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CASE REPORTS

Two cases of exploding head syndrome documented by polysomnography that improved after treatment

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Exploding head syndrome is a rare sleep disorder, characterized by an explosive feeling in the head, that occurs during the sleep-wake transition. Usually the attacks are painless, but the fear caused by the attack can result in awakening and insomnia when it is generated frequently. It has been suspected that exploding head syndrome is related to emotional stress, because most patients report stressful life situations in periods when attacks are intense and frequent. The benign character and good prognosis of exploding head syndrome are the most likely reasons why it has not become a subject of more extensive neurologic research. Moreover, most of the articles reported symptomatic episodes but a lack of objective physiologic examinations, such as polysomnography, and effective treatment. Here, we report two cases of exploding head syndrome with the attacks documented by polysomnography and our trial treatment.

Keywords: exploding head syndrome; tinnitus; PSD; electroconvulsive treatment; Ménière's disease

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INTRODUCTION

Exploding head syndrome (EHS) is a rare sleep disorder, characterized by an explosive feeling in the head that occurs during the sleep-wake transition.[1,2] The attacks are characterized by sudden loud noises, described as being like a bomb explosion, gunshot, door slamming, roar, or buzzing noise. Usually the attacks are painless, but the fear caused by the attack can result in awakening and insomnia when it is generated frequently. It has been suspected that EHS is related to emotional stress, because most patients report stressful life situations in periods when attacks are intense and frequent. The benign character and good prognosis of EHS are the most likely reasons why it has not become a subject of more extensive neurologic research. In a search of PubMed with the key phrase of "exploding head syndrome" in January 2020, there were only 44 references up to that time. Moreover, most of the articles reported symptomatic episodes but a lack of objective physiologic examinations, such as polysomnography (PSG), and effective treatment. Here, we report 2 cases of EHS with the attacks documented by PSG and our trial treatment.

REPORT OF CASE

PSG

The 2 participants underwent full-night PSG, and a sleep medicine physician interpreted the results at the Good Sleep Center, Nagoya City University Hospital. The participants were continuously observed by a PSG technician and were recorded on video with the use of an infrared video camera.

An Alice 5 PSG system (Respironics, Inc., Tokyo, Japan) was used to record the following: electroencephalogram, electro-oculogram, chin electromyogram, airflow, nasal pressure, rib cage and abdominal movements, arterial oxygen saturation, snoring, electrocardiography, anterior tibialis electromyogram, and body position. The records were scored according to standard criteria.[3]

The various indices of sleep architecture analyzed were total sleep time, sleep efficiency (total sleep time divided by time in bed), wake after sleep onset, and arousal index. Sleep disturbance events were scored apnea, hypopnea, snoring, and leg movement.

Additionally, arousals were classified in related events: respiratory events (apnea, + hypopnea, + snoring), leg movement, and spontaneous arousal without any event.

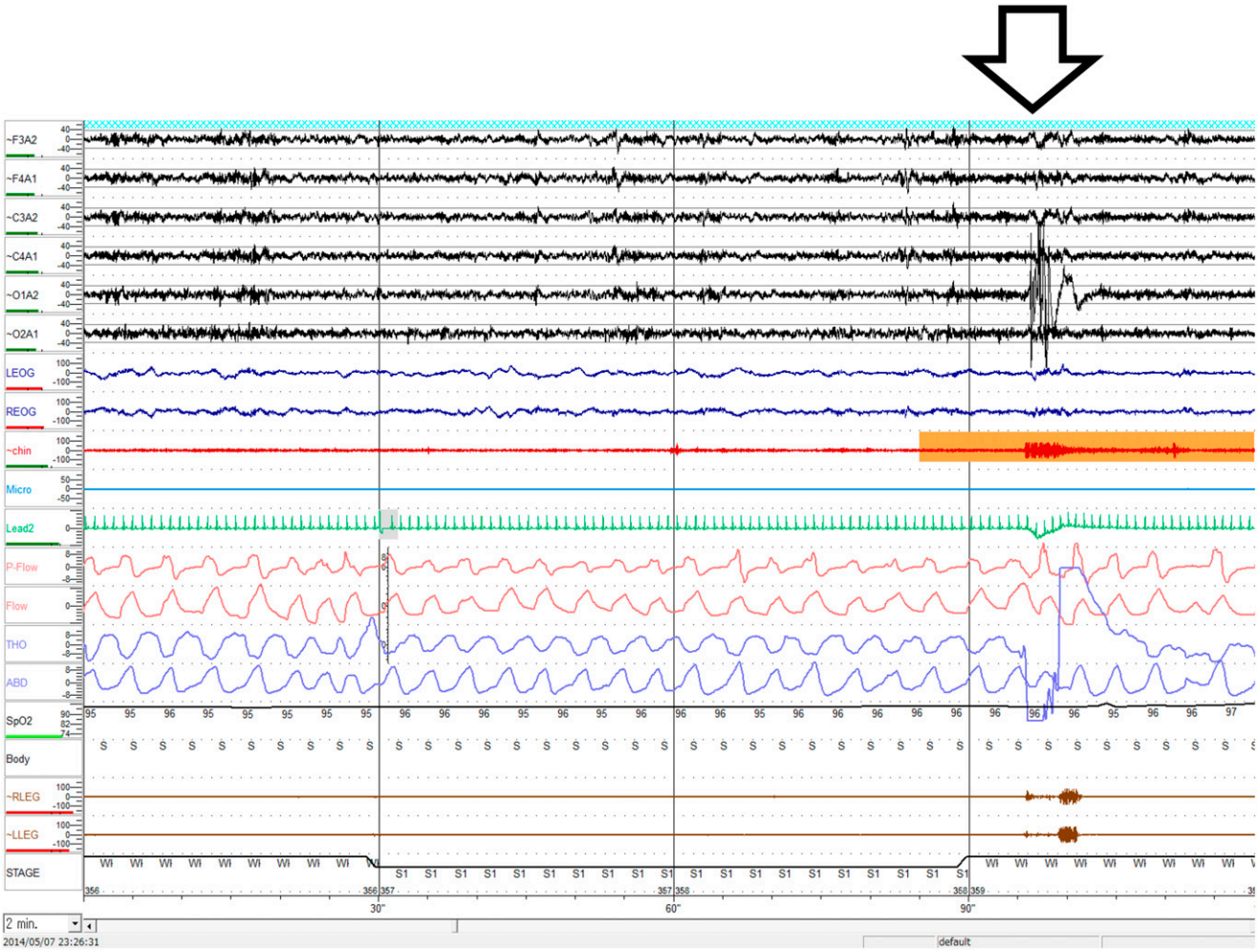
Case 1

Clinical consequence

A 71-year-old woman with a body mass index of 38.1, who had been medicated for left Ménière's disease at Osaka University, was referred to us because she had jerking movement of legs and was occasionally awaked by an explosive noise at the time she fell asleep. Although she had left tinnitus, with a constantly "zee" sound, the tinnitus did not disturb her daily life or her sleep. The explosive noise appeared in the late night and was completely different from the tinnitus. She reported that the tinnitus presented inside of the left ear, but the explosive noise was inside of her brain. The explosive noise attacks occurred approximately once a week, and she had a severe attack, with 4–5 explosions, the day before her full-night PSG.

By the time she first visited us, her Ménière's disease was well controlled. Other conditions including primary and secondary headache disorders and nocturnal seizures were excluded.

Figure 1—Moment of awake by EHS in case 1.



An explosive noise (a loud banging sound) attacked her around 2230 hours, correlating with the transition from wakefulness to non–rapid eye movement stage 1.

PSG result

An explosive noise (a loud banging sound) attacked her around 2230 hours, correlating with the transition from wakefulness to non–rapid eye movement (REM) sleep stage 1 (**Figure 1**). Sleep latency was 82.0 minutes and might have been prolonged because of the restless legs syndrome. Wake time after sleep onset was 155.0 minutes, sleep efficiency was 46.3%, and arousal index was 30.3. Moderate sleep apnea syndrome with an apnea-hypopnea index of 17.9 events/h was observed. Respiratory event with arousal index was 17.4, and spontaneous arousal index was 8.2. No evidence of seizure activity was noted during monitoring.

Effective oral appliance treatment

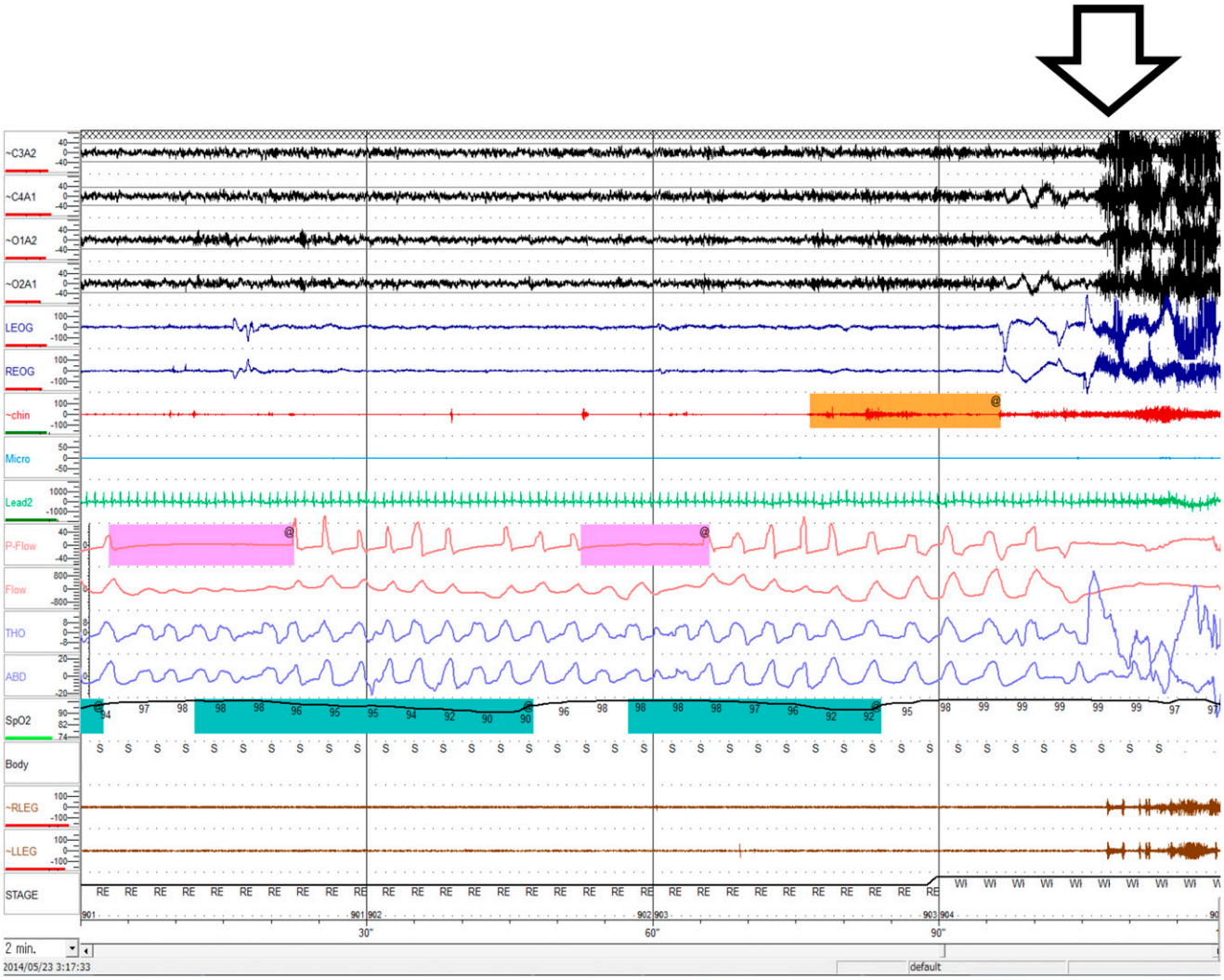
Rotigotine 2.25 mg/d was prescribed from the day after the tests, and there were no side effects such as hallucinations, delusions, or nausea. She reported that her restless legs syndrome symptoms improved remarkably, but there was no difference in the frequency of the EHS attacks. After starting an oral appliance in the next month, she reported a lower frequency of the explosive noises; 4–5 EHS attacks before treatment decreased to 1 for the first month and completely disappeared after 3 months.

Case 2

Clinical consequence

A 55-year-old woman with a body mass index of 28.9 kg/m^2 had frequent excessive daytime sleepiness, and her husband complained of her snoring during sleep. During the previous 2 years, she had been occasionally awakened by a sound like the popping of a balloon during sleep. These attacks occurred 3 times per week, often around 0400 hours. She was referred to us by the Department of Psychiatry of our hospital because of daytime sleepiness and suspected sleep apnea syndrome. Before she visited our Department of Psychiatry, she had severe depression for 7 years while being treated at another psychiatry clinic. She was medicated at our Department of Psychiatry with sertraline hydrochloride 25 mg 4 times a day, duloxetine hydrochloride 20 mg 3 times a day, lithium carbonate 200 mg and clonazepam

Figure 2—Moment of awake by EHS in case 2.



There was video confirmation of attack correlating with the transition from rapid eye movement sleep to awake around 0330 hours.

0.5 mg twice a day, and etizolam 1 mg, brotizolam 0.25 mg, and loxoprofen 60 mg once a day.

PSG result

There was video confirmation of attack correlating with the transition from REM sleep to awake around 0330 hours (**Figure 2**). Sleep onset was 17.5 minutes, sleep efficiency was 89.2%, and arousal index was 16.0. Mild sleep apnea syndrome with an apnea-hypopnea index of 11.3 events/h was observed. Respiratory arousal index was 4.1. Spontaneous arousal index was 11.7. No evidence of seizure activity was noted during monitoring.

Effective electroconvulsive treatment

She was only prescribed an oral appliance, and an oral appliance evaluation with PSG was done 4 months after the first PSG. Her sleep breathing disorder was shown to have improved from an apnea-hypopnea index of 11.3 to 1.2 events/h on the second PSG. However, she had another EHS attack during the second PSG, similar to the first PSG, at around 0345 hours when awakening from REM sleep. Unfortunately, her EHS attacks were not reduced even though her sleep apnea improved with the oral appliance.

Nine months after her last PSG, she underwent electroconvulsive treatment 8 times at our Department of Psychiatry for her depression. Surprisingly, this improved not only her depression but also the EHS. Although we were unable to perform PSG again after her improvement, she reported the EHS had completely disappeared soon after the first electroconvulsive treatment, and the quality of her entire sleep was much improved compared with before the electroconvulsive treatment.

DISCUSSION

EHS is a rare sleep disorder, and only a small number of PSG studies of EHS have been reported. Electroencephalogram data indicate that patients with EHS often misperceive when their

episodes actually occur. Although often reporting the noises when they are "asleep," electroencephalograms indicate that alpha activity predominates along with short episodes of theta.[4,5] No studies to date have observed EHS during verified sleep but only in periods of relaxed (yet awake) drowsiness. In our study, the EHS in case 1 had onset during the transition from awake to non-REM sleep stage 1, and in case 2, it was during the transition from REM sleep to awake around 0330 hours. This is the first report to indicate that EHS may happen during REM. The conflict between patients' and physicians' reports as to whether EHS occurs during sleep or not may be because of the small number of cases along with insufficiency of PSG documentation. There are also problems in that patients may not understand why PSG is necessary and in insurance coverage. However, scientific elucidation of EHS with PSG is needed for more detailed investigation of this condition in the future.

In 1989, Pearce[6] reported a remarkably large number of EHS cases in 54 patients, and that article increased interest in EHS among physicians. Most patients in that report were diagnosed with EHS from episodes only, and 4 patients were excluded because of inadequate historical data or features suggestive of primary otologic disease with prominent tinnitus. In our study, the patient in case 1 had Ménière's disease, and she did not complain of EHS until we asked about her sleep quality. She did not associate the EHS with the Ménière's disease because the 2 had completely different noises and different times of onset. In addition, EHS as classified in ICSD III[2] is not ruled out by the presence of otologic disease. Thus, we are uncertain as to why Pearce excluded EHS patients with concurrent otologic diseases. Because patient episodes involve various sounds, such as loud bangs, explosions, shotgun sounds, thunderclaps, and loud metallic noises, there is a possibility that patients may visit otolaryngologists. In fact, a Japanese group reported that 4 EHS patients visited otolaryngologists because they thought the EHS was related to the inner ear or because EHS occurred alongside tinnitus.[7] EHS does not directly occur from the auditory system, but we could not find any reason to rule out EHS only because it occurred in combination with otologic diseases. The relation between EHS and the auditory system is controversial, and we wish to call attention to the fact that EHS patients may complain of it as an auditory problem to physicians, especially those with specialties related to the auditory system. EHS is rare and not well known even to sleep physicians. It is our hope that all physicians will pay more attention to elucidating this unknown sleep disorder.

No open or controlled clinical trials on EHS have yet been conducted. However, several case studies have documented effective pharmacologic treatments, such as the tricyclic antidepressant clomipramine, calcium channel blockers, or anticonvulsants.[4] Nonpharmacologic treatments may also be effective. Several researchers have noted the effectiveness of education and reassurance to patients that EHS is a fairly benign condition.[8] It is also possible that treating other sleep disorders first may result in reductions of EHS. Okura et al[9] described the case of a patient with comorbid obstructive sleep apnea. When the patient began using an oral appliance, no further EHS attacks were reported. We strongly support the approach of treating other sleep disorders as the first line of treatment for EHS, as in our first case, in which a cure was achieved with an oral appliance before pharmacologic treatment, because evidence for pharmacologic treatments is lacking. The ETC in the second case is the first description of ETC as being possibly effective for EHS. However, ETC in this case was the treatment for depression, not for EHS, and it was found by chance that EHS improved with the resolution of depression. Although we have no evidence to prove whether ETC directly affected EHS, this may indicate a possible association between depression and EHS, and that by treating concurrent diseases, EHS may also be cured.

ABBREVIATIONS

EHS, exploding head syndrome
PSG, polysomnography
REM, rapid eye movement

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DISCLOSURE STATEMENT

All authors have seen and approved the manuscript. Work for this study was performed at Good Sleep Center, Nagoya City University Hospital. This is a case report but not a clinical trial and received ethic permission from patients and the facility (NCU-44-01). The authors report no conflicts of interest.