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Video-Clinical Corners

Characteristic symptoms of exploding head syndrome in two male patients

Xiaoli Wang, Wenjuan Zhang, Na Yuan, Ying Zhang, Yonghong Liu*

Epilepsy and Sleep Disorders Unit, Department of Neurology, Xijng Hospital, The Fourth Military Medical University, Xi'an, 710032, China

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1. Introduction

Exploding head syndrome (EHS) is a rare sensory parasomnia, characterized by a sudden loud noises or sense of explosion in the head either at the wake–sleep transition or upon waking during the night [1,2]. The attacks are painless but unpleasant, and can cause arousal from sleep. Moreover, these attacks are often associated with a sense of fright and may sometimes be terrifying [1]. A review of the literature reveals that the core features, risk factors, accompanying symptoms are well established, while the effective treatments of EHS are seldom reported [1,3,4]. We present two male patients diagnosed with EHS, of whom the attacks were highly associated with stress, one of the patient's attacks could also be induced by intermittent photic stimulation (IPS). Duloxetine hydrochloride diminished the EHS episodes in one patient. Our study suggests that stress is a high risk factor of EHS, and anti-anxiety therapy may be effective.

2. Cases presentation

2.1. Case 1

A 26-year-old man was admitted to our hospital to investigate episodic 'sudden sensations of explosion inside the head, awakening him from sleep', which were occurring with greater frequency. Attacks were highly associated with emotional stress. The episodes were occurring within an hour of falling asleep and reoccurred several times within a single night's sleep. They lasted from a few seconds to less than 1 min on a nightly basis for nine years. The patient was previous misdiagnosed as epilepsy, with anti-epileptic drugs prescribed without any improvement.

The patient had become anxious about these events and psychological examination revealed generalized anxiety disorder (GAD). His neurological exam, laboratory test results, and neuroimaging were normal. There was no previous history of regular daytime headaches, migraines, or cluster headaches. His medical history and family history were non-contributory.

2.2. Case 2

A 30-year-old man experienced three separate episodes of a sensation of a flashing light and electric shock, with associated visual phenomena for the past two months, similar to "a positive and negative pole of an electrical circuit contacting each other". The feeling occurred inside his head, awakening him from sleep, lasting for a few seconds and was occasionally accompanied with a sensation of nausea. None of the attacks were painful or related to an obvious precipitant. The patient was able to fall asleep immediately after attacks and to recall the event in the morning. He described these feelings as an unpleasant experience but not a fearful one. The patient was concerned that a more serious disorder was responsible for his symptoms. Detailed neurological examination, imaging studies (brain magnetic resonance imaging) and electroencephalogram were all normal.

The main clinical features of the two patients are described in Table 1.

3. Video analysis

3.1. Patient 1

Two 24 h V-PSG with a full-scalp electroencephalogram were performed in 2015 and 2017, respectively. No epileptic abnormalities were detected during these monitoring periods. A total of six episodes were captured, four were awakenings from NREM sleep (video 1), and two from wakefulness during the IPS procedure (video 2). The patient suddenly awakens from sleep (body position changed from lying to sitting), with the sensation of explosion inside

* Corresponding author.
E-mail address: liuyhong@fmmu.edu.cn (Y. Liu).

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Table 1
The clinical features of two male patients presenting with EHS.

Patient Age/sex/onset age	Symptoms/sensations	Duration of episodes	Timing of episodes	Comorbidity
26/M/17	Explosion-like feeling Fear Muscle jerks/twitches Breathing difficulties Sweat Visual phenomena	20–50s	NREM IIstage IPS	Generalized anxiety disorder
30/M/29	Flash light Short circuit/electrical circuit nausea Tachycardia	10s	Sleep	None

the head associated with fear and respiratory arrest. He then buries his head in his hands, with brief muscle jerks/twitches and sweating, each episode lasts 20–50 s. The patient falls asleep within a few minutes after each episode without difficulty and recalls the experience in the morning. The awakenings did not cause any daytime sleepiness. PSG showed normal sleep duration and architecture.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.sleep.2019.02.002.

Reassurance was given after the correct diagnosis, but without any improvement, then duloxetine hydrochloride was prescribed. A routine follow up visit was made six months later, and reported the frequency of episodes and duration of the events had decreased significantly.

3.2. Patient 2

Unfortunately, during the 24 h PSG-EEG monitoring, we were unable to capture the patients described events due to their infrequency. Pharmacological treatment is typically not needed in this sort of patient. Improved sleep habits and reassurance were given for the prevention of events.

4. Brief discussion

In this study, we describe two cases of EHS in where one patient's events not only occurred in NREM sleep, but also could be induced by IPS, the other patient perceived visual phenomena and an electrical sensation during the attacks. Specifically, none of the attacks in both two patients were painful, which is clear different from hypnic headache (Table 2) [5]. During sleep, we speculate that the primary visual areas of the brains may not be completely inactive as usual, resulting in a prominent burst of neuronal activity initially perceived as light flashes, and subsequently rapidly spreading to an electrical or exploding sensation.

Studies linking EHS and IPS have not been reported to date. Several previous studies indicate that the occurrence of visual disturbances in EHS cases range from 10% to 27% [6,7]. Patients perceived lightening, light flashes or visual static, indicating involvement of the primary visual cortex [1,4]. IPS usually precipitates seizures in photosensitive epilepsy patients, along with the associated EEG feature of a photoparoxysmal response with epileptic discharges [8]. There was no epileptic discharge during 24 h monitoring in our two patients. The mechanism of IPS related attacks in our patient remain speculative. Several studies have demonstrated that bright light exposures at the wrong circadian time can lead to circadian disruption and disease risks [9]. Circadian rhythms are regulated by an internal biological clock located in the suprachiasmatic nuclei (SCN) contained within the hypothalamus region of the brain. The eyes' photoreceptors in the retina, can convert light signals into electrical signals, which reach the suprachiasmatic nuclei (SCN) via the retinohypothalamic tract (RHT) [9,10]. Specifically, light–dark cycle of intermittent photic stimulation induced circadian disruption, which results in the occurrence of EHS. It raises the question of further study of the pathophysiology of visual cortex in EHS to gain better future understanding of this rare disorder.

Psychological factors such as stress and anxiety are reported to often trigger EHS [1]. Patient 1 reported his attacks increased as soon as he felt anxious about his work, and the EHS began after he witnessed the death of his father in a car accident, which also triggered several psychogenic non-epileptic spells in him during the past nine years. Patient 2 reported attacks were always associated with increased stress, which caused insomnia and retiring to sleep late after midnight. It is possible that the increased stress result in disrupted sleep, which in turn may easily trigger EHS symptoms. Duloxetine hydrochloride did not result in complete remission of EHS in patient one, but reduced the intensity and frequency of events. It is possible that treating generalized anxiety disorders may result in reductions of EHS. The underlying pharmacological mechanism of drugs remains poorly understood. Detailed psychological exploration of EHS cases in the future could reveal important mechanisms in EHS pathophysiology.

These cases emphasize that stress plays an important role in the occurrence of EHS, and anti-anxiety therapy may be effective. The influences of light exposures on circadian rhythms may also cause the occurrence of EHS. However, present evidence is from a single center and further exploration of EHS cases is needed to understand the complex mechanisms in EHS pathophysiology, and

Table 2
The differentiation between EHS and hypnic headache.

	Painful	Symptom/sensation	Population–>	Duration of episodes	Sleep stage	Frequency	Classification
EHS	No	Explosion-like feeling visual phenomena Fear Muscle jerks/twitches Breathing difficulties Sweat	n/a	Brief duration	NREM sleep	Variable	Sleep Disorders (a sensory parasomnia)
Hypnic headache	Yes	Recurrent headache	Elderly	Lasting ≥ 15 min and for up to 4 h after waking	n/a	Occurring on ≥10 days per month for >3 months	Headache Disorders

long-term follow-up of case records could help in the selection of therapeutic strategies.

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Conflict of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2019.02.002.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.sleep.2019.02.002.

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