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REVIEW ARTICLES

Catathrenia (Nocturnal Groaning): A Social Media Survey and State-of-the-Art Review

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Study Objectives: Catathrenia is an underrecognized nocturnal vocalization phenomenon that can be a source of perplexity to patients, bed partners, and medical providers. Catathrenia is distinct from both sleep talking (a parasomnia with loud talking during sleep) and snoring (noise due to vibration of upper airway soft tissues related to variations in airway resistance). The objective of this review is to provide an evidence-based resource to help the practitioner reliably evaluate and manage patients with this condition.

Methods: Data were gathered from: (1) PubMed, Scopus, Web of Science, and Google Scholar; and (2) catathrenia social media groups (Yahoo and Facebook).

Results: Data collected were (1) 15 case reports and 17 case series describing 191 patients with catathrenia; (2) questionnaires from 47 catathrenia subjects; (3) 5 audio files.

Conclusions: Catathrenia is a noise produced during sleep (distinct from snoring) with identifiable harmonics, a computable main frequency, and high-decibel intensity that involves active adduction and vibration of the vocal cords during expiration. The quality of groaning in catathrenia is monotone, and often presents with a morose or sexual connotation, causing a significant social problem for patients. Although there is no association with risk of physical harm, catathrenia does present a significant disturbance to the bed partner and has been associated with subjective impairments to sleep quality, including unrefreshing sleep and fatigue. Polysomnography can be useful if performed properly to confirm the diagnosis and to evaluate for comorbid sleep disturbances, such as obstructive sleep apnea or parasomnia. Directions for further research could involve consideration of deep breathing exercises, yoga, meditation, or myofunctional therapy to help abate symptoms.

Keywords: catathrenia, nocturnal groaning, nocturnal moaning, sleep-disordered breathing, vocalization

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INTRODUCTION

Catathrenia is a sleep-disordered breathing pattern characterized as expiratory groaning or moaning during sleep.[1] Catathrenia is not usually noticed by the person producing the sound but can be extremely disturbing to sleep partners. Bed partners generally report hearing the person take a deep breath, hold it, then slowly exhale; often with a high-pitched squeak or groaning sound. Once aware of it, people with catathrenia tend to be awakened by their own groaning as well.[2–5] Due to limited familiarity with the condition, catathrenia is often confused with sleep talking or snoring.[6]

Although catathrenia was originally described as a single case report by De Roeck et al. in 1983,[7] there have since been several reports from various institutions around the world. However, the disorder is sufficiently rare that many otolaryngologists and even sleep specialists remain unfamiliar with this atypical breathing pattern. In a series by Jaar et al. in 2009, the incidence of catathrenia was identified as 25 of 15,052 patients (0.17%) who presented with sleep and/or wake problems at a sleep center over a 10-year period in Japan.[8] Overland et al. state that the incidence of catathrenia was 4 of 1,004 patients (0.4%) undergoing polysomnography over a 1-year period at their institution in Norway.[9] Unfortunately, however, catathrenia may be underrecognized despite a comprehensive sleep evaluation. As Okura and Muraki explain, the polysomnographic pattern of catathrenia so closely resembles that of central sleep apnea that catathrenia may be erroneously scored as central sleep apnea unless a detailed analysis of video and audio recordings is performed by properly trained technicians.[10]

Patients have taken to online forums to help each other fill in the gap of knowledge for this unusual sleep-disordered breathing pattern. In 2009, a Yahoo group called "Catathrenia (nocturnal groaning)" was developed as a discussion forum for catathrenia sufferers from all over the world. The group has collected approximately 600 members. In 2013, a Facebook support group called "catathrenia (night groaning)" was also developed with 67 highly active members to date. A common theme on the forums is frustration with the medical communities' lack of knowledge and familiarity with the disorder.

The aim of our current research was to utilize the Yahoo and Facebook catathrenia groups to further understand and characterize this underrecognized sleep disorder. We also aimed to perform a systematic review of the literature to address

questions or issues identified as areas of concern by patients from this population.

METHODS

Survey Study

Subject recruitment

This study was approved by the Institutional Review Board at University of California, Los Angeles. A posting was placed on two online support groups for patients with catathrenia: (1) Yahoo Groups "Catathrenia (nocturnal groaning)" and (2) Facebook "catathrenia (night groaning)." The posting was placed on June 18, 2015; patients with catathrenia were recruited to complete an online survey and/or provide audio recordings of their unusual nighttime groaning.

Inclusion criteria

(1) Age older than 18 years, (2) diagnosis of catathrenia by a medical professional *or* have been told by a family member or friend that they produce unusual expiratory moaning and groaning during sleep.

Exclusion criteria

All subjects meeting the aforementioned inclusion criteria were invited to participate in the survey, which included a thorough sleep and medical history questionnaire. Survey submissions that did not meet the International Classification of Sleep Disorders, Third Edition (ICSD-3) definition of catathrenia (ie, nocturnal vocalizations during sleep) were excluded from the analysis.[1]

Survey questionnaire

Patients were asked to respond "yes" or "no" and provide an open-ended response "If so, please explain" to a series of questions regarding their sleep and medical history. This included the following: description of unusual breathing; how long they have been aware of it; how often they experience it; family history; prior diagnosis or evaluation for a sleep disorder; diagnosis of obstructive sleep apnea (OSA); if they have tried continuous positive airway pressure (CPAP); results of prior sleep study; prior recommendations of medical or dental specialist for the unusual breathing.

The following data were also collected: age; sex; height; weight; history of thyroid disorder, hypertension, anxiety, depression, psychiatric disorders, and parasomnias (such as teeth grinding, sleep walking, and sleep talking); current medications; use of alcohol, tobacco, and recreational drugs; snoring; excessive daytime sleepiness; unrefreshing sleep; feelings of fatigue; insomnia; sudden waking with breath holding; gasping or choking; nighttime waking for urination; number of hours of sleep per night; decrease in concentration or memory; witnessed apneas or choking episodes; prior stroke, myocardial infarction or heart problems as a result of lung disease (cor pulmonale); motor vehicle accident; and decrease in daytime alertness. Fatigue and level of distress were rated on a scale from 1 (none) to 10 (worst). The Epworth Sleepiness Scale Questionnaire was administered.[11] Swimming history was collected (based on the recommendation of authors MC and CG), including: whether the patient was a competitive swimmer (up to the ages of 13 years, 18 years, 25 years, and current); history of recreational swimming; number of days per week swimming.

Data analysis

JMP 12 Pro (SAS Institute Inc, 2015) was used for quantitative descriptive statistics (mean, standard deviation) and analytical statistics.

Systematic Literature Review

Patient concerns were identified and a systematic review of the literature was performed to provide evidence-based responses to patient questions regarding the condition. The PRISMA (Preferred Reporting Items for Systematic Reviews and Meta-Analysis) guidelines were followed.[12]

Search strategy and selection criteria

A comprehensive search was initiated on August 23, 2013 with completion and update through May 1, 2016 by authors SZ, MC, CG, and JA. The following databases were searched from inception: PubMed, Scopus, Web of Science, and Google Scholar. The databases were searched using a combination of keywords and phrases to include: "catathrenia," "nocturnal vocalization," "nocturnal groaning," "nocturnal moaning." An example of a search on PubMed is: ("Catathrenia" OR "nocturnal vocalization" OR "nocturnal groaning" OR "nocturnal moaning"). The authors independently reviewed the titles and abstracts and then downloaded relevant studies for further evaluation. The reference lists for each study were then reviewed to assess for additional studies to include in this systematic review.

Study selection

Inclusion criteria: Studies needed to report original data for adult patients who presented with features of nocturnal groaning, vocalization, or moaning consistent with ICSD-3 diagnosis of catathrenia. Exclusion criteria: Review and commentary only. No original studies were excluded.

Data abstraction and study quality assessment

After obtaining the articles for inclusion, the data collected from the studies was reviewed for quality assessment based on the NICE (National Institute for Health and Clinical Excellence) checklist. The instrument consists of eight items that are assessed for each individual study: (1) Was the case series collected in more than one center, ie, multicenter study? (2) Is the hypothesis/aim/objective of the study clearly described? (3) Are the inclusion and exclusion criteria (case definition) clearly reported? (4) Is there a clear definition of the outcomes reported? (5) Were data collected prospectively? (6) Is there an explicit statement that patients were recruited consecutively? (7) Are the main findings of the study clearly described? (8) Are outcomes stratified? (eg, by abnormal results, disease stage, patient characteristics)?

RESULTS

Reponses were collected from June 18, 2015 to November 7, 2015. Fifty surveys were collected. Forty-seven patients provided

Table 1—"Describe your unusual breathing."

I breathe in then hold my breath (with my mouth open) for quite some time and then finally let it out with a grunt or groan.
I hold my breath and make a groaning noise
I hold my exhalation. I typically make a long whining sound before a "burst" of exhaled breath when I need to breathe again.
Heavy sigh followed by a deep inspiration held for about 10 seconds; expiration is slow and halfway through the exhale becomes a low groan or growl. We have noticed that high stress/anxiety worsens it as does sleeping in a warm environment.
I inhale normally, then hold my breath (I think by closing the back of my throat). I then slowly let out a very small amount of air with a croaking/groaning/vibrating sound, for maybe 5 seconds. I then quickly exhale the remaining air, as a kind of sigh, then inhale and the process repeats.
I stop breathing and begin to moan loud and monotonous up to one minute. Sometimes these moans are just like sighs but mostly they are very loud and long. After a few silence seconds the moaning begins again. This can lasts up to one or two hours. In good nights these episodes appears 2–3 times. In bad nights nearly the whole night.
According to my husband, I hold my breath for 3–4 seconds then breathe out very slowly while groaning
I hold my breath for an unusually long time, then slowly and laboriously exhale with a slight noise—like my throat is closed.
I take a very deep breath and then hold it for quite some time, then slowly "squeak" to let that breath out.
My husband says I hold my breath then make a loud groan. My daughter only mentioned the groaning.
Typically hold my breath for a long time then slowly exhale making groaning/squeaking sounds. Occasionally this is followed by a deep gasp for breath.
Very prolonged exhale of breath producing a very loud groan. Repeats.
It happens in the early hours of the morning. I hold my breath for at least 10–15 seconds and then groan while exhaling. Sometimes I catch myself holding my breath while I'm awake too.
It seems as if I were underneath water, holding my breath and exhaling air very slowly. And groaning as I do it.
My husband said it's almost like I am having an orgasm. That's the only way he could describe the breathing noises.
Deep and very loud moaning, groaning.
I breathe in, hold my breath and slowly release it over a minute or so. This will be repeated until I am woken up. It can happen any time during the night but tends to happen during light sleep.
I take a deep breath and hold it…then slowly let it out creating a high-pitched squeaky noise.
It sounds like I am slowly releasing air from my throat which makes a groaning noise and at times a higher pitched noise in between the groaning.
I hold my breath, then exhale whilst making a groaning sound.

Twenty demonstrative responses by survey participants as reported in their own words are included.

descriptions of nocturnal vocalizations during sleep that were consistent with catathrenia and included in the analysis; three surveys were excluded. **Table 1** provides characterization of the nocturnal groaning as reported by patients in their own words. **Table 2** illustrates survey responses from subjects. There were five audio files submitted and all five were consistent with catathrenia (see supplemental material for audio examples).

Diagnosis and Prior Evaluation

In **Table 2**, 35 patients reported an evaluation by a medical or dental sleep specialist; 29 patients had level I in-laboratory polysomnogram performed, 2 patients had an ambulatory sleep study performed, 2 patients were provided with an oral appliance for snoring by a dentist without sleep study, and 1 patient was recommended a sleep study to confirm a diagnosis of catathrenia and rule out sleep apnea but did not pursue it further. Twelve patients in this series made a self-diagnosis of catathrenia through online search and did not choose to seek medical evaluation.

Polysomnography

Of 29 patients who underwent in-laboratory overnight polysomnogram, a diagnosis of mild OSA was confirmed in 10 cases, and 6 of 10 patients were recommended to try CPAP; the other 4 patients were reassured the apnea was not severe enough to require treatment as described in **Table 2**. Three patients received a specific diagnosis of catathrenia (no OSA diagnosis) and recommended to try CPAP; two patients were told they did demonstrate an unusual vocalization during rapid eye movement (REM) sleep but without mention of catathrenia or OSA diagnosis and both were recommended to try CPAP. The remaining 14 patients who underwent in-laboratory overnight polysomnography were reassured that they had a "normal" sleep study. Of the two patients who underwent ambulatory sleep study, they were both found to be within normal limits and did not identify OSA or oxygen desaturations. In the systematic review, for all studies a standard in-laboratory polysomnography was performed in the diagnostic evaluation of subjects. There were 14 patients in whom OSA was

Table 2—Demographic factors and survey results.

	Number Responding "Yes"	% of Total
Sex		
Male	20	42.6%
Female	27	57.4%
Occurrences, distress, concerns		
Distress to bed partner	22	46.8%
Poor quality or unrefreshing sleep	21	44.7%
Health concerns	14	29.8%
Social embarrassment	7	14.9%
Effect on relationship	5	10.6%
Effect on sex life	1	2.1%
Method of catathrenia awareness		
Other Observers	44	93.6%
Self-aware	3	6.4%
Sleep parameters		
Unrefreshing sleep	39	83.0%
Fatigue	36	76.6%
Reduced daytime alertness	33	70.2%
Reduced concentration or memory	29	61.7%
Witnessed gasping or choking episodes	24	51.1%
Nocturia	25	53.2%
Insomnia	21	44.7%
Sudden wakening with breath holding, gasping, or choking	15	31.9%
Parasomnias	27	57.4%
Comorbid medical conditions		
Anxiety/depression	21	44.7%
Gastroesophageal reflux disease	3	6.4%
Hypothyroidism	2	4.3%
Asthma	2	4.3%
Psychiatric disorder	2	4.3%
Hypertension	1	2.1%
Migraine headache	1	2.1%
Diabetes	1	2.1%
Social history		
Ever used tobacco	13	27.7%
Active tobacco use	6	12.8%
Alcohol use	36	76.6%
Recreational drug use	6	12.8%
Swimming		
Active swimmer	9	19.1%
Competitive participation	9	19.1%
Up to age 13 y	7	14.9%
Up to age 18 y	1	2.1%
Up to age 25 y	1	2.1%

diagnosed with mean ± standard deviation apnea-hypopnea index 13.1 ± 8.4 events/h (n = 4 studies).[3,13–15]

Demographic Data

There were 27 female and 20 male participants whose surveys were included in the analysis (**Table 2**). Although there was a slight female predominance (57.4% female versus 42.5% males), the differences in the distribution of gender were not statistically significant (P = .1487). Participants had mean ± standard deviation age of 40.2 ± 10.9 years and body mass index of 24.8 ± 4.6 kg/m^2 (normal range of weight to height ratio). Interestingly, the systematic review shows a very similar sex distribution: 58.3 % female (n = 94), 41.6% male (n = 67), sex not reported (n = 30) (see **Table 3**).

Catathrenia Onset

Participants reported having been aware of their unusual nocturnal groaning for an average of 14.9 ± 9.5 years. The majority of patients were made aware of the problem through other observers (94%, **Table 2**). Only 6% of subjects (3 of 47) reported becoming self-aware of the breathing pattern: (Patient A) "I would hear myself making noises while half-asleep. People sleeping near me would also tell me what I was doing"; (Patient B) "I woke myself up moaning. My dad was looking at me"; (Patient C) "Was told by a roommate, but also I am sometimes semiconscious of doing it." The other 94% of participants with catathrenia were made aware through spouse/significant other/bed partner (62%), friends during sleepover (13%), parent (11%), roommate (9%). In the systematic review, age at onset of catathrenia ranged from 4 to 36 years.[3,16]

Occurrences, Distress, Concerns

Subjects reported almost nightly occurrences of catathrenia with average 6.56 ± 1.15 nights per week. Level of distress associated with the condition is reported as 6.3 ± 2.3 (moderate distress, **Table 2**). Reasons for distress and concern include: disturbance to bed partner (22 of 47, 46.8%), poor quality or unrefreshing sleep (21 of 47, 44.7%), worry about health consequences (14 of 47, 30.0%), concern for social embarrassment: (7 of 47, 14.9%), effect on relationship because partner sleeps in separate room (5 of 47, 10.6%), self-consciousness that affects sex life and/or opportunity for intimacy (1 of 47, 1.5%). In the systematic review, two articles reported average occurrences of catathrenia of 3.6 and 5.9 days per week,[2,17] and one article described an occurrence of almost every night.[4]

Sleep Parameters

As shown in **Table 2**, the following number of participants responded "yes" when asked if they ever experienced: unrefreshing sleep (39 of 47, 83%); fatigue (36 of 47, 76.5%); decreased daytime alertness (33 of 47, 70.2%), decreased concentration or memory (29 of 47, 61.7%); witnessed gasping or choking episodes (24 of 47, 51.1%); waking up at night to urinate (25 of 47, 53.2%); insomnia (21 of 47, 44.7%), sudden waking with breath holding, gasping, or choking (15 of 47, 31.9%). Among patients reporting "yes" to ever feeling fatigued, the level of fatigue was rated as 5.89 ± 1.79 (moderate fatigue). Total Epworth Sleepiness Scale score was 8.3 ± 5.0 (mild to moderate sleepiness). Typical amount of sleep each night was reported as 7.0 ± 0.85 hours per night. Incidence of motor vehicle accident involvement was 17 of 47, or 36.2%. There were 27 of 47 participants (57.5%) who reported experiencing parasomnias (including teeth grinding, sleep walking, sleep talking), and there were 18 of 47 participants (38.3%) who reported a family history of parasomnia. In the systematic review, 5 articles[4,16–19] report a history of parasomnia among study subjects. A positive family history of parasomnia has also been reported[2–4,14,18]; interestingly, 1 article reported a positive family history of catathrenia.[14]

Table 3—General characteristics and National Institute for Health and Clinical Excellence quality criteria of studies identified by systematic literature review.

Catathrenia: Systematic Review - Study General Characteristics							NICE: Quality Assessment Checklist							
Study, Year (Design)	**Study Site**	**n (sex)**	**Age, mean ± SD (range)**	**Age of Onset (y)**	**Variables Analyzed**	**Treatment**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**
Abbasi et al., 2012 (retrospective review)[5]	Minnesota, United States	10 (F:5)	46.2 ± 22.5 (4–76)	–	PSG, BMI, ESS, EEG, Acoustic Analysis	CPAP (effective in 4/4)	N	Y	Y	Y	N	N	Y	Y
Bercovici and Mirsattari, 2013 (case report)[28]	London, Canada	1 (M:1)	28	–	PSG, MRI	–	N	Y	N	Y	N	N	Y	N
Bar et al., 2016 (case report)[29]	Bordeaux, France	1 (M:1)	4	–	PSG	–	N	Y	Y	Y	N	N	Y	N
Brunner and Gonzalez, 2004 (retrospective case series)[30]	Zurich, Switzerland	8 (F:2)	–	–	PSG	–	N	Y	N	Y	N	N	Y	N
De Roeck et al., 1983 (retrospective case report)[7]	Edegem, Belgium	1 (M:1)	35	15	PSG	–	N	-	-	-	-	-	-	N
Guilleminault et al., 2008 (case series)[18]	California, United States	7 (F:7)	26.7 ± 4.6 (23–34)	Child-hood	ESS, craniofacial exam, BMI, neck circumference, PSG, EEG	CPAP (effective in 7/7); Oral appliance in 3/7 (effective in 3/3)	N	Y	Y	Y	Y	Y	Y	Y
Hao et al., 2015 (retrospective case-control study)[31]	Beijing, China	22 (M:7; F:15)	34.9 ± 12.2 (22–69)	–	PSG, Cephalometric analysis	–	N	Y	Y	Y	N	N	Y	Y
Hong et al., 2008 (case series)[19]	Seoul, South Korea	8	22–23	–	BMI, PSG	CPAP: Effective in 3 of 3 patients.	N	Y	Y	Y	N	N	Y	N
Iriarte et al., 2006 (retrospective case report)[15]	Pamplona, Spain	1 (F:1)	62	"many years ago"	PSG, Laryngoscopy	CPAP, effective	N	Y	Y	Y	N	N	Y	N
Iriarte et al., 2011 (retrospective case series)[25]	Pamplona, Spain	2 (F:1)	63.5 ± 2.1 (62–65)	Child-hood	PSG, Acoustic analysis	–	N	Y	Y	Y	N	N	Y	Y
Jaar et al., 2009 (retrospective case series)[8]	Osaka, Japan	8 (F:4)	33.4 ± 11.3	–	PSG	–	N	Y	Y	Y	N	N	Y	N
Jankovic et al., 2013 (case report)[32]	Belgrade, Serbia	1 (F)	37	–	PSG	–	N	Y	Y	Y	N	N	Y	N
Kapur et al., 2012 (retrospective case report)[33]	New York, United States	1 (M)	27	–	PSG, laryngoscopy	–	N	Y	Y	Y	N	N	Y	N
Koo et al., 2012 (case series)[17]	Seoul, South Korea	5 (F:5)	31 ± 6.9 (22–39)	14–32	PSG acoustic analysis, ESS, Mallampati, tonsil size	–	N	Y	Y	Y	N	N	Y	Y
Manconi et al., 2008 (retrospective case report)[34]	Milan, Italy	1 (M:1)	31	29	PSG	CPAP, ineffective	N	Y	Y	Y	N	N	Y	N
Muza et al., 2012 (retrospective case series)[23]	London, United Kingdom	18 (M: 10 F: 8)	19–61	Years to decades	PSG, ESS	CPAP: ineffective (0 of 4 patients improved); Zopiclone: ineffective (0 of 3 patients improved); Clonazepam: ineffective (0 of 1 patients improved).	N	Y	Y	Y	N	N	Y	N

BMI = body mass index, CPAP = continuous positive airway pressure, CT = computed tomography, EEG = electroencephalogram, ESS = Epworth Sleepiness Scale, MRI = magnetic resonance imaging, NICE = National Institute for Health and Clinical Excellence, PSG = polysomnogram, SD = standard deviation.

Table 3 continues on the following page

Table 3 (*continued*)—General characteristics and National Institute for Health and Clinical Excellence quality criteria of studies identified by systematic literature review.

Catathrenia: Systematic Review - Study General Characteristics							NICE: Quality Assessment Checklist							
Study, Year (Design)	**Study Site**	**n (sex)**	**Age, mean ± SD (range)**	**Age of Onset (y)**	**Variables Analyzed**	**Treatment**	**1**	**2**	**3**	**4**	**5**	**6**	**7**	**8**
Neutel et al., 2012 (case report)[20]	Lisbon, Portugal	1 (M:1)	32	–	PSG, EEG	CPAP (effective)	N	Y	Y	Y	N	N	Y	N
Oldani et al., 2005 (retrospective case series)[2]	Milan, Italy	21 (M:13, F:8)	31.4 ± 8.1 (8–36)	21.7 ± 7.0	PSG, ESS, Family History	Clonazepam: ineffective (0 of 2 patients improved); gabapentin: effective transiently for 6 mo and then returned (1 of 1), pramipexole: ineffective (0 of 1); trazodone: ineffective (0/1)	N	Y	Y	Y	N	N	Y	Y
Okura and Muraki, 2006 (retrospective case series)[10]	Osaka, Japan	18 (M:4, F:14)	22–59	–	PSG	–	N	N	N	N	N	N	N	N
Ott et al., 2011 (case report)[35]	Berne, Switzerland	1 (F)	29	14	Laryngoscopy, PSG	CPAP: effective	N	Y	Y	Y	N	N	Y	N
Overland et al., 2012 (prospective cohort study)[9]	Oslo, Norway	4	34.5 ± 4.7 (29–40)	–	BMI, ESS, PSG	CPAP: effective (1 of 4 improved)	N	Y	Y	Y	Y	N	Y	Y
Pevernagie et al., 2001 (retrospective case series)[3]	Gent, Belgium	10 (M:7, F: 3)	32.3 ± 8.8 (20–49)	12–36	PSG, MRI Brain (n = 7), CT Brain (n = 1)	CPAP: ineffective (0 of 2 patients improved, clonazepam: ineffective (0 of 3); trazodone, paroxetine, dosulepine: ineffective (0/5).	N	Y	N	Y	N	N	Y	Y
Prihodova et al., 2009 (case series)[16]	Prague, Czech Republic	8 (M:5, F:3)	22.6 ± 7.6 (11–32)	4–21	PSG	–	N	Y	Y	Y	N	N	Y	Y
Ramar et al., 2008 (case report)[36]	Rochester, Minnesota	1 (M:1)	41	36	BMI, PSG, ESS	–	N	Y	Y	Y	N	N	Y	N
Rogers et al., 2008 (case series)[14]	Cambridge, United Kingdom	13 (M:5, F:8)	34 ± 9.7	13–35	PSG, ESS, Family History	CPAP (attempted in 6; effective in 2 of 6)	N	Y	Y	Y	N	N	Y	N
Romigi et al., 2014 (case report)[37]	Rome, Italy	1 (F:1)	15	–	PSG	–	N	Y	Y	Y	N	N	Y	N
Siddiqui et al., 2008 (case report)[22]	Ohio, United States	1 (M)	13	8	PSG	–	N	Y	Y	Y	N	N	Y	N
Songu et al., 2008 (case report)[13]	Izmir, Turkey	1 (F)	40	7	BMI, Laryngoscopy, PSG	CPAP: effective	N	Y	Y	Y	N	N	Y	N
Steinig et al., 2008 (case report)[38]	Gottingen, Germany	1 (M)	33	–	BMI, PSG, ESS	CPAP: (mild improvement, but eventually rejected)	N	Y	Y	Y	N	N	Y	N
Vetrugno et al., 2001 (retrospective case series)[39]	Bologna, Italy	4 (M:3, F:1)	22 ± 4.8 (15–25)	5–16	PSG, family history,	–	N	Y	Y	Y	N	N	Y	N
Vetrugno et al., 2007 (case series)[4]	Bologna, Italy	10 (F:5)	27 ± 7.4 (15–41)	5–19	Family history, BMI, ESS, PSG, CT/MRI brain and neck, spirometry, laryngoscopy	–	N	Y	Y	Y	Y	N	Y	Y
Zinke et al., 2010 (retrospective case report)[21]	Jena, Germany	1 (M)	22	21	PSG	–	N	Y	Y	Y	N	N	Y	N

BMI = body mass index, CPAP = continuous positive airway pressure, CT = computed tomography, EEG = electroencephalogram, ESS = Epworth Sleepiness Scale, MRI = magnetic resonance imaging, NICE = National Institute for Health and Clinical Excellence, PSG = polysomnogram, SD = standard deviation.

Comorbid Medical Conditions

Table 2 summarizes the number of participants who responded "yes" when asked if they had any of the following conditions: anxiety or depression (21 of 47, 44.7%), gastroesophageal reflux (3 of 47, 6.38%), hypothyroidism (2 of 47, 4.3%), asthma (2 of 47, 4.3%), psychiatric disorder (2 of 47, 4.3%), high blood pressure (1 of 47, 2.1%), migraine (1 of 47, 2.1%), diabetes (1 of 47, 2.1%). In addition, 19 of 47 participants responded that they had no medical problems whatsoever (40.3%). None of the participants had any prior history of stroke, myocardial infarction, or heart problems as a result of lung disease (cor pulmonale). In the systematic review, three articles describing a history of epilepsy in their study subjects were identified.[16,20,21] Two articles described a history of allergic rhinitis.[16,22] In addition, a history of narcolepsy,[23] pulmonary hypertension,[13] mild mental retardation,[16] and depression[16] have also been described. One article described a patient with vocal cord paralysis and another with a vocal cord papilloma.[10]

Social History

Of the 47 participants, 13 (27.7%) reported that they have ever smoked tobacco and 6 (12.7%) were active smokers with mean use of 0.70 ± 0.65 packs of cigarettes per day (range, 1 cigarette per month to 2 packs per day, **Table 2**). There were 36 of 47 participants (76.6%) reported drinking alcohol with mean of 4.5 ± 4.6 drinks per week (range, once every few months to 20 drinks per week). Recreational drug use was reported by 6 of 47 participants (12.7%): 2 patients reported distant history of marijuana use, 2 reported continued occasional use of marijuana, 1 patient reported continued occasional use of ecstasy, 1 patient had a history of smoking methamphetamines. In the systematic review, we did not identify any articles that reported pertinent social history data regarding tobacco, alcohol, or recreational drug use.

Swimming

Of the 47 participants, 9 (19.2%) reported participation in recreational swimming; they reported an average 0.91 ± 0.60 days swimming per week (**Table 2**). These respondents all had a prior history of competitive swimming: 7 up to the of age of 13, 1 up to the age of 18, and 1 up to the age of 25. In the systematic review, none of the articles reported data on the prevalence of swimming in patients with catathrenia.

Systematic Literature Review

Fifteen case reports and 17 case series describing 191 patients with catathrenia were identified (aged 4 to 65 years, no significant sex predilection). We did not identify any reports that suggested catathrenia to be associated with significant risk of physical harm or long-term morbidity. Prior reports have attempted treatment with positive airway pressure with some limited success; surgical intervention and treatment with various medications (benzodiazepines, antidepressant, antiepileptic) has been attempted. See **Figure 1** and **Table 3**.

DISCUSSION

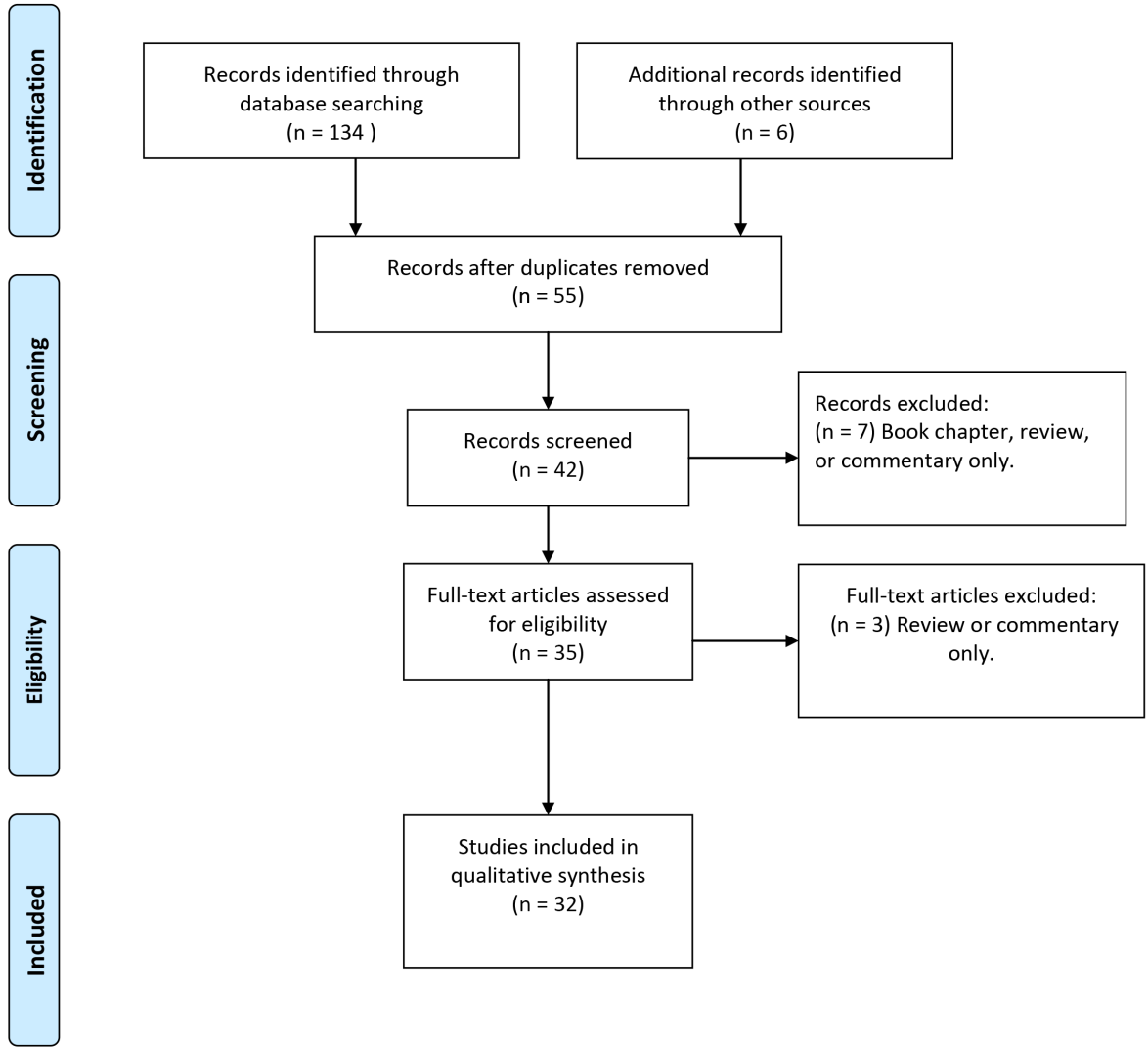
Catathrenia was formally classified as a parasomnia in the second edition of the International Classification of Sleep Disorders[24] but has more recently been classified among sleep-related breathing disorders in the third edition.[1] The classic polysomnographic description is a deep inhalation followed by a protracted exhalation during which moaning or groaning sounds are produced, usually lasting between 2 and 49 seconds.[25] Catathrenia occurs predominantly during REM sleep,[2,3,7,16,35,39] but has also been identified in non-REM sleep[39]; these episodes occur without observed respiratory distress and the arterial oxygen saturation remains normal.[26]

Catathrenia is distinct from both sleep talking (a parasomnia with loud talking during sleep) and snoring (noise due to vibration of upper airway soft tissues related to variations in airway resistance). It has been emphasized that catathrenia cannot be considered as expiratory snoring.[6] Acoustic analysis shows that the origins of the sounds in catathrenia and snoring and clearly different: catathrenia is laryngeal, whereas snoring is guttural. Iriarte et al. demonstrated the ability to calculate a main frequency, presence of harmonics, and a greater relative decibel intensity as features characteristic of catathrenia compared to the absence of these during snoring.[25] Indeed, the presence of harmonics supports sound of vocal origin—as this is distinct from the chaotic sound produced during snoring. In addition, Ott et al. identified active adduction and vibration of the vocal cords during expiration via laryngoscopy in a sedated patient with catathrenia, thus providing similarities between groaning and phonation.[35] The duration of the noise and the absence of any concomitant abnormal motor phenomena distinguish this expiratory sound from the moaning that occurs during epileptic seizures.[4] Interestingly, however, the REM predominance of catathrenia and the increased laryngeal motor activity seems to suggest that catathrenia has features of both a sleep-related breathing disorder and parasomnia (ie, REM Sleep Behavior Disorder).

Although the mechanism of action of CPAP treatment in patients with catathrenia is not well understood, Ott et al. demonstrated reductions in nocturnal groaning and improved daytime well-being following CPAP possibly from a reduction in active adduction during expiration.[35] Pharmacotherapy and cognitive behavioral therapy are other modalities others have used to manage catathrenia, though with varied results.[39] Although catathrenia has not been associated with significant risk of physical harm or long-term morbidity, it remains a significant cause for concern among patients and their bed partners. In this series, episodes of catathrenia were reported to occur on an almost nightly basis (6.6 ± 1.1 nights per week) and were associated with a moderate level of distress; by comparison, Oldani et al. reported a similar a frequency of 5.4 ± 1.5 nights per week.[2] The reasons for distress and concern include: disturbance to bed partner, worry about health consequences, effect on sleep quality, social embarrassment, and effect on relationship. Indeed, it has been described that the quality of groaning in catathrenia is monotone, and often presents with a morose or sexual connotation, causing a social problem for patients.[18]

Specific concerns offered by the participants in the series included the following: What causes catathrenia? Are there any associated health consequences? Is it associated with sleep apnea? What can be done to improve the condition? Is catathrenia

Figure 1—Flow diagram demonstrating systematic literature search results for original studies reporting cases of catathrenia.



n = number of articles.

associated with stress or anxiety? Is there an association with swimming? Could changing the sleep position help? Are there other natural forms of therapy to help reduce the chance of noisy breathing (ie, meditation, yoga?) Is treatment necessary, or can we just continue with the abnormal breathing as long as we are not bothered by the condition? Some of these concerns, it seems, may be amenable to more reliable information for patient and bed partner education.

Catathrenia has been described to occur in patients of all ages. The patients in this series were middle-aged, normal weight, otherwise healthy individuals who reported having experienced the unusual breathing pattern for many years. The majority of participants (94%) were unaware of their disorder and were made aware via bed partners. Men and women seem to be equally affected. The reported incidence of mild OSA diagnosed by polysomnogram was 34% in this series. Most patients did report experiencing unrefreshing sleep (83%), fatigue (77%), decreased daytime alertness (70%), decreased concentration or memory (61%), or insomnia (45%); however, it is unclear whether these symptoms are directly related to catathrenia.

These findings are similar to those summarized by Pevernagie et al.[3]:

> "Patients are unaware of their disorder, but may complain about restless sleep and mild daytime fatigue. The range of burden of insomnia or sleepiness complaints ranges from minimal to severe with most studies reporting burdens in the intermediate to severe range. For example, Drakatos et al. described moaning (52%), snoring (18%), and daytime sleepiness (45%) as presenting complaints in their cohort of 38 patients. Typically, the bed partner is the one who is most troubled by the nocturnal vocalization, as this occurs very often, if not every night. As a rule, there is no association with abnormal motor behavior, sleep talking, or recall of vivid dreams. There may be coincidental occurrence of snoring. Findings of clinical and neurological

examination are generally normal, or nonspecific if abnormal. There is at present no evident association with any predisposing factors or underlying disease."

In this review, we did identify an interesting relationship between catathrenia and swimming, as 20% of participants were active swimmers (approximately once a week) with a history of prior competitive swimming. By comparison, approximately 2% of high school students in the United States actively participate in competitive swimming.[40] In the words of one of the patients in the series, "I was a competitive swimmer in high school and college and the other day when I was swimming, it dawned on me that I hold my breath at night exactly how I hold my breath with swimming." We did not explore the effect of yoga or meditation but it is certainly possible that this could potentially ameliorate the condition. Sleep position (supine versus nonsupine) is unlikely to be related to the production of the expiratory groaning, based on prior literature review.

We did not identify any significant health consequences associated with catathrenia and most patients with an otherwise normal sleep study could be reassured that further treatment is not necessary. Although catathrenia may coexist with OSA, the prolonged groaning sounds occur during central apneas or the central component of mixed apneas. Prior reports do suggest successful treatment with CPAP.[18,19] However, surgical intervention does not appear to be of value. Treatment with various medications (benzodiazepine, antidepressant, antiepileptic) has also not been shown to be particularly effective. See **Table 2**. A study by Poli et al. showed catathrenia to be exacerbated by sodium oxybate.[27]

Implications for Practice

Although there is no association with risk of physical harm, catathrenia does present a significant cause for concern and anxiety among patients and their bed partners. Polysomnography can be useful if performed properly to confirm the diagnosis and to evaluate for comorbid sleep disturbances, such as OSA or parasomnia. Direction for further research could involve consideration of deep breathing exercises, yoga, meditation, or myofunctional therapy to help abate symptoms. Otherwise, bed partners may consider utilizing earplugs to minimize disturbance.

ABBREVIATIONS

BMI, body mass index
CPAP, continuous positive airway pressure
CT, computed tomography
EEG, electroencephalogram
ESS, Epworth Sleepiness Scale
ICSD, International Classification of Sleep Disorders
MRI, magnetic resonance imaging
NICE, National Institute for Health and Clinical Excellence
OSA, obstructive sleep apnea
PRISMA, Preferred Reporting Items for Systematic Reviews and Meta-Analysis
PSG, polysomnogram
REM, rapid eye movement
SD, standard deviation

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