁸	Quasi experimental study investigating the effects of vestibular stimulation on motor development in infants	Quasi experimental study	III	26 normal infants aged between 3 and 13 months	Aetiology	N/A
Drake (1986)⁴⁸	Case report on the development of head-banging in a 16 year old female after a head injury	Case report	VII	16 year old female with RMD	Co-morbidities and associated factors Persistent RMD Treatment	Yes
Dyken et al. (1997)⁴³	Case series evaluating the use of split-screen polysomnography in RMD	Case series	VI	7 children with RMD aged 1-12 years	Co-morbidities and associated factors Classification Identification and diagnosis Persistent RMD Sleep stage distribution	No
Etzioni et al. (2005)⁶¹	Case series evaluating the effects of controlled sleep restriction in the Treatment	Case series	VI	6 children aged 3.5 to 12 years with RMD	Aetiology Persistent RMD Treatment	Children had diagnosis of RMD
Evans (1961)⁷	Case report of RMD in a 17 year old male with learning difficulties and developmental delay	Case report	VII	17 year old male with RMD	Aetiology Consequences of RMD Persistent RMD	Yes
Golbin et al. (2013)⁴⁹	Expert opinion article performing clinical and psychological analysis of	Expert opinion	VII	Children (age range unspecified)	Aetiology	No

	parasomnias in children using data from three different institutions	article				
Groswasser et al. (1995)⁵⁹	Quasi-experimental study investigating the effects of body rocking in infants on respiratory behaviour during sleep	Quasi-experimental study	III	18 infants with documented obstructive sleep apnoea	Aetiology	N/A
Happe et al. (2000)³⁵	Case report on two cases of persistent RMD evaluated using video polysomnography	Case series	VI	2 cases of RMD (aged 15 and 18 years old)	Persistent RMD Treatment	No
Hashizume et al. (2002)³¹	Case report on a male with persistent head-banging	Case report	VII	15 year old male with RMD	Aetiology Persistent RMD Sleep stage distribution Treatment	Yes
Haywood et al. (2012)¹¹	Practical guidance on the diagnosis and management of RMD in children	Expert opinion article	VII	Children (age range unspecified)	Aetiology Co-morbidities and associated factors Classification Identification and diagnosis Persistent RMD Treatment	Yes
Jeannet et al. (2005)³⁶	Case report on the potential association between a case of Hirayama disease and severe RMD	Case report	VII	10 year old female with RMD	Consequences of RMD Persistent RMD Treatment	Yes
Kaneda et al. (2000)¹⁵	Case report on a male with sleep related body-rolling	Case report	VII	12 year old male with RMD	Identification and diagnosis Persistent RMD Sleep stage distribution	No
Klackenberg (1971)⁴	Cohort study investigating the prevalence and natural history of head-banging, head-turning and rocking	Cohort study	IV	203 infants	Aetiology Co-morbidities and associated factors Classification Consequences of RMD Prevalence of RMD Sleep stage distribution	No
Kohyama et al. (2002)⁴²	Case series reporting the relationship between RMD and sleep stages using two original and 31 published cases of RMD	Case series	VI	33 cases of RMD (age range 2-56 years, 18 children)	Neurophysiology Persistent RMD Sleep stage distribution	No
Kuhn et al. (2003)⁴⁷	Review of the efficacy of the psychological Case report for paediatric behavioural sleep conditions using the Chambless criteria	Review article	V	Children (age range unspecified)	Aetiology Consequences of RMD Treatment	No

Laberge et al. (2000) ²⁶	Cohort study examining the prevalence and development of parasomnias in childhood and early adolescence and investigating associations with anxiety, family adversity and gender	Cohort study	IV	1353 children, parents completed questionnaires between ages of 3-10 years	Co-morbidities and associated factors Persistent RMD Prevalence	No
Lee (2013) ⁸	Case report of the treatment of repetitive head punching with dopamine antagonists	Case report	VII	17 year old male with RMD	Persistent RMD Treatment	Yes
Lindsay et al. (1982) ³⁹	Case report on the use of behavioural approaches in the treatment of head-rolling in a 13 year old male	Case report	VII	13 year old male with RMD	Aetiology Consequences of RMD Persistent RMD Treatment	No
Liukkonen et al. (2008) ⁴⁴	Cross sectional study determining the prevalence of snoring in young children and assessing other related factors	Cross sectional study	VI	2100 children aged between 1-6 years old	Co-morbidities and associated factors	No
Manni and Terzaghi (2010) ⁵⁰	Review of the comorbidity between epilepsy and sleep disorders	Review article	V	All ages (no specified age range)	Co-morbidities and associated factors Identification and diagnosis	No
Manni et al. (2004) ⁹	Case report evaluating a male with multiple forms of RMD using videosomnography and cyclic alternating pattern analysis	Case report	VII	9 year old male with RMD	Persistent RMD	Yes
Manni et al. (2005) ²³	Review of the clinical and polysomnography aspects of RMD using both published and unpublished literature and data	Review article	V	All ages (no specified age range)	Co-morbidities and associated factors Classification	No
Martin and Conway (1976) ⁵¹	Case report on the use of aversive stimulation to eliminate infant nocturnal rocking	Case report	VII	25 month old female with RMD	Treatment	No
Mattewal et al. (2010) ³⁸	Case report of a child with RMD associated with rapid eye movement sleep	Case report	VII	8 year old male with RMD	Co-morbidities and associated factors Consequences of RMD Neurophysiology Persistent RMD Sleep stage distribution	Yes
Mayer et al. (2007) ³	To classify RMD based on clinical, polysomnographic and videometric evaluation in a predominantly adult population	Case series	VI	24 patients aged 11-67 years old	Co-morbidities and associated factors Persistent RMD Sleep stage distribution	No
Nevés et al. (2001) ²⁵	Cross-sectional study describing the sleep habits and sleep problems in a sample of school children and	Cross-sectional study	VI	1413 children aged 6.2-10.9 years old	Co-morbidities and associated factors Consequences of RMD Persistent RMD Prevalence	No

	evaluating the association between different sleep problems					
Petit et al. (2007) ²⁴	Cross-sectional study to determine the prevalence of dysomnias and parasomnias in early childhood and investigating their natural history and any gender differences	Cross-sectional study	VI	1997 families completed questionnaires at various intervals (between ages of 2.5 to 6 years)	Co-morbidities and associated factors Consequences of RMD Persistent RMD Prevalence	No
Reimao and Lefevre (1982) ⁵²	Report on the use of flurazepam in the treatment of childhood sleep disorders	Case series*	VI	2 children with head-banging	Treatment	Children had diagnosis of RMD
Ross et al. (1971) ⁵²	Case report on the treatment of RMD using behaviour modification	Case report	VII	16 year old female with RMD	Aetiology Persistent RMD Treatment	Yes
Sadeh et al. (2005) ⁵³	Review of the cognitive-behavioural interventions proposed for the treatment of childhood sleep disorders	Review article	V	Children (age range unspecified)	Aetiology	No
Sallustro and Atwell (1978) ²⁹	Cross-sectional study determining the incidence and demographics of body-rocking, head-banging and head-rolling in normal children and investigating the effects of large amounts of vestibular stimulation on attainment of developmental milestones	Cross-sectional study	VI	525 healthy children aged 3 months to 6 years	Co-morbidities and associated factors Classification Persistent RMD Prevalence of RMD Sleep stage distribution	No
Silberstein et al. (1966) ⁵⁴	Expert opinion article describing the psychoanalytic approach to interpreting head-banging	Expert opinion article	VII	Children (age range unspecified)	Aetiology	No
Stepanova et al. (2005) ¹⁸	Cohort study evaluating rhythmic movements in sleep stages of children and young adults and investigating whether persistent rhythmic movement disorder (RMD) is associated with any daytime symptoms or psychopathology	Cohort study	IV	10 individuals aged 7-24 years with RMD (7 children)	Co-morbidities and associated factors Classification Neurophysiology Persistent RMD Sleep stage distribution	Yes
Stores et al. (2009) ²²	An article describing aspects of common sleep problems in children using the International Classification of Sleep Disorders	Expert opinion article	VII	Children (age range unspecified)	Aetiology Consequences of RMD Identification and diagnosis Prevalence of RMD	No
Strauss et al. (1983) ¹⁰	Case report on the elimination of head-banging in a 7 year old female	Case report	VII	7 year old female with RMD	Persistent RMD Treatment	Yes

	using overcorrection and rewards					
Su et al. (2009) ³⁴	Case report on the existence of multiple forms of rhythmic movements in an adolescent male with RMD	Case report	VII	15 year old male with RMD	Classification Persistent RMD Sleep stage distribution Treatment	Yes
Thorpy et al. (1987) ¹⁶	Review of parasomnias in line with the International Classification of Sleep Disorders	Review article	V	All ages (no specified age range)	Aetiology Co-morbidities and associated factors Classification Consequences of RMD Sleep stage distribution	No
Vetrugno and Montagna (2011) ⁴¹	Review of three motor phenomena linked with sleep-wake transition	Review article	V	All ages (no specified age range)	Aetiology Co-morbidities and associated factors Classification Sleep stage distribution	No
Vrugt et al. (1973) ⁶⁰	Randomised controlled trial investigating the effects of vertical rocking frequencies on the arousal level in 2 month old infants	Randomised controlled trial	II	64 infants with mean age of 64 days	Aetiology Consequences of RMD	N/A
Walsh et al. (1981) ³⁷	Case report of head-banging in a 8 year old female	Case report	VII	8 year old female with RMD	Aetiology Classification Consequences of RMD Persistent RMD	No
Walters (2007) ⁵	Review of the sleep-related movement disorders and guidance on forming a more narrowly focused differential diagnosis	Review article	V	All ages (no specified age range)	Identification and diagnosis Treatment	Yes
Walters et al. (2008) ⁴⁶	Review of the possible relationship between attention deficit hyperactivity disorder and the simple sleep-related movement disorders	Review article	V	All ages (no specified age range)	Co-morbidities and associated factors	Yes
Walters et al. (2007) ¹⁹	Review article on the scoring of movements during sleep with the aim of establishing scoring criteria for each of the simple sleep-related movement disorders	Review article	V	All ages (no specified age range)	Identification and diagnosis	Yes
Wiggs et al. (2009) ⁵⁵	Review of key aspects of behavioural therapies used in children's sleep	Review article	V	Children (age range unspecified)	Co-morbidities and associated factors	No
Yeh et al. (2012) ³³	Case series on atypical presentations of head-banging with evaluation using video polysomnography	Case series	VI	3 cases of RMD (aged 11-22 years old)	Aetiology Classification Consequences Identification and diagnosis Persistent RMD Sleep stage distribution Treatment	Yes

ISCD = International Classification of Sleep Disorders, RMD = rhythmic movement disorder

* Although this study was a randomised controlled trial of Flurazepam in sleep disorders in childhood, only two cases of RMD were identified and no statistical analysis was conducted. Thus the evidence is considered to be equivalent to a case series.

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Appendix 2: Summary of case reports and small case series

Where possible, measures recorded using PSG have been reported rather than parental observations

Author (Yr of study)	Age of child (yrs)	Gender	Sub-type of rhythmic movement	Onset of rhythmic movements (yrs)	Frequency of movements	Number of episodes per night	Duration of episodes	Total time performing movements (mins)	Timing: sleep onset or in sleep	Clinical consequences
Balaschak and Mostofsky (1980) ⁴⁰	16	Male	Head-banging	3.5	Not reported	1-8	2-20 minutes	Not reported	Both	Affecting parents' sleep
Blunden et al. (2009) ³⁰	6	Male	Body-rocking ^a	Not reported	Not reported	1	30-75 minutes	30-75 minutes	Sleep onset	Sleep disturbance Daytime sleepiness
Bramble (1995) ⁶	11	Male	Head-banging Body-rocking	'mid toddlerhood'	Not reported	Not reported	Not reported	Not reported	During sleep	Sleep disturbance Affecting family's sleep
Bramble (1995) ⁶	12	Male	Head-banging	3	Not reported	Not reported	Not reported	Up to 4 hours	Sleep-onset	Social implications Injury Time consuming
Drake (1986) ⁴⁸	16	Female	Head-banging 'Body-thrashing'	16 ^c	Not reported	Not reported	1-3 minutes	Not reported	During sleep	Risk of injury
Evans (1961) ⁷	17	Male	Body rocking Head-banging	2 years 3 months	70-100 per minute	>36	Not reported	Average of 1 minute	>2 hours	Injury Insomnia
Happe et al. (2007) ³⁵	15	Male	Head-banging	'Early childhood'	Not reported	1	1 minute	1 minute	Sleep-onset	Not reported
Hashizume et al. (2002) ³¹	15	Male	Body-rolling Head-rolling Head-banging	Not reported	Not reported	Not reported	Not reported	'Few minutes'	During sleep	Risk of injury
Jeannet et	10	Female	Head-	'Infancy'	0.5-1 per	'Several'	15 minutes	Not reported	During	Hirayama

al. (2005) ³⁶			rocking 'Violent neck flexions'		second				sleep	disease ^b
Kaneda et al. (2000) ¹⁵	12	Male	Body-rolling	4	1 per second	4-6	20-30 seconds	Not reported	During sleep	Not reported
Kohyama et al. (2002) ⁴²	13	Female	Head-banging	1	1.6 per second	49	2-93 seconds	Not reported	During sleep	Not reported
Kohyama et al. (2002) ⁴²	9	Male	Head-rolling	Since 27 days old	1 per second	38	2-92 seconds	Not reported	During sleep	Not reported
Lee (2013) ⁸	17	Male	Head-punching	5	2Hz	Not reported	5 seconds - 10 minutes	Not reported	During sleep	Injury
Lindsay et al. (1982) ³⁹	13	Male	Head-rolling Body-rolling	18 months	1 per second	4-5	Not reported	Not reported	Both	Parents concerned about effect on growth Social implications
Manni et al. (2004) ⁹	9	Male	Head-banging Body-rocking Body-rolling	0.5	Not reported	10 ^d	5.1-20.9 minutes	Not reported	During sleep	Injury Sleep disturbance Impaired daytime functioning Morning fatigue
Martin and Conway (1976) ⁵¹	2	Female	Body-rocking	Not reported	Not reported	Not reported	Not reported	Not reported	Not reported	Affecting family's sleep
Mattewal et al. (2010) ³⁸	8	Male	Head-banging Body-rocking	Not reported	1 per second	Not reported	Not reported	Not reported	Both	Daytime sleepiness
Ross et al. (1971) ³²	16	Female	Head-banging	Several years previously	1-13 headbangs per episode	1-3	1-15 seconds	Not reported	During sleep	Sleep disturbance
Strauss et al. (1983) ¹⁰	7	Female	Head-banging	3	Not reported	8-32 (average 18)	Not reported	Not reported	During sleep	Injury
Su et al. (2009) ³⁴	15	Male	Body-rocking Head-	3	0.5-2Hz	78	1.1-27.4 minutes ^e	82.7	Both	Sleep disturbance Daytime

			banging Leg-rolling Arm- rocking 'Rhythmic feet movements ' 'Rhythmic hand movements '							sleepiness
Walsh et al. (1981)³⁷	8	Female	Body-rocking	7 months	1 per second	6	Up to 23 minutes	25-39 minutes	During sleep	Social implications
Yeh et al. (2012)³³	13	Male	Head-punching	3	0.6-1 per second	4	14-47 seconds	3.69 minutes	During sleep	Injury risk Affecting parents' sleep
Yeh et al. (2012)³³	11	Male	Head-punching	5	1.6 per second	1	5 seconds	5 seconds [†]	During sleep	Injury risk Affecting parents' sleep

^a body rocking over a soft toy positioned under groin

^b authors speculate that Hirayma disease may have been caused by the repeated neck flexions performed each night

^c following accident resulting in head injury and coma

^d pre-treatment figures reported

^e see research paper for full breakdown according to rhythmic movement performed

^f based on only one night of PSG which recorded only one episode - parents reported 'several' episodes per night

Rhythmic movement disorder: Diagnostic criteria

Criteria A-D must be met for diagnosis

A) Patient displays repetitive, stereotyped and rhythmic movements involving large muscle groups

B) Movements are predominately related to sleep

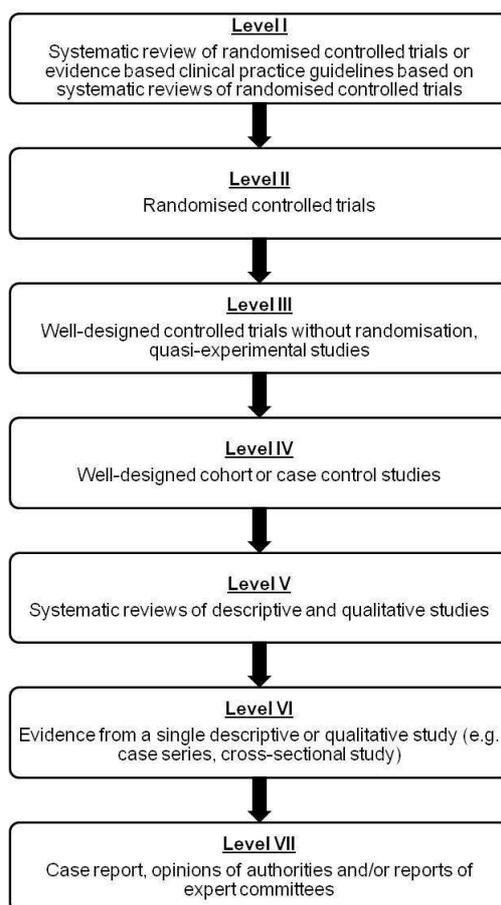
C) The repetitive movements results in a significant complaint. This may involve at least one of the following:

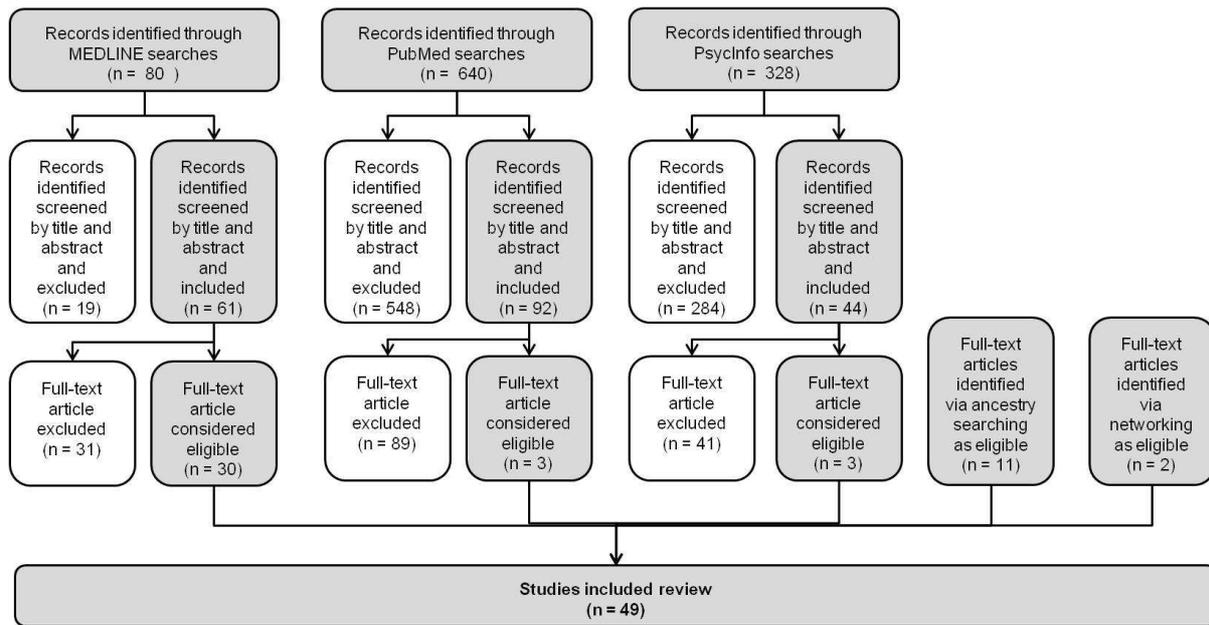
1) Normal sleep interference

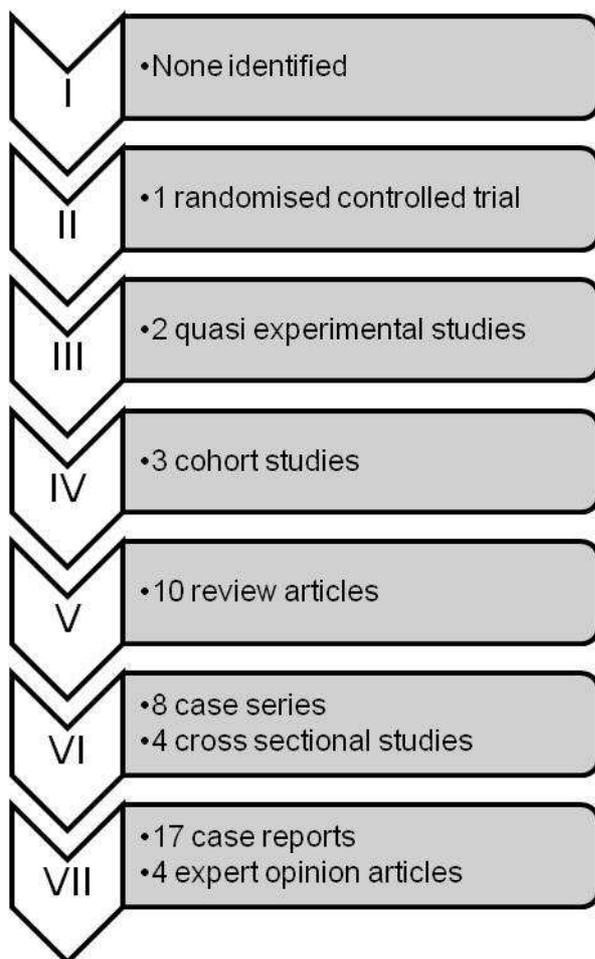
2) Significant impairment of daytime functioning

3) Actual or potential self-inflicted body injury

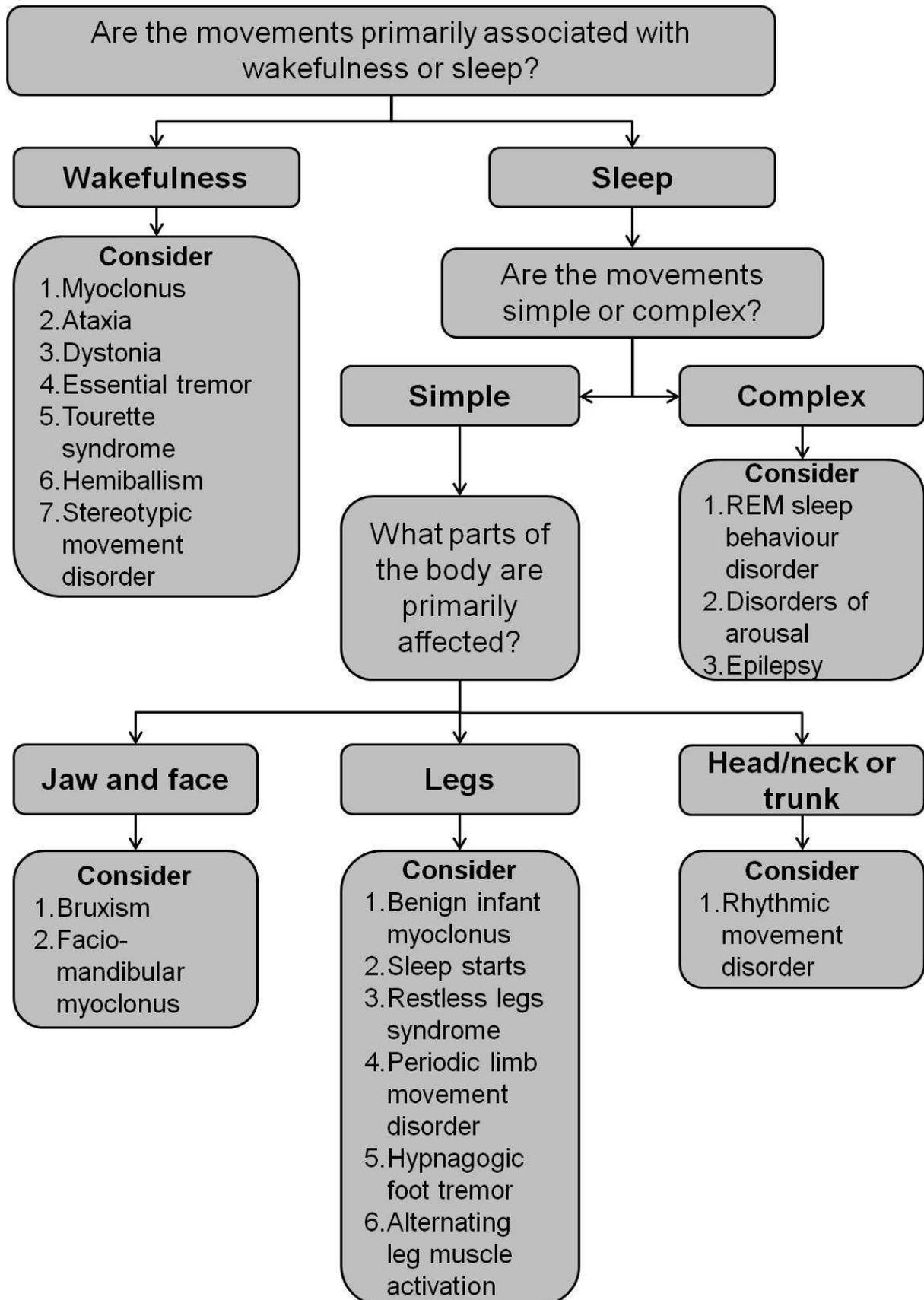
D) The movements are not explained by epilepsy or another movement disorder (diagnosis of elimination)







Movement Disorders



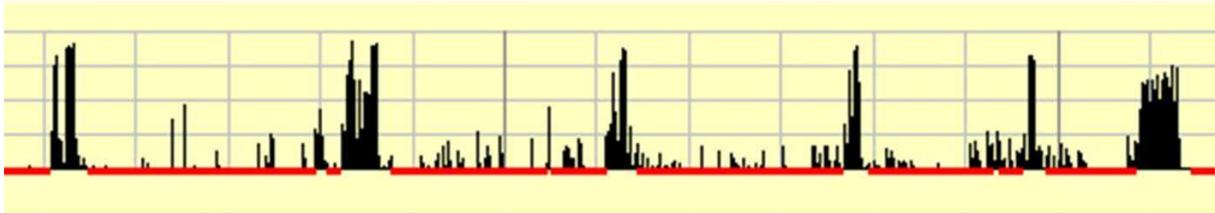
Polysomnographic assessment of rhythmic movement disorder**Set-up:**

- For monitoring RMD, bipolar surface electrodes should be placed to record electrical activity of the large muscle groups involved, in addition to standard leg and chin EMG electrodes. This is likely to involve placement of electrodes on the neck paraspinal muscles.
- Time-synchronized video PSG is necessary to accurately characterize the disorder, in addition to polysomnographic criteria.

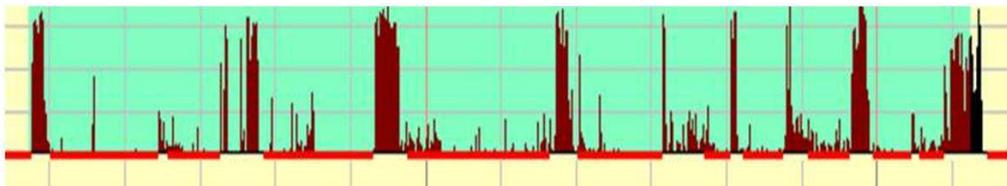
Scoring criteria:

- a) The minimum frequency for scoring rhythmic movements is 0.5 Hz.
- b) The maximum frequency for scoring rhythmic movements is 2.0 Hz.
- c) The minimum number of individual movements required to make a cluster of rhythmic movements is 4 movements.
- d) The minimum amplitude of an individual rhythmic burst is 2 times the background EMG activity.

a) Actigraphy in a toddler where simultaneous videosomnography did NOT confirm clinical history of rhythmic movements



b) Actigraphy in a toddler where simultaneous videosomnography confirmed clinical history of rhythmic movement disorder



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