CASE REPORT

Central sleep apnoea syndrome with upper airway collapse

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Central sleep apnoea syndrome with upper airway collapse. J.L. Pépin, A. Pajon, D. Veale, G. Ferretti, P. Lévy. ©ERS Journals Ltd 1993.
ABSTRACT: We report on an 83 yr old man with hypersomnia and central sleep apnoea (CSA). He had several possible causes for CSA, including a central nervous system lesion, hypocapnia and anatomical narrowing of the airway at the hypopharyngeal level. We postulate that reduced central respiratory drive occurring in conjunction with upper airway narrowing may have led to central apnoeas. These in turn could have facilitated a complete passive hypopharyngeal collapse at the end of each apnoea, as visualized by somnofluoroscopy. The CSA could also have been favoured by respiratory instability due to chronic hypocapnia.
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Apnoeas during sleep can be obstructive (OSA) or central (CSA), or a combination of both (mixed sleep apnoea (MSA)). In OSA there is a cessation of airflow but respiratory efforts continue. In CSA airflow ceases for at least 10 s and there are no thoracoabdominal movements. CSA may have a variety of causes, such as a failure of central respiratory generation with lesions of the central nervous system (CNS) [1]. Alternatively, there may be reflex inhibition of ventilation through activation of upper airway receptors [2], or CSA may occur in response to instability of respiratory control, as occurs at altitude [3], with metabolic disorders [4], and with heart failure [5, 6].

We report a patient with a number of possible causes for CSA. Lateral fluoroscopy of the pharynx with polysomnography during sleep allowed visualization of pharyngeal collapse at the end of central apnoeas. Continuous positive airway pressure applied through the nose (nCPAP) prevented the apnoeas. This stimulated closer examination of the possible causes of CSA in this patient.

Case report

An 83 yr old man presented with severe daytime hypersomnolence. He had a pacemaker *in situ* for atrioventricular block with Stokes-Adams attacks. Because of iatrogenic hypothyroidism due to amiodarone therapy, he was also on adequate thyroid replacement therapy, with 100 µg thyroxine daily. He further complained of brief episodes of nocturnal dyspnoea, and a recent loss of intellectual capacity. He also admitted to mild infrequent snoring in recent months, as described by his wife. Clinical examination quickly revealed daytime somnolence, as the patient fell asleep during the consultation!

Physical examination showed a respiratory rate of 25 breaths·min⁻¹ at rest. The patient had a broadly based gait, and was unable to maintain his equilibrium with his eyes shut. Cranial nerve examination was normal, with no nystagmus, and normal gag reflex. Position and touch sensations were normal. He had mild pyramidal weakness in the right arm, and slightly reduced ankle tendon jerks, but other tendon reflexes were normal. The patient was neither obese nor hypertensive. Computed tomography (CT) scan of the brain showed cerebello-bulbar atrophy. Auditory evoked responses were normal. Cardiological review showed no evidence of cardiac failure or structural abnormalities on echocardiography.

On spirometry, forced expiratory volume in one second (FEV_1) was 1.86 l and vital capacity 2.59 l (both 82% of predicted). Blood gas analysis, breathing air, seated at rest showed: pH 7.46, arterial oxygen tension (Pao_2) 102 mmHg (13.6 kPa), arterial carbon dioxide tension ($Paco_2$) 32 mmHg (4.3 kPa), and arterial oxygen saturation (Sao_2) 97%.

Standard polysomnography was performed with recording of electroencephalogram (EEG), electrooculogram (EOG), and chin electromyogram (EMG). Airflow was assessed using three thermistors mounted on nasal spectacles. Respiratory efforts were monitored using uncalibrated inductive plethysmography. Sao_2 was measured using a Biox-Ohmeda 3700 oximeter. The polysomnogram was scored manually, according to standard criteria [7].

The patient slept in the dorsal decubitus position. There were 98 apnoeas+hypopnoeas·h⁻¹ of sleep, of which 86% were central (fig. 1), 4% hypopnoeas, and 10% mixed apnoeas. Mean nocturnal Sao_2 was 94%, with continuous Sao_2 oscillations (from 97 down to 90%), corresponding to repeated short central apnoeas, which

were nearly always followed by an arousal evident on EEG. Thus, sleep was very fragmented, with 211 changes of sleep stage; there was a predominance of stage I sleep, with no stage III or IV or rapid eye movement (REM) sleep.

Cephalometric roentgenograms were performed, and analysed following the technique described by RILEY et al. [8]. These measurements showed a reduction of posterior airway space (PAS 6 mm). The length of the soft palate was within the normal range at 37 mm. The uvula was angulated. CT-scanning of the pharynx showed a reduction of the luminal cross-sectional area of the airway at the level of hypopharynx (cross sectional area 168 mm²).

Because of the reduction of the hypopharyngeal lumen, lateral fluoroscopy of the pharynx with polysomnography during sleep was performed and recorded on video. The records were analysed focusing on: area of commencement of obstruction of the airway, extent of the collapse, and morphology of this collapse. Somnofluoroscopy also allowed visualization of events occurring outside the pharyngeal airway during apnoeas, such as movements of the cervical spine and the hyoid bone. This showed hypopharyngeal collapse at the end of the central apnoeas, without any movement of the soft palate, cervical spine or hyoid bone (fig. 2). The collapse tended to extend upwards to the oropharynx. At the time of the
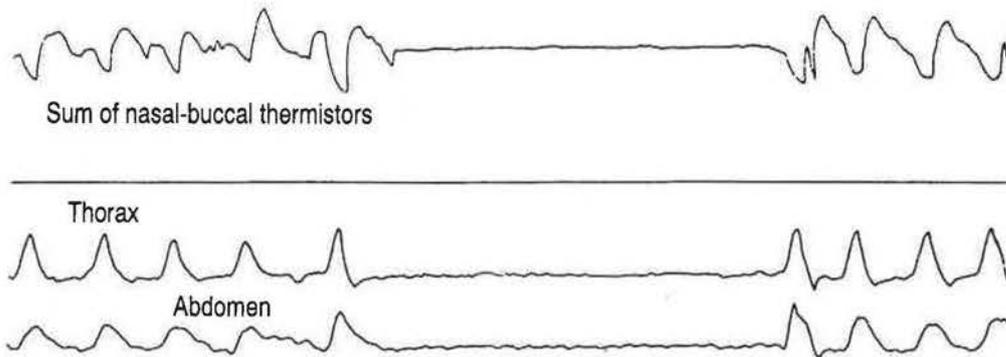

Fig. 1. – Typical tracing of a central apnoea (CSA). The duration of this event is 13 s. There is no thoracic or abdominal movement. One can see cardiac oscillations on the inductance plethysmography lines.
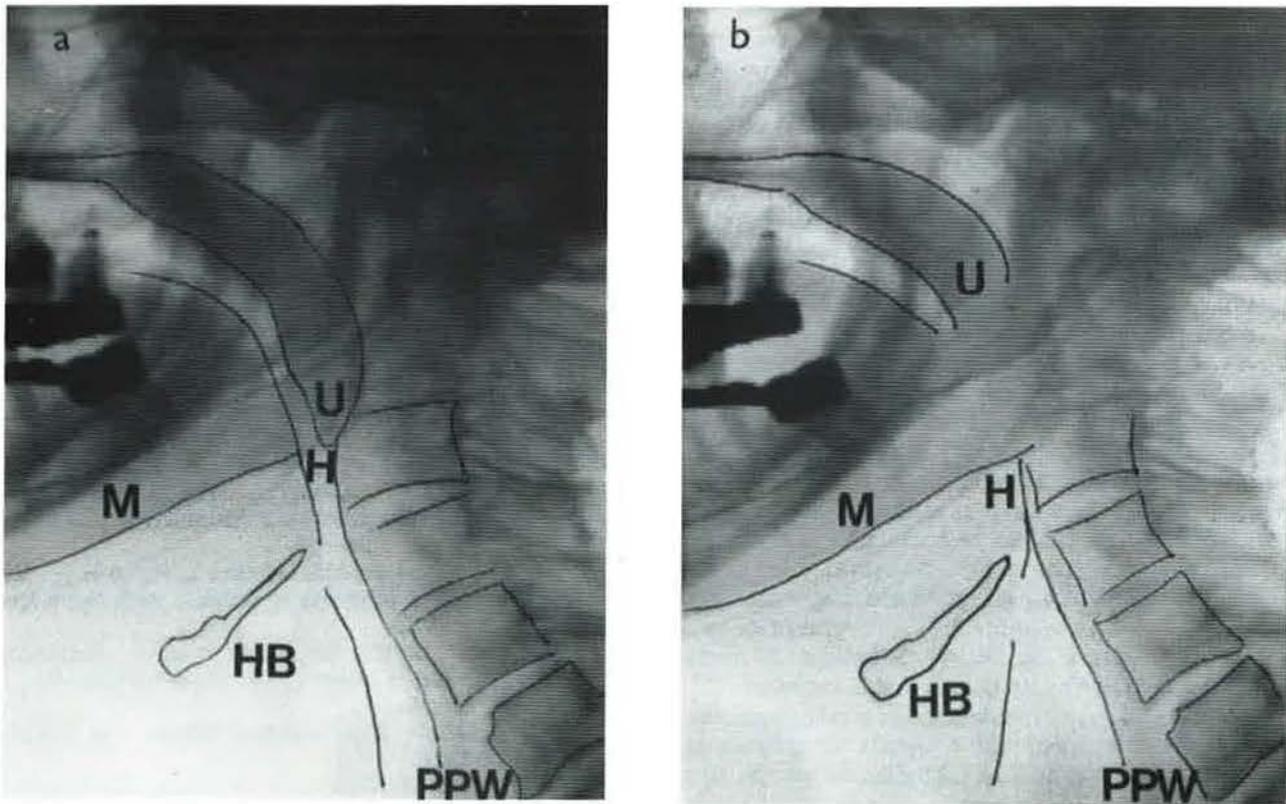

Fig. 2. – Fluoroscopic images: a) at the beginning; and b) at the end of the CSA, showing the hypopharyngeal collapse whilst the oropharynx is still open. U: uvula; H: hypopharynx; HB: hyoid bone; PPW: posterior pharyngeal wall. The total time between the two fluoroscopic images was 10 s.

collapse, we observed the previously described angulation of the uvula, but no hooking of the soft palate. There was no movement of the hyoid bone, nor cervical spine movements as we have observed in obstructive sleep apnoea syndrome (OSAS) [9].

Continuous positive airway pressure at 10 cmH$_2$O applied through the nose prevented CSA, as documented by polysomnography showing 30 apnoeas and hypopnoeas (5·h^{-1}) and a recovery of a nearly normal sleep structure: 4 sleep cycles with 4 REM epochs and 2 epochs at stage II and IV. Hypersomnia was dramatically improved under treatment.

Discussion

The distinction between CSA and OSA can be made by non-invasive techniques, such as inductive plethysmography, without the use of oesophageal balloon monitoring, as this is adequate to detect respiratory effort, particularly in non-obese patients as in our case [10, 11]. Furthermore, a catheter through the nose and pharynx to measure oesophageal pressure, may interfere with sleep and alter the type of apnoea observed [12].

This patient had CSA of multiple possible causes, which allowed examination of the interaction of causal factors and mechanisms of development of this disorder. Although a number of CNS lesions have been described as causes of CSA, this is unlikely to be the case in our observation. From clinical presentation and CT scan results, no clear cerebello-bulbar lesion could be inferred. A magnetic resonance scan could have given a more precise localization of the patient's anatomical lesions, but it was impossible because of his pacemaker. The auditory evoked responses were normal. Moreover, the most frequently associated abnormalities are an alteration of ventilatory responses to hypoxia and hypercapnia [13]. We have not examined the ventilatory responses in this 83 yr old man with poor coordination. His low Paco$_2$ and normal Pao$_2$ do not suggest depression of CO$_2$ response, at least whilst awake [14].

Hypocapnia and alkalaemia may, however, be important factors in the development of CSA in our patient. The Paco$_2$ level normally present during wakefulness is lower than that prevailing during sleep. Thus, with the onset of sleep, ventilation may cease, transiently, until Paco$_2$ rises to the sleeping set-point value. Our patient had frequent changes from sleep to wakefulness, which could have generated fluctuations between apnoeic and hyperpnoeic ventilatory patterns [14]. Moreover, when hyperventilation and alkalosis are present during wakefulness (analogous to high altitude [3]), this phenomenon is exaggerated because the difference between the awake Paco$_2$ and the sleeping set-point value is increased [15].

There was no clinical, radiological or echographic evidence of left ventricular failure, despite the patient's age and pacemaker requirement. Cardiac failure [5, 6] is thought to cause CSA by means of an increased circulation time between peripheral chemoreceptors and the respiratory centres, thereby leading to a delay in respiratory response to CO$_2$.

Although we lack direct evidence, we hypothesize a role for local factors in the genesis of CSA in this case. As postulated by BRADLEY et al. [16], partial or complete collapse of the pharynx, facilitated by increased pharyngeal compliance, may stimulate mechanoreceptors in the airway and reflexly inhibit central respiratory drive. In a patient with an anatomically narrow upper airway, instability of ventilatory control may become critical, and lead to central apnoea. Furthermore, ISSA and SULLIVAN [2] have shown that the application of local anaesthetic to the pharynx diminished the number of central apnoeas or transformed CSA into OSA.

Thus, we speculate that our patient had a narrowed hypopharynx, which became more compliant during sleep. Local reflexes were then initiated, which induced central inhibition of respiration. As a consequence of the concomittant upper airway muscle tone reduction, the hypopharyngeal collapse could occur progressively as a passive mechanism until the end of the central apnoea. In OSAS, in contrast, airway collapse occurs rapidly, with increased transpharyngeal pressure related to persistent respiratory efforts.

The effectiveness of nCPAP supports the suggestion of a role for local upper airway factors in the genesis of CSA in this case. Reflex central stimulation of breathing in response to local pressure has not been demonstrated [6]. The effects of nCPAP on respiratory instability related to hypocapnia remain unknown.

In conclusion, local factors in the upper airway, and hypocapnia, may play combined or complementary roles in the development of an instability of central respiratory control and, thereby, facilitate the development of CSA. Correction of the peripheral disturbance may break the cycle of events and reverse the disorder.

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