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Sensory stimulation in the treatment of children with sleep-related rhythmic movement disorder: a feasibility and acceptability study

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Abstract

Background: Sleep-related rhythmic movement disorder is characterized by repetitive gross-motor movements at sleep onset or during sleep, which result in clinical consequences such as impact on daytime functioning and injury. No well-established therapies exist today. Substituting the patient's movements with external sensory stimulation may offer a treatment modality. The aim of the current study was to test the feasibility and acceptability of vestibular stimulation using a rocking bed (Somnomat) in children with rhythmic movement disorder and to assess children's movement preference.

Methods: Children with rhythmic movement disorder ($n = 6$, Age: 5–14 years) were studied over three nights in a sleep laboratory: adaptation night (normal bed) and randomised-order baseline (Somnomat) and intervention nights (Somnomat). Child's preferred movement direction (head-to-toe or side-to-side) and frequency (between 0.25 and 2 Hz), determined during an afternoon protocol, were applied using the Somnomat for 1 h after lights out, and in response to subsequent episodes of rhythmic movement during intervention nights. Comfort assessed using a questionnaire, and objective sleep parameters assessed using videosomnography, were compared.

Results: The participants' sometimes violent rhythmic movements did not disturb device performance. All children rated intervention nights equally or more comfortable than baseline nights. Self-reported sleep quality, as well as the number and duration of movement episodes did not significantly differ between baseline and intervention nights.

Conclusions: Providing rocking movements using the Somnomat is both technically feasible and acceptable to the target population. The therapeutic value of this novel stimulus substitution for rhythmic movement disorder should now be evaluated in a larger sample over a longer period in the home setting.

Trial registration: The trial was retrospectively registered at clinicaltrials.gov (NCT03528096) on May 17th 2018.

Keywords: Pediatrics, Jactatio capitis nocturna, Rythmie du sommeil, Head banging, Sensory substitution, Vestibular stimulation

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Background

Sleep-related rhythmic movement disorder (RMD) is a childhood-onset sleep disorder characterized by gross motor movements occurring at sleep onset and during sleep (Manni and Terzaghi 2005) (Supplemental Information Documents 1 and 2). These nocturnal episodes of rhythmic movement may disrupt the sleep of both the sufferer and other household members with impact on daytime functioning, and can result in injury (Sateia 2014). Even though clinical consequences of sleep-related RMD can be severe, there are no well-established therapies (Gwyther et al. 2017). While psychological, behavioural and pharmacological treatments have been reported in case studies and small observational studies (Gwyther et al. 2017), the most consistent treatment recommendations for children are based on reassurance and safety advice to reduce injury risk (Mahowald 1995; Mindell and Owens 2015; Shneerson 2009) with no quality treatment trial data to support clinical management.

Understanding what causes RMD is limited to hypotheses (Manni and Terzaghi 2005). As rhythmic sensory stimulation is known to be soothing and relaxing, (Vrugt and Pederson 1973; Grabherr et al. 2015; Omlin et al. 2016) and promotes sleep in infants and young adults, (Barnard and Bee 1983; Bayer et al. 2011; Shibagaki et al. 2017) it has been suggested that RMD might be a learned soothing, (Haywood and Hill 2012) or self-stimulating, behaviour that becomes habitual and compulsive (Clark and Chee 1977; Sallustro and Atwell 1978). At the same time, rhythmic movements might also promote early motor development (Clark and Chee 1977; Sallustro and Atwell 1978). Sallustro and colleagues found that children reported to perform daytime rhythmic movements reached motor milestones earlier, compared to peers who did not (Sallustro and Atwell 1978). Furthermore, a controlled experiment showed that infants who received experimental vestibular stimulation in a rotating chair reached motor milestones earlier than a control group (Clark and Chee 1977). While daytime rhythmic movements may have developmental advantages, when rhythmic movements disrupt sleep and result in clinical consequences this is considered a sleep disorder. Common consequences such as local injury and daytime cognitive and behavioural impairments could be minimised if the number, duration or intensity of rhythmic movements were reduced. Evaluation of therapeutic interventions for this poorly researched sleep disorder are overdue.

Sensory systems stimulated by the patients' rhythmic movements include the visual, proprioceptive, vestibular and, in the case of head banging, the auditory system (Golbin et al. 2013). Thus, one therapeutic approach could be to substitutes the sensory experience of rocking with an external source of rhythmic movement, which may down-regulate the internal drive to rock. We previously reported the benefits of vestibular stimulation, in the form of a swinging motion passively generated when sleeping in a hammock, in five children with RMD treated in the Southampton Children's Hospital sleep clinic (Hewertson and Hill 2018). Although promising, the passive movement of a hammock cannot be reliably reproduced. An automated bed that generates rocking movements offers the advantage that stimulation intensity can be controlled and manipulated (Crivelli et al. 2016). Furthermore, stimulation can be deployed at sleep onset (aiming to prevent episodes of rhythmic movement) and can be applied during the night in response to rhythmic movements.

We have developed and tested the rocking Somnomat bed in adults, but have not tested its technical capabilities with participants who may move violently in the device. The present study explored the stimulation settings that are perceived as most comfortable and likely to promote sleep by children with RMD. Furthermore, the feasibility and acceptability of rocking movements provided on demand during the night were assessed.

Methods

Recruitment

Six typically developing children with RMD (2 female) aged 5 to 14 years (Median: 8.5 years, IQR: 5.25 to 12.5 years) were included in the study (Table 1). Participants were recruited from a social media group of affected families in the UK. Eligible participants were children between 5 and 18 years of age with self-reported RMD and no self-reported sensitivity to motion sickness (Henriques et al. 2014). Diagnosis according to the International Classification of Sleep Disorders III, was confirmed by a European certified somnologist (CMH) based on a structured interview (Supporting Information Document 3) and a home video recording (Sateia 2014; Gogo et al. 2018).

Rocking bed

Sensory stimulation was provided by the Somnomat, an actuated bed previously validated for use in healthy adults (Crivelli et al. 2016). In summary, the bed provides sinusoidal rocking movements along the trajectory of a pendulum with a centre of rotation 4 m above the bed. It moves in two directions: head-to-toe and side-to-side (Fig. 1). For children under 10 years of age only head-to-toe direction was used for safety reasons. Pressure sensitive floor mats initiated a safety stop to prevent participants from approaching the moving mechanism. Cushioning of the bed prevented contact injuries.

The control software was adapted so that the bed frequency could be varied between 0.25 and 2.0 Hz at a

Table 1 Demographics

Case	Sex	Age	Rhythmic movement semiology	Habitual movement frequency	Injury	Influence on daytime functioning	Influence on sleep	Onset Age	Medication prior to study
		years		Hz				months	
1	male	5	head rolling, body rolling	1.4	none	no	not sure	12	none
2	male	5	head banging, body rocking	1.2	callus, bleeding	yes	a little	9	none
3	female	6	body rocking, head rolling	0.6	bruising, bleeding, hair loss, neck pain	yes	very much	6	Melatonin
4	male	11	head banging, head rolling	2.3*	bruising, bleeding, hair loss	no	a little	6	none
5	female	13	body rocking, body rolling	0.9	hair loss	yes	a little	24	none
6	male	14	body rocking	0.7	none	yes	very much	24	none

Habitual movements were determined based on video recordings and in the one child showing no movements (*) by voluntary demonstration of symptoms during daytime.

resolution of 0.25 Hz. This range encompassed the full range of typical rhythmic movement frequencies (Sateia 2014). The trajectory amplitude was scaled to the frequency to achieve a maximum velocity of 0.1 m/s resulting in amplitudes between 0.008 and 0.67 m. As rhythmic movements can be forceful, the impact of these movements on the provided stimulation was of interest. Accuracy of the provided stimulation was assessed by calculating the root mean square error between the reference velocity and the measured velocity of the bed along its trajectory, during the first hour of the intervention for each participant.

Movement preference protocol

A strict protocol assessed participant's preference for Somnomat movement direction and intensity using a

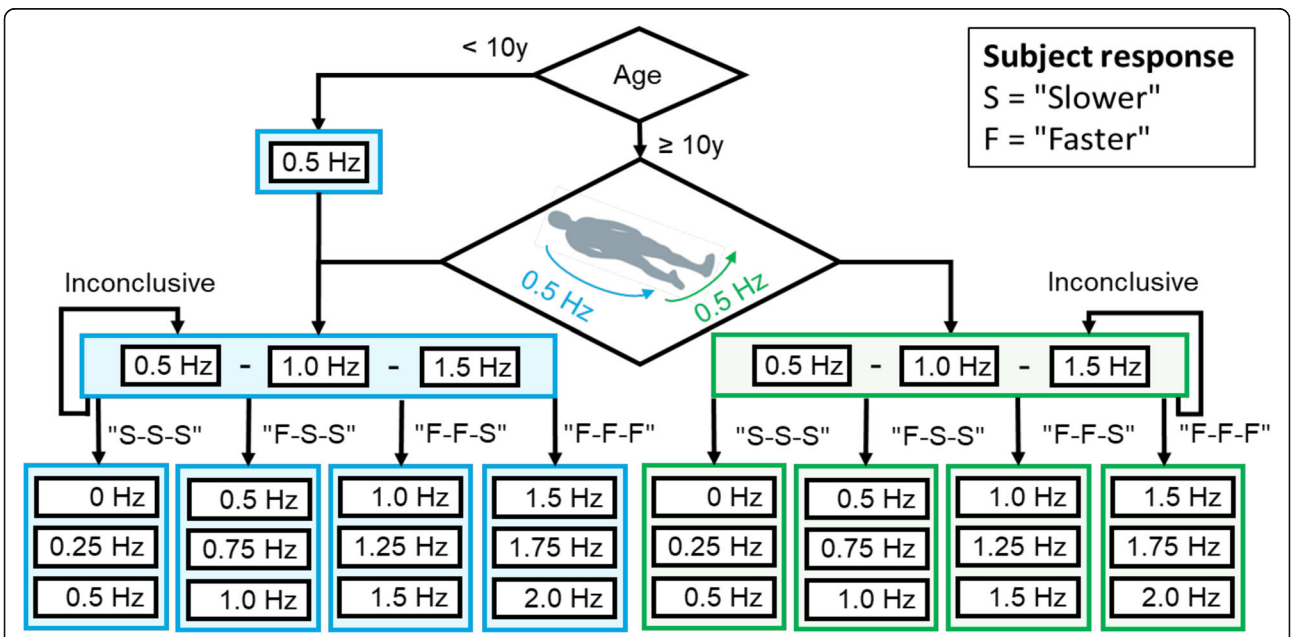

Fig. 1 Stimulation preference selection protocol flow chart. First participants of 10 years and older were presented two motion direction (side-to-side or head-to-toe) while the bed was moving with a frequency of 0.5 Hz, and were asked to choose a preferred direction. For safety reasons participants under 10 years tried only the head-to-toe direction. All participants were then presented three frequencies and were instructed to indicate whether for the bed to promote sleep it should move faster (F) or slower (S). These responses were taken into account to narrow down the stimulation frequency for a second round of three frequencies. All conditions were presented for 1 min. Amplitude and frequency were set to result in a maximum velocity of 0.1 m/s. Outcome of the stimulus preference protocol was a direction and frequency to be used during the intervention night

combination of settings, each for 1 min, with participants lying in their preferred sleeping position (Fig. 1). Participants rated comfort of the bed, and how sleepy they felt after each bout of stimulation using a custom-designed five-point visual rating scales (5 = maximum comfort and sleepiness) (Supporting Information Document 4). For children over 10 years, both movement directions (head-to-toe and side-to-side) were presented in a random order at a frequency of 0.5 Hz. For children under 10 years of age only head-to-toe was presented for safety reasons. Preferred movement direction was then used to assess preferred the preferred frequency. Three frequencies (0.5, 1.0 and 1.5 Hz) were presented in random order and participants indicated whether the bed should move faster or slower to promote sleep. Depending on their responses, further frequencies were presented in increments of 0.25 Hz to narrow down the preferred stimulation setting.

Night protocol

Following a sleep laboratory acclimatisation night (normal bed), participants spent the intervention and baseline night (order randomised) in the Somnomat. Rocking movements provided by the Somnomat (intervention night) or a sound recording of the Somnomat (baseline night) were activated for 1 h after lights out and again upon detection of RMs in a live video stream (Supporting Information Documents 1) until 10-min after RMs had stopped. After the hour of movement at sleep onset the amount of consecutive stimulation was restricted to 30 min per episode and a total of 3 h per night. During the baseline night, a recording of the sound of the moving bed was played to the participants to control for the influence of auditory stimulation generated by the bed (Crivelli et al. 2014). The same criteria were used for activating the audio as were used for activating the bed. Participants were instructed not to consume caffeine in the 5 h prior to the agreed bedtime.

Measurement and analysis

Participants wore an actigraphy watch on the non-dominant wrist, and parents completed a sleep diary, to assess whether sleep in the laboratory was representative of sleep at home during the week prior to the study (Basic Mini Motionlogger watch, Ambulatory Monitoring Inc., New York). The device was set to zero crossing mode and sleep duration (minutes) was calculated using the Sadeh algorithm validated for use with children (ActionW2 software) (Sadeh et al. 1994).

To check compliance with caffeine intake guideline, as well as sleepiness and comfort, questionnaires were filled out prior to lights off and upon awakening by the participant and their parent. In the sleep laboratory, we further recorded video-somnography in the form of whole night 2D video recordings, as well as 3D videos, which have been reported elsewhere (Gall et al. 2019).

Video recordings (2D) were visually analysed according to the following criteria: An episode of rhythmic movement was defined as three or more consecutive rhythmic movements. The end of an episode was defined as a pause longer than the duration of two movements. Movements were scored based on visual inspection by two researchers: once online during the experiment and once offline. Number and duration of episodes were calculated over the common length of baseline and intervention night of each participant.

The habitual movement frequency was determined by dividing the number of rhythmic movements of one episode by the duration of the episode. For one participant the habitual movement frequency was determined based on a voluntary demonstration of his rhythmic movements prior to his third evening in the laboratory, since no episodes were recorded at night.

Statistics

Descriptive statistics for all variables are reported as medians and interquartile ranges (IQR), due to the small sample size. Comfort and sleepiness ratings upon awakening, total time in bed, total duration of rhythmic movement, number of episodes and the average duration of episodes during the baseline and intervention night were compared numerically. Statistics was performed using IBM SPSS version 24.

Results

Stimulation preference

Participants chose frequencies both within their characteristic movement range (0.5, 1.0 and 2.0 Hz; n = 3), as well as below (0.25 Hz; n = 3). The median habitual movement frequency was 1.1 Hz (IQR: 0.8 to 1.3 Hz, Table 1) and this did not correlate with preferred Somnomat frequency (Fig. 2a). Median levels of reported comfort and sleepiness at the child's preferred bed frequency were 3.5 (IQR: 3 to 4.5) and 4 (IQR: 4 to 5) out of 5 respectively, with 5 indicating optimal comfort and maximal sleepiness (Fig. 2b).

Of the three participants that were older than 10 years and therefore eligible to choose a movement direction, two chose the side-to-side direction. The chosen Somnomat movement direction matched the habitual rhythmic movement direction for one participant, one chose a direction that was not in line with their habitual direction, and one participant had a habitual movement direction (head banging on pillow) not simulated by the Somnomat.

To assess reliability, the responses to two trials of the preference protocol during which the children were provided with the same direction and frequency were

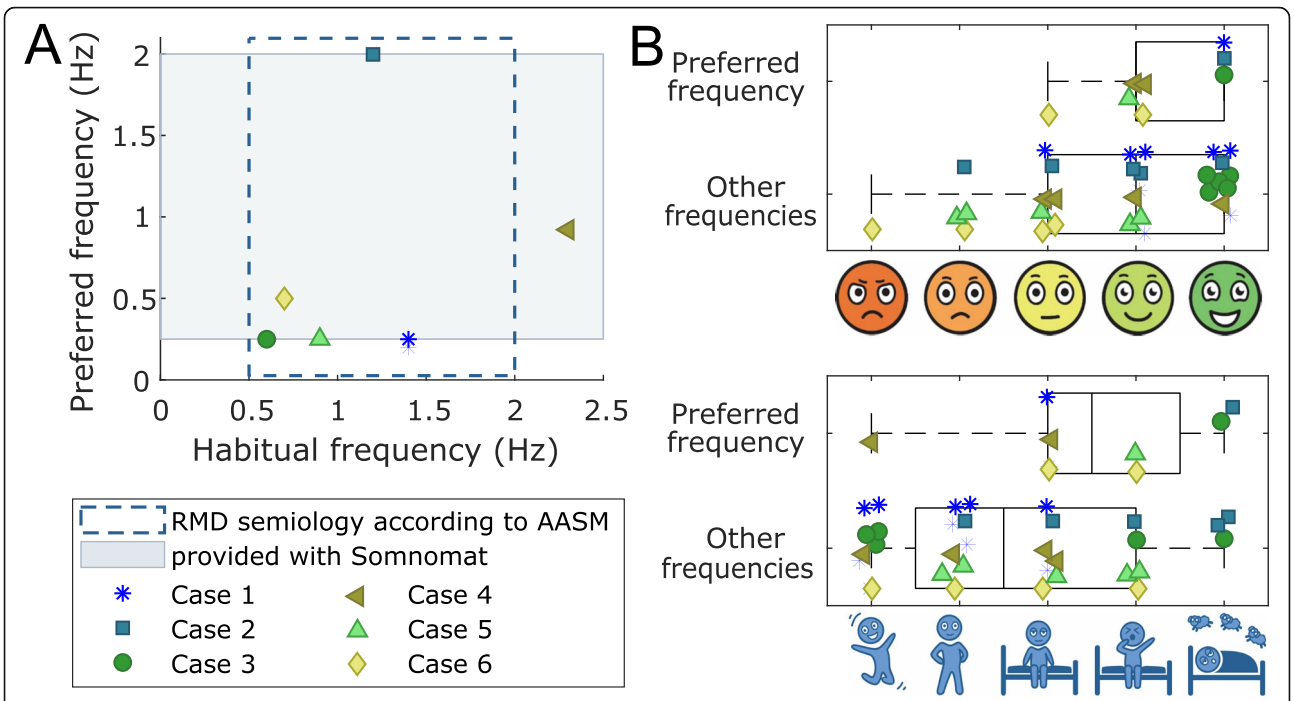

Fig. 2 Outcome of the movement preference protocol regarding frequency of the bed movement ($n = 6$). A. Habitual movement frequency while making rhythmic movements vs. the preferred bed frequency as determined during the preference protocol. Area indicated in blue shows the frequency range of rhythmic movement disorder (RMD) reported by the American Academy of Sleep Medicine (AASM). Area shaded grey shows the range of frequency provided using the Somnomat. B. Boxplots of comfort (top) and sleepiness (bottom) at the preferred bed frequency and all other frequencies combined

compared. Five out of six children responded consistently to the question 'For the bed to help you sleep should it move faster or slower?'. Comfort was rated more consistently ($Median_{difference}$: 0, $IQR_{difference}$: 0 to 1) than sleepiness ($Median_{difference}$: − 1, $IQR_{difference}$: − 1.5 to − 0.5).

Performance of the rocking bed

The accuracy of the stimulation, in the form of the root mean square error in velocity of the Somnomat, during rhythmic movements (Median: 0.0023 m/s; IQR: 0.0019 to 0.0092 m/s) was lower than while participants were lying still in the bed (Median: 0.0015 m/s; IQR: 0.0014 to 0.0028 m/s). In both cases, the error was minimal compared to the intervention maximum velocity (0.1 m/s). For one participant with two movement semiologies, a larger velocity error in the movement direction of the bed was observed when rhythmic movement occurred in the direction of the bed (0.0033 m/s) compared to when movement was perpendicular to the movement of the bed (0.0015 m/s).

During one intervention night the variability induced by the moving participant led to a mismatch between the bed reference and target position, twice leading to a safety stop. At the frequency of 2.0 Hz (upper limit) the amplitude was 0.001 m, a displacement very close to the resolution of the safety sensor used to monitor bed motion. By adapting the safety configuration to trajectories with small amplitudes this problem can be avoided. Researcher error resulted in the control intervention starting 12 min late on one occasion (Case 5) and ending 8 min late on another occasion (Case 4). No episodes of rhythmic movements were missed by the experimenters based on the comparison of the online scoring and offline scoring of the video data.

Acceptability and self-reported feedback on night protocol

All children reported feeling safe in the Somnomat and could imagine having a rocking bed at home. Parents also reported feeling their child was safe and five of six parents could imagine having the rocking bed at home. Four children thought the bed could help their RMD, the other two were unsure. After trying out the bed overnight, four out of six children thought it could help them sleep, one was unsure and one did not think it would be helpful.

The children reported feeling more rested upon awakening after the intervention night (Median: 1.5, IQR: 1 to 2) than after the baseline night (Median: 2, IQR: 1.25 to 2.75). Participants also felt more comfortable upon awakening after the intervention night (Median: 4.5,

IQR: 4 to 5) than after the baseline night (Median: 4, IQR: 3.25 to 4.75).

Three participants (case 2, 3 and 5) rated their time to fall asleep more positively during intervention nights than during baseline nights. However, one participant thought they woke up more frequently (case 1) and one felt a stronger urge to move (case 1).

Effect of bed movement on symptoms

Sleep duration based on actigraphy was similar during the baseline night (Median: 503 min, IQR: 446 to 532 min, $n = 3$) compared to the average of the nights at home (Median: 477 min, IQR: 446 to 497 min). Based on videosomnography analysis the children spent between 0 and 40% of the time in bed engaged in rhythmic movements. The duration of rhythmic movement did not differ between baseline (Median: 3.3%, IQR: 0.4 to 17.1%) and intervention nights (Median: 3.2%, IQR: 0.7 to 17.9%). Neither did time in bed, number of episodes and the average duration of episodes (Fig. 3).

Case 5 showed symptoms during the baseline, but not during the intervention night (preferred movement in habitual movement direction). Case 2 was able to turn 90 degrees in the bed and was stimulated in his habitual movement direction for 27 min and perpendicular to his habitual movement direction for 130 min. When stimulated in his habitual movement direction, episodes were shorter and less frequent (5.11 episodes/hour; $\text{Median}_{\text{duration}}$: 0.80 min; $\text{IQR}_{\text{duration}}$: 0.73 to 1.83 min) compared to other directions (17.6 episodes/hour; $\text{Median}_{\text{duration}}$: 1.56 min; $\text{IQR}_{\text{duration}}$: 0.63 to 2.04 min). Case 4 did not show symptoms on either night. For the other three participants no effect of bed movement was observed (Fig. 4).

Discussion

The primary aim of this study was to test the feasibility and acceptability of rhythmic vestibular stimulation as therapy for sleep-related rhythmic movement disorder, which could be shown based on device performance and participant feedback. With our small sample, measured during one night in the laboratory, no significant clinical outcomes were expected. Indeed, unlike the case series reporting on use of hammocks in the home environment (Hewertson and Hill 2018), we could not show a positive effect on rhythmic movements using a rocking bed for one night in the laboratory. For all five reported cases a resolution of rhythmic movements whilst using the hammock in the home environment was observed (Hewertson and Hill 2018). Timing and intensity of hammock use was not recorded, but the hammock was available for use on a daily basis and over a longer period, which might be essential for clinical efficacy. One of the five children who returned to a normal bed after 5 weeks of hammock use did not relapse, suggesting that effects of sensory stimulation might have lasting effects.

A limitation of the current study was that the available movement directions of the bed were limited to head-to-toe and side-to-side and, therefore, did not include the habitual directions of movement of all children. Furthermore, for safety reasons, participants under 10 years of age were exposed to only head-to-toe movement. This could explain why the movement direction and frequency preferred by children did not reliably match that of their habitual rhythmic movements. Moreover, for one participant who moved 90 degrees in his bed during the night, the bed movement direction seemed to effect treatment outcome. When stimulated in the direction of habitual movement, the episodes of rhythmic movement were shorter and less frequent compared to movements perpendicular to his habitual movement direction. It has been suggested that the frequency of vestibular stimulation modulates the influence of the stimulation on sleep (Vrugt and Pederson 1973). For future trials it might be of interest to see if emulating the direction and frequency of habitual rhythmic movements or rather

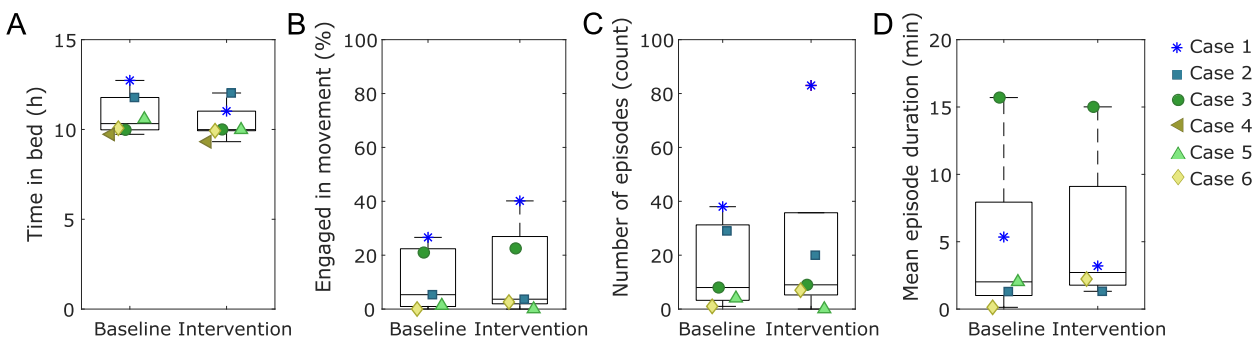

Fig. 3 Objective measures collected during the baseline and intervention night based on the video recordings ($n = 6$). A. Time in bed. B. Time spent engaged in rhythmic movement calculated over the common length of baseline and intervention night. C. Number of episodes calculated over the common length of baseline and intervention night. D. Mean episode duration calculated over the common length of baseline and intervention night

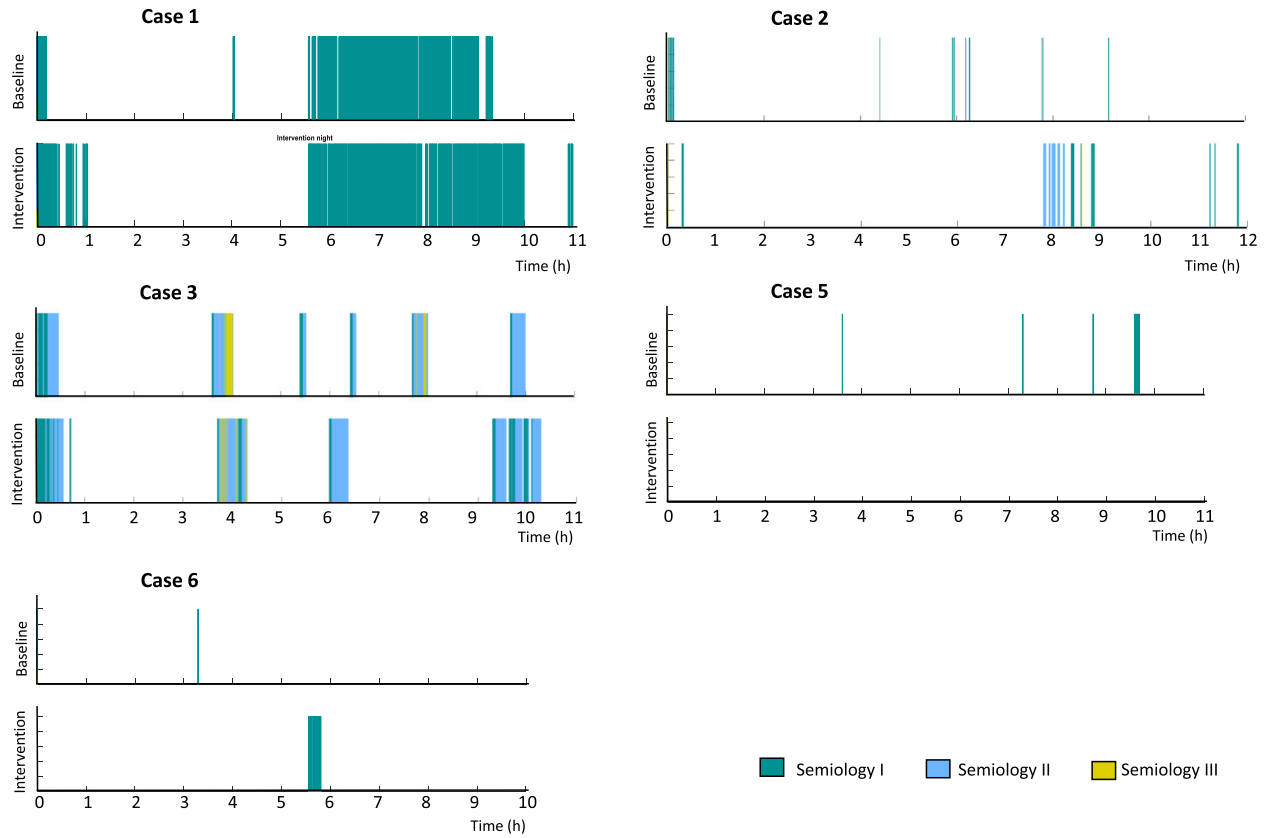

Fig. 4 Episodes of rhythmic movement during the baseline and intervention night for each case that showed rhythmic movement in the laboratory. Observed semiologies for case 2 were head banging (I: while bed was moving in habitual direction, II: bed moving perpendicular to habitual movement) and body rocking. Observed semiologies for case 3 were I: body rocking sideways and II: body rocking forward, furthermore lying still on all fours was observed (III). Case 4 did not show any movements in the two nights

finding the most comfortable setting reported by the participants, as was done in this study, has higher therapeutic value. If the first is true, than it seems the vestibular stimulation generated by the patient's movements is effectively inducing sleep, where if the latter is true it might be likely that the patient's movements are a self-soothing behaviour.

A challenge when studying RMD is that not all patients engage in rhythmic movement during polysomnographic recordings in a laboratory setting (Stepanova et al. 2005). This is confirmed by our clinical experience. In this study, with only contactless video-somnography recording, five out of six participants engaged in rhythmic movements in the laboratory setting. Solely one child (case 4), who was included in the study based on video evidence of rocking in the home environment, did not rock in the sleep laboratory. To increase the proportion of participants that engage in rhythmic movement in an experimental setting, moving the treatment and measurements to the home environment is recommended.

We see significant within child night-to-night variability of both number and duration of episodes of rhythmic movements during clinical home video recordings. Thus, to study the efficacy of our proposed therapy, evaluation over a longer exposure period and in the home setting would be ideal. Domiciliary rocking therapy would require an automated system. Two previous studies reported on acoustically based online applications potentially suited to RMD therapy, in one case supplemented by body worn sensors. In the present study the intervention was controlled by experimenters continually observing a 2D video stream. Contactless technologies, such as radar and 3D video, have the potential to automatically detect abnormal movements during sleep (Rahman et al. 2015; Garn et al. 2016). For future studies a closed loop system based on movement-sensitive acoustic sensors, radar or 3D video, would allow this promising new therapy to be studied in the ecologically relevant home setting.

Conclusion

Application of rocking movements in children with rhythmic movement disorder using the Somnomat was technically feasible. The sometimes violent movements

generated by the participants only minimally perturbed the movement of the bed, resulting in reliable execution of the planned bed movement. After trying out the bed, children reported the bed movements to be comfortable. Furthermore, children were more comfortable and were less sleepy upon awakening after the intervention night than after the baseline night. All children and their parents perceived the bed to be safe and the majority of children and parents believed the bed could help reduce rhythmic movements and promote sleep.

Considering the limited scientific evidence on therapies for sleep-related rhythmic movement disorder, a theoretically driven potential therapy substituting patient rhythmic movements with an external source of sensory stimulation, deserves further evaluation.

Supplementary information

Supplementary information accompanies this paper at https://doi.org/10.1186/s41606-020-00049-9.

Additional file 1. Supplemental information document 1. Video showing the start of an episode of rhythmic movement.

Additional file 2. Supplemental information document 2. Video of an episode of rhythmic movement in a patient with two movement semiologies. The patient switches from one semiology (side-to-side rocking) to another semiology (forward rolling movement) without pause. Often this occurs multiple times in an episode of rhythmic movement.

Additional file 3. Supplemental information document 3. The Rhythmic Movement Disorder Questionnaire. A structured interview for diagnosis of sleep-related rhythmic movement disorder in children

Additional file 4. Supplemental information document 4. Pediatric visual rating scales for comfort and sleepiness.

Abbreviations

IQR: Interquartile range; RMD: Rhythmic movement disorder

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Authors' contributions

The study was designed by RS, EW, LJ, OJ, and CH. Device and sensor set-up were developed by RS, EW, QR, MG, PA, and RR. Data was collected by RS, EW, QR, and CH. Data processing and analysis was performed by RS, EW, MG, and PA. The manuscript was written by RS and CH. The manuscript was critically reviewed by all co-authors. The author(s) read and approved the final manuscript.

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Availability of data and materials

The video data generated and/or analysed during the current study are not publicly available, because this data could not be anonymized. The manual scoring of the video data, supporting the conclusions of this article, is available in the ETH research collection repository: DOI https://doi.org/10.3929/ethz-b-000392724, http://hdl.handle.net/20.500.11850/392724.

Ethics approval and consent to participate

The UK National Research Ethics Committee and Health Research Authority (IRAS 234505), the Cantonal Ethics Committee of Zurich (KEK 2017–01880) and Swissmedic (2017-MD-0035) approved the study. All children gave informed assent and the parent gave informed consent.

Consent for publication

The parents of the participant visible in the supplemental video signed a consent form for clinical photography, stating that the video material can be used for presentation at scientific meetings, teaching purposes and publication in a scientific journal.

Competing interests

Authors report no conflicts of interest.

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