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Case Report & Case Series

Glioblastoma in the limbic system presenting as sustained central hypopnea

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ABSTRACT

A 71-year-old woman was transferred to our hospital after experiencing an epigastric sensation followed by unconsciousness. On arrival, the patient showed impaired consciousness without convulsive movement, cyanosis and shallow breathing, arterial O_2 desaturation, and increased PCO_2. Artificial respiration improved CO_2 accumulation and consciousness, but interruption of artificial respiration returned the patient to her former state. Computed tomography of the head showed a mass around the left corpus callosum. The patient's hypopnea followed by unconsciousness suggested sustained nonconvulsive epilepsy manifesting in central hypopnea and subsequent unconsciousness due to CO_2 narcosis. Intravenous (IV) anticonvulsants promptly improved the respiratory condition, and the patient started to regain consciousness. Magnetic resonance imaging revealed a lesion involving the bilateral limbic systems. To our knowledge, limbic seizure manifesting with hypopnea causing unconsciousness due to CO_2 narcosis has not previously been reported, despite evidence of a strong relationship between the limbic and respiratory systems. The current case suggests that sustained limbic seizure can manifest as hypopnea. Since emergency EEG can be difficult to perform, IV anticonvulsant treatment is an appropriate diagnostic therapy.

1. Introduction

The limbic system has been suggested to have a strong relationship with the respiratory system [1,2]. A case of partial epilepsy presenting as episodic dyspnea caused by glioblastoma multiforme (GBM) was previously reported as limbic epilepsy [3]. However, to our knowledge, no case of sustained limbic epilepsy manifesting as hypopnea accompanied by unconsciousness due to CO_2 narcosis has previously been reported. Here, we report a case suspected to be limbic epilepsy manifesting as hypopnea, and discuss the treatment of this condition.

2. Case report

A 71-year-old woman was transferred to our hospital after experiencing an epigastric sensation and unconsciousness. On admission, the patient had a Glasgow Coma Scale score of 7 points (E2-V1-M4), with no convulsive movement. The patient exhibited cyanosis and shallow breathing. The saturation O_2 monitor under room air at the time was 80%. Arterial blood during oxygen mask therapy showed increased PCO_2 up to 79.1 mm Hg, and base excess −2.3. Artificial respiration assistance improved CO_2 accumulation and consciousness. However, interruption of artificial ventilation returned the patient to her former state. Computed tomography (CT) of the head showed a slightly high density mass around the left corpus callosum (Fig. 1). We considered that the patient's hypopnea associated with impaired consciousness may have been caused by sustained nonconvulsive epilepsy manifesting as central respiratory failure; therefore, we administrated intravenous (IV) diazepam and phenytoin. IV anticonvulsants promptly improved the respiratory condition and the patient started to regain consciousness after her respiratory condition improved. However, the full recovery of consciousness took several hours because of the sedative effects of diazepam. Electroencephalography (EEG) 6 days after admission did not show epileptic discharge.

A fluid-attenuated inversion recovery (FLAIR) image on a magnetic resonance imaging (MRI) scan of the patient's head showed a high-intensity lesion in the bilateral hippocampus (Fig. 1a), and the left side of the corpus callosum (Fig. 2b). A T1-weighted image with gadolinium showed heterogeneous enhancement around the left retrocommissural hippocampus (Fig. 2c). Subtotal removal of the enhanced lesion was performed. The pathological diagnosis was GBM. At 12-month follow-

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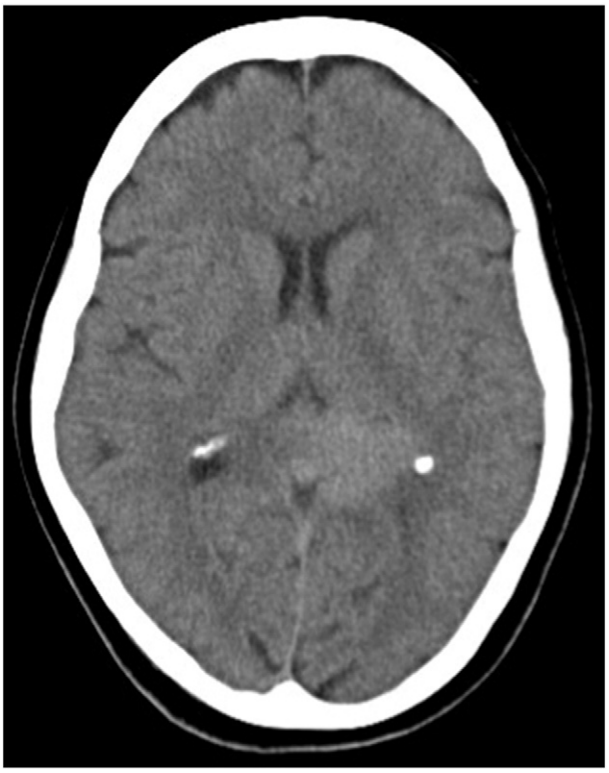

Fig. 1. CT on admission showing slightly high-density mass around the left corpus callosum.

up, the patient remained free of hypopneic episodes, with ongoing oral anticonvulsant treatment.

3. Discussion

We reported a case of GBM, causing a hypopnea attack accompanied by unconsciousness due to CO_2 narcosis. Dyspnea was resolved by administration of anticonvulsants. Thus, we consider that this patient's condition was a type of sustained nonconvulsive epilepsy.

To our knowledge, no case of sustained nonconvulsive epilepsy manifesting as hypopnea accompanied by CO_2 narcosis and unconsciousness has been previously reported. It is important to discuss forms of epilepsy that may manifest as hypopnea, and potential treatments.

Studies of the relationship between central apneas or hypopneas and epilepsy have reported an association with sudden unexpected death in epilepsy (SUDEP) [1,4]. Bateman et al. [1] examined ictal hypoxemia and SUDEP, reporting that central apneas or hypopneas occurred in 50% of seizures. They found that desaturation was significantly correlated with seizure localization in the temporal lobe. It has been reported that stimulation of the hippocampal formation in rats evoked decreases in respiration [2]. In the clinical setting, one case of partial epilepsy presenting as episodic dyspnea caused by GBM in the medial temporal lobe was reported [3]. These findings support the hypothesis that the limbic system lesion in the present case caused epileptic discharge and subsequent hypopnea.

EEG recording is often difficult to perform during sustained hypopnea, because patients typically require artificial ventilation. If ictal EEG was recorded, it may have classified the present case as nonconvulsive status epilepticus. As a supplemental diagnostic measure, IV antiepileptic treatment is recommended [5]. In the present case, IV anticonvulsants promptly resolved the patient's respiratory failure. However, IV diazepam can have significant adverse effects in cases of respiratory depression [6]. Thus, in patients with hypopnea and hypercarbia, special care must be given to IV administration of diazepam and risk-benefit should be carefully estimated to prevent respiratory conditions from worsening.

Tumors in the limbic system can cause hypopnea as a symptom of epilepsy. Because obtaining EEG in the ictal phase is often difficult, IV antiepileptic drug treatment is an appropriate diagnostic therapy. We propose that sustained limbic epilepsy manifesting as hypopnea leading to CO_2 narcosis is possible, and that IV anticonvulsant treatment is an appropriate diagnostic therapy.

Conflicts of interest/disclosures

The authors declare that they have no financial or other conflicts of interest in relation to this research and its publication.

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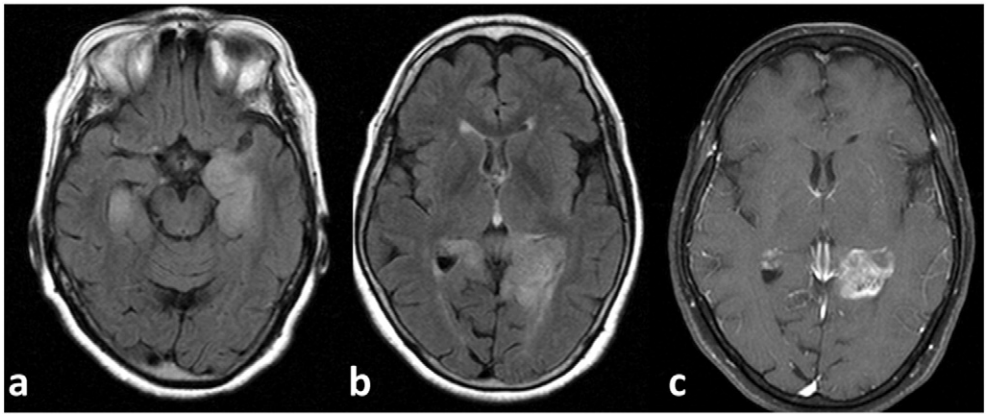

Fig. 2. MRI FLAIR image on admission showing high-intensity lesion in bilateral hippocampus (A) and around left corpus callosum (B). Contrasted T1WI showing heterogeneous enhancement around the left retrocommissural hippocampus (C).