Atypical Headbanging Presentation of Idiopathic Sleep Related Rhythmic Movement Disorder: Three Cases with Video-Polysomnographic Documentation

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SCIENTIFIC INVESTIGATIONS

Study Objectives: To describe three cases of sleep related, idiopathic rhythmic movement disorder (RMD) with atypical headbanging, consisting of head punching and head slapping.

Methods: Three consecutive patients (2 males [11 and 13 years old] and one female [22 years old]) presented with atypical headbanging of 6 years, 7 years, and 17 years duration. In 2 cases, typical rhythmic headbanging (with use of the head) shifted after 3-4 years to atypical headbanging, with frontal head punching that was quasi-rhythmic. In one case, atypical headbanging (head-slapping) was the initial and only RMD. There was no injury from the headbanging. Prenatal, perinatal, developmental, behavioral-psychological, medical-neurological, and family histories were negative. Clinical evaluations and nocturnal video-polysomnography with seizure montage were performed on all patients.

Results: Atypical headbanging was documented in all 3 cases; episodes always emerged late in the sleep cycle: from N2

sleep in 11 episodes, from REM sleep in 4 episodes, and from N1 sleep in 1 episode. Epileptiform activity was not detected. Clonazepam therapy was substantially effective in 1 case but not effective in 2 cases.

Conclusions: These 3 cases of idiopathic atypical headbanging expand the literature on this RMD variant, as to our knowledge only one previously documented case has been reported.

Keywords: Headbanging, jactatio capitis nocturna, sleep related rhythmic movement disorder, RMD, clonazepam, parasomnia, video-polysomnography, handedness, periodic limb movements/PLMs, central pattern generators, cyclic alternating pattern (CAP)

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Headbanging is a subtype of sleep related rhythmic movement disorder (RMD) and is classified in the Sleep Related Movement Disorder section of the International Classification of Sleep Disorders, 2nd Edition (ICSD-2).¹ It mainly affects infants and children, and involves large muscle groups (especially neck and trunk muscles) that engage in repetitive, stereotyped, and rhythmic movements that emerge predominantly during drowsiness or sleep, with a frequency of 0.5-2 sec, and with clustered episodes usually lasting less than 15 min. For diagnosis, there must also be a complaint of sleep interference, impaired daytime function, or self-inflicted bodily injury.¹ Headbanging typically involves repetitive lifting of the head or entire upper torso, and forcibly banging the head into the pillow or mattress (frontal banging); or slamming the back of the head against a headboard or wall (occipital banging). Headbanging can be combined with other forms of RMD, such as body rocking and head rolling.² Less common rhythmic movements include body rolling, leg banging, or leg rolling. We now report 3 cases of atypical headbanging with head punching and head slapping. We are aware of only one previously reported case, with video-polysomnographic (vPSG) documentation, of this self-hitting variant of *jactatio capitis nocturna* (headbanging).³

BRIEF SUMMARY

Current Knowledge/Study Rationale: 3 cases of atypical headbanging with head punching and head slapping are reported since there has only been one previously reported case, with video-polysomnographic documentation, of this self-hitting variant of *jactatio capitis nocturna* (headbanging). Typical headbanging involves banging of the head with the head forwards or backwards against the bed or bedside wall.

Study Impact: These 3 cases, together with a previously reported case, should encourage inclusion of the distinction between typical vs. atypical headbanging in the Sleep Related RMD section for the revision of the ICSD-2 that has recently been initiated by the American Academy of Sleep Medicine.

METHODS

Over 2 years, a series of 3 patients was gathered; the patients had presented to the sleep center of S-BY on account of longstanding nightly punching or slapping of the head that emerged mainly during nocturnal sleep and rarely from daytime naps. These patients were unaware of what was happening during sleep, and had no impairment of daytime function. Their headbanging made the parents concerned about the risk

for brain injury, which eventually prompted referral to the sleep clinic.

A comprehensive questionnaire covering lifetime sleep-wake, medical and behavioral-psychiatric history, and review of systems was completed by the parents for these 3 patients. Neurological examinations and psychiatric interviews were conducted. Neuropsychological testing was not performed, since there was no suspicion of at least mild pervasive developmental disorder or other problem in any patient, based on intact psychosocial functioning at home and at school, including interactions with classmates and friends. Brain MRI and awake and sleep EEG studies were performed. Serum liver function tests, BUN, creatinine, and fasting blood sugar levels were routinely performed; testing for heavy metals or other toxins was not done. There was no suspicion of substance abuse in any patient.

Overnight, hospital-based, vPSG monitoring, utilizing standard recording and scoring methods,⁴ and with EEG seizure montage, was then performed for the 3 patients. All medications were stopped ≥ 8 weeks prior to vPSG. The PSG monitoring included eye movements (electrooculogram); expanded EEG (12-channel seizure montage) with fast paper speed (1 cm/sec), submental and anterior tibialis electromyograms (EMG); oral-nasal airflow, chest and abdomen respiratory effort; electrocardiogram; and continuous, time-synchronized, audiovisual recording. Patient #1 had his parents sleep in the same room in a separate bed during his vPSG study. Patients #2 and 3 had their mothers sleep in the same room in a separate bed during their vPSG studies.

RESULTS

Case Presentations

Patient 1

A 13-year-old boy suffered from banging of his head into his pillow while sleeping in the prone position since the age of 3 years. These events usually happened in the early morning (4-6 a.m.) nearly every night and several times per night. His father was afraid that the frequent headbanging might cause brain injury, so he purchased a softer bed and softer pillow for his son. At the age of 6 years, without any identified trigger, the patient shifted his headbanging into the pillow to frontal head punching, for which he was always amnesic. The patient was referred to several pediatric neurologists. Neurologic histories and exams were normal. Awake and sleep EEG, and brain MRI were also normal. He was prescribed clonazepam for what was regarded as bizarre nocturnal behavior, with little or no effect. The patient considered his sleep to be normal, and he and his parents denied any daytime somnolence or functional impairment. Seven years after the emergence of nocturnal frontal head punching, there was a spontaneous increase in its severity for 6 months leading up to the time the parents brought him to the sleep clinic. Medical history was benign. He was a normal full-term baby with excellent lifelong health, including after the onset of headbanging. There was no history of behavioral problems at home or at school. Family history was negative. Physical and neurological exams were normal.

Patient 2

An 11-year-old boy suffered from head-slapping during his night sleep since he was 5 years old. Episodes occurred as clustered events several times nightly during nearly every night, with each event lasting several seconds, with either or both hands slapping his parietal area. The patient was never aware of these events, but his parents were disturbed by the sharp and clearly slapping sounds during his night sleep. They were particularly concerned about whether brain injury would result from the strong and repeated head slapping. He was evaluated by a pediatric neurologist soon after the onset of the sleep related head slapping. Physical and neurological exams were normal. Awake and sleep EEG and brain MRI were unremarkable. Nevertheless, a nocturnal seizure disorder was diagnosed and sodium valproate was prescribed, with poor response, and then various other anti-epileptic medications were prescribed (including clonazepam) during the subsequent 5-6 years, without benefit. Another pediatric neurologist eventually diagnosed a sleep disorder rather than a seizure disorder. The patient was then brought to the sleep clinic for evaluation. Medical history was benign. He was a full-term baby. His general health was excellent, he achieved developmental milestones normally, and there were no behavioral or emotional problems. Family history was non-contributory.

Patient 3

A 22-year-old female suffered from headbanging since she was 1 year-old. Headbanging episodes always occurred during transitions from sleep to wake and were accompanied by crying, but there was never dream recall, nor recall for the events. She would turn to the prone position and bang her head into the pillow for several minutes. When her mother picked her up from bed and held her, she would continue to bang her head into her mother's shoulder. Consultation with a pediatric neurologist did not identify any neurological disorder, but the sleep problem was not addressed. Head-punching during sleep began spontaneously at the age of 5 years, with episodes gradually becoming concentrated into nocturnal sleep, most often around midnight (and occasionally during daytime naps). She usually punched the front of her head with either hand, but mostly with the right hand. At times her mother observed that she would still bang her head into the bedside wall. Also, if her mother held her hand to prevent head-punching, she would then bang her head into the wall by her bed. Subsequently, the emergence of the episodes migrated into the early morning hours. When her mother observed that this problem had been escalating since age 16 years, she sought a neurologic consultation for her daughter. Physical and neurologic exams, brain MRI and EEG were unremarkable. Clonazepam 0.5 mg at bedtime reduced the frequency of these episodes from 10+ times nightly to 2-3 times nightly. When the neurologist increased the clonazepam dose to 2 mg at bedtime, the patient felt excessive daytime sedation, so the dose was reduced back to 0.5 mg. She then presented to the sleep clinic. Medical history was benign. She was a full-term baby, without any abnormal developmental, medical, neurological or behavioral-emotional history. Family history was negative. Physical

and neurological exams were unremarkable and the patient denied any daytime sleepiness.

In 2 of the cases (#1 and #3), typical headbanging emerged first and subsequently evolved after a number of years—for no identified reason—into almost exclusively atypical headbanging consisting of head-punching. Case #2 had a history of exclusively atypical headbanging manifesting as head-slapping, but without ever demonstrating any typical headbanging (with use of the head). Pt. #3 had (non-rhythmic) crying that accompanied her headbanging episodes. In none of the 3 cases was there reported or elicited symptoms suggestive of other sleep pathology, such as sleepwalking, sleep terrors, dream-enactment, restless legs syndrome, propriospinal myoclonus, excessive movements, sleep bruxism or tongue-biting, sleep related scratching, snoring, non-restorative sleep, or daytime somnolence. None of the children considered his or her sleep to be disturbed or abnormal. They were all healthy children with normal IQs who were devoid of medical, neurological or psychological-behavioral problems. All brain MRIs and blood test results were unremarkable. There was no history of head injury or loss of consciousness. There was also no history of any wakeful tendency for verbal or physical aggression directed towards others or themselves (e.g., self-hitting, self-biting, self-cutting), and the patients were well-adjusted in all spheres of their lives. They all came from upper middle class, university-educated families. The female patient is currently a university student, and both boys are junior high school students. The uncle of one boy is a physician and Chief of Family Medicine at the hospital in Chiayi, Taiwan where the sleep center that evaluated the 3 patients is located.

All 3 cases were right-hand predominant, and although the head-punching and slapping were observed by the parents to occur with either hand, it was predominantly with the dominant right hand. With patient #2, the mother would occasionally observe simultaneous slapping of the head with both hands. With patient #1, one episode of strictly left-handed headbanging was followed by an episode with strictly right-handed headbanging during the vPSG study.

Parental concerns over brain injury from the repeated head-punching and head-slapping that occurred for years during sleep had prompted medical evaluation in the first 2 cases, and parental concern over intensification of the headbanging at age 16 in the third case had prompted medical evaluation. However, none of the patients complained of headaches, or had sustained head injury or other injury from their atypical headbanging. In contrast, the parents of all 3 patients, especially those of patient #2, complained of having their sleep repeatedly interrupted by their child's head-slapping, even when they slept in a separate bedroom (as with patient #2). They would wake up several times nightly to check on the status of their children. Nevertheless, none of the parents complained of non-restorative sleep upon arising in the morning.

Misdiagnosis of nocturnal seizures occurred in the second case, with multiple unsuccessful antiepileptic medication trials lasting years that ultimately resulted in parental concern, when the diagnosis of a sleep disorder was established, about possible adverse long-term effects from these medications. Clonazepam therapy, often effective in RMD, was not effective in the first 2 cases but was substantially effective in the third case.

Video-Polysomnographic Results

During their vPSG monitoring, patients #1 and #3 demonstrated repetitive head punching/hitting during their single vPSG studies. Patient #2 had 2 vPSG studies, since the first night was devoid of any episode; rhythmic head-slapping appeared during the second vPSG. **Table 1** contains the clinical and vPSG data.

Patient #1 had normal sleep latency, moderately increased N1 sleep, and a substantial reduction of REM sleep of about 50%. Otherwise, sleep was unremarkable, apart from 4 episodes of quasi-rhythmic fist-punching over the frontal head. **Figure 1** shows the PSG correlates of the first episode that emerged from REM sleep.

Patient #2 had normal sleep latency, substantially increased N2 sleep, and substantially reduced N3 sleep and REM sleep (by about 50% each stage). Periodic leg movements (PLMs) unassociated with arousals were present during both vPSGs. Sleep was otherwise unremarkable, apart from one precipitous episode of violent, rhythmic head slapping emerging from N2 sleep. **Figure 2** shows the PSG correlates of the episode of violent head-slapping.

Patient #3 had a normal sleep latency, and a substantial reduction (by > 50%) of REM sleep (9.7%). Otherwise, sleep was unremarkable, apart from 11 episodes of head-hitting that emerged mainly from N2 sleep, but also from REM sleep and N1 sleep. Events emerging from N2 sleep (8 episodes) usually were immediately preceded by a K-complex. **Figure 3** shows the PSG correlates of the final (11th) episode of head-hitting.

Table 2 contains the vPSG data for the documented headbanging episodes. During the vPSG studies, patient #2 showed the most rhythmic headbanging, whereas patients #1 and #3 showed both rhythmic and quasi-rhythmic headbanging. The hand or any other part of the body was never bitten during any episode. The EEG accompanying the atypical headbanging episodes remained consistently asleep and without post-episode arousals. This EEG finding is typical for RMD.

Patient #1 RMD Headbanging Episodes

Time of onset of episodes #1-4 was as follows: (03:36:33) (03:38:41) (05:10:02) (05:12:47). Onset-to-onset intervals for the successive episodes were: 2 min 8 sec; 1 h 31+ min; 2 min 45 sec. The first 3 episodes involved strictly right-handed headbanging and other hitting, and the last episode involved strictly left-handed headbanging. The same strength was shown in headbanging with either the right or left hand, as demonstrated by the video examples for the second (right hand) and fourth (left hand) episodes.

Description of the second episode (uploaded video): At 03:38:41 during REM sleep, while supine and with knees drawn up, the *right hand* in a fist punches the forehead with the back of the hand moderately hard 5 times in 8 sec, then there is a 5-sec pause, followed by one hit, then a 7-sec pause, then one hit, followed by a 12-sec pause, and finally he lifts the right hand as if ready to come down with another hit, but instead he puts the hand on his face briefly and then moves his hand over his chest. Episode ends at 03:39:15. The parent sleeping in a nearby bed aroused during this episode from the noise of the hitting, and upon cessation of the episode, the child and parent started talking.

Table 1—Clinical and video-polysomnographic (vPSG) data in 3 atypical headbanging patients

	Patient #1	Patient #2 ¹	Patient #3
Gender	Male	Male	Female
Age, years	13	11	22
Age, onset of RMD ² , years	3	5	1
Age, onset of head-punching/head-slapping RMD, years	6	5	5
vPSG Data			
Sleep-onset time	23:56:22	00:17:49	23:41:32
Total sleep time (TST), min.	410.5	(377) ³ 453.0	421.5
Sleep efficiency, %	92.5	(98.0) ³ 97.8	92.3
Sleep latency, min	20.0	9.5	5.0
REM sleep latency, min	123.0	(102.5) ³ 122.5	91.0
N1% TST	10.9	(1.2) ³ 1.2	4.2
N2% TST	57.0	(74.1) ³ 76.6	58.5
N3% TST	21.0	(6.0) ³ 7.7	19.7
REM% TST	11.1	(15.8) ³ 12.2	9.7
REM sleep without atonia	—	—	—
Periodic Limb Movements, index	—	(13.7) ³ 8.5/h	—
Periodic Limb Movements, total	(86) ³ 64		
Snoring/Apnea-hypopnea/O ₂ desaturations/arrhythmias	—	—	—
EEG epileptiform activity	—	—	—
Movement time, %TST	0.5	(0.9) ³ 1.0	0.7
Event time (RMD), %TST	0.9	0.1	1.7
# RMD episodes	4	1	11
Total NREM/REM cycles	4	4	5
RMD in NREM/REM cycle #	2, 3	3	4, 5

¹Patient #2: data for the second night of 2 consecutive vPSG studies, as there was no RMD during the first vPSG study. ²Patients #1 and #3 had onset of typical frontal-occipital headbanging as the initial form of RMD before the atypical head-punching variant of RMD subsequently emerged. Patient #2 had head-slapping as the initial and only form of (atypical) RMD. ³First night PSG study, devoid of RMD, for patient #2. (36 night separation between vPSG #1 and #2).

Description of the fourth (final) episode (uploaded video):

At 05:12:47 during N2 sleep, while supine and knees drawn up, the *left hand* in a poorly formed fist hits the forehead with the back of the hand moderately hard 4 times in 4 sec, then there is a 6-sec pause, followed by one more hit, and the left arm is lifted up, extended in the air for 5 sec before coming back down. Episode ends at 5:13:01.

Patient #2 RMD Headbanging Episode

Description of the episode (uploaded video): At 04:53:38, during N2 sleep (**Figure 2**), while supine with knees drawn up, the right hand hits the right parietal skull with 8 very hard and fast slaps in 5 sec, ending at 04:53:43. The open hand was rotated so that the palm was pounding the head with full aggression. Early in the episode, the left arm drifts to the left.

Patient #3 RMD Headbanging Episodes

Time of onset of episodes was as follows: (05:26:27) (05:28:40) (05:32:28) (05:33:53) (05:42:55) (05:48:25) (06:37:49) (06:40:32) (06:41:45) (06:43:05) (06:46:44). Onset-to-onset intervals for each of the successive episodes were: 2 min 13 sec; 3 min 48 sec; 1 min 25 sec; 9 min 2 sec; 5 min 30 sec; 49 min 24 sec; 2 min 43 sec; 1 min 13 sec; 1 min 20 sec; 3 min 39 sec. Range: 1 min 13 sec to 49 min 24 sec.

Description of Episode #11 (uploaded video):

At 06:46:44, during N2 sleep (**Figure 3**), while supine and the right knee drawn up, the right hand, without a fist being made, hits the forehead with the back of the hand moderately hard 6 times in 6 sec, followed by a 4-sec pause, then he hits the head 4 times in 3 sec, then a 4-sec pause, then hits herself 3 times in 3 sec, then a 3-sec pause, then hits herself 3 times in 2 sec, and the episode ends at 06:47:09. In this sequence, she never made a fist and never rotated her arm while hitting, so it appeared that she was hitting herself without purpose or without aggression with the back of the forearm/wrist—while the right wrist was extended (“cocked”) in a peculiar manner. During this episode, the left arm was fully extended and motionless. This form of hitting was uniformly present in all 11 headbanging episodes during the vPSG study.

DISCUSSION

Clinical Findings

Our 3 RMD cases with atypical headbanging involved the use of the hand for either striking the head with the fist/knuckles, slapping the head with an open palm, or striking the head with the back of the hand/wrist/forearm. In 2 cases, typical rhythmic headbanging (with use of the head) shifted spontaneously to

Figure 1— PSG correlates of repetitive head-punching by patient #1

Red arrows indicate the multi-channel artifacts caused by repetitive head-punching during REM sleep beginning at 03:36:33 a.m. EEG: channels 2-5; 12-19. EOG: channels 7-8. EMG: submental (channel 6); left/right anterior tibialis (channels 9-10). ECG: channel 11. Oral-nasal airflow: channel 20. Chest/abdominal respiratory effort: channels 21-22. SpO₂: channel 23.

atypical headbanging after 3-4 years, with decay in rhythmicity. In one case, head-slapping was the initial and only form of atypical headbanging. Therefore, atypical headbanging reflects a dynamic process in which multiple forms of head-hitting with the hands during sleep can coexist, and can also shift from one form into another form during the long-term evolution of the RMD, and can shift from rhythmicity to quasi-rhythmicity.

The bizarre clinical presentation confused primary care physicians and pediatric neurologists, indicating how headbanging can be prone to misdiagnosis as a nocturnal behavior problem or a seizure disorder, with resulting inappropriate treatment. An important point for differential diagnosis is that punching or slapping the head with either hand, or both hands simultaneously, during the episodes speaks strongly against seizures because of the lack of stereotypy. At times RMD is the sole manifestation of nocturnal seizures.⁵

We consider that the ICSD-2 diagnostic criteria for RMD in these 3 cases were satisfied in regards to “self-inflicted bodily injury,” since repeated (hard) blows to the head on a nightly basis for years posed a serious risk for injury. However, they did not satisfy either of the other 2 diagnostic criteria for RMD, involving complaints of sleep interference or impaired daytime functioning. All parents complained about their own sleep interference from their child’s headbanging, but without daytime

impact. In regards to risk of injury from typical or atypical headbanging, not only the level of force and violence should be considered, but also “victim vulnerability factors” should be considered,⁶ as less forceful blows to the head could cause injury in the setting of hemophilia or other bleeding disorder; anticoagulant therapy; prior head injury⁷; CNS disorder; or other medical vulnerability factors.⁶ The threshold for initiating treatment should be influenced by both these clinical risk-related factors. In this regard, the natural history of atypical headbanging in our 3 cases is unknown, and may involve progressive or episodic increase in severity over time, as shown by patient #3, with increased risk for injury.

Therefore, given the lack of any neuropsychiatric disorder that could be linked with atypical headbanging, such as autism or mental retardation,^{8,9} and given the lack of daytime aggressive tendencies or any psychodynamic factors that could promote sleep related self-hitting, as was previously reported in one case,³ our 3 cases represent idiopathic, sleep related RMD. There was never self-awareness during the episodes and no subsequent recall for the episodes. There was also no associated dreaming even though RMD emerged from REM sleep at times. The underlying mechanisms would most likely involve rhythmic (or non-rhythmic) activation during sleep and sleep-wake transitions, of subcortical central (motor) pattern genera-

Figure 2—PSG correlates of violent repetitive head-slapping by patient #2

Multi-channel artifacts caused by rhythmic, violent head-slapping from N2 sleep (4:53:37 a.m. to 4:53:43 a.m.). Polygraphic channels: same as for **Figure 1**.

Table 2—Atypical headbanging episodes: vPSG data

	Patient #1	Patient #2 ¹	Patient #3
Time, sleep onset	23:56:22	00:17:49	23:41:32
Time, onset of first episode	03:36:33	04:53:38	05:26:27
Latency, sleep onset to onset of first episode	3 h 40+ min	4 h 35+ min	5 h 44+ min
RMD episode #	Sleep stage (duration)		
#1	REM (47 sec)	N2 (5 sec)	N2 (35 sec)
#2	REM (34 sec)		N2 (38 sec)
#3	N2 (38 sec)		REM (20 sec)
#4	N2 (14 sec)		N1 (69 sec)
#5			REM (25 sec)
#6			N2 (39 sec)
#7			N2 (25 sec)
#8			N2 (33 sec)
#9			N2 (15 sec)
#10			N2 (18 sec)
#11			N2 (28 sec)
Mean $\pm$ SD, duration of RMD episode (sec)	33.2 $\pm$ 13.9	5.0	31.4 $\pm$ 14.8
range (sec)	14-47	N/A	15-69

Figure 3—PSG correlates of repetitive head-hitting by patient #3

An episode of recurrent head-hitting initiated by a K-complex (channels 13-14 & 17-18) during N2 sleep beginning at 06:46:44. Red arrows indicate the multi-channel EEG artifacts caused by the head-hitting. Polygraphic channels: same as for **Figures 1, 2**.

tors (CPGs) located in the brainstem and spinal cord.^{10,11} It is unknown how certain muscle groups are preferentially selected, and how vigorous and potentially injurious self-hitting behaviors (focused exclusively on the head) are repetitively activated.

Video-Polysomnographic Findings

The vPSG results are compatible with those from 2 previous studies: (i) preponderance of atypical headbanging arising from N2 sleep was also found by Dyken et al.¹²; (ii) a study by Mayer et al. of 24 patients with typical RMD found that episodes were not restricted to sleep-wake transition, and in fact occurred most frequently in wake, N1, and N2, but also in REM sleep and N3 sleep.¹³ All 24 cases were free of brain pathology. Their findings were highly congruent with the findings in our 3 cases, apart from the difference between typical and atypical headbanging.

The PSG data for all 3 patients indicated that they were good sleepers, with normal sleep latency and high SE%. However, in all 3 patients, REM sleep% was consistently reduced by half of normal (9.7%-15.8%). Patient #2 (with PLMs on both nights): had low N3 and low REM sleep% both nights—with a robust increase of N2 (to 74%-77%), but no increase in N1 (1.2%-1.2%). The alterations in sleep architecture just described for the 3 patients may reflect a “first night effect” in the sleep lab. Nevertheless, there was probably no clinical consequence

from these sleep architecture alterations, especially since none of the patients (or their parents) had a complaint about their sleep or waking state. The finding of a K-complex signaling the onset of all 8 episodes coming during N2 sleep in patient #3 is most likely explained by the onset of the activated phase of the “cyclic alternating pattern,” i.e., “A phase of the CAP” that facilitates release of abnormal behaviors.¹⁴ This K-complex finding was also reported in a 41-year-old man with leg-kicking RMD, whose episodes during all sleep stages and wake-sleep transitions were often preceded by high-voltage K-complexes¹⁵ (PLMs were absent in this patient).

Previously Reported Case of Atypical Headbanging

To our knowledge, there is only one previously reported case involving the same atypical variant of headbanging RMD as with our 3 cases, with self-punching of the forehead and face.³ A 24-year-old man had rhythmically pounded himself with his knuckles during sleep since the age of 2 years. From the ages of 2 to 15 years, he had placed one hand on his forehead and then repeatedly hit that hand with the other hand. At age 15 years, he started to directly pound his forehead without the protection of the other hand, and these episodes occurred multiple times nightly throughout sleep during virtually every night. Episodes were often brief, but could last as long as 45 minutes. He was always amnesic for these events. In contrast to our 3 cases,

this man sustained serious injuries, such as bleeding, suppurative wounds, and facial scar-related disfigurement. Psychiatric evaluation uncovered major psychodynamic problems, with his harboring persistent rage towards his parents.

Two consecutive vPSGs (without mention of a seizure montage) documented 20 episodes (18 during REM sleep) of atypical headbanging that was “at all times rhythmic and stereotypical” with variable strength “and at times were quite impressive in their force; each strong blow produced a distinctive hollow thud.” Although the man was right-handed, he struck himself with both hands, “and at one point switching hands in the middle of an episode.” The episodes from REM sleep were marked by the head-pounding beginning 1-2 minutes after REM-onset and persisting into N1 sleep in 11/18 episodes. EEG during the episodes was unremarkable, sleep architecture was intact, and there was no comorbid sleep disorder. There was a positive family history for RMD involving an 18-year-old cousin with persistent, typical headbanging.

Given this family history, together with the clinical and vPSG findings, the authors concluded that their patient had a variant of typical headbanging in which the hand rather than the head was mobilized for banging. The unique self-destructive quality of the head-pounding was speculated to result from a powerful psychodynamic drive in which the patient’s latent rage towards his parents was released during sleep, taking the form of the familial predisposition to headbanging RMD. (Presumably in this construct, if there were a family history of sleepwalking or sleep terrors, then this psychodynamic rage would have been released as a NREM parasomnia).

Final Comments

A number of findings and observations from our 3 cases of atypical headbanging, together with the previously reported case, should be highlighted:

- (1) Five forms of sleep related motor dyscontrol were documented:
 - (i) the head bangs itself rhythmically, frontwards and/or backwards
 - (ii) the head is banged rhythmically/quasi-rhythmically by the upper extremity
 - (a) fist/knuckles punching
 - (b) slapping with the open palm
 - (c) hitting with the back of the hand/wrist/forearm
 - (iii) crying during RMD events
 - (iv) PLMs during NREM sleep (on 2 consecutive vPSG studies in one patient)
 - (v) shifts in handedness in the self-hitting
 - (a) during the same episode
 - (b) across consecutive episodes
 - (c) simultaneous bilateral head-punching during atypical headbanging episodes
- (2) The shifting dominance of handedness emerging with RMD, including atypical headbanging, calls attention to shifting language dominance emerging with sleeptalking (another form of motor dyscontrol in sleep) in some bilingual sleeptalkers (Spanish, Euskera).¹⁶
- (3) The spontaneous shifting from typical to atypical headbanging, with change in rhythmicity, after 3-4 years in 2 patients was also present in a case of complex RMD

involving a 16-year-old male who had spontaneous shifts across 4 different types of RMD, including typical headbanging.¹⁷

- (4) The long latencies from sleep onset to onset of headbanging in all 3 patients during their vPSG studies, ranging from 3 h 40+ min to 5 h 44+ min, together with the absence of wake-sleep transitional RMD, should be noted and may suggest a unique profile for late nocturnal sleep cycle atypical RMD without transitional RMD wake-sleep release.
- (5) Aggressive and non-aggressive atypical headbanging, in various forms, were documented by the vPSG studies.
- (6) The strong suppression of REM sleep (by > 50% of normal) linked with normal (or slightly prolonged) REM latency, normal number of NREM/REM sleep cycles (4-5 cycles/patient), and very high sleep efficiency is rare, and may be another feature of a unique profile for this group of patients. Substantial REM sleep suppression is usually a medication effect, or a result of a neurologic disorder, sleep disruption related to sleep disordered breathing or other factors, or very prolonged REM latency.
- (7) The breakdown in rhythmicity of RMD evident in the evolution of typical headbanging (highly rhythmic) to atypical headbanging (quasi-rhythmic, non-rhythmic) suggests reduced precision of the CPG pacemakers in the brainstem and spinal cord, by unknown mechanisms.
- (8) The knees were always drawn up during the atypical headbanging episodes, which may be part of the symptom complex for atypical headbanging, or may be a common supine sleeping posture for these patients.
- (9) Lack of neuropsychiatric disease with adult persistence of atypical headbanging in patient #3 was also found in various cases of adult persistence of typical headbanging¹⁸⁻²¹ (and in other cases, as reviewed).²²
- (10) Clonazepam therapy was reported to be effective in isolated cases of typical headbanging^{17,20,23} but was effective in only 1 of our 3 patients. Imipramine was effective in 2 typical headbanging cases.^{7,21}

In conclusion, our 3 cases together with the previously reported case³ should encourage inclusion of the distinction between typical vs. atypical headbanging in the Sleep Related RMD section for the revision of the ICSD-2¹ that has recently been initiated by the American Academy of Sleep Medicine.

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