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Nocturnal frontal opercular seizures masquerading as catathrenia: a case report and review of the literature

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Abstract

A 33-year-old man with two bilateral tonic-clonic seizures of unknown onset 12 years earlier was admitted to our epilepsy monitoring unit for characterization of his nocturnal groaning spells suspicious for catathrenia. Video-scalp electroencephalogram (EEG) was suggestive of drug-resistant non-lesional localization-related focal epilepsy in the right hemisphere. Intracranial EEG showed right frontal opercular origin of seizures with insular spread as the cause of his spells. This illustrative case shows the importance of video-EEG monitoring to distinguish nocturnal frontal opercular seizures from catathrenia. We review the evidence on distinguishing the two disorders.

Keywords: *Catathrenia. Nocturnal vocalization. Frontal lobe epilepsy. Frontal opercular seizures. Electroencephalogram.*

Crisis epilepticas frontales operculares nocturnas enmascaradas como catatrenia: reporte de caso y revisión de la literatura

Resumen

Un hombre de 33 años con dos crisis epilépticas tónico-clónicas bilaterales de inicio desconocido quien 12 años antes ingresó en nuestra unidad de monitorización de epilepsia para la caracterización de sus ataques de gemidos nocturnos sospechosos de catatrenia. El video-electroencefalograma (EEG) de piel cabelluda sugirió epilepsia focal relacionada con la localización no lesional resistente a los medicamentos en el hemisferio derecho. El EEG intracraneal mostró un origen opercular frontal derecho de las crisis epilépticas con extensión insular como causa de sus ataques. Este caso ilustrativo muestra la importancia de la monitorización por video-EEG para distinguir las crisis epilépticas operculares frontales nocturnas de la catatrenia. Revisamos la evidencia para distinguir los dos trastornos.

Palabras clave: *Catatrenia. Vocalización nocturna. Epilepsia del lóbulo frontal. Crisis operculares frontales. Electroencefalograma.*

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Introduction

A previously health 33-year-old man without a family history of epilepsy or sleep disorder was admitted to our epilepsy monitoring unit (EMU) for the characterization and management of his nocturnal groaning spells. At the age of 21 years, he had had two brief (i.e., < 1 min each) episodes of nocturnal whole-body tonic-clonic movements without auras or postictal features occurring in the context of extreme emotional stress and sleep deprivation. He was treated with phenytoin until age 23 years, at which point it was discontinued because he had no further recurrences. At age 24 years, he developed exclusively nocturnal episodes consisting of moaning and groaning sounds that would occasionally wake him up and be followed by a sensation of cold "chills" in the right shoulder which would then migrate across his chest. He frequently turned to the right side during these spells. These spells recurred up to 15 times per night and each lasted approximately 2-3 seconds. He had no known daytime events and the spells were not associated with loss of awareness. He tried multiple antiseizure medications (ASMs) to abate these events (phenytoin, valproic acid, levetiracetam, and lacosamide) but had to discontinue them due to undesirable adverse effects. He was finally successfully treated with carbamazepine. He had no further spells on low-dose carbamazepine monotherapy until age 30 years, when the same spells recurred nearly every night basis and became resistant to therapy despite taking lamotrigine, phenobarbital, and clobazam. At age 33 years, he was admitted to the EMU. At first, there were no abnormal electrographic activities or clinical events in his continuous video-electroencephalograms (VEEGs) for several days, and given the semiology of his spells, the diagnosis of catathrenia was raised by the consultant sleep medicine specialist (GFPG).

Results of investigations

His brain magnetic resonance imaging (MRI) was normal. Sleep study revealed mild to moderately severe sleep-disordered breathing but it did not capture the spells (Fig. 1).

Continuous VEEGs using scalp electrodes showed normal background activity in wakefulness and normal sleep potentials. Eventually, frequent interictal epileptic discharges (IEDs, a.k.a. epileptic spikes) from the right temporal-central-frontal region were captured on EEG during sleep (Fig. 2).

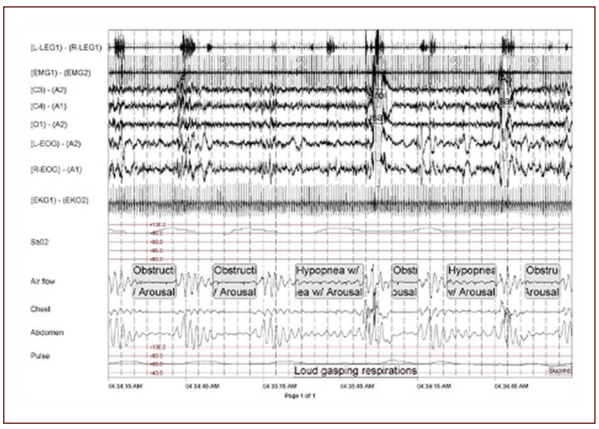

Figure 1. Sample polysomnogram from patient. Patient did not produce catathrenia during this recording although he did demonstrate obstructive sleep patterns.

Several typical nocturnal spells were captured that were consistent with right hemispheric seizures with predominant involvement of the right frontotemporal regions. Significant muscle and movement artifacts during them distorted the EEG signals that prevented so a very precise localization. On a subsequent admission, intracranial EEG electrodes using stereoencephalography (SEEG) revealed the seizure onset zone within the right frontal opercular region with rapid spread to the right insular region during his typical nocturnal spells (Fig. 3).

His neuropsychological assessment revealed frontal dysfunction. He is currently waiting for resective surgery of the seizure onset zone in the right frontal opercular region.

Discussion

Catathrenia was first described De Roek *et al.* in 1983 as the production of nocturnal groaning during episodes of prolonged expiration[1]. Catathrenia is classified as a "sleep-related breathing disorder" by the international classification of sleep disorders diagnostic and coding manual and consists of repeated groaning during exhalation, mainly in rapid eye movement (REM) sleep[2]. The groaning quality of catathrenia is described as "morose" or containing "sexual connotations" which can cause social problems for many patients[3,4]. Typically, the patients do not have any subjective complaints of sleep disturbances or excessive daytime sleepiness (Fig. 4). The referral to a sleep clinic is generally made by family or bed partners. Some case series have described

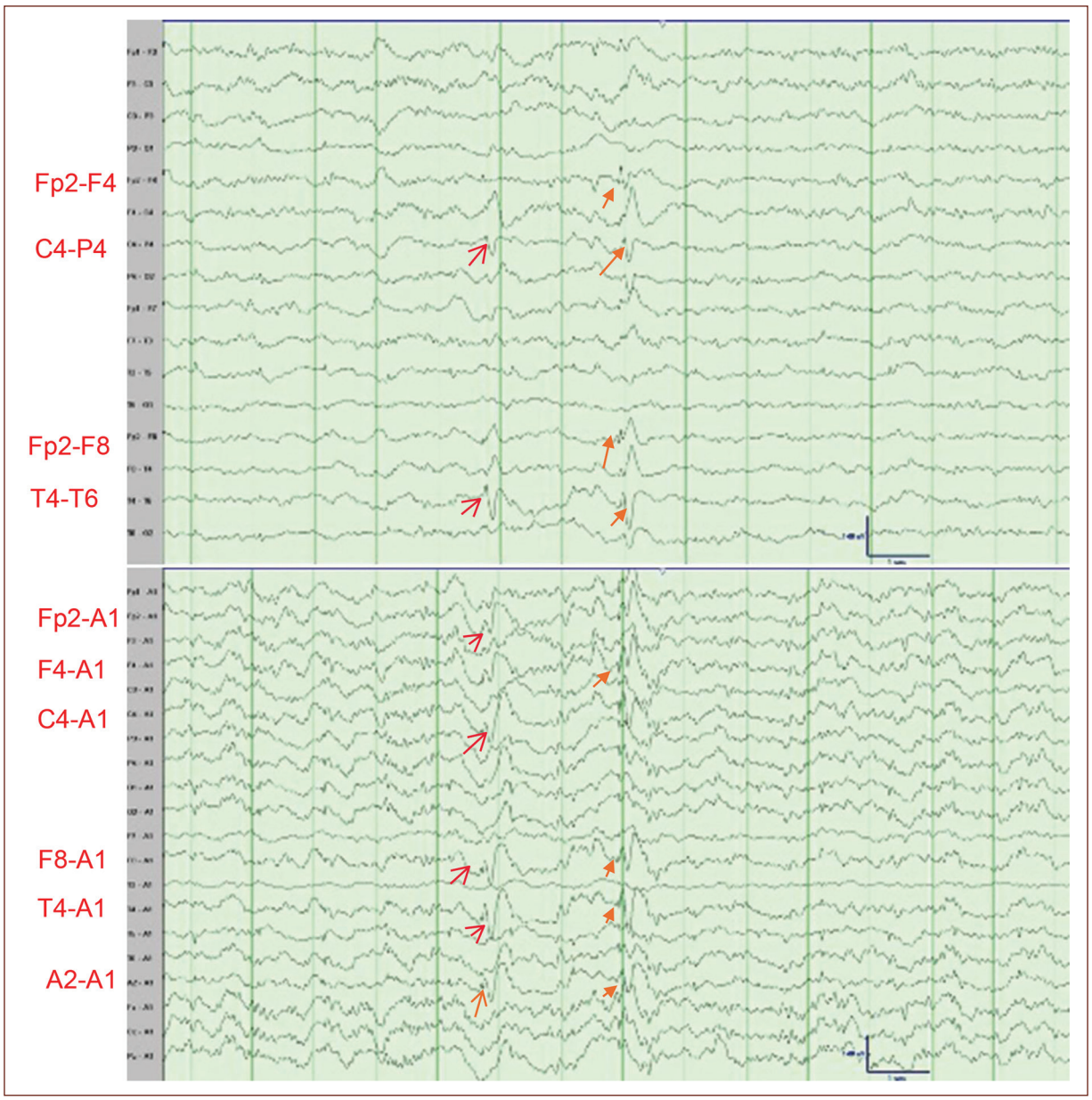

Figure 2. EEG recording shows the right temporal-central-frontal spikes. 1A bipolar montage; 1B ipsilateral ear montage. Settings: LFF = 1 Hz, HFF = 75 Hz, Notch Filter Off, Sensitivity = 10 µv/mm.

frequent epochs during non-REM sleep[3,5,6] but most describe a predominance during REM sleep, especially during awakening from REM sleep[4,7-14]. Groaning is not typically associated with significant oxygen desaturation, although concurrent obstructive sleep apnea has been reported[6,8-10] (Table 1)[3-6,8-18].

The differential diagnosis for catathrenia includes sleep talking (somniloquy), stridor, laryngospasm, snoring, and, notably, nocturnal epilepsy-related vocalization[2,5,11]. It is crucial to distinguish the latter from catathrenia as their treatments differ markedly. There is indeed some evidence supporting the use of continuous positive airway pressure (CPAP) in patients with catathrenia[5-8,10,12-14,19]. There is a paucity of the literature that focuses on the topic of distinguishing between these two disorders (Table 2).[11] Zinke et al.[11] described a 22-year-old male who developed catathrenia 10 years after being diagnosed and successfully treated with ASMs for

Table 1. Review of published literature on catathrenia

Study	Sample size (n)	Age at onset (mean)	Gender (M, F)	Brain MRI	EEG	Initial diagnosis	PSG	Treatment outcomes (responders*/cases tried)
Pevernagie et al. 2001[15]	10	23.4	7.3	8/10 normal, 1/10 parietal cortical atrophy, 1/10 left frontal meningioma	7/10 normal, 3/10 intermittent slow wave activity	N/A	93% in REM sleep	Trazodone (0/2), clonazepam (1/3), paroxetine (1/1), dosulepine (1/3), nasal CPAP (2/2)
Vetrugno et al. 2001[4]	4	10.8	3.1	N/A	Normal	N/A	Especially in REM sleep, often with EEG arousal, normal sleep structure 3/4	N/A
Iriarte et al. 2006[8]	1	62	0.1				Epochs associated with oxygen desaturations, more common in REM sleep	CPAP (1/1)
Oldani et al. 2005[9]	12	31.4	10.2	N/A	Normal	N/A	No significant apneas, 89% in REM	Clonazepam (0/2), gabapentin (1/1), pramipexole (0/1), Trazodone (0/1)
Steinig et al. 2010[10]	1	33	1.0	N/A	Normal	Central sleep apnea	Epochs occur during REM sleep with cessation of breathing without oxygen desaturation	CPAP (0/1)
Guilleminault et al. 2008[5]	7	26.7	0.7	N/A	Normal	N/A	Most expiratory groaning occurs in NREM, with diminished intensity in REM sleep	CPAP (7/7)
Siddiqui et al. 2008[3]	1	8	1.0	Normal	Normal	Central sleep Apnea	Groaning mostly in stage 2	None
Songu et al. 2008[6]	1	40	0.1	N/A	Normal	N/A	3/9 in NREM sleep, severe OSA	CPAP (1/1)
Zinke et al. 2010[11]	1	22	1.0	N/A	Generalized slowing without epileptiform activity	Seizures (has concurrent generalized seizure disorder diagnosed 10 years before catathrenia)	REM-associated episodes of nocturnal groaning	None
Abbasi et al. 2012[12]	10	46.2	5.5	N/A	Normal	N/A	Most episodes occurred during REM sleep. 9.4% of epochs associated with awakening	CPAP (4/4)

(Continues)

Table 1. Review of published literature on catathrenia (continued)

Study	Sample size (n)	Age at onset (mean)	Gender (M, F)	Brain MRI	EEG	Initial diagnosis	PSG	Treatment outcomes (responders*/cases tried)
Iriarte et al. 2011[17]	2	63.5	1.1	N/A	N/A	Oscilogram: woman is 8.5 waves in 25 ms = 340 Hz. is 6 waves in 25 ms = 240 Hz. Yanagihara classification Type I, II	PSG in patient 1	N/A
Neutel et al. 2014[13]	1	32	1.0	Perisylvian polymicrogyria	Normal	66% of respondents correctly identified the disorder	Occur during awakening from REM sleep	CPAP (1/1)
Hao et al. 2015[16]	22	35	7.15	N/A	N/A	Cephalograms, anatomical gaps between OSAS and Catathrenia PAR index (p = 0.048), and lower arch length (p = 0.021), upper arch length (p = 0.030)	N/A	1 patient without antidepressant and other patients with irregular Hypnotics not excluded
Drakatos et al. 2016[14]	38	33.1	23.15	N/A	Normal	N/A	Most events occurred in REM sleep and with arousal	CPAP (3/9), MAD (3/6), prevent (1/1), clonazepam (2/3), trazodone (0/1), melatonin (1/1), zopiclone (2/4), CBTi (0/1)
Alonso et al. 2017[5]	47	40.2	20.27	N/A	N/A	35 patients reported evaluation by medical or dental sleep specialist, 29 PSG, 12 self-diagnosis and did not choose medical evaluation	29 patients: 10 OSA diagnosis, 3 catathrenia, 14 "normal" study underwent in-laboratory or ambulatory diagnosis	CPAP= all patients independent of the diagnosis anxiety/ depression = 21 (44.7%) with antidepressant or anxiolytics

*A responder here is defined as a patient who noted a decreased in their groaning after initiation of treatment[18].
MRI: magnetic resonance imaging; NREM: non-rapid eye movement; EEG: electroencephalogram; CPAP: continuous positive airway pressure; MAD: mandibular advancement device; CBTi: cognitive behavioral therapy for insomnia.

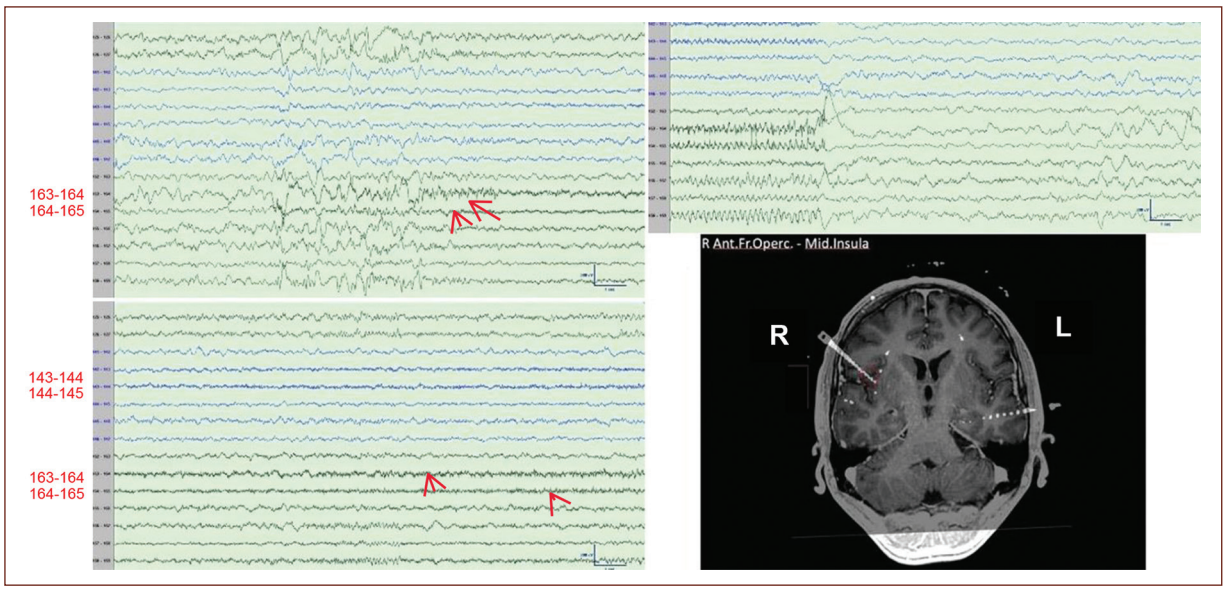

Figure 3. Intracranial electrode recording (left) shows a seizure focus within the right frontal opercular area (see MRI correlate on the right). Note seizure onset at electrodes 163, 164, and 165. Settings: LFF = 1 Hz, HFF = 75 Hz, Notch Filter Off, Sensitivity = 10 µv/mm.

Table 2. Clinical characteristics distinguishing catathrenia from nocturnal seizures[11]

Clinical characteristics	Catathrenia	Nocturnal seizures
Frequency	Daily	Variable but can be daily
Sleep stage	Any stage, but a predilection for REM sleep	Usually, NREM
Clustering of episodes	Yes	Yes
Semiology	Inspiration, loud prolonged expiration; vocalization	Can be stereotyped hypermotor or dystonic/tonic may be associated with somatosensory or viscerosensorial symptoms
Vocalization	Expiratory monotonous groans or moans	Non-specific groans → speech
Awakening	Not usually	Yes
Patient recall	None	Usually
Duration	6-30 s but can cluster for up to 1 h	Average 5-60 s
EEG	Non-epileptic arousals	± Epileptiform activity
Excessive daytime sleepiness	Not usually	Common

NREM: non-rapid eye movement; REM: rapid eye movement; EEG: electroencephalogram.

daytime seizures secondary to generalized epilepsy. He had no improvements in his nocturnal spells from catathrenia despite multiple adjustments to his ASMs until he was treated effectively for his coexisting presumed catathrenia. Neutel et al.[13] describe the case of a 32-year-old man with somatosensory seizures due to perisylvian polymicrogyria successfully controlled with valproic acid who later developed catathrenia. His nocturnal groaning spells responded to treatment with CPAP. Neither of the above-mentioned reported epilepsy cases had

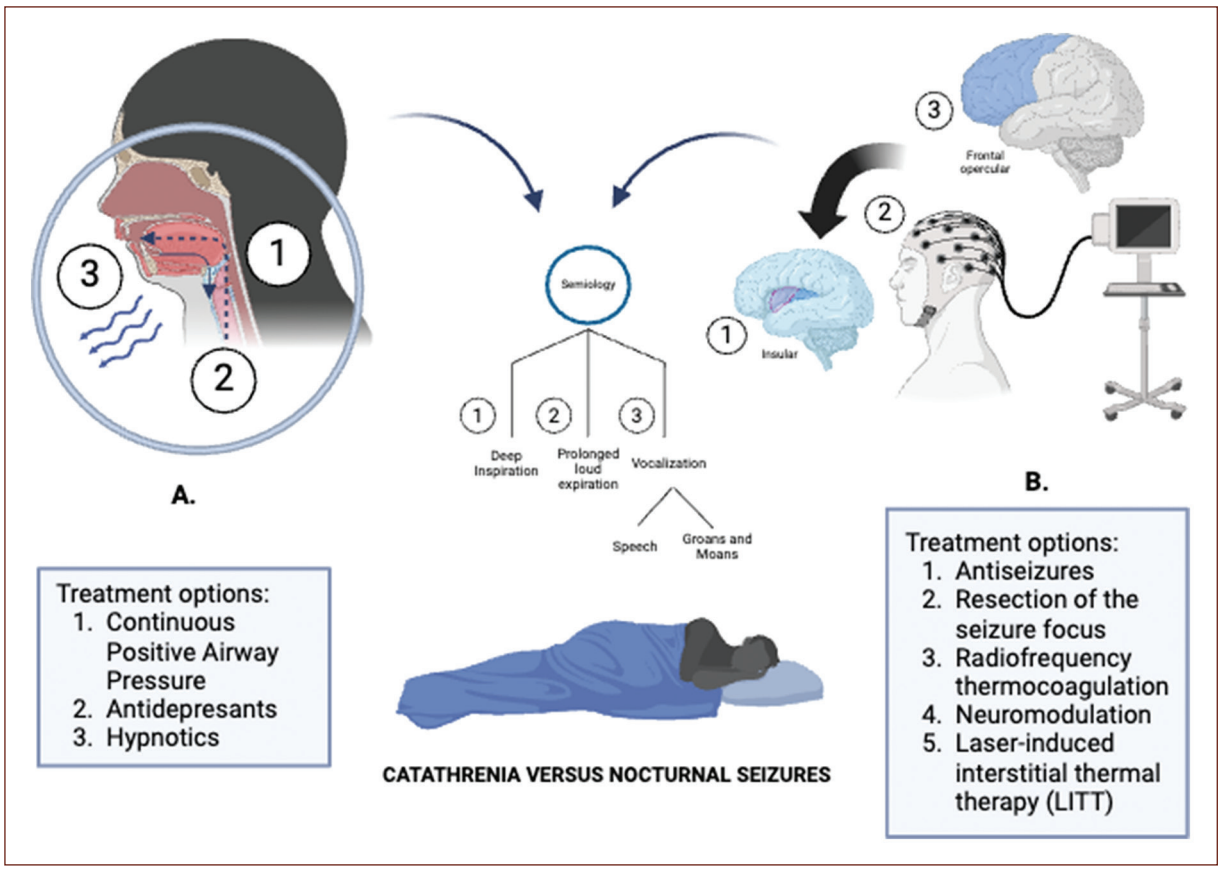

Figure 4. A: illustrative sequence of the anatomical structures involved in Catathrenia (top) and its treatment (bottom). **B:** nocturnal seizures of the frontal opercular origin with spread to the insula (top) and its treatment (bottom).

intracranial EEG recording. To the best of our knowledge, this is the first case report of frontal opercular epilepsy masquerading as catathrenia as confirmed by SEEG recordings.

Ictal screaming or crying is more commonly associated with generalized seizures[20]. The seizure semiology of our case closely mimicked the breathing patterns seen in catathrenia. The opercular onset is in keeping with early vocalization while the subsequent somatosensory semiology across the chest is compatible with spread to the insula seen in his SEEG[21-23]. This patient could have been misdiagnosed as catathrenia due to his presenting nocturnal grunting and given the inherent challenges of capturing frontal opercular and insular seizures on scalp EEG. However, our continuous VEEG in the EMU did eventually demonstrate that these events were epileptic in origin and SEEG confirmed the localization so that he could be treated accordingly. This case highlights the importance of longer VEEG monitoring and possible SEEG in patients in whom there is a higher clinical suspicion of epileptic seizures as the cause of their nocturnal vocalizations.

Conclusion

Catathrenia and frontal opercular epilepsy may share overlapping symptoms such as nocturnal screaming, grunting, or moaning during deep expirations. Catathrenia is a respiratory disorder that does not respond to ASMs. Seizures of the frontal opercular origin can be difficult to detect with scalp EEGs. Intracranial EEGs (especially SEEGs) are required to identify frontal opercular seizures and their patterns of spread to the adjacent brain areas including insula. VEEG telemetry to exclude epileptic seizures is necessary before diagnosing a patient with catathrenia because treatments of the two conditions are vastly different. Close collaborations between sleep medicine specialists and epileptologists are needed to work up such potentially overlapping cases.

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Conflicts of interest

The authors declare that they have no conflicts of interest.

Ethical disclosures

Protection of human and animal subjects. The authors declare that the procedures followed were in accordance with the regulations of the relevant clinical research ethics committee and with those of the Code of Ethics of the World Medical Association (Declaration of Helsinki).

Confidentiality of data. The authors declare that they have followed the protocols of their work center on the publication of patient data.

Right to privacy and informed consent. The authors have obtained the written informed consent of the patients or subjects mentioned in the article. The corresponding author is in possession of this document.

Use of artificial intelligence for generating text. The authors declare that they have not used any type of generative artificial intelligence for the writing of this manuscript, nor for the creation of images, graphics, tables, or their corresponding captions.

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