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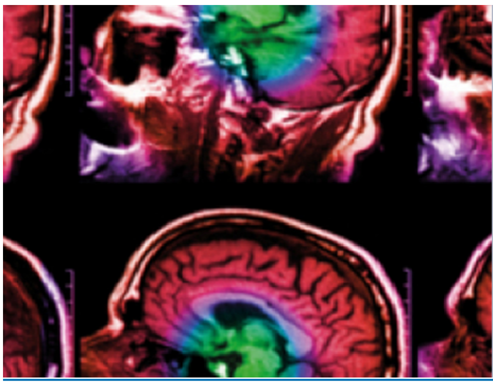

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Polysomnographic characteristics and acoustic analysis of catathrenia (nocturnal groaning)

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Abstract

Objective: Catathrenia is a sleep disorder characterized by nocturnal groaning sounds emitted during prolonged expiration. As a rare condition, its polysomnographic findings were inconsistent. We aimed to present polysomnographic characteristics of catathrenia patients and perform acoustic analysis of groaning sounds. *Approach*: Twenty-three patients (eight males and 15 females) diagnosed with catathrenia by video-polysomnography were included. They underwent clinical evaluation and physical examination, and answered a questionnaire. Acoustic analyses (oscillograms and spectrograms) of catathrenia and snoring signals were performed by Praat 6.1.09. Sounds were classified according to Yanagihara criteria. *Main results*: The average age of catathrenia patients was 29.6 $\pm$ 10.0 years, with a body mass index of 22.3 $\pm$ 5.1 kg m^{-2}. A total of 3728 groaning episodes were documented. Catathrenia events of 16 patients (70%) were rapid eye movement (REM)-predominant. The average duration of groaning was 11.4 $\pm$ 4.6 s, ranging from 1.3 to 74.9 s. All signals of groaning were rhythmic or semi-rhythmic, classified as type I and type II, respectively, with formants and harmonics. Snoring events were observed in nine patients. Snoring mainly occurred in the non-REM stage, with a duration of less than 1.5 s. Signals of snoring were chaotic, classified as type III, without harmonics. *Significance*: Catathrenia occurred in all sleep stages but mainly in REM. Durations of groaning varied greatly across patients. Acoustic characteristics of catathrenia were typical. Groaning had rhythmic or semi-rhythmic waveform, formants and harmonics, indicating vocal origin, while snoring had chaotic waveform.

1. Introduction

Catathrenia, also known as nocturnal groaning, is a rare condition classified as an isolated symptom and normal variant under sleep-related breathing disorders (American Academy of Sleep Medicine 2014). Patients with catathrenia represent approximately 0.063% to 0.54% of the population admitted to sleep centers (Oldani *et al* 2005, Abbasi *et al* 2012). The distinctive characteristic of groaning is a large inspiration followed by a protracted expiration during which a monotonous vocalization is produced.

The polysomnography (PSG) findings of catathrenia are inconsistent, including its distribution of sleep stages, duration of groaning sounds, onset time of groaning episodes, and association with other findings in PSG. Iriarte *et al* reviewed different reports of catathrenia and proposed two subtypes: type I—a long sound emitted mainly in rapid eye movement (REM) sleep; and type II—a very intense short sound emitted in both non-REM (NREM) and REM sleep (Iriarte *et al* 2015).

Catathrenia events might be confused with expiratory snoring (Guilleminault *et al* 2008, Vetrugno *et al* 2008), and these events can be discriminated by analysis of sound signals. Acoustic analysis has been performed in recent decades in differential diagnosis between simple snoring and obstructive sleep apnea (OSA), and in the detection of snoring sites (Agarwal and Mcguiness 1998, Hara *et al* 2006, Herzog *et al* 2008). As for catathrenia, sound analyses such as spectrograms and oscillograms have been carried out on seven patients, suggesting that the generator of the monotonous sound might be adjacent to the vocal cord region (Iriarte *et al* 2011, Koo *et al* 2012). Therefore, the aims of this study were to (1) present the clinical and PSG characteristics of catathrenia patients and (2) verify previous findings of the acoustic characteristics of groaning sounds with a larger sample size.

2. Methods

2.1. Subjects

This study is a retrospective consecutive case series. Twenty-three patients diagnosed with catathrenia were recruited from March 2016 to August 2019. Past medical history and family history were obtained. Standard clinical evaluations, Epworth sleepiness scale (ESS) assessments, physical exams, and craniofacial evaluations were carried out. The study was registered in the Chinese Clinical Trial Registry (No. ChiCTR-COC-17013239). It was approved by the committee of the Institutional Review Board of Peking University School of Stomatology (No. 201631128), and written informed consent was obtained from all subjects. The procedures used in this study adhere to the tenets of the Declaration of Helsinki.

2.2. Polysomnography

All patients underwent all-night video-PSG using the Compumedics E-series at Peking University People's Hospital. Recordings included a six-channel electroencephalogram (F4-M1, F3-M2, C4-M1, C3-M2, O2-M1, and O1-M2), a two-channel electrooculogram (E1-M2 and E2-M2), a two-channel electromyogram (submental and anterior tibialis muscles), and an electrocardiogram with surface electrodes. Nasal air pressure monitors, pulse oximeters, piezoelectric bands, and body position/respirator sensors were also attached to patients. Time-synchronized video was recorded, and audio was captured by a microphone positioned slightly below the patient's chin to reduce airflow effects.

All the records were scored manually by certified technicians and verified by a researcher according to the American Academy of Sleep Medicine (AASM) scoring manual (Berry *et al* 2012). Events of apnea and hypopnea were scored to calculate apnea hypopnea index. A groaning episode was defined as a deep inspiration followed with a prolonged expiration during which a monotonous sound was produced or an exhalation like a sigh was produced at the end of expiration. There should be no paradoxical movement of chest and abdomen during groaning. Groaning events were scored separately but not in clusters under the guideline of the International Classification of Sleep Disorders, 2nd edn (ICSD-2) in order to calculate groaning index (GI, groaning episodes per hour of sleep) (American Academy of Sleep Medicine 2005). Snoring was assessed by listening to the microphone, and sounds with an intensity exceeding 55 dB were scored as snoring (Hoffstein *et al* 1995, Arnardottir *et al* 2016). The technician/researcher who scored the PSG records did not participate in data measurement.

2.3. Acoustic analysis

If the total groaning episodes in REM or NREM sleep stages were less than ten, all the episodes were selected for analysis. If not, ten episodes of nocturnal sound were randomly selected for each sleep stage, respectively. The same method was used for the analysis of snoring events. All sound samples were digitized at 44 100 Hz, with a level of 16-bit quantization in mono. Acoustic analyses were performed using Praat 6.1.09 designed by Paul Boersma and David Weenink (Praat), including the oscillograms and spectrograms. The oscillogram consists of the analysis of sound intensity in a selected section and a microstructure of the sound. And the spectrogram is the analysis of the frequencies in the selected section, from 0 to 4500 Hz. The following measurements calculated by the analysis program were considered for the purposes of the present investigation: mean fundamental frequency (mean F_0, called 'mean pitch' in Praat), relative intensity, jitter (measure of frequency perturbation, called jitter [local] in Praat), and the presence of harmonics and formants. The analysis of the harmonics and formants spectrogram was conducted using the criteria of Yanagihara (Yanagihara 1967), according to which type I and II correspond, respectively, to periodic or almost regular signals generated by the vocal cords, while type III corresponds to non-periodic signals without harmonics.

2.4. Statistical analysis

The statistical analysis was performed using SPSS 26.0 (IBM Corp. IBM SPSS Statistics for Mac, Version 26.0, Armonk, NY). Normal distribution was verified by the Shapiro–Wilk test, and parameters were summarized as mean $\pm$ standard deviation. A paired-samples t-test was performed for inter-group comparison of each patient. Parameters of skewed distribution were summarized as median (interquartile range) and were analyzed using the Wilcoxon signed ranks test. Statistical significance was considered when $p < 0.05$.

3. Results

3.1. Clinical characteristics

Eight men and 15 women ranging in age from 14 to 56 years were included in the study. Detailed information on the subjects is shown in table 1. The mean age of recalled symptom onset was 18.3 ± 6.5 years. The most frequent complaints were disturbance of bedpartners or roommates (65%) and concern from family members (43%). Other complaints included dry mouth in the morning, unrefreshing sleep, and a lack of concentration during the day. Patient #19 had undergone bilateral tonsillectomy and pharyngoplasty, along with septoplasty. Patient #23 had Ménière's disease. Patient #17 and #20 reported a family history of snoring of their father. Other patients' family histories were unremarkable. None of the patients took medication regularly and they were all medication-free during the diagnostic PSG. Most of the patients had a normal body mass index, except for patients #1, #2, #12, #16, and #19. The Mallampati and the Friedman tonsil scales were within normal range. Only two patients had an ESS score of more than ten.

3.2. PSG characteristics

The detailed PSG record is shown in table 1. Patients had overall well-preserved sleep architecture and normal cyclic pattern. Sleep efficiency was greater than 75% in all but patient #12. Sleep latency was prolonged in some patients because of the first-night effect according to self-reports. Three patients (#1, #3, and #12) were diagnosed with mild OSA and patients #21 and #19 were diagnosed with moderate and severe OSA, respectively. Bruxism was observed in four patients (#4, #9, #10, and #14).

The characteristics of groaning and snoring episodes are shown in table 2. Groaning started between 20 and 195 min after sleep onset. A total of 3728 groaning episodes were recorded, among which 1493 events (40%) occurred during REM sleep and 2235 (60%) during NREM sleep. However, a REM-predominant distribution of catathrenia events was present in 16 patients (70%). The mean duration of all catathrenia events was 11.4 ± 4.6 s, ranging from 1.3 to 74.9 s. For REM sleep, it was 10.9 ± 4.1 s and for NREM sleep it was 11.6 ± 6.1 s, without a significant difference between sleep stages ($p = 0.444$). The median GI was 7.8 (4.1, 19.0) events per hour. REM-GI was 30.0 (19.7, 64.6) and NREM-GI was 2.2 (0.9, 10.7) events per hour, with a significant difference ($p < 0.01$). Snoring events were observed in nine patients. A total of 4577 snoring episodes were documented, of which 4190 (91.5%) occurred in the NREM stage. The duration of snoring was short, being less than 1.5 s.

The median number of arousals of all patients was 76 (65, 156) events and median arousal index (ArI) was 11.2 (9, 19.4) events per hour. Comparing to arousal norms by age (Bonnet and Arand 2007), five patients (#3, #10, #17, #20, and #23) had increased ArI, among whom arousals of patients #17 and #20 could be derived from groaning. Overall, EEG arousals preceded or coincided with the onset of 30.6 ± 20.3% of groaning events.

3.3. Sound analysis

Nocturnal groaning sounds on oscillograms and spectrograms had distinctive characteristics (figure 1 and table 3). The mean fundamental frequency (F_0) was 195 ± 46 Hz for all sleep stages, 195 ± 40 Hz during REM, and 196 ± 52 Hz during NREM, without significant difference between sleep stages ($p = 0.564$). The average jitter was 4.5 ± 1.4% and average intensity was 66 ± 4 dB. No statistical difference was present between REM and NREM sleep in jitter or mean intensity. All the episodes of groaning sound had harmonics and formants. The sounds of 13 patients (56.5%) could be classified as type I with sinusoidal waveform, whereas the sounds of ten patients (43.5%) could be classified as type II with semi-rhythmic sawtooth waveform. F_0 of type I was slightly higher than that of type II, but without statistical difference ($p = 0.749$). Fundamental frequency and jitter of snoring events could not be found. Thus, snoring was classified as type III, an irregular sound with formants but no harmonic reinforcement.

4. Discussion

The latency of groaning events from previous studies varied greatly, from 3 min to 8 h (Songu *et al* 2008, Koo *et al* 2012). In our study, the onset of groaning episodes was between 20 and 195 min after falling asleep. Some patients' groaning episodes in PSG were not as typical as during home recording, and some patients reported first-night effect, which affected indicators including sleep latency. The ICSD-2 established sound duration between 2 and 49 s (American Academy of Sleep Medicine 2005). However, episodes as short as 0.4 s were also reported (Abbasi *et al* 2012). We found that the average duration of groaning episodes was 11.4 s, ranging from 1.3 to 74.9 s. Patient #17 had a groaning episode in the N3 stage which lasted for as long as 74.9 s, but without oxygen desaturation. On the other hand, the durations of snoring events were all short, being less than 1.5 s.

Table 1. Demographic and PSG characteristics of patients with catathrenia.

Patient no.	Sex	Age (years)	Onset age (years)	BMI (kg m^{-2})	ESS	SL (min)	Stage R LAT (min)	SE (%)	AHI	Lowest SpO_2	ArI	Other findings
1	F	56	25	27.5	12	1.5	89.0	92.1	9.8	87	16.5	
2	M	36	21	29.7	8	30.5	52.0	89.8	2.9	93	15.1	
3	F	46	30	22.2	9	7.5	110.5	77.8	7.0	94	30.7	
4	M	34	25	22.2	5	8.5	57.0	92.2	0.0	98	7.4	Bruxism
5	F	43	28	21.0	9	16.0	74.0	93.9	0.0	98	12.8	
6	F	19	16	19.8	5	19.5	157.5	94.9	0.0	81	5.7	
7	F	30	22	21.6	6	49.0	108.0	84.3	0.4	79	9.7	
8	F	24	16	19.9	8	5.5	69.5	96.2	0.6	84	7.3	
9	F	23	19	21.6	8	1.5	69.5	95.1	0.9	87	10	Bruxism
10	F	25	18	16.8	10	0.5	207.5	91.7	2.3	87	42.1	Bruxism
11	F	14	10	17.6	6	2.5	80.5	96.5	0.7	93	7.4	
12	M	26	15	39.7	9	82.5	112.0	74.0	13.9	82	10.4	
13	F	18	18	19.0	5	18.0	106.0	95.9	3.9	92	9.5	
14	F	21	19	16.9	9	4.5	187.0	87.7	0.0	97	11.2	Bruxism
15	F	33	26	18.8	10	8.5	203.5	80.2	4.2	95	15.8	
16	M	36	20	25.5	11	8.5	91.0	93.6	0.0	91	10	
17	M	25	18	19.3	11	55.5	71.5	81.2	1.0	94	22.5	
18	F	32	23	21.3	6	10.5	162.0	96.0	4.7	94	9	
19	M	35	31	25.3	10	9.0	159.0	88.7	57.4	79	19.3	
20	M	33	18	24.5	13	5.5	127.0	95.0	0.1	92	26.3	
21	M	32	18	23.2	8	2.5	112.5	94.9	15.2	90	19.4	
22	F	19	5	22.5	5	4.0	180.5	93.8	0.6	94	7.6	
23	F	20	17	17.5	8	2.5	55.0	95.9	0.3	95	21.4	
Total M±SD/MED(IQR)	8/15	29.6 ± 10.0	18.3 ± 6.5	22.3 ± 5.1	8.3 ± 2.3	15.4 ± 20.6	114.9 ± 49.2	93.6 (87.7, 95.1)	0.9 (0.1, 4.7)	92 (85.5, 94)	11.2 (9, 19.4)	

Note: BMI, body mass index; ESS, Epworth sleepiness scale; SL, sleep latency; Stage R LAT, sleep onset to first epoch of REM sleep; SE, sleep efficiency; AHI, apnea hypopnea index; Lowest SpO_2, lowest oxygen saturation; ArI, arousal index; M±SD/MED(IQR), mean ± standard deviation/median (interquartile range).

Table 2. Characteristics of groaning and snoring events.

Patient no.	Groaning episodes			Groaning duration (s)					Snoring events			
	Total	REM	NREM	Total	Min	Max	REM	NREM	Total	REM	NREM	Durations (s)
1	50	40	10	18.0	4.1	39.9	18.4	16.4	681	174	507	<1
2	41	37	4	12.4	4.0	50.4	11.3	22.5	281	4	277	<1.5
3	119	85	34	12.4	2.2	32.8	11.4	14.8	232	39	193	<0.5
4	24	8	16	18.1	4.9	36.7	12.9	20.0				
5	191	182	9	5.4	2.6	9.5	5.4	6.5	28	4	24	<1
6	8	2	6	18.3	11.6	32.4	15.1	19.4				
7	111	77	34	9.5	1.9	39.6	7.5	14.0				
8	32	32	0	10.2	4.1	29.1	10.2	0.0	26	2	24	<0.5
9	94	78	16	14.2	3.8	31.1	14.3	13.2				
10	179	119	60	10.7	4.1	27.6	10.5	10.8				
11	42	34	8	12.1	3.5	27.5	12.5	9.1	92	0	92	<1
12	108	22	86	10.1	2.0	27.6	7.2	10.8	1415	132	1283	<0.5
13	765	114	651	3.5	2.2	20.7	4.4	3.3				
14	35	25	10	14.6	8.7	25.7	14.3	13.7				
15	12	10	2	4.8	2.4	10.1	5.1	3.6				
16	50	34	16	14.4	3.6	44.6	15.4	11.9				
17	131	46	85	17.6	9.7	74.9	16.2	17.8				
18	30	26	4	14.8	6.7	25.9	14.1	19.1				
19	329	231	98	5.7	1.3	25.1	6.2	4.6	1131	10	1121	<1.5
20	1271	201	1070	4.8	2.1	42.5	8.6	4.1				
21	6	2	4	6.2	4.8	7.0	5.8	10.0	691	22	669	<1
22	43	41	2	13.8	4.9	56.9	14.1	8.9				
23	57	47	10	9.6	4.3	25.5	8.9	13.1				
Total M±SD/MED(IQR)	50 (32, 131)	40 (25, 85)	10 (4, 60)	11.4 ± 4.6	1.3	74.9	10.9 ± 4.1	11.6 ± 6.1	4577	387	4190	<1.5

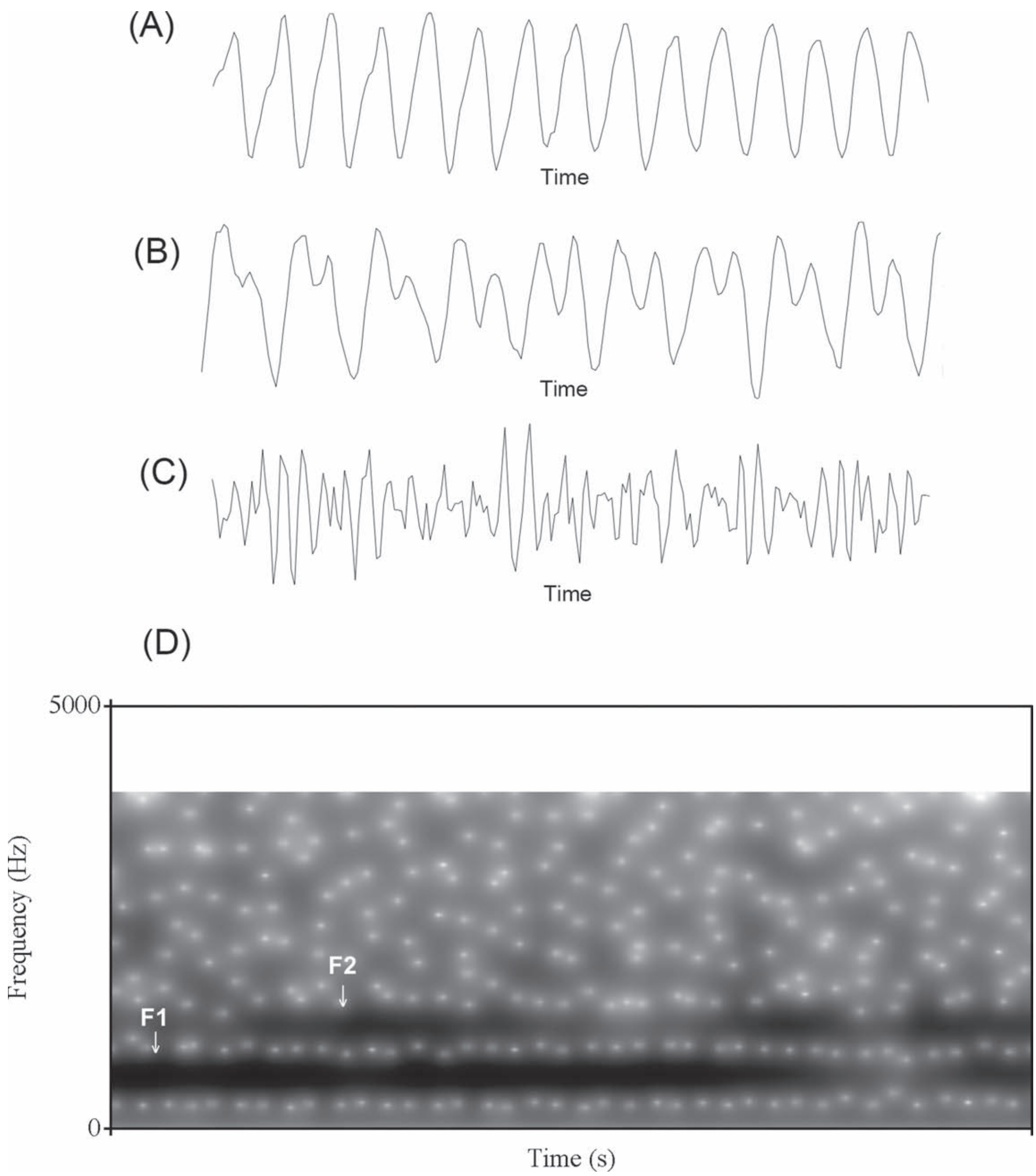

Figure 1. Oscillograms of groaning and snoring sounds. The visible part is 25 ms. (A) This signal of a catathrenia episode is type I, which is a rhythmic sinusoidal waveform. (B) This signal of a catathrenia episode is type II, which is a semi-rhythmic and sawtooth waveform. (C) This signal of snoring is type III, which is a non-periodic and chaotic waveform. (D) Spectrogram of a catathrenia episode, which shows two formants (F1 and F2), represented by white arrows.

Catathrenia events of most patients were REM-predominant in our study, which was consistent with previous findings (Pevernagie *et al* 2001, Vetrugno *et al* 2001, Oldani *et al* 2005). However, neither sound duration nor acoustic analyses were different between REM and NREM sleep. On the other hand, snoring tended to occur in the NREM stage. Guilleminault *et al* reported seven patients with short-duration groaning events present through all stages of sleep, raising the question of subtypes of catathrenia (Guilleminault *et al* 2008). A similar non-REM type was supported by other researchers' reports (Songu *et al* 2008, Abbasi *et al* 2012). However, for the seven patients with non-REM distribution in our study, the average sound duration was 11.2 s, without a significant difference to that of REM-predominance.

Muraki *et al* documented that EEG arousal occurred before the onset of 90.5% of groaning events (Muraki *et al* 2015). And Darakatos *et al* also found EEG arousals preceded or coincided with the onset of 84% of catathrenia events (Drakatos *et al* 2017). However, such a close association between EEG arousal and groaning episode was not observed in our study. An average of 30.6% of catathrenia events occurred in tandem with an arousal, and only patient #20 had 84.9% of groaning events occurring right after EEG arousal.

Table 3. Acoustic analyses of groaning and snoring sounds.

Patient no.	Sleep stage	F_0 (Hz)	Jitter	Mean intensity (dB)	Harmonics	Formants	Waveform	Yanagihara classification
1	R	152	5.3%	65	Yes	Yes	Sawtooth	II
	N	167	5.6%	65				
	Snore	Undefined	Undefined	65	No	Yes	Irregular	III
2	R	224	4.4%	67	Yes	Yes	Sawtooth	II
	N	269	4.7%	63				
	Snore	Undefined	Undefined	65	No	Yes	Irregular	III
3	R	282	7.0%	58	Yes	Yes	Sawtooth	II
	N	258	9.1%	63				
	Snore	Undefined	Undefined	65	No	Yes	Irregular	III
4	R	257	5.2%	64	Yes	Yes	Sinusoidal	I
	N	351	6.0%	66				
5	R	213	4.7%	77	Yes	Yes	Sawtooth	II
	N	158	1.2%	72				
	Snore	Undefined	Undefined	67	No	Yes	Irregular	III
6	R	129	8.5%	66	Yes	Yes	Sinusoidal	I
	N	140	2.9%	67				
7	R	193	4.5%	69	Yes	Yes	Sinusoidal	I
	N	228	4.2%	74				
8	R	154	4.1%	67	Yes	Yes	Sawtooth	II
	Snore	Undefined	Undefined	62	No	Yes	Irregular	III
9	R	142	3.6%	74	Yes	Yes	Sawtooth	II
	N	119	2.2%	72				
10	R	174	4.8%	66	Yes	Yes	Sinusoidal	I
	N	219	4.3%	63				
11	R	150	3.7%	64	Yes	Yes	Sawtooth	II
	N	144	4.8%	60				
	Snore	Undefined	Undefined	58	No	Yes	Irregular	III
12	R	221	4.7%	66	Yes	Yes	Sinusoidal	I
	N	221	3.2%	67				
	Snore	Undefined	Undefined	65	No	Yes	Irregular	III
13	R	238	2.6%	68	Yes	Yes	Sinusoidal	I
	N	181	3.1%	68				
14	R	210	4.2%	65	Yes	Yes	Sinusoidal	I
	N	189	3.8%	69				
15	R	205	5.2%	61	Yes	Yes	Sawtooth	II
	N	193	5.3%	65				
16	R	194	4.5%	59	Yes	Yes	Sinusoidal	I
	N	180	4.6%	62				
17	R	237	5.0%	64	Yes	Yes	Sinusoidal	I
	N	210	4.6%	64				
18	R	173	5.8%	65	Yes	Yes	Sinusoidal	I
	N	143	4.0%	66				
19	R	200	4.9%	67	Yes	Yes	Sinusoidal	I
	N	192	3.8%	69				
	Snore	Undefined	Undefined	66	No	Yes	Irregular	III
20	R	235	4.4%	63	Yes	Yes	Sinusoidal	I
	N	131	2.9%	63				
21	R	189	4.2%	74	Yes	Yes	Sawtooth	II
	N	217	5.5%	74				
	Snore	Undefined	Undefined	58	No	Yes	Irregular	III
22	R	153	2.8%	65	Yes	Yes	Sawtooth	II
	N	198	4.0%	68				
23	R	162	3.0%	64	Yes	Yes	Sinusoidal	I
	N	204	3.5%	67				
Total	All	195 ± 46	4.5 ± 1.4%	66 ± 4				
	R	195 ± 40	4.7 ± 1.3%	66 ± 4				
	N	196 ± 52	4.2 ± 1.6%	67 ± 4				

Note: F_0, mean fundamental frequency; R, REM sleep; N, NREM sleep.

Groaning sounds had distinctive characteristics on acoustic analyses. Signals were organized, periodic, or almost regular on oscillograms, and fundamental frequency could be defined on spectrograms. All these signals had formants and harmonics, indicating that the sound was produced by the vocal folds (Pevernagie *et al* 2010). In contrast, signals of snoring were non-harmonic, even chaotic, and fundamental frequency could not be found.

The average F_0 of catathrenia in our study was 195 ± 46 Hz, which was lower than the 285.2 ± 100.1 Hz of the report by Koo *et al* (2012) and the 240 to 396 Hz found by Iriarte *et al* (2011). Differences might derive from the different demographic characteristics of these studies. In addition, different recording procedures, data acquisition systems, protocols for processing sound, and the software used for data analysis might also lead to these differences (Dalmasso and Prota 1996). Nevertheless, the findings of acoustic characteristics of groaning sounds and snoring events were consistent between these studies and ours.

5. Conclusion

In conclusion, catathrenia occurred in all sleep stages, but mainly in REM. Latency and duration of groaning varied greatly across patients. Acoustic characteristics of catathrenia were typical. Groaning had rhythmic or semi-rhythmic waveform, formants, and harmonics, indicating vocal origin, while snoring had chaotic waveform.

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