Received: 13 November 2023 | Accepted: 11 January 2024

DOI: 10.1111/jsr.14153

RESEARCH ARTICLE

Nocturnal vestibular stimulation using a rocking bed improves a severe sleep disorder in a patient with mitochondrial disease

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Funding information
Stiftung KinderInsel Bern; Promedica

Summary

Mitochondrial diseases are rare genetic disorders often accompanied by severe sleep disorders. We present the case of a 12-year-old boy diagnosed with a severe primary mitochondrial disease, exhibiting ataxia, spasticity, progressive external ophthalmoplegia, cardiomyopathy and severely disrupted sleep, but no cognitive impairment. Interestingly, his parents reported improved sleep during night train rides. Based on this observation, we installed a rocking bed in the patient's bedroom and performed different interventions, including immersive multimodal vestibular, kinesthetic and auditory stimuli, reminiscent of the sensory experiences encountered during train rides. Over a 5-month period, we conducted four 2-week nocturnal interventions, separated by 1-week washout phases, to determine the subjectively best-perceived stimulation parameters, followed by a final 4-week intervention using the optimal parameters. We assessed sleep duration and quality using the Mini Sleep Questionnaire, monitored pulse rate changes and used videography to document nocturnal interactions between the patient and caregivers. Patient-reported outcome measures, clinical examinations and personal outcomes of specific interests were used to document daytime sleepiness, restlessness, anxiety, fatigue, cognitive performance and physical posture. In the final 4-week intervention, sleep duration increased by 25%, required caregiver interactions reduced by 75%, and caregiving time decreased by 40%. Subjective fatigue, assessed by the Checklist Individual Strength, decreased by

Alexander Breuss and Marco Strasser contributed equally to this study.

Robert Riener and Matthias Gautschi are the shared last authors, both are equal sponsors of the study.

40%, falling below the threshold of severe fatigue. Our study suggests that rocking beds could provide a promising treatment regime for selected patients with persistent severe sleep disorders. Further research is required to validate these findings in larger patient populations with sleep disorders and other conditions.

KEYWORDS
alternative treatment, auditory stimulations, kinesthetic stimulation, mitochondrial disease, mitochondrial disease management, mitochondrial disease with an associated severe sleep disorder, robotic bed, rocking bed, sensory experiences, sleep disorder, Somnomat Casa, stimulation, vestibular stimulation

1 | INTRODUCTION

Mitochondrial diseases (MDs) are genetic disorders that are characterized by a defect in the oxidative phosphorylation resulting in a deficiency of cellular ATP synthesis. They show a wide range of clinical presentations, which may affect any organ, in any combination, at any age, from infancy to adulthood (Gorman et al., 2016). Approximately 45% of paediatric patients with MD suffer from neuromuscular problems (Munnich et al., 1996; Trenell et al., 2007). Fatigue is seen in 71%–100% of patients with primary MD (Brunetti et al., 2021; Parikh et al., 2019) and, in many cases, the severity of fatigue correlates with the severity of the MD (Gorman et al., 2015). Although severe sleep disorders are often associated with MD, they are not well understood, often underdiagnosed (Ramezani & Stacpoole, 2014), and difficult to treat. MDs may be associated with any type of sleep disorder (Sateia, 2014). Their overall incidence in MD has been estimated at 33%–75% (Parikh et al., 2019; Smits et al., 2012). Most often reported are sleep-related breathing disorders, including central versus peripheral (i.e. obstructive) sleep apnoea and depressed ventilatory response to hypoxia/hypercapnia; less often reported are sleep-related movement disorders, such as restless legs syndrome, sleep–wake rhythm disorders or hypersomnolence (Mosquera et al., 2014; Příhodová et al., 2021; Primiano et al., 2021; Smits et al., 2012). Surprisingly, insomnia has only rarely been reported in MD (Příhodová et al., 2021; Suzuki et al., 1997).

Vestibular stimulation (VS) could be a promising additional option to classical pharmaceutical treatments (e.g. dopamine agonists, benzodiazepines or melatonin) as it is known to have positive effects on respiration, heart rate and blood pressure (Monahan & Ray, 2002; Omlin et al., 2016; Thurrell et al., 2003), on sleep consolidation and sleep onset (Bayer et al., 2011; van Sluijs et al., 2020; Woodward & Tauber, 1990), on psychological (Cross et al., 2018; Sailesh et al., 2014), neurological (Dave, 1992), neurodevelopmental (Ottenbacher et al., 1981) and neurodegenerative disorders, and on Parkinson's disease (Grabherr et al., 2015; Khoshnam et al., 2018; Wilkinson et al., 2019). However, to our knowledge, nocturnal VS has never been investigated in patients suffering from MD.

We present the case of a 12-year-old male patient with a severe MD due to a complex I deficiency of the respiratory chain diagnosed in his first year of life. His sleep was characterized by fragmented sleep of 3–5 hr a night, which had a direct impact on his quality of life. He had no obstructive sleep apnea, seizures or central hypoventilation, factors frequently present in MD. Interestingly, his parents reported that on night train rides their son enjoyed recreative sleep.

The Somnomat Casa (Figure 1) is a rocking bed developed at the Sensory-Motor Systems Lab of ETH Zurich, Switzerland, and is capable of applying translational movements in the longitudinal axis (Breuss, Fujs, & Riener, 2023; Breuss & Riener, 2023; Breuss, Suter, et al., 2023; Crivelli et al., 2016). Moreover, the motion can be overlaid with various motion disturbances and auditory stimuli displayed by speakers, allowing to imitate immersive train rides. The goal of this individual therapeutic trial is to apply VS generated by the rocking bed and observe whether the aforementioned positive effects of nocturnal VS may also apply to a patient with MD.

FIGURE 1 Somnomat Casa placed in the bedroom of the patient. It has dimensions comparable to a standard single bed.

2 | METHODS

2.1 | Subject

At the age of 10 months, our patient presented an infection-triggered nystagmus as the first manifestation of his MD. Brain imaging was suggestive of a mitochondrial encephalopathy, and a biopsy of the muscle showed a severe complex I deficiency of the respiratory chain, confirming the diagnosis of a primary MD. In the course, he developed a chronic progressive external ophthalmoplegia, severe ataxia and tetraspasticity with motor developmental delay and microcephaly, a severe skeletal myopathy, as well as a cardiomyopathy with a Wolf–Parkinson–White syndrome. Recurrent supraventricular tachycardias had to be cardioverted repeatedly with adenosine (the last event occurred at the age of 9 years). His myopathy and movement disorder with ataxia and spasticity were progressive, and secondary orthopaedic complications such as scoliosis and progressive hip-luxation allowed him in the course to only move around with an electric wheelchair. He was fed by a gastrostomy tube since his second year of life. He lived with his parents and needed constant care, but he had no cognitive impairment. One major burden with an important impact on his quality of life was his sleep: the nocturnal sleep was short, with a duration of 3–5 hr and fragmented, with most sequences of only 15 min. While lying awake, he was anguished and ridden by pain and itchiness. Moreover, during these phases, he was restless, needed a lot of care, and was most of the time in a sitting/rolled position worsening his scoliosis (Figure 2). Every night, either a parent or a caregiver was in the bedroom with him. Drug therapy with melatonin or cannabidiol (CBD) had failed, as well as alternative aroma or audio stimulation; no other drug therapies, including benzodiazepines and others, had been used due to fear of excessive respiratory repression in this patient. During the study phase, the patient did not take any sleep medication. Because of his sleep disorder, he had severe mental and physical fatigue, affecting his and his surrounding's quality of life.

2.2 | Device

The Somnomat Casa (Sensory-Motor Systems Lab, ETH Zurich, Switzerland) is a robotic bed that provides translational movements along the longitudinal axis (Figure 1; Breuss, Fujs, & Riener, 2023; Breuss & Riener, 2023; Breuss, Suter, et al., 2023; Crivelli et al., 2016). It has the dimensions 90 × 200 × 44 cm (width × length × height, excluding mattress), can be started and stopped using a single button (Figure 3), and requires a standard wall power outlet (230 V) for operation. Operational noise emission is below 30 dB (LAeq), which allows the bed to be used in long-term sleep studies in a private home setting. An accelerometer, as well as a photodetector integrated into the bed, report bed usage, movements on the bed, as well as day–night patterns of the users. In addition to the integrated sensors, polysomnography (PSG) devices, as well as other recording devices, can be connected directly to the bed, allowing for time-synchronized measurements. Data aggregation of device usage and the sensors connected, as well as data upload to a server is performed automatically. Using an external computer, motion parameters can be adjusted.

The amplitude of the translational movements can be adjusted but has been fixed to 10 cm for this study. The Somnomat Casa has three integrated operational modes:

1. Constant frequency: motion follows a sinusoidal trajectory with a frequency to be chosen between 0.04 Hz and 0.4 Hz, corresponding to accelerations between 0.006 and 0.64 m s^{-2}. From the literature, accelerations that showed the largest sleep-related benefits were between 0.2 m s^{-2} (0.23 Hz) and 0.3 m s^{-2} (0.28 Hz; Omlin et al., 2016). The relationship between frequency and accelerations can be expressed using $f = \frac{1}{2\pi}\sqrt{\frac{a}{A}}$, where A corresponds to the amplitude of 10 cm.
2. Frequency schedule: a list of frequencies and desired durations can be provided. The bed then iterates through this list of frequencies, before repeating the entire schedule. As an example, the frequency list {(0.25 Hz, 3600 s), (0.2 Hz, 1800 s), (0 Hz, 1800 s)} would result in the bed moving at 0.25 Hz during the first hour, followed by 0.2 Hz for 30 min, and 0 Hz for the next 30 min before repeating.
3. Train simulator: the bed replays motion disturbances, hence simulating a train ride. Additionally, the background sound of the train can be played time-synchronized to the motion disturbances of the movement. The motion disturbances can be varied in three different intensities (low, medium, high).

The Somnomat Casa was installed in the bedroom of the patient for 5 months (02–06/2022). Any non-expert was able to operate the bed after a short instruction. Moreover, redundant mechanical, electrical and software safety mechanisms ensured safe operation.

Ethical approval for this study was waived by the ethics committee of the canton of Bern (Req-2021-01096), and informed

FIGURE 2 The patient spent the nights either in a "rolled" position (left) or flat position (right). To transition from flat to rolled position, assistance from the attending caregiver is required.

consent was obtained by the legal representative before the start of the study.

2.3 | Study protocol and parameter settings

Before the Somnomat Casa was installed in the patient's private bedroom, baseline assessments were conducted. After the bed was delivered, there was an initial 2-week familiarization phase without any nocturnal interventions. Moreover, during the days of this period, the patient was asked to try all operational modes and report on settings that he deemed inadequate. In the study phase that then followed the familiarization phase, four intervention configurations that were perceived as most comfortable by the boy were selected based on the three available modes (Table 1). Each intervention configuration lasted for 2 weeks, separated by 1-week washout phases in between. After completing the four intervention configurations, a final intervention of 4 weeks was reapplied for 1 month using the movement profile that was perceived most promising for improving sleep quality. The final clinical follow-up examination was performed 3 months after the cessation of the treatment (Figure 4).

FIGURE 3 Smart Handle to turn bed on and off, illuminate the surrounding, and record bed and user movement, as well as day- and night-patterns.

Out of the three available operational modes, the subjectively most comfortable mode, as reported after the familiarization phase, was the train simulation with motion disturbances of medium strength. The patient felt nausea with frequencies about 0.3 Hz (equalling 0.35 m s^{-2}). Therefore, the frequencies were bound between 0.196 Hz (0.15 m s^{-2}) and 0.25 Hz (0.25 m s^{-2}), which is also in line with the literature (van Sluijs et al., 2020). For the first intervention, the frequency of 0.196 Hz was tested without auditory stimulation, and in the second intervention, acoustic feedback was added. In the third intervention, the frequency of 0.25 Hz was applied without auditory stimulation. Seeing that the frequency of 0.19 Hz subjectively led to a faster sleep onset and the frequency of 0.25 Hz enabled him to stay longer in bed, we combined these two movements in a fourth intervention configuration: after the first movement was applied using a frequency of 0.19 Hz for the first 30 min, it was followed by the frequency of 0.25 Hz for the rest of the night. This movement profile was then also reapplied in the final 4-week intervention phase, as the subjective perception was also confirmed by an increased sleep duration obtained from a preliminary analysis after the first four intervention phases.

TABLE 1 Motion parameters that were investigated during the study.

Intervention	Mode	Frequency (Hz)	Acceleration (m s^{-2})	Motion disturbances	Sound
1st	Train ride	0.19	0.15	Medium	No
2nd	Train ride	0.19	0.15	Medium	Yes
3rd	Train ride	0.25	0.25	Medium	No
4th	Train ride	0.19 to > 0.25 (after 30′)	0.15 to > 0.25	Medium	No
Final	Train ride	0.19 to > 0.25 (after 30′)	0.15 to > 0.25	Medium	No

Note: Each intervention lasted for 2 weeks, and was succeeded by 1 week of washout (Figure 4). After the four, 2-week interventions, a final 4-week intervention of the subjectively best movement profile was reapplied to investigate the effects of varying intervention duration.

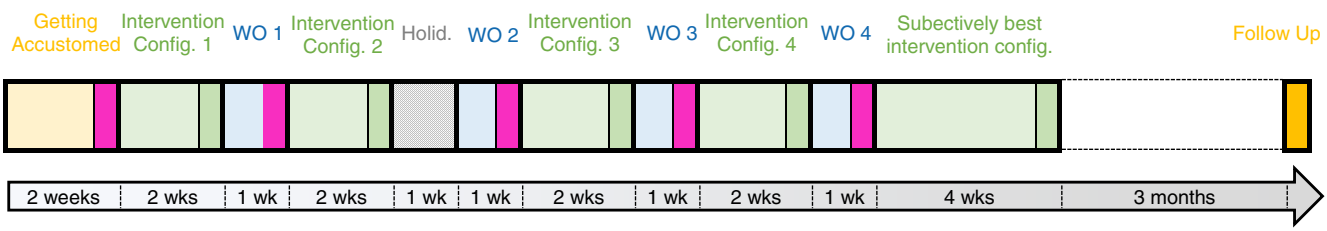

FIGURE 4 Schedule of the interventions. Interventions lasted for 2 weeks each and were separated by 1-week washout phases (WO). The dark-shaded cells correspond to the last 3 nights of every phase, and indicate three consecutive measurement nights. The last 3 nights of all non-intervention phases are combined into a rolling baseline, denoted in pink.

2.4 | Recordings and assessments

In the pre-study and follow-up phases, physicians completed the Newcastle Paediatric Mitochondrial Disease Scale (NPMDS; Phoenix et al., 2006), and a global clinical impression (GCI) was gathered. A physiotherapist performed joint measurements, and the Gross Motor Function Measure-66 (GMFM-66; Russell et al., n.d.) was scored, from which 13 items were selected. The test was held under video recording for interrater reliability, rated by the physiotherapist and a paediatrician. Additionally, a neuropsychologist conducted the Test of Nonverbal Intelligence 4th Edition (TONI-4; Brown et al., 2010). A neurological clinical examination was performed by a neuropaediatrician to gather the Manual Ability Classification System (MACS; Eliasson et al., 2006) and the Gross Motor Function Classification System (GMFCS; Palisano et al., 1997). To ensure comparability, subsequent tests were conducted under standardized conditions at the same locations, whereby the same physiotherapist and neuropsychologist administered the tests on the same day of the week, as well as at the same time of day.

During the study, all nights were recorded with an infrared camera (Reolink E1) in the bedroom of the patient. Using these video files, the interactions of the parents and caregivers were manually counted, and two metrics were defined. The *Normalized Interaction Time* denotes the aggregated timespan of the night during which assistance was required, divided by the total time in bed (Equation 1). The *Interaction Frequency* represents the mean number of interactions divided by the total time in bed (Equation 2). Moreover, the time in bed, as well as the onset and duration of two distinct positions (rolled, flat; Figure 2), were manually extracted from the video data. Using the time in bed and time in the flat position, we calculated the *Relative Time in Flat Position* (Equation 3), an indicator of how restful the night was perceived.

$$\text{Normalized Interaction Time} = \frac{\text{Summed Interaction Times}}{\text{Time in Bed}} \quad (1)$$

$$\text{Interaction Frequency} = \frac{\text{Number of Interactions}}{\text{Time in Bed}} \quad (2)$$

$$\text{Relative Time in Flat Position} = \frac{\text{Time in Flat Position}}{\text{Time in Bed}} \quad (3)$$

The parents or caregivers filled out the adapted Mini Sleep Questionnaires (MSQ) after every night, as well as a sleep diary throughout the day, including the estimated sleep duration. At the end of the study, the patient furthermore summarized his experience with the rocking bed in an unstructured written report.

Patient-reported outcome measures (PROMs) were collected in the final 3 nights of every intervention and washout phase: on a night after the patient visited school, after a mid-day off, and after a whole day off. To increase objectivity and whenever possible, each of the 3 nights was reported by the mother, the father, as well as a caregiver before being averaged. Additionally, on the final night, the adapted questionnaires, Checklist of Individual Strength (CIS; Vercoulen et al., 1994), Pittsburgh Sleep Quality Index (PSQI; Buysse et al., 1989), Epworth Sleepiness Scale (ESS; Johns, 1991) and the Fatigue Severity Scale (FSS; Krupp et al., 1989), were administered and filled out by the father and mother together over the last period. The subjective changes based on the personal outcomes of specific interests (POSIs) were filled out by the mother and father separately.

Saturation, heart rate and respiratory rate were collected in the three assessment nights using a PSG (Nox A1, Nox Medical with Nonin WristOx2 3150, Nonin Medical). During the other nights, the patient used his pulse oximeter (Bitmos sat 801+, Bitmos GmbH), which he had become familiar with for years. After every phase, a clinical examination and a global clinical impression were performed by a physician.

For a better understanding of the sleep architecture and sleep-related breathing disorders of the patient, a full PSG (including electroencephalogram [EEG] and airflow) was originally planned during the last 3 nights of every phase. Despite a multi-week acclimatization phase before the study where three different EEG devices (Nox A1, Nox Medical; Dreem 2, Dreem; SleepLoop, HMZ, Zurich) of varying obtrusiveness were evaluated, the patient neither tolerated the patches of the EEG, electrocardiogram nor the nasal cannula. Therefore, sleep duration and quality could only be approximated using the data from the MSQ, as well as the data from the video (i.e. time in bed and time in flat position).

The primary outcome of the study was the sleep duration, approximated using the MSQ and videography. As a recommendation from the parents of the child, we also included a pulse rate threshold as a proxy to decide between sleep and wake. According to their observation, their son's pulse was below 90 bpm whenever he was asleep. Secondary outcomes were subjective fatigue captured through the CIS, as well as the number and duration of interactions between the patient and the caregivers, extracted from the videography.

Statistical Analysis was performed using GraphPad Prism 9.2.0 (GraphPad Software, LLC, San Diego, USA). Phases were compared

TABLE 2 Sleep durations reported through the MSQ for every study phase.

Study phase	Mean sleep duration (min)
Baseline	260.8
1st Intervention	250.6
2nd Intervention	253.8
3rd Intervention	254.4
4th Intervention	302.9
Final Intervention	321.3

TABLE 3 Sleep durations for every study phase compared against the baseline.

Comparison	Difference (min)	*p*-Value
Baseline versus 1st intervention	−10.2	0.9938
Baseline versus 2nd intervention	−7.0	0.9986
Baseline versus 3rd intervention	−6.3	0.9989
Baseline versus 4th intervention	+42.1	0.2223
Baseline versus final intervention	+60.6	0.0053 (**)

Note: The patient slept on average 4.3 hr during the baseline period. During the last interventional period, a statistically significant increase ($p \leq 0.01$) of 25% to an average sleep duration of 5.4 hr sleep could be reported.
**$p \leq 0.01$.

using an unpaired one-way ANOVA with $\alpha = 0.05$ (95% confidence interval) against the baseline. A Dunnett test was used to correct for multiple comparisons using statistical hypothesis testing. Each p-value is then adjusted to account for the multiple comparisons. Residuals are checked for Gaussian distribution and overall data for equal SD.

3 | RESULTS

3.1 | Sleep duration and time in bed

The daily sleep duration filled in the MSQ showed a significant ($p < 0.005$) increase in mean sleep duration between the baseline and the best intervention configuration (Table 2). At the end of the final intervention, the patient showed an increase in sleep duration exceeding 25% from 4.3 to 5.4 hr from the baseline to the final intervention configuration (Table 3).

During the study, not only did the overall time in bed increase (Figure 5) in every intervention phase compared with baseline, but also the time spent in a flat position increased gradually in all intervention nights from below 80% during the baseline nights to over 95% during the final intervention (Figure 6).

We could not confirm the observation from the parents that a pulse of 90 bpm can be used as a decision boundary for sleep and wake. Although the MSQ and the relative time spent in a flat position indicate a significant increase in sleep duration, the duration of the pulse below 90 bpm was not significantly different between the different phases (Figure 7).

3.2 | Interaction

The mean relative interaction time, the time when the caregivers or parents provided support for the patient, decreased from 35% at the

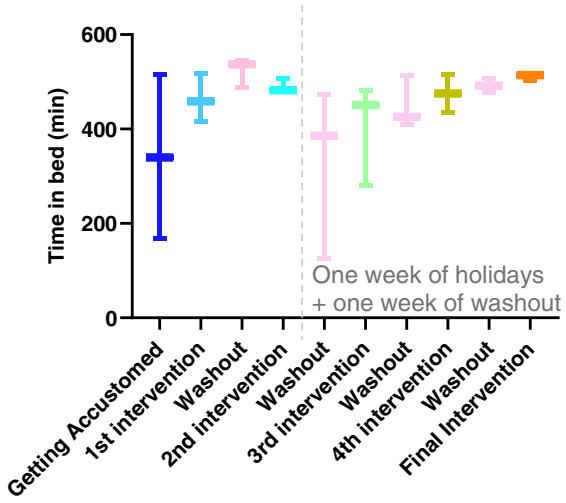

FIGURE 5 Overall time in bed over all phases.

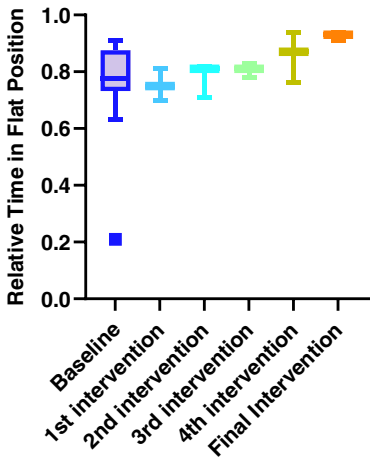

FIGURE 6 Relative time spent in flat position as defined in Equation (3).

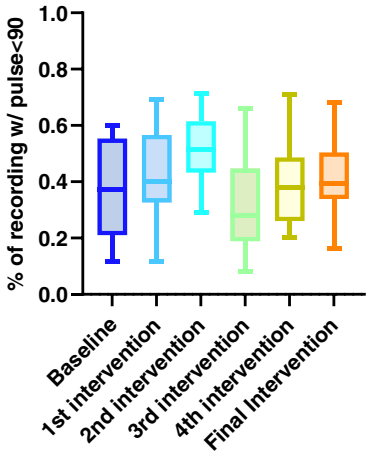

FIGURE 7 Percentage of the night with a pulse rate below 90 bpm. There were no significant differences between the different phases.

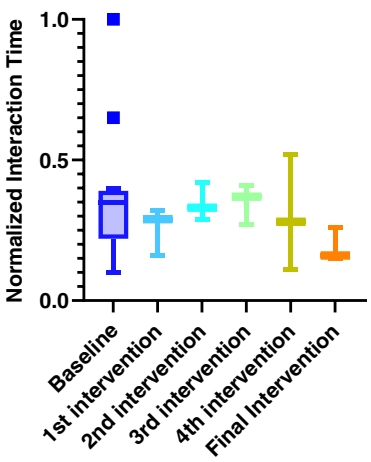

FIGURE 8 The mean time required for assistance was reduced from baseline nights (35%) to final intervention nights (19%).

beginning of the study to 19% (Figure 8) after the final intervention. Furthermore, the mean number of caregiver interactions per hour was reduced from 3.7 to 1.0 (Figure 9).

3.3 | Questionnaires and clinical impression

Of the collected questionnaires, the CIS showed a constant improvement from the pre-study stage to the end of the study (Figure 10). Before the study, a total score of 105 points was reported, with the Subjective Fatigue, Concentration, Motivation, and Activity domains being at 56, 24, 12 and 13 points, respectively. During the study, all four domains of the CIS improved: after the final intervention, a total score of 65 points was observed, with the subdomains Subjective Fatigue, Concentration, Motivation, and Activity domains being at 33, 14, 8 and 10 points, respectively. The 33 points scored for Subjective Fatigue are 2 points below the cutoff of severe fatigue (Fnm & Lamm, 2017). The PSQI, FSS and ESS confirmed a chronic sleep disorder and fatigue, but no excessive daytime sleepiness. These results did not significantly change from the pre-study state to the final intervention.

Also, these results were separately but consistently reported by the parents and caregivers through the PROMs, and by physiotherapists, neuropsychologists and physicians in the free text of the questionnaires. The reported overall impression was that the patient was more active and awake, and the voice was stronger during day-time. Activities like going to an indoor pool, which were too exhausting before the study, were possible already after the first intervention block. In an unstructured report formulated by the patient, he expressed significant improvements, including better sleep, increased communication ability, extended wheelchair use, enhanced enjoyment of school, improved learning speed and reduced itching, attributing these positive changes to the calming and relaxing effects of the bed.

The physical condition was tested objectively with the GMFM-66, of which 13 items could be tested. The patient scored 31.19 at the pre-study stage and 36.79 after the final intervention, with a possible score of 100. The joint measurements showed in the right and left elbow a range of motion increase in flexion of up to 15° and 5°, respectively. No changes in the other joints were detected.

The results of the non-verbal IQ (TONI-4) showed an IQ of 114 in the pre-study and an IQ of 137 after the study. At the follow-up examination, 3 months after the end of the study period, the non-verbal IQ dropped again to 115, corresponding to the pre-study level (Table 4).

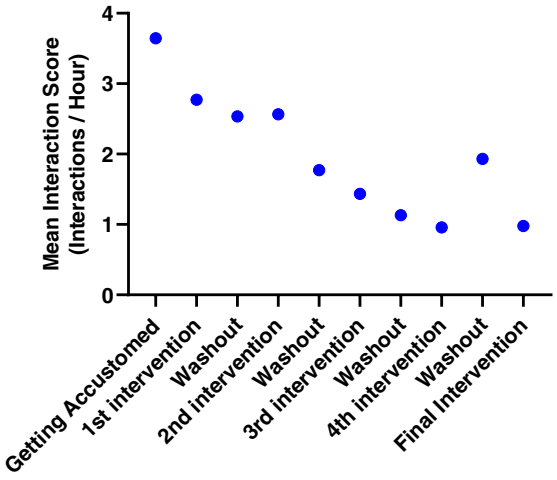

FIGURE 9 Mean number of interactions per hour over the course of the study. The number of interactions decreased from 3.65 interactions per hour during the pre-study state to less than one interaction per hour after the final intervention.

4 | DISCUSSION

Our patient presented with a primary MD characterized by severe predominantly motor symptoms and an associated severe sleep disorder. In the course of 5 months, he received nocturnal rocking

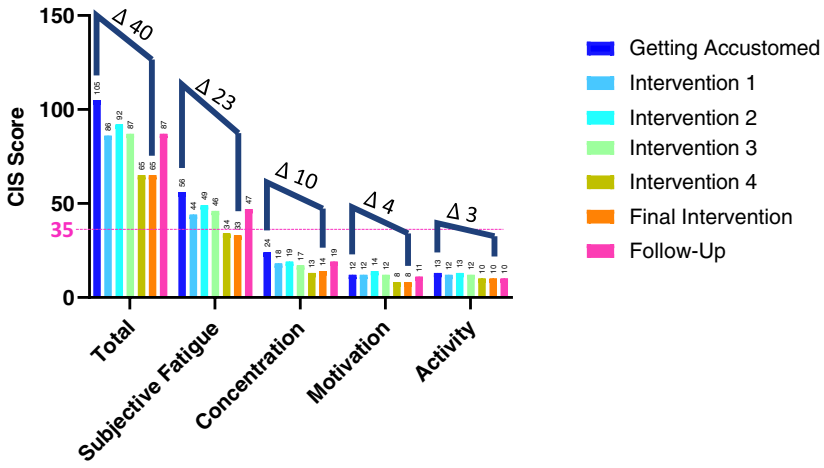

FIGURE 10 The Checklist of Individual Strength (CIS) showed a constant improvement during the entire study. The total score improved by 40 points from 105 points at the pre-study state to 65 points at the end of the study. The Subjective Fatigue decreased by 23 points and was last reported at 33 points, 2 points under the cutoff of severe fatigue. Three months after cessation of the treatment ("Follow-Up"), the total score worsened to 87 points, and the subjective fatigue increased significantly.

interventions from the Somnomat Casa, a rocking bed capable of applying nocturnal translational VS. After combining the frequencies 0.19 and 0.25 Hz, where the lower frequency was changed to the higher frequency 30 min after the intervention start, and applying motion disturbances of medium intensity that simulate a train ride, his sleep duration and quality improved significantly (Table 3). The rationale for changing the frequency after 30 min was mainly based on feedback from the parents and caregivers in which they reported a faster sleep onset with the slower frequency but a longer sleeping duration with the higher frequency. This is in line with the literature, where personalized treatments were recommended for nocturnal interventions (Crivelli et al., 2016; Omlin et al., 2016; Omlin et al., 2018; Perrault et al., 2019). However, to the best of our knowledge, none of the previous studies changed rocking frequencies during the intervention.

In our patient, the previously attempted behavioural, drug (melatonin or CBD) or alternative (aroma or acoustical) treatments for improving his sleep had failed due to habituation effects already after 2 weeks. No such effect was seen even after 4 weeks of the same intervention with the rocking bed. VS could, therefore, also be a potential new therapeutic approach for patients with MD with severe sleep disorders if prior behavioural or pharmaceutical approaches

TABLE 4 Summary of the subjective questionnaires and the clinical assessments at the pre-study state, after the final intervention, and 3 months after the treatment (i.e. follow-up).

		Pre-study	Final intervention	Follow-up	Interpretability (◇... worst, ↗...best)
Subjective questionnaires	PSQI	17	**11**	16	**0**↗–21◇ 0–5: good
	FSS	62	**51**	54	**9**↗–63◇
	ESS	**7**	**6**	**4**	0–24 **0–10: normal** 10–24: pathological
	CIS	109	**65**	87	**20**↗–140◇
	POSIs				
	Fatigue	–	↘	↗	
	Sleepiness	–	↘	↗	
	Restlessness	–	↘	↗	
	Sleep quality	–	↗	↘	
Clinical assessments	GMFCS	**4**	**4**	**4**	1◇–**5**↗
	MACS	**4**	**4**	**4**	1◇–**5**↗
	Range of motion				
	Neutral zero method				
	Right elbow	135°/70°/0°	**150°/45°/0°**	Data not collected	150°/0°/10°
	Left elbow	135°/20°/0°	**140°/35°/0°**		Flexion/0/Extension
	Hand	Big clench	**Big and small clench**		Big and small clench
	GMFM-66*				
	Gross motor function Measure, adapted with 13 items tested	31.19	**36.79**	Data not collected	0◇–**100**↗
	Non-verbal IQ	114 (P83)	**137 (P99)**	115 (P84)	**> 132 (P99)**
	TONI-4				100 (P50)
	Test of Nonverbal Intelligence				
	Global clinical impression				
	Overall impression	–	↗	↘	
	Voice	–	↗	↘	
	Physical posture	–	↗	↘	
	Thought process	–	↗	↘	

Note: Best patient outcomes are highlighted in bold.

Abbreviations: CIS, Checklist of Individual Strength; ESS, Epworth Sleepiness Scale; FSS, Fatigue Severity Scale; GMFCS, Gross Motor Function Classification System; GMFM-66, Gross Motor Function Measure-66; MACS, Manual Ability Classification System; POSI, personal outcomes of specific interests; PSQI, Pittsburgh Sleep Quality Index; TONI-4, Test of Nonverbal Intelligence 4th Edition.

were ineffective. Interestingly, we first expected in the washout phase a renewed deterioration of the sleep duration. However, this could not be objectively confirmed as none of the scores returned to baseline levels (Figures 5 and 9) after the 1-week washout phases, which led us to assume a sustained effect of the rocking bed. Only after an extended washout phase of 2 weeks or after the 3 months period between the last intervention and the follow-up examination, we could see washout effects (Figure 5).

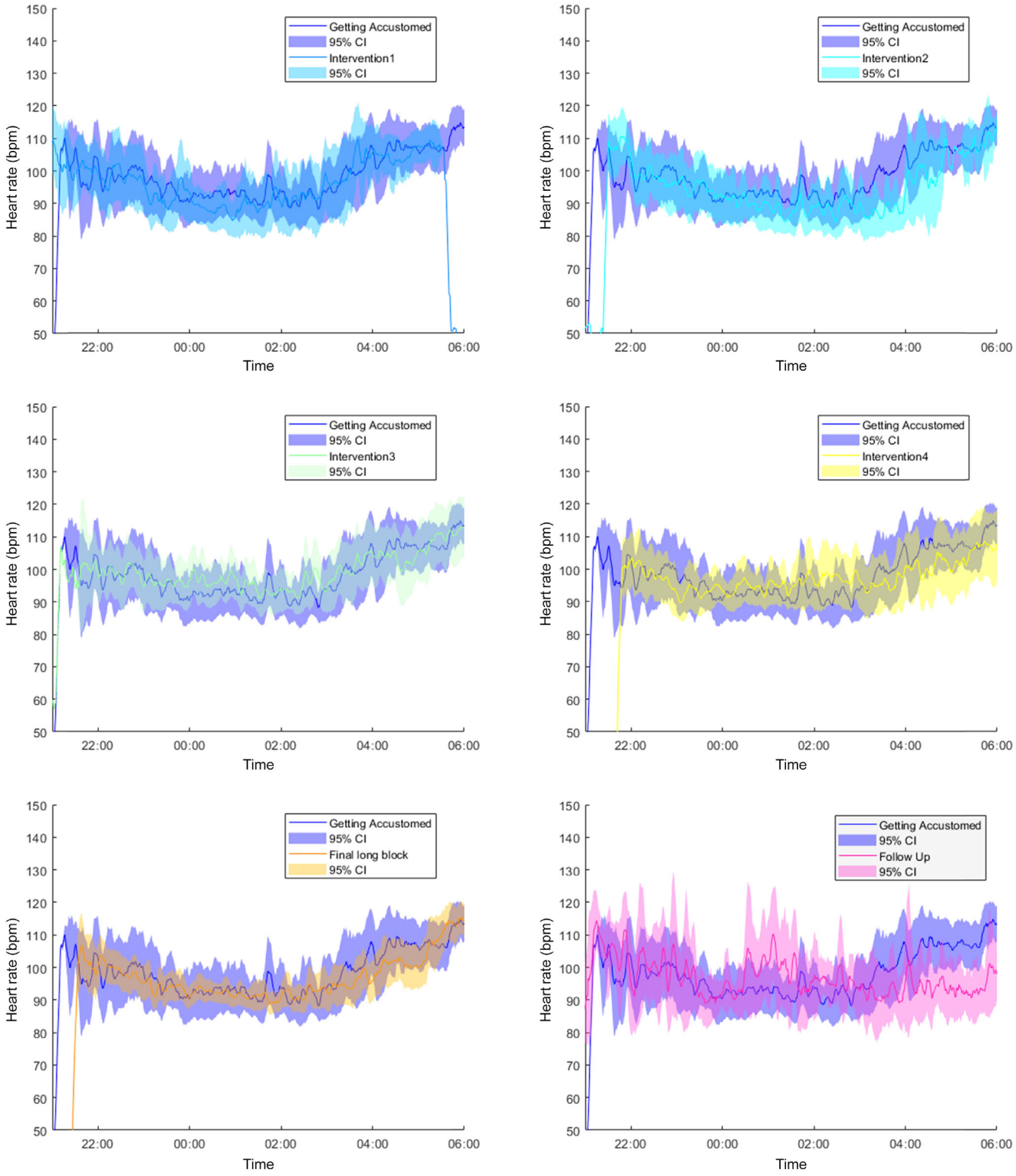

FIGURE 11 Aggregated pulses over each study phase. We plotted the mean and the 95% confidence interval of all nights in the respective study phase, and compared its area with the pre-study state.

For further studies where carry-over effects need to be prevented, an extension of the washout phase and the intervention phase should be taken into consideration.

No side-effects related to the rocking bed were reported during the study. Also, during the study period, the patient experienced no episode of supraventricular tachycardias due to the Wolf–Parkinson–White syndrome. And because the bed was compact enough, it could be installed in the bedroom of the patient. This circumvented any laboratory confounder usually present in sleep studies.

The entire study lasted 5 months and, therefore, seasonal effects might have played a role. However, the study took place between February and July 2022 and, for seasonal effects, we would expect a natural decrease in sleep duration towards the summer months (Allebrandt et al., 2014; Hjorth et al., 2013), which was not the case. In fact, sleep quality was significantly improved and, taking into account seasonal effects, the outcome might have been even better if the final intervention had occurred during the winter months.

The increased sleep duration also had a positive effect on the subjective fatigue of the patient, as clinically evaluated through the CIS: while the patient was scored as severely fatigued before the study, he was below the threshold for severe fatigue after the final intervention (Figure 10). Unfortunately, there are no minimal clinically important differences available for the total CIS score, making statements about the significance of the total CIS score difficult (van Dijk et al., 2022). Throughout the study, both objective clinical assessments and subjective data from the PROMs constantly indicate a more awake and more active patient during the daytime with better cognitive performance and better physical posture, compared with the baseline without the Somnomat Casa. This agreement between the objective and subjective measures also helps alleviate concerns that the parents might have been biased to favour the intervention. Most importantly, the patient himself described these substantial improvements during the daytime in writing at the end of the study, calling the rocking bed calming and relaxing.

Consequently, the improved quality of sleep also led to fewer interactions between the parents or caregivers and the patient during the night. This led to the conclusion that the patient has a more calm and more peaceful time in bed with fewer restless phases. Also, the care effort was reduced, allowing his caregivers to have better sleep, too.

Although we could not confirm the observation from the parents that a pulse rate of 90 bpm can be used as a decision boundary for sleep and wake, we observed less variance in the pulse during the interventional phases (Figure 11). This could indicate a more restful and undisturbed sleep during the interventional phases compared with the pre-study state due to reduced physiological stress, fewer nocturnal arousals, and a more balanced autonomic nervous system.

The main limitation of this study was the lack of PSG recordings. Despite attempting various minimally obtrusive measurement devices, none was tolerated by the patient, allowing only for an approximation of the effective sleep duration through questionnaires and videography. Therefore, no statement about the impact of the rocking bed on the patient's sleep architecture can be made. To minimize the bias, we used different methods to compare the sleep duration, and tried to objectify it using input from different raters, pulse oximetry and videography. Furthermore, this study primarily served as a pilot study for a larger study planned in the future. Such a larger study will need to encompass a patient population capable of undergoing PSG and should allow for sufficiently-powered statistical tests.

To our knowledge, this is the first report of a successful treatment of a severe sleep disorder in a patient with MD using a rocking bed. Overall, the presented results with respect to increased sleep duration, improved sleep quality, reduced physical and mental fatigue, as well as reduced care effort are highly promising and warrant further studies with larger patient cohorts. Moreover, it was the first time that the intervention device (Somnomat Casa) was used in a private bedroom for several months. No safety or operational issues were reported during this time, proving the applicability of the Somnomat Casa in long-term studies in home environments.

AUTHOR CONTRIBUTIONS

Alexander Breuss: Conceptualization; investigation; methodology; validation; visualization; writing – review and editing; software; formal analysis; project administration; data curation; writing – original draft. **Marco Strasser:** Conceptualization; investigation; writing – original draft; methodology; validation; visualization; writing – review and editing; formal analysis; project administration; data curation; software. **Jean-Marc Nuoffer:** Writing – review and editing; supervision. **Andrea Klein:** Writing – review and editing; supervision. **Eveline Perret-Hoigné:** Writing – review and editing; investigation. **Christine Felder:** Investigation; writing – review and editing. **Ruth Stauffer:** Writing – review and editing; investigation. **Peter Wolf:** Methodology; conceptualization; writing – review and editing; supervision. **Robert Riener:** Conceptualization; writing – review and editing; methodology; supervision; resources. **Matthias Gautschi:** Resources; supervision; project administration; writing – review and editing; writing – original draft; funding acquisition; investigation; conceptualization; methodology; validation; visualization; formal analysis.

ACKNOWLEDGEMENTS

The authors sincerely thank the patient and his family for their willingness to participate in this study and for their valuable contributions and feedback. The authors extend their gratitude to the dedicated caregivers from Spitex who provided exceptional support and care of the patient throughout the study. Furthermore, the authors would like to acknowledge the Inselspital and specifically Sebastian Grunt and the Sleep Wake Epilepsy Center for their assistance in facilitating this research. Their expertise has been instrumental in the execution of this study. Deep appreciation goes to the members of the SMS-Lab at ETH Zurich for their invaluable contributions, especially Michael-Herold Nadig, Paul Schürmann and Lars Peter for their work on the electronics of the rocking bed, Marco Bader for the fabrication of the bed, and the civil servants Nicola Wyss, Patrick Bösch, Christoph Ungricht, Joel Flückiger and Thiemo Frei for their support during the

study. The authors are also grateful to Giulia Da Poian, Oriella Gnarra and Manuel Fujs for their critical and valuable feedback throughout the study. Finally, the authors gratefully acknowledge the financial support provided by the Stiftung KinderInsel Bern, and Alexander Breuss thanks the Promedica Foundation for supporting his research. Open access funding provided by Eidgenossische Technische Hochschule Zurich.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

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How to cite this article: Breuss, A., Strasser, M., Nuoffer, J.-M., Klein, A., Perret-Hoigné, E., Felder, C., Stauffer, R., Wolf, P., Riener, R., & Gautschi, M. (2024). Nocturnal vestibular stimulation using a rocking bed improves a severe sleep disorder in a patient with mitochondrial disease. *Journal of Sleep Research*, e14153. https://doi.org/10.1111/jsr.14153