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Objectively confirmed prevalence of sleep-related rhythmic movement disorder in pre-school children

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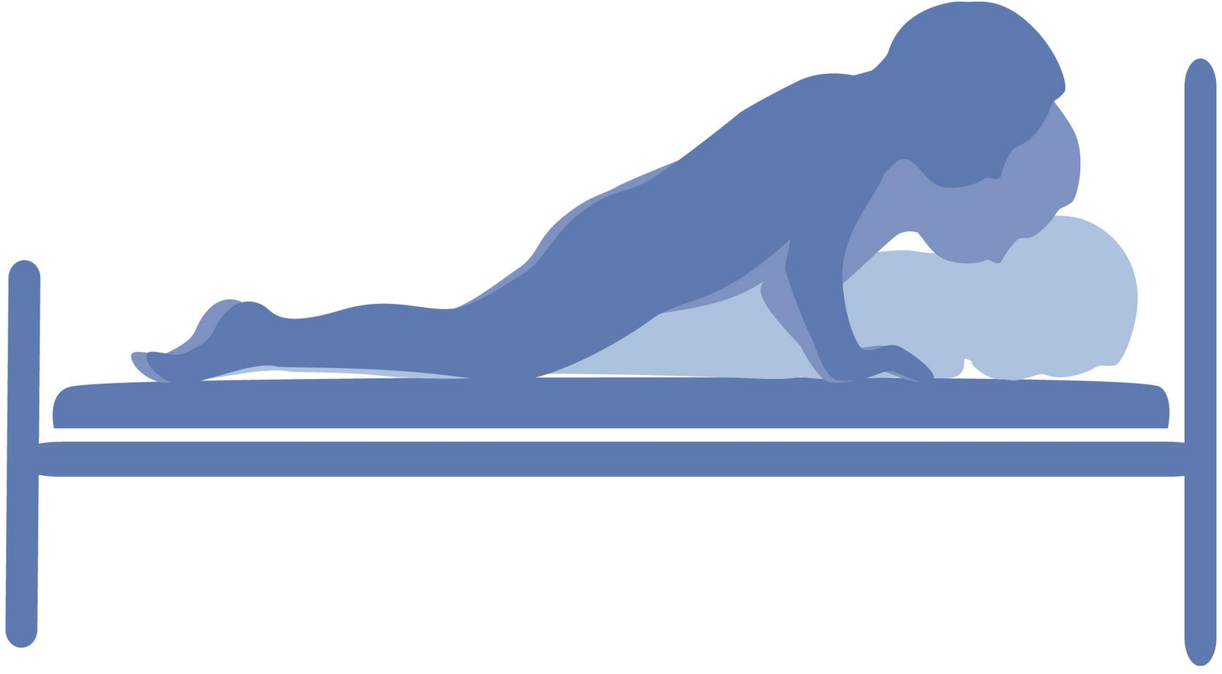
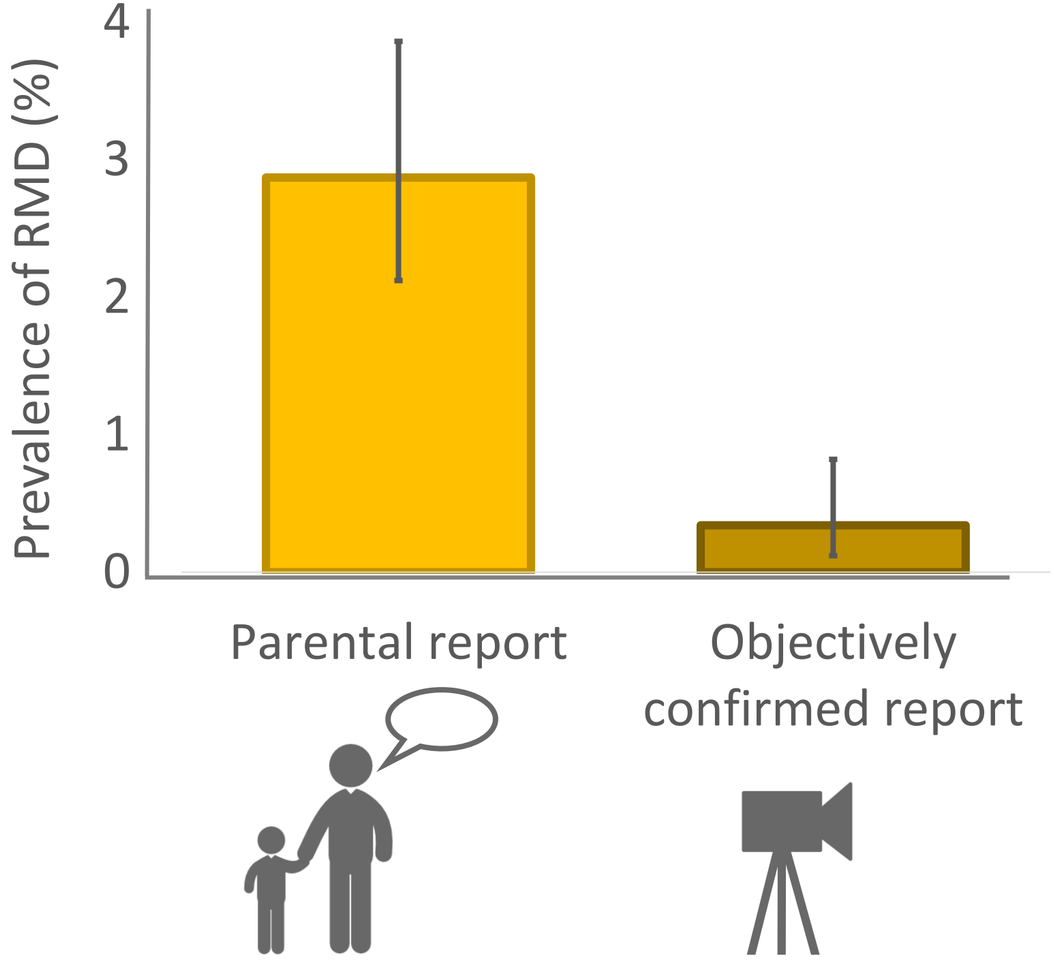
Prevalence in 1464 children recruited at one- and two-year health check





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Abstract

Objective

Childhood sleep-related rhythmic movement disorder (RMD) – sleep-related repetitive movements involving large muscle groups -can impair sleep quality, cause local injury and disturb household members. Previous parental reports indicate prevalence rates in children under 3 years of age between 5.5 and 67%. We studied the prevalence of RMD with objective home videosomnography.

Methods

Parents of 707 children having their one-year routine health check (357 male), 740 children having their two-year health check (395 male), and 17 children of unknown age (9 male), were asked if their child showed sleep-related rhythmic movements. If telephone interview confirmed likely RMD, parents completed a standardised clinical questionnaire and three nights of home videosomnography.

Results

At the one-year health check, 31/707 possible cases of RMD were identified (maximal prevalence: 4.38%; 95% CI [2.81, 5.89]) compared to 11/740 at the two-year check (maximal prevalence: 1.49%, 95% CI [0.61, 2.36]). Of 42 possible cases, 9 had resolved; 14 were uncontactable, or did not wish to participate, and 4 did not complete the study protocol. In four of ten remaining one-year olds and four of five remaining two-year olds parental report was objectively confirmed by videosomnography. Minimal prevalence based on objective observation was therefore 0.28% (95% CI [0.08, 1.30]) at one-year check and 0.41% (95% CI [0.08, 1.24]) at two-year check.

Conclusions

Prevalence of RMD in a large population of infants and toddlers was lower than previously reported (maximum prevalence 2.87%, minimum prevalence 0.34%). It is important to confirm parental report using objective measures.

Key words

Epidemiology

Rhythmic movement disorder

Jactatio capitis nocturna

Rhytmie du sommeil

Head banging

Videosomnography

List of Abbreviations

RMs: rhythmic movements

RMD: sleep-related rhythmic movement disorder

ICSD III: International Classification of Sleep Disorders III

Statement of Significance

To our knowledge, this is the first study assessing prevalence of rhythmic movement disorder using objective measures to confirm parental report of symptoms. In our sample of infants and toddlers from the South of England prevalence was lower than previously reported. Furthermore, parental report in this sample was sometimes unreliable, stressing the importance of objective data to improve diagnostic accuracy.

1. Introduction

Sleep-related rhythmic movements (RMs) are repetitive, stereotypic, large muscle group movements that occur at a frequency of 0.5-2.0 Hz, usually prior to sleep onset and sometimes during sleep[1-6]. Onset is typically during the first year of life. Semiology of movements varies between children. Examples include: body rolling, body rocking, head rolling and, the striking of a body-part against a surface: head banging and limb banging[7] (Figure 1). Importantly, when RMs are accompanied by clinical consequences, such as local trauma and/or impaired sleep quality and daytime functioning, this leads to a diagnosis of sleep-related rhythmic movement disorder (RMD).

RMD remains one of the most poorly understood sleep disorders. It is speculated that RMD is exploited as a conditioned behaviour to induce sleep, both at the beginning of the night and after natural night waking[8]. However, this does not explain why some children perform the movements during all stages of sleep in polysomnographic recordings[2, 4, 9]. Secondly, it is assumed that the condition is common in the infant and spontaneously resolves in early childhood. This underpins popular treatment recommendations, which are largely based on reassurance[10-12].

The first step in understanding any condition is to determine its true prevalence in the population. Klackenberg (1970) studied parental account of rhythmic head or body movements in 212 typically-developing infants from Stockholm, Sweden[13] followed up to 5 years of age. A prevalence of 67% at 9 months; 12% by three years and 6% by 5 years of age was reported. In 2000 Laberge and colleagues collected retrospective parental report of body rocking in 843 children[14]. 15.3% of parents reported body rocking between the age of 3-10 years old, decreasing to 3.1% by the age of 11 and 13 years. In 2001 Nevéus and colleagues studied 1407 school aged children (6.2 to 10.9y) from Uppsala, Sweden[15]. Children were asked 'Do you rock or sway back and forth before falling asleep?', and 8.3% responded positively. Most recently in 2007 Petit and colleagues, studied 1058 children followed up from the age of 2.5 to 6 years old in Quebec, Canada[16]. They reported a significant drop in prevalence of 'body-rocking and head-banging' from 5.5% at 2.5 years to 2.0% at 6 years.

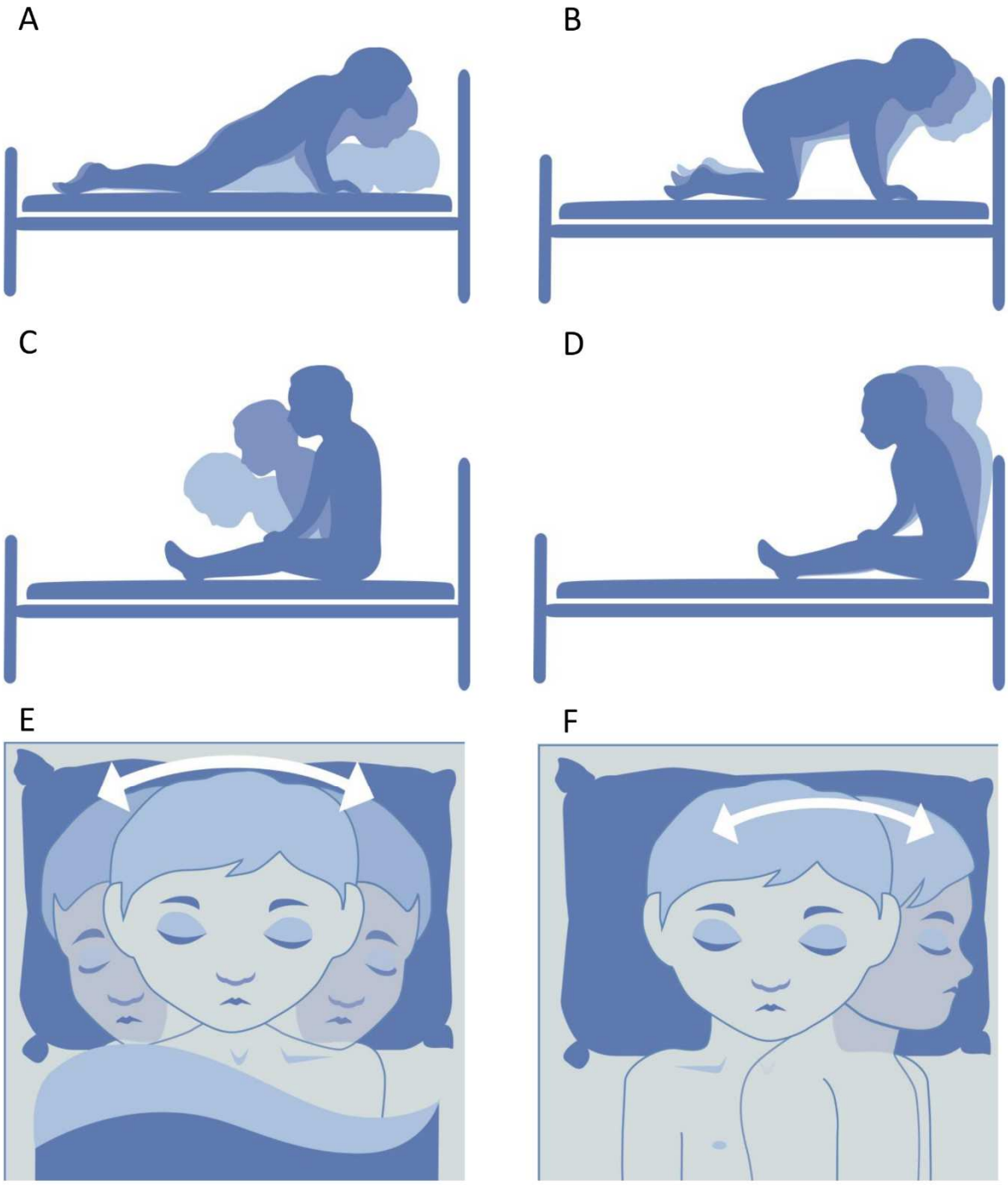

Figure 1. Different forms of rhythmic movements. A. Upper body movement that occurs with and without head banging on pillow or mattress. B. Full body movement that occurs with and without head banging on headboard or wall. C/D. Body rocking and banging E. Head rolling F. Body rolling. All movements may be accompanied by rhythmic vocalisations. E/F can include striking body part (e.g. head/limb) against hard surface.

All four studies relied on a single questionnaire item response from the child or parent. Contemporary non-invasive sleep monitoring technology, such as videosomnography (video recording of the sleep period), allows objective confirmation of RMD in the child's natural sleep environment. The International Classification of Sleep Disorders (ICSD III) has updated the diagnostic criteria for RMD since the publication of these prevalence studies with the criteria that these repetitive sleep-related movements should result in significant complaint[17] defined as an interference with normal sleep, significant impairment of daytime function, self-inflicted body injury or a combination of the previous. In this study we adopted the ICSD III criteria and investigated the prevalence of RMD using objective measures to confirm parental report.

2. Materials and Methods

2.1 Recruitment procedure

Children were recruited from Solent National Health Service Trust, in the coastal city of Portsmouth, United Kingdom via health visitor child health checks offered to all children registered with local general practitioners. The health checks are during infancy (around 1st birthday) and toddlerhood (around 2nd birthday). Parents were asked: 'Does your child usually show repetitive actions such as body rocking or head banging, while falling asleep, or during the night?'. This question was designed to capture the typical movement pattern described in the ICSD III in lay terms. Health visitors received training on RMD prior to data collection. Irrespective of response, child gender, age and the first part of the postcode of the home address were collected for all respondents. Parents who responded positively to the question were provided with further information about the study. The study was approved by the UK National Research Ethics committee (14/SW/1169). Parents signed a consent form on behalf of their child.

2.2 Telephone interview

Interested families were contacted by telephone by TC or EG and inclusion criteria were confirmed, namely that an adult in the family could speak English and that a more detailed description of the nature, timing and onset of their child's movements was suggestive of the condition. Where symptoms resembled RMs or RMD, families were invited to participate in a home visit, a standardised clinical questionnaire and videosomnography.

2.3 Standardised clinical questionnaire

A parent-completed 36 item questionnaire was developed to explore age of onset, detailed movement semiology, timing and duration of movements, as well as clinical consequences for the child and household members. The researcher was available to support parents with questionnaire completion.

Cognitive interviews were conducted with two parents of pre-school children to check the questionnaire was understandable prior to the study. "Thinking aloud" techniques were used where parents read the questionnaire items aloud and commented as they answered each question, so guiding the final phrasing of questions.

Parental education levels were translated to International Standard Classification of Education values for report[18].

2.4 Videosomnography

While attended polysomnography is the traditional approach to confirmatory diagnosis, clinical experience suggests that many children suppress movements in the laboratory setting. To capture sleep behaviour in an ecologically valid setting we used home videosomnography in the child's bedroom. Subjects were monitored from sleep onset to morning waking during three consecutive nights solely using an infrared camera (Wansview NCM624W H.264 Mega Pixel Indoor Wireless WIFI IP Camera). Video data was downloaded in Windows Media Player and visually inspected by the

researchers. Where movements were suspected to fulfil RMD criteria this was confirmed by a certified somnologist (CMH) using ICSD III criteria.

2.5 Prevalence analysis

Prevalence levels of both RMs and RMD were determined at three thresholds

- Maximal prevalence was calculated as all potential cases, defined as any child where the family initially reported YES to the screening questionnaire unless the symptom description during the telephone interview excluded RMs (e.g. other sleep disorder), divided by all families who were asked the screening question during a health check.

$$Prevalence_{max} = \frac{n_{potential\ cases}}{n_{total}} \quad (1)$$

- Minimum prevalence was calculated as only cases where RMs were confirmed by videosomnography, divided by all families who were asked the screening question during a health check.

$$Prevalence_{min} = \frac{n_{objectively\ confirmed}}{n_{total}} \quad (2)$$

- Likely prevalence was calculated as potential cases, i.e. screening questionnaire positively answered, and symptom described during the telephone interview did not excluded RMs, multiplied by the proportion of parentally reported cases that were confirmed by videosomnography. This accounted for attrition due to families not contactable or unwilling to have their child studied at home.

$$Prevalence_{likely} = \frac{n_{potential\ cases} * \frac{n_{objectively\ confirmed}}{n_{potential\ cases}}}{n_{total}} \quad (3)$$

2.6 Statistical analysis

The maximal, minimal and likely prevalence of RMs and RMD in children having their one-year check and two-year check were compared using chi-square for independence or where appropriate Fisher's exact tests. 95% confidence intervals were calculated according to Agresti-Coull's calculation methods[19]. Statistical analysis was performed using IBM SPSS version 24.

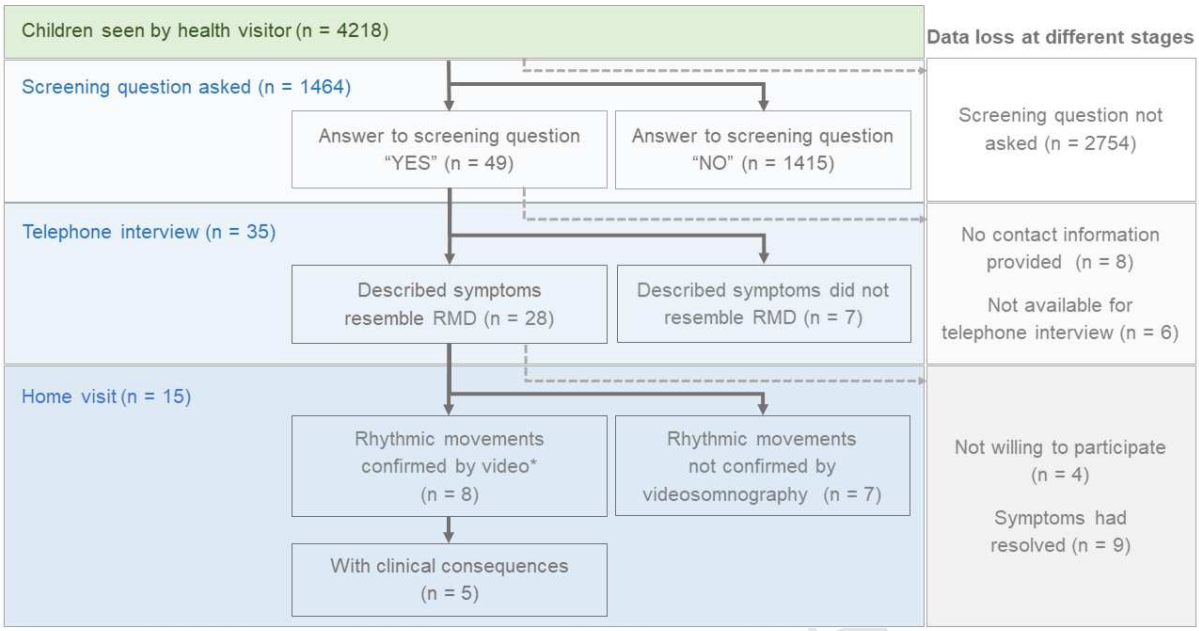

Figure 2. Consort diagram. The population of interest contained 4218 children living in Portsmouth, United Kingdom, who were seen by a health visitor for a routine health check. During the health check 1464 parents were asked a screening question. Parents answering yes were given participant information and were invited to provide contact details. Researchers contacted the parents who provided contact information and asked the same standardised question, allowing parents to describe the symptoms. If described symptoms resembled symptoms of RMD a researcher visited the family at home and parents completed a standardised clinical questionnaire and three days of home videosomnography. Drop out for each stage is described on the right. *No three-day videosomnography available, but symptoms confirmed based on video captured by parents.

3. Results

3.1 Study sample

Home visits were conducted between February 2015 and March 2016. In this period, 4218 children were seen by health visitors (Figure 2). Due to incomplete compliance of health visitors with the study protocol, 1464 families were asked the standardised question (34.7%). These 1464 children are the study sample. Data from 707 children were collected at the one-year check (age: M = 10.76 mo, SD = 1.33 mo; 357 males) and data from 740 at the two-year check (Age: M = 25.39 mo, SD = 1.96 mo; 395 males). For 17 children age was not recorded, nor was it known if the child was recruited during a one- or two-year check.

The distribution of the sample over the six postcode regions of Portsmouth was similar to the distribution of the city's population as reported by the Hampshire County Council [20].

3.2 Reliability of parental report

Characteristic rhythmic movements were observed in the videosomnography data in four of ten studied one-year olds and four of five studied two-year olds (Table 1). Thus, reliability of parental report in the overall sample was 53.3%.

Table 1. Overview of individual cases. International Classification of Education Levels (ISCED) goes from 0=pre-primary education to maximally 8=doctoral or equivalent, with 4 being the first level of post-secondary education. *No three-day videosomnography available, but characteristic rhythmic movements confirmed based on smaller amount of video data.

ID	*Gender*	*Age in months*	*Movement types parental report*	*Confirmed by video*	*Clinical consequences*	ISCED Parent 1	ISCED Parent 2
1	male	24	Head banging, body rocking	Yes	Affects sleep and daytime functioning of child and others	4	n. a.
2	male	25	Head banging and rolling, body rocking and rolling, limb banging	Yes	None for child, but affects sleep and daytime functioning of others	3	3
3	male	13	Body rocking	Yes	Affects sleep of child	7	6
4	female	25	Head banging, body rocking	Yes	Injury	6	5
5	male	24	Head banging and rolling, limb banging	Yes*	Minor injury	5	4
6	male	13	Body rocking	Yes	Minor injury	6	6
7	female	10	Body rolling, head rolling, limb banging	Yes	None	7	6
8	female	12	Head rolling, limb banging	Yes*	None	5	3
9	female	14	Head banging and rolling, limb banging	No	Affects daytime functioning of child and sleep of others	6	3
10	male	16	Head banging and rolling, limb banging	No	Affects sleep and daytime functioning of child and others	3	n. a.
11	male	12	Body rocking, limb banging	No	Affects sleep of child	6	6
12	female	12	Head banging and rolling, body rocking and rolling, limb banging	No	None for child, but affects sleep of others	6	6
13	male	11	Head banging	No	Minor injury, affects sleep and daytime functioning of others	5	5
14	female	10	Body rocking, head rolling	No	None	6	6
15	male	27	Body rocking, other	No	None	3	7

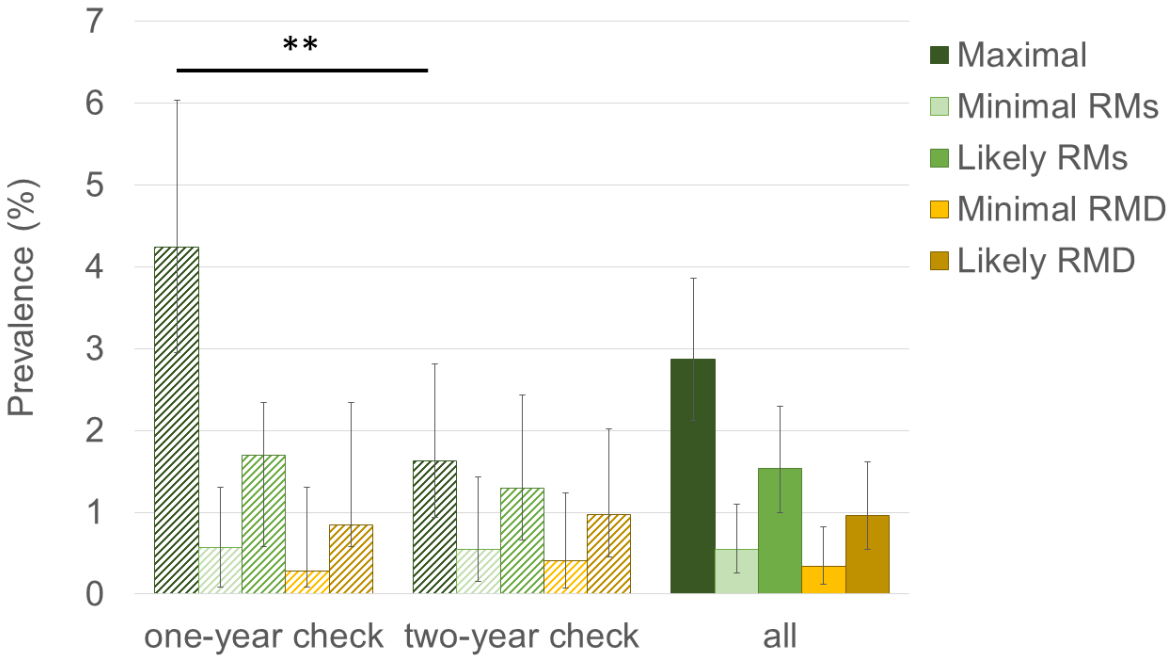

Figure 3. Estimated prevalence of sleep-related rhythmic movements (RMs) and sleep-related rhythmic movement disorder (RMD) at one-year and two-year health checks, as well as for both age groups combined. Maximal estimate relates to both RMs and RMD, since estimate is solely based on response to a single question that did not mention clinical consequences. Error bars indicate 95% confidence intervals. **indicated p=0.001

3.3 Prevalence of rhythmic movements

During the health check, 49 families gave a positive reply to the screening question. A possible RMD diagnosis was excluded in seven cases during the telephone interview. False positives included sleep apnea, benign myoclonic jerks and sleep terrors. The remaining 42 possible subjects, including nine resolved cases, were considered potential cases of RMD.

In this sample the maximal estimated prevalence of RMD based on parental report alone was significantly higher in children at the one-year health check (4.38%, 95% CI [3.09, 6.18]) than in children at the two-year health check (1.49%, 95% CI [0.80, 2.68]), (χ^2=10.78, p=0.001), (Figure 3). However, the minimal prevalence did not differ between age groups, both when looking at RMs (one-year: 0.57%, 95% CI [0.08, 1.30]; two-year: 0.54%, 95% CI [0.16, 1.43]) and when looking at RMD (one-year: 0.28%, 95% CI [0.08, 1.30]; two-year: 0.42%, 95% CI [0.08, 1.24]).

3.4 Male female ratio

The male to female ratio was 5:3 in cases of confirmed RMs, 4:3 in cases that were not confirmed using videosomnography and 5:4 in cases where the symptoms described during the telephone interview resembled those of RMD, but had resolved at the time of the interview (Table 1).

4. Discussion

To our knowledge, this is the first prevalence study of sleep-related rhythmic movement disorder in infants and toddlers using both parental report and confirmatory home videosomnography. Across our sample the maximal prevalence, based on parental report, was 2.87% with a likely prevalence of 0.96%. This is lower than the 5.5% prevalence Petit and colleagues reported in their sample of 2.5-year-olds, and significantly lower than the 67% prevalence in nine-month-olds and 12% in three-year-olds reported by Klackenberg.[13, 16]. In our study only four in five cases identified using a screening question were confirmed at telephone interview suggesting that parents often misunderstand simple screening questions. Previous studies relying on parental report are therefore likely to have significantly over-estimated prevalence. However, a sensitive screening questionnaire is important to avoid missing potential cases. In our study the likelihood of false negative responses was low as health visitors who administered the screening question received training to recognise sleep-related rhythmic movements. Therefore, it is reasonable to assume that the prevalence of RMD is genuinely lower than previously supposed.

A common assumption is that RMD is disruptive during early childhood, but spontaneously resolves with age[21]. Based on this assumption, reassurance and safety advice is often recommended[10-12]. Our sample included nine cases where historic symptoms resembled RMD, but were no longer present at the time of the interview. Furthermore, the likely prevalence of RMs was slightly, albeit not significantly, lower at the two-year check than at the one-year check (1.30% and 1.70% respectively). This is in line with previous longitudinal studies that reported a decrease in prevalence with age[13, 16]. However, the likely prevalence of RMD in our sample did not differ between the one-year check and

two-year check groups (0.85% and 0.97% respectively). This suggests that although benign rhythmic movements might disappear with age, RMD might not. Importantly, rhythmic movement disorder may persist with age, the oldest reported case in literature being 67 years[9]. There is a pressing need for research into the natural history of the condition using accurate case definition, rather than unreliable parental report, to determine the frequency of spontaneous resolution of RMD and hence guide treatment approaches.

Previous research data by Sallustro suggest a male:female ratio of 3:1 [22]. Others report no such trend[13, 16]. The male female ratio in the resolved cases and those studied with videosomnography in our study was 14:10. This supports our clinical experience (current clinic caseload 25 boys:13 girls). Interestingly, of the six cases in this study where rhythmic movements had no clinical consequences, four were female. Although numbers are small it could be that males experience more severe clinical consequences of RMD and their parents are more likely to seek help.

There is conflicting data on the influence of socio-economic status and ethnicity on the prevalence of sleep disorders in children[23]. Parents who participated in the home visit in this study were of mixed educational levels and were of Caucasian descent. Based on the national Income Deprivation Affecting Children Scale, Portsmouth is socioeconomically diverse with a slight tendency towards deprivation, with 33/125 layer super output areas in the UK's bottom 20%[24]. If deprivation contributes to the development of RMD then our data may have overestimated the national prevalence.

Clinical history is the bedrock of any diagnosis and parental report is relied on heavily in paediatric medicine. However, parental report may be unreliable for sleep disorders where symptoms may not be observed. In this study videosomnography was only undertaken where parents gave a convincing history of RMD movements in the standardised RMD questionnaire. Surprisingly, parental report was not confirmed by videosomnography for 47% of children. It is possible that 3 nights of recording failed to capture the child's typical movements. However, this does not fit with our current understanding of RMD which presumes that episodes occur every night or almost every night[3, 4, 25-27]. Present diagnostic criteria do not stipulate the number of nights per week required to qualify for a

RMD diagnosis but the requirement for clinical consequences implies frequent disruption. Future studies should include longer periods of videosomnography to clarify this point.

Two children with rhythmic movements on videosomnography were reported by parents to have no clinical consequences. For a condition that occurs every night it may be difficult for parents to discern subtle differences in their child's daytime behaviours or function. Future work should study associations between the severity of this disorder (for example percentage of time in bed disrupted by rhythmic movements) and its consequences.

Only 35% of the children attending health checks were screened for RMD. It is unclear why screening rates were so low; though high workload, primary focus on clinical care rather than research and changes in head office location are likely explanations. The fact that our data show lower prevalence rates than previous publications suggest no inherent sampling bias towards positive cases. Furthermore, this study represents the largest published sample of RMD prevalence in pre-school children, reducing the chance of sampling bias.

5. Conclusion

Limitations of existing research, specifically failure to objectively substantiate parental reports and the use of outdated diagnostic criteria, underpinned the need for a new prevalence study. This study estimates prevalence of rhythmic movement disorder across the sample to be within the range of 0.3 to 2.9%, with a likely prevalence of 1.0%. Our findings support and do not contradict the idea that prevalence is higher in males. While prospective studies are needed to confirm the previously reported drop in prevalence with age, our data suggest this may not be the case. Furthermore, an important finding in this study is that parental report of symptoms of RMD can be unreliable and that objective confirmation of diagnosis is therefore important.

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Contributors' Statement

Dr Gogo, assisted in the project design, collected data, carried out data analysis, drafting of the initial manuscript, reviewed the final manuscript as submitted
Ms van Sluijs carried out data analysis and wrote the final manuscript as submitted
Dr Chueng designed and prepared the videosomnography measurement setup, contributed to data collection and approved the final manuscript as submitted
Ms Gaskell collected data and approved the final manuscript as submitted
Dr Jones collected data and approved the final manuscript as submitted
Dr Alwan critically reviewed the methods and approved the final manuscript as submitted
Dr Hill conceptualized and designed the study, contributed to data collection and analysis, critically reviewed and approved the final manuscript as submitted.

Competing Interests Statement

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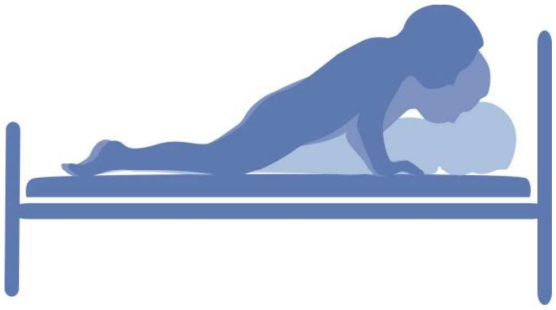

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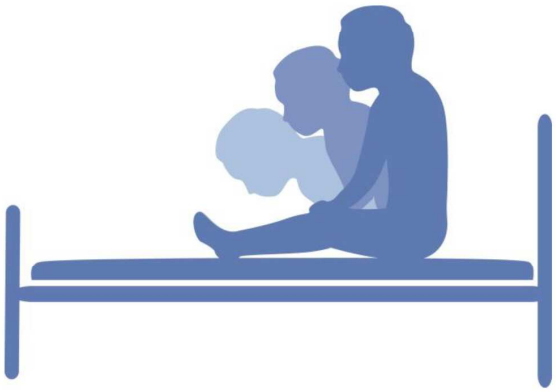

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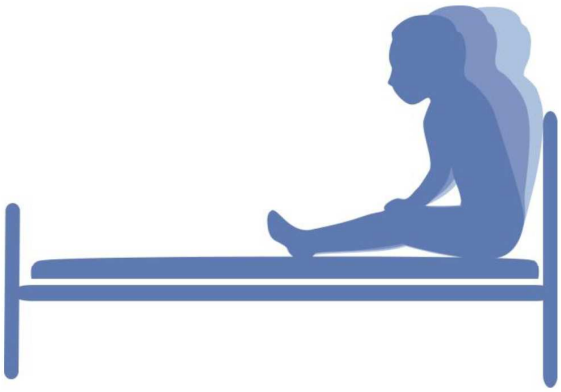

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Highlights

- Sleep-related rhythmic movements may have severe clinical consequences.
- Prevalence was assessed in a large cohort (*N*=1464) of infants and toddlers.
- Parental report of symptoms could be objectively confirmed in 53.3% of cases.
- The prevalence in our sample is much lower (maximal 2.87%) than previously reported.