

Idiopathic central sleep apnea with periodicity presenting as Cheyne-Stokes breathing – Case report

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ABSTRACT

Cheyne-Stokes breathing (CSB) is described by a cyclic variation in breathing with periods of central apneas or hypopneas alternating with periods of hypopneas in a gradual waxing and waning manner. It is usually seen in patients with congestive cardiac failure or neurologic disorders. We describe this rare case of idiopathic central sleep apnea coming in a periodic fashion presenting as CSB pattern without specific causes. Overnight polysomnography test showed central sleep apnea; but cardiac evaluation and magnetic resonance imaging of the brain could not find any cause of this sleep-disordered breathing. Idiopathic central sleep apnea usually presents with poor quality of sleep, but many of them easily misdiagnosed and do not get any proper treatment.

Keywords: Sleep Apnea Syndromes. Cheyne-Stokes Respiration. Polysomnography

BACKGROUND

A central sleep apnea (CSA) is defined as pause in airflow for more than 10 seconds in the absence of any respiratory effort (1). CSA is characterized by repeated apneas during sleep with no associated respiratory attempt. A frequency of more than five per hour is considered abnormal. The pathogenesis and etiology of CSA is still vague. There are several manifestations of CSA; which includes high altitude periodic breathing, narcotic-induced central apnea, obesity hypoventilation syndrome, idiopathic central sleep apnea (ICSA) and Cheyne-Stokes breathing (CSB). ICSA is comparatively rare form of central sleep apnea (2). Cheyne-Stokes breathing (CSB) is defined as periods of central apneas or hypopneas alternating with periods of hypopneas with a cyclic fluctuation in breathing, usually occurs in a gradual waxing and waning style (3). It is most commonly seen in patients with central nervous system dysfunction and congestive cardiac failure (4). Thus we report a patient; whose overnight polysomnography (PSG) showed central sleep apnea with a crescendo-decrescendo appearance; resembles CSB pattern.

Patient complained sleep fragmentation and excessive daytime sleepiness; but detailed neurologi-

cal and cardiac evaluation did not show any significant abnormalities to support CSB.

Poor sleep quality in elderly patients may not always suggest primary insomnia. Early identification and proper treatment may helpful for good prognosis. There are only few case reports were published, so we think more studies required to describe this phenomena.

Case Report

Seventy two year old male (body mass index: 21.9 kg/m²) visited our neurology outpatient department, tertiary level hospital of South Indian capital city, with the complaints of excessive daytime sleepiness and fatigue for about 2 years. He was not on any medications. His usual bed time is 10pm to 6am. Even though he has slept 8-9 hours every day; he never gets satisfaction in sleep. No neurological deficit was seen in clinical evaluation. On detailed sleep history patient denied snoring, cataplexy, hypnogogic hallucination, sleep paralysis and sleep attack. ENT and cardiac evaluation was normal. Blood tests, including thyroid function tests were normal except for slight elevation in liver enzymes. Arterial blood gas analysis showed pH 7.47, PaCO₂ 37.0mmHg,

PaO₂ 72.9 mmHg, HCO₃⁻ 27.3 mmol/L, Base excess 3.5 mmol/L.

An overnight PSG was done. PSG showed total sleep time of 443 minutes, sleep latency of 19.0 minutes, and sleep efficiency of 70.0%. Sleep architecture for stage 1 sleep was 31.7%, stage 2 sleep was 52.4%, slow wave sleep was 12.9%, REM sleep was 3.1%. Total arousal index was 21.8/hr and Apnea Hypopnea Index was 48 events/hour. Mean SaO₂ was 96.4%, the lowest SaO₂ was 82.8%, and SaO₂ below 90% was 0.1%. Snoring was not observed and periodic limb movement index was 8.8/hr. PSG also showed cyclic fluctuation in breathing with periods of central apneas or hypopneas alternating with periods of hyperpneas in a gradual waxing and waning fashion (Figure 1). We evaluated the patient to find out the cause of CSB pattern. There was no ventricular hypertrophy or dysfunction and ejection fraction was 70% on two dimensional echocardiography. No abnormal findings were found on magnetic resonance imaging of the brain.

DISCUSSION

The PSG in this case showed cyclic fluctuations in breathing with periods of apneas or hypopneas irregular with periods of hypopneas in a steady waxing and waning manner, with lacking ventilatory

effort as well as recurrent arousals from sleep at the peak of hyperpnea. But, this CSB prototype in our case is not classic. The length of the ventilatory apnea sequence in is almost longer than 45 seconds in CSB model⁵. The duration of the apneic cycles in our case were usually less than 35 seconds (Figure 2). We did not find the specific causes of CSB pattern and so we entitled it periodic breathing pattern with central sleep apnea presenting CSB. Esophageal pressure or end-tidal CO₂ monitoring sensors were not used in this case, but there was no obstructive apnea by nasal pressure sensor throughout the overnight monitoring. His arterial blood gas analysis showed mild respiratory alkalosis and metabolic alkalosis. Although PaCO₂ during sleep was not checked, there was no constant oxygen desaturation during sleep that was inexplicable by isolated apnea and hypopneas on PSG. His body mass index was within normal range and neurological clinical examination was normal. Sleep related hypoventilation/hypoxemic syndrome could be easily rule out in this case. The CSB pathophysiology of congestive heart failure can be explained with elevated ventilatory drive, small difference between the apneic threshold and sleeping eupneic PaCO₂, extended circulation time, and raise in reflex stimulation of breathing, but the CSB pathophysiology of brain vessel diseases and renal failure are not yet clearly de-

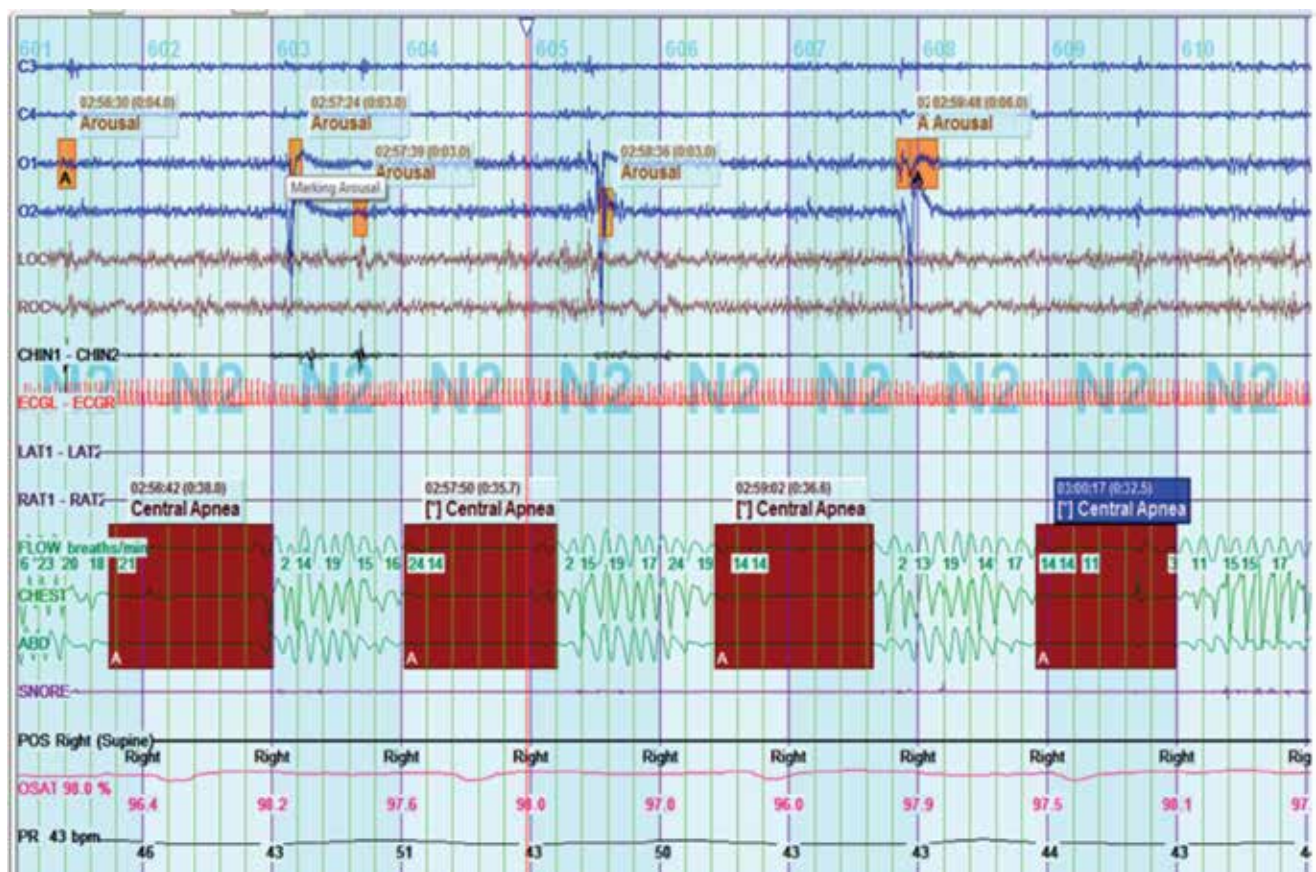


FIGURE 1. Cyclic fluctuation in breathing with periods of central apneas (5 minutes/page)

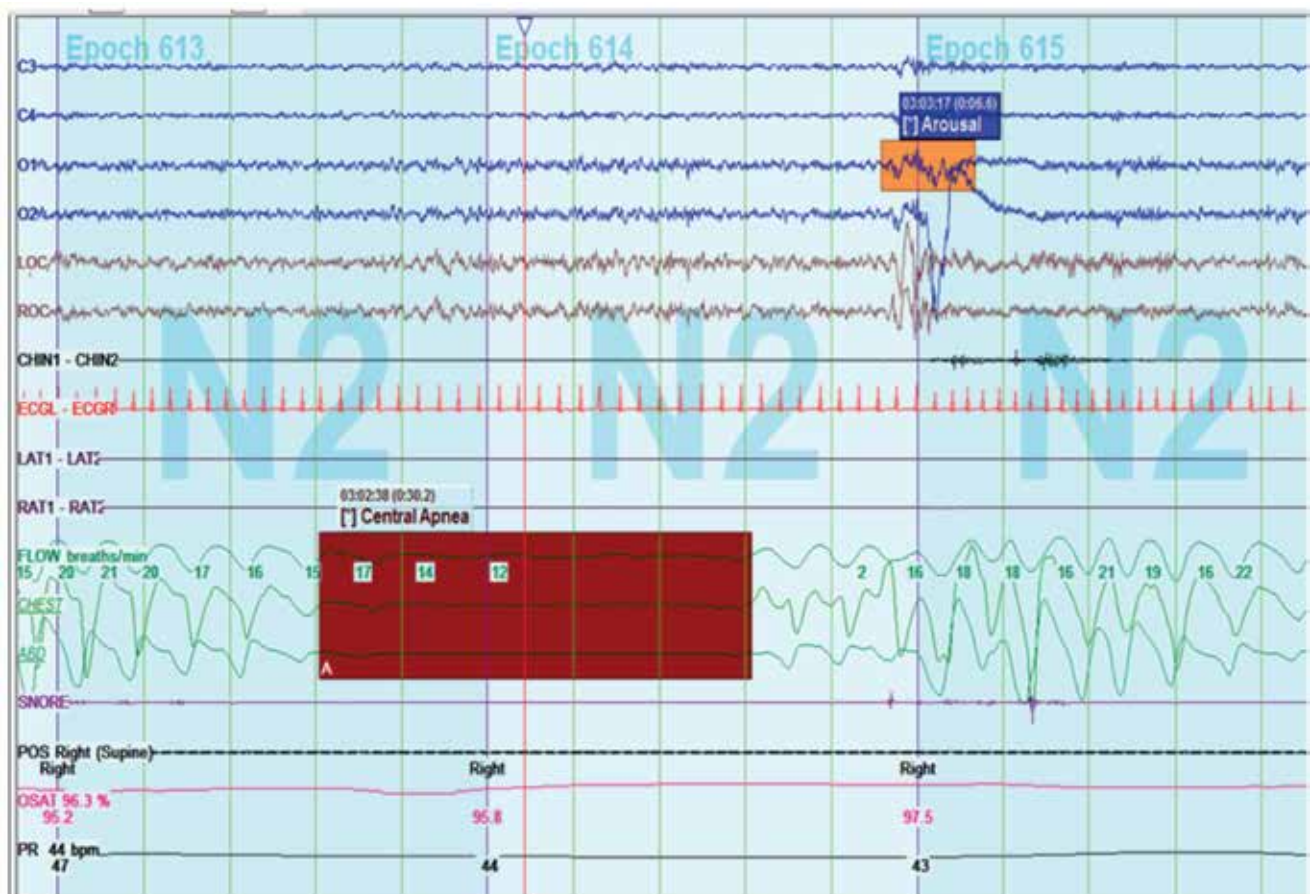


FIGURE 2. Central apnea lasting about 30 seconds (90 minutes/page)

fined (6). This case might also have ventilatory instability although the cause of ventilatory instability was not detected.

A previous case report by Cormican et al; male patient presented with apparent idiopathic central sleep apnea but after 4 years developed features of multiple system atrophy (7). Only few studies were available in Indian sub-continent. Ravi et al. reported as case regarding ICSA in 2015 (8). His study restates the importance of diagnosis of ICSA in patients with chronic non-refreshing sleep and not improving to any treatment to any treatment. Similarly to our case report; Kang et al. also reported a

patient with idiopathic central sleep apnea; presenting as CSB pattern (9). We think that further studies will be required to determine whether idiopathic central sleep apnea presenting as CSB pattern is the initial indication of certain diseases. Due to the poor recognition of ICSA; many of them do not get proper treatment which may lead to stroke and cardiac dysfunctions. An overnight polysomnography test is very useful in this condition. We advised continues positive airway pressure therapy and patient is on close follow up. So we concluded that this case was an idiopathic central sleep apnea with periodic pattern; presenting as CSB pattern.

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