Cluster Breathing Associated With Bihemispheric Infarction and Sparing of the Brainstem

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**Objective:** To report cluster breathing pattern associated with a nonbrainstem lesion.

**Design:** Case report.

**Setting:** Neurointensive care unit, St Mary’s Hospital, Rochester, Minn.

**Patient:** A patient with subarachnoid hemorrhage developed severe, diffuse, distal bilateral middle cerebral artery vasospasm with resultant cortical laminar necrosis and transient cluster breathing.

**Intervention:** Magnetic resonance imaging revealed bihemispheric lesions but no brainstem lesion.

**Conclusion:** Cluster breathing may occur with nonbrainstem lesions.

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VARIOUS FORMS OF BRAIN INJURY, including subarachnoid hemorrhage, are associated with abnormal breathing patterns.1,2 Biot,3 in 1876, described an abnormal breathing pattern in a patient with tuberculous meningitis, which was later interpreted by Fisher4 to be an ataxic respiratory pattern. Biot's name was later ascribed incorrectly to breathing abnormalities seen in cat experiments.5 Despite the confusion over the use of Biot's name, the pattern of cluster breathing was clearly defined in 1982 by Plum and Posner6 as irregular clusters of breaths followed by apneic episodes of variable duration. Cluster breathing is invariably described in the literature as associated with upper medulla or lower pons (pneumotaxic center) lesions.5,6 Cluster breathing has not been described, to our knowledge, in bihemispheric lesions without brainstem abnormality.

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REPORT OF A CASE

A 66-year-old, right-handed woman was brought to our emergency department after she experienced sudden loss of consciousness. On arrival, she was comatose (Glasgow Coma Scale score, 3) with intact brainstem reflexes. An endotracheal tube was inserted for airway protection. An emergent external ventriculostomy was performed after a non–contrast medium–enhanced computed tomographic imaging of the head showed diffuse subarachnoid hemorrhage and acute hydrocephalus. A cerebral angiogram revealed an anterior communicating artery aneurysm that was occluded by endovascular coiling. The patient's neurologic status improved, and the endotracheal tube was removed on hospital day 2. On hospital day 7, right-sided hemiparesis developed, which, on day 10, worsened, with development of expressive aphasia. Cranial nerve examination yielded normal findings with the exception of right lower facial weakness (ipsilateral to hemiparesis). Pupils were 4 mm in diameter, equal, and reactive to light bilaterally, with intact ocular motility and corneal, cough, and gag reflexes. The endotracheal tube was reinserted secondary to neurologic deterioration and lack of airway protection. Repeat non–contrast-enhanced computed tomography of the head revealed evolving hypodensity in the left middle cerebral and left anterior cerebral artery territories. A second cerebral angiography failed to show significant vasospasm of the large intracranial arteries, with the ex-
ception of the left anterior cerebral artery, and decreased flow in the distal middle cerebral artery territories. Verapamil, 5 mg, was administered intra-arterially in the left anterior cerebral artery. Because of the patient’s decline in consciousness and worsened right hemiparesis and ischemic changes seen on computed tomographic images, hemodynamic augmentation therapy was initiated. The ventricular drain was replaced with a lumbar drain on hospital day 14 to prevent ventricular drain infection. Tracheostomy was performed by an otolaryngologist on hospital day 19. Cluster breathing became evident (Figure 1) the next day with use of a humidified tracheostomy collar during the day and persisted with continuous positive airway pressure overnight. Neurologic examination revealed global aphasia, intact cranial nerves except for right lower facial weakness, severe right-sided hemiparesis with hyperreflexia, and a positive right-sided extensor toe sign. The patient’s respiratory pattern was characterized by approximately 6 to 8 rapid, large tidal volume breaths in approximately 14 seconds, followed by apneic pauses of approximately 10 seconds. This pattern was repetitive but was not uniformly periodic. The patient’s vital signs were otherwise unremarkable. On hospital day 21, magnetic resonance imaging was performed because of concern that a pontine lesion might be responsible for the cluster breathing (Figure 2). The magnetic resonance images demonstrated extensive laminar cortical necrosis of the hemispheres (left side greater than the right side) and no brainstem ischemia or infarcts. Clinical pulmonary examination yielded normal findings; a chest x-ray film was normal. On hospital day 25, cluster breathing stopped. The patient underwent percutaneous gastrostomy, and the lumbar drain was removed. The patient remained

Figure 1. Respiratory monitoring. The patient’s respiratory excursions are shown in the bottom tracing in each group. The patient’s cluster breathing was irregular with an approximate 13-second apneic pause preceded by a cluster of breaths (6-8) over approximately 14 seconds. Several respiratory cycles are shown. Other intensive care unit vital sign monitoring is shown and includes an electrocardiographic tracing (A), arterial blood pressure (B), and pulse oximetry (96%-97%) (C). Noninvasive blood pressure determination (157/75 mm Hg) is shown at the lower left in each image.
neurologically impaired during hospitalization, with global aphasia and right-sided hemiparesis, and was discharged to home under her husband’s care after 44 days of hospitalization.

Laboratory values during cluster breathing included stable anemia (hemoglobin level, 89 g/L) and normal levels of serum urea nitrogen, creatinine, bicarbonate, and serum electrolytes. Arterial blood gas values were as follows: pH, 7.54; PCO₂, 29 mm Hg; and PO₂, 93 mm Hg, with room air. Cerebrospinal fluid obtained from the lumbar drain (not the reservoir bag) under normal cerebrospinal fluid pressure included the following values: lactic acid, 28 mg/dL (reference, <23 mg/dL); glucose, 38 mg/dL (2 mmol/L); protein, 67 g/dL; white blood cell count, 1.6 × 10⁶/µL, with 74% lymphocytes, 24% monocytes, 2% neutrophils; and red blood cell count, 9.9 × 10⁶/L. Gram stain and cultures of cerebrospinal fluid were negative for bacteria and fungal organisms.

The importance of respiratory patterns and their localizing value in the diagnosis of neurologic disorders was described by Plum and Posner.6 The various breathing abnormalities in stupor and coma, with associated localization, potential causes, and description, are given in the Table. Cluster breathing is characterized by clusters of breaths followed by apneic episodes of variable duration, typically caused by low pontine or high medullary lesions.6 Other reports of cluster breathing include cerebellar hemorrhage with brainstem compression,9 Shy-Drager disease,7 and anoxic encephalopathy with ocular bobbing.10 Our patient developed transient cluster breathing with evidence at magnetic resonance imaging of bilateral cortical laminar necrosis that resulted from severe vasospasm after subarachnoid hemorrhage. Cortical lesions may be associated with the Cheyne-Stokes

Figure 2. Magnetic resonance images. A, Diffusion-weighted image reveals no areas of restricted diffusion in the pons or brainstem. Areas of T2 shine-through appear as restricted or hyperintense areas in the inferior and anterior temporal lobes but did not appear as hyperintense areas on apparent diffusion coefficient images. B, Diffusion-weighted image shows areas of restricted diffusion predominantly in the left hemisphere along gyri and sulci. C, Coronal T1-weighted gadolinium-enhanced sequence shows no signal abnormality of brainstem but demonstrates increased signal in a gyral-sulcal pattern of the left sylvian fissure. Trace, similar signal changes are seen in the right sylvian cortex. D, Sagittal T1-weighted image shows signal hyperintensity along the left perisylvian fissure and along other left hemisphere gyri and sulci, consistent with cortical laminar necrosis.

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breathing pattern; however, our patient did not demonstrate a crescendo-decrescendo (spindle) breathing pattern. Central neurogenic hyperventilation with posthyperventilation apnea is another possible cause of the breathing pattern in our patient; however, central neurogenic hyperventilation is thought to be rare and is typically associated with midbrain lesions, although bihemispheric lesions have been described.6,8 Diagnostic criteria for central neurogenic hyperventilation include hyperventilation with marked respiratory alkalosis (high arterial pH, low arterial PaCO₂, and high PaO₂).8 While our patient had arterial blood gas values consistent with respiratory alkalosis, the pattern of inspiration (6 to 8 rapid, deep breaths followed by intervening apnea) was not consistent with the pattern seen in central neurogenic hyperventilation or the patterns seen in short-cycle Cheyne-Stokes respiration described by Fisher.4 In addition, we found no literature, except our own, that describes arterial blood gas values in cluster breathing. Further, our patient was asleep with cluster breathing, which is indicative of disrupted automatic breathing and eliminates volitional hypopnea as a potential cause.11

The cluster breathing pattern in our patient was related to bihemispheric cortical damage seen on magnetic resonance images, which, to our knowledge, has not been previously described. We are uncertain as to how cortical lesions, which classically are lesions of the pontomedullary region, cause cluster breathing; however, we believe that the case we describe may provide evidence for cortical system integration into brainstem mechanisms that govern respiration.

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