

Pseudohypoxic Brain Swelling after Unilateral Burr Hole Drainage: A Novel Case Report

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Abstract

An 85-year-old man underwent emergency right trepanation and drainage for a symptomatic chronic subdural hematoma. Pseudohypoxic brain swelling (PHBS) was suspected because magnetic resonance imaging revealed diffuse brain swelling. Although cerebrospinal fluid (CSF) leakage was not obvious during or after surgery, most of the reported PHBS cases have leaked CSF during craniotomy or spine surgery. PHBS has not been previously reported in patients without obvious CSF leakage or after unilateral burr hole drainage. Herein, we report an extremely rare case with a literature review investigating its pathogenesis and clinical features.

Keywords: pseudohypoxic brain swelling, chronic subdural hematoma, burr hole drainage, postoperative intracranial hypotension-associated venous congestion, cerebrospinal fluid leakage

Introduction

First reported by Van Roost et al. in 2003, pseudohypoxic brain swelling (PHBS) is extremely rare and potentially leads to fatal post-surgical complications following even uneventful brain or spine surgery. PHBS is characterized by an unexpected postoperative deterioration of consciousness, with magnetic resonance imaging (MRI) showing radiological changes in the basal ganglia, thalamus, brainstem, and cerebellum similar to those of hypoxic encephalopathy but without radiological changes in the cerebral vessels.¹ An English literature search in the PubMed database revealed that most cases follow craniotomy and spine surgery. The cause of PHBS is speculated to be cerebrospinal fluid (CSF) leakage. There was one report of excessive CSF drainage following bilateral chronic subdural hematoma (CSDH) burr hole drainage,² but no case without obvious CSF leakage or after unilateral burr hole drainage has been reported. Herein, we report an extremely rare case with a literature review investigating its pathogenesis and clinical features.

Case Report

An 85-year-old man with unremarkable medical history had a head injury 51 days before admission and was being followed up with computed tomography (CT) for a small CSDH. On the day of admission, the patient had mild gait instability and drowsiness. The head CT scan revealed a right-sided frontal subdural hematoma with mass effect and midline shift (Fig. 1A&B). No base of skull fracture or pneumocephalus was noted, and CSF leakage was not obvious (Fig. 1C&D). No laboratory findings suggestive of abnormalities, such as dehydration or infection, were found. On the first day of admission, the patient underwent emergency right trepanation and drainage under local anesthesia. The total volume drained was 30 mL, and there were no findings suggestive of decreased intracranial pressure. The burr hole was tightly closed with the temporalis muscle and subcutaneous tissue, with a drainage tube left *in situ*. Postoperatively, the patient drained a total of 55 mL with no obvious abnormalities over 19 h, but his consciousness level declined to a Glasgow Coma Scale (GCS) score of 8. Subsequently, another head CT scan was performed, which confirmed that the subdural hematoma had

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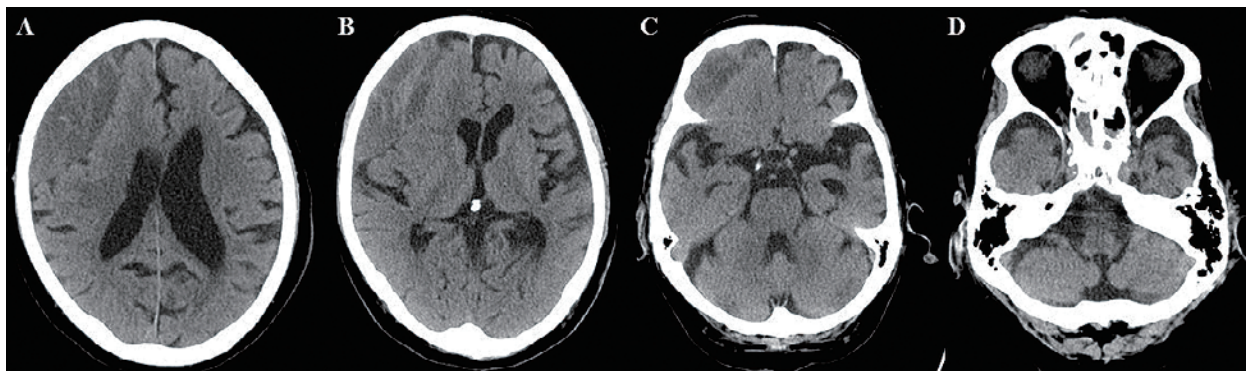


Fig. 1 Computed tomography (CT) scan upon admission revealed a right-sided frontal subdural hematoma with mass effect and midline shift (A, B). No base of the skull fracture or pneumocephalus was noted, and CSF leakage was not obvious (C, D).

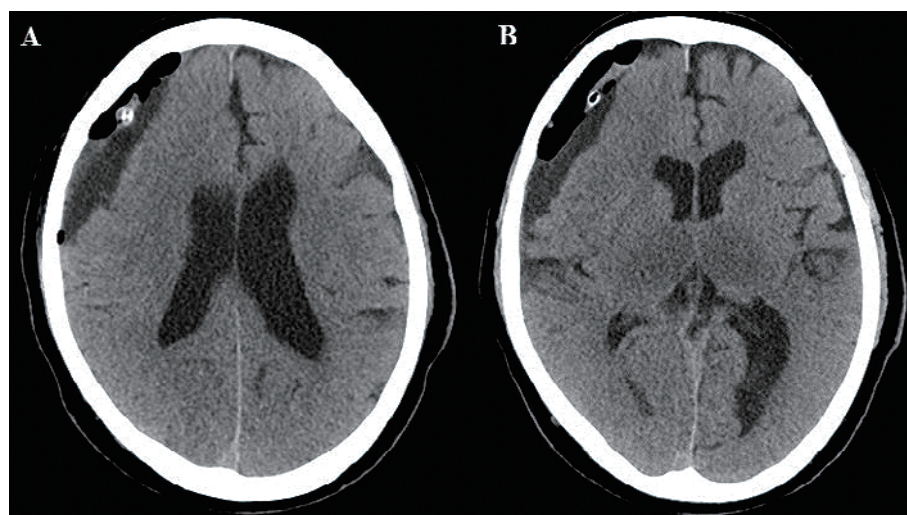


Fig. 2 CT scan obtained after the operation revealed that the subdural hematoma was successfully removed (A), with no obvious abnormalities, aside from low-density lesions in both thalami (B).

been successfully removed (Fig. 2A&B), with no other obvious abnormalities noted aside from low-density lesions in both thalami (Fig. 2B). The patient was observed for possible prolonged effects of local anesthesia, which may alter his consciousness. On postoperative day (POD) 1, his consciousness was still impaired, and he developed new-onset seizures; therefore, a head MRI was conducted, which revealed bilateral thalamic hemorrhages (Fig. 3A-C) and fluid-attenuated inversion recovery hyperintensity in the basal ganglia, thalamus, brainstem, and cerebellum similar to those in hypoxic encephalopathy (Fig. 3D&E). The intracranial and extracranial arteries and cerebral veins exhibited no apparent changes radiologically (Fig. 3F-H), and no dilation of the cortical veins suggestive of venous infarction was observed (Fig. 3C). Arterial spin labeling (ASL) showed an increase in perfusion of the right frontal lobe and brainstem on POD 1 (Fig. 3I), but ASL on POD 4 revealed no difference in perfusion between the left and right sides (Fig. 3J). During the intraoperative and postop-

erative periods, the patient remained hemodynamically stable with a normal sinus rhythm and stable respiratory function. Laboratory parameters, including glucose, ammonia, electrolytes, and autoantibodies, were within normal limits, and there were no signs of infection. A lumbar puncture was performed, and CSF examination revealed no obvious abnormalities.

We administered anti-hypertensive, anti-epileptic, and anti-edema treatment, including steroids. The patient's level of consciousness gradually improved; thus, steroids were continued until POD 14, and he was transferred to a convalescent hospital for rehabilitation after his GCS score reached 14 and his modified Rankin scale score reached 3. Informed consent was obtained from the patient for the publication of this case report and accompanying images.

Discussion

PHBS is a rare condition that usually occurs after brain

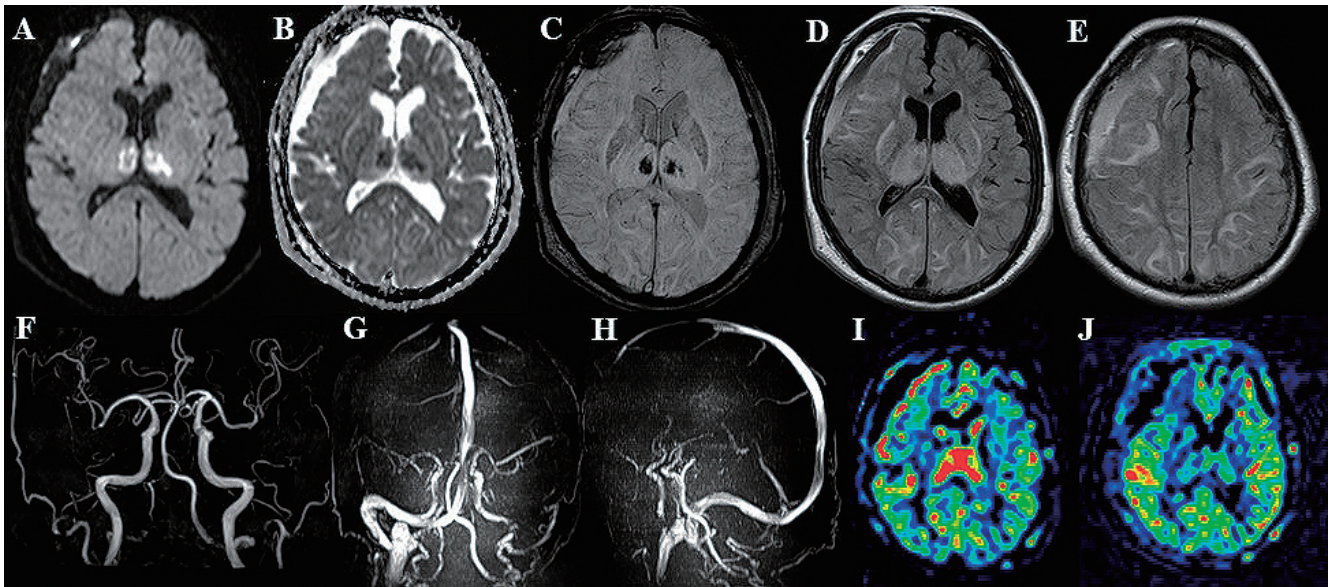


Fig. 3 Magnetic resonance imaging revealed areas of high intensity on diffusion-weighted imaging (A), whereas apparent diffusion coefficient showed low intensity in both thalami (B), and susceptibility-weighted imaging showed bleeding in the same area. Dilatation of the cortical veins suggestive of venous infarction was not observed (C). There was fluid-attenuated inversion recovery hyperintensity in the basal ganglia, thalamus, brainstem, and cerebellum, similar to hypoxic encephalopathy (D, E). Magnetic resonance angiography and venography revealed no abnormalities (F, G, H). Arterial spin labeling (ASL) showed an increase in perfusion of the right frontal lobe and brainstem on postoperative day (POD) 1 (I), but the ASL imaged on POD 4 showed no difference between the left and right sides (J).

or spine surgery, with sudden neurological decline, new-onset seizures, and brainstem dysfunction. Its clinical course varies from full recovery to death. It was first reported by Van Roost et al. in 2003 and is sometimes called postoperative intracranial hypotension-associated venous congestion (PIHV). In this paper, this entity is referred to as PHBS. Typical findings on MRI and/or CT are radiological changes in the basal ganglia, thalamus, brainstem, and cerebellum, similar to hypoxic encephalopathy, with no apparent changes in the cerebral vessels.¹⁾

The pathophysiology is neither well understood nor completely elucidated. The Monro-Kellie doctrine³⁾ has been proposed as a pathophysiological hypothesis, stating that the volume within the skull is constant, consisting of the sum of the volumes of the brain itself, CSF, and intracranial blood. Most of the reported PHBS cases had CSF leakage. During a brain or spine surgical procedure, it has been speculated that a rapid reduction in CSF volume may initiate a series of events, including a downward descent of the brainstem and a compensatory increase in cerebral blood volume with venous congestion.⁴⁾

However, in this case, CSF leakage was excluded based on the amount of CSF drained intraoperatively and postoperatively. Thus, PHBS may occur even without CSF leakage and in trepanation, which usually has no tendency for CSF leakage. Although there have been reports in which CSF leakage was not confirmed, this is the first report of PHBS following burr hole drainage for CSDH without CSF

leakage.

We speculate the following regarding possible mechanisms by which PHBS occurred in this particular case. First, after draining the hematoma, the compressed brain may re-expand; at that time, the cerebral veins and sinuses may be towed, which causes physical damage to or obstruction of the veins, leading to venous congestion. Second, it has been reported that CSDH presents with circulatory and venous perfusion disorders due to chronic pressure on the brain, affecting the brain's autoregulation. Moreover, it is known that local cerebral blood flow increases immediately after drainage of CSDH,⁵⁾ as demonstrated by this case's ASL findings of an increase in perfusion of the right frontal lobe and brain stem. However, if the autoregulatory ability is compromised, as aforementioned, the veins may not be sufficiently dilated for hyperperfusion, resulting in venous congestion. Third, according to the Monro-Kellie doctrine,³⁾ the increase in cerebral parenchyma and cerebral blood flow causes a relative decrease in intracranial CSF, resulting in brain sagging similar to that caused by CSF leakage, leading to venous congestion. Although the volume of removed hematoma was small relative to the intracranial volume, the aforementioned mechanism may have altered the cranial environment.

During the intraoperative and postoperative periods, respiratory and circulatory dynamics were stable, and laboratory parameters and CSF examination revealed no obvious

abnormalities. There are no robust studies linking specific anesthetic agents or techniques to PHBS; thus, PHBS in this case is likely due to intracranial decompression by burr hole drainage. The limitation of this study is that CSF leakage could not be completely excluded, though the leakage of a large volume of CSF was ruled out. Regarding treatments, although anti-edema therapy using steroids might have been effective, the number of reported cases is extremely small, so further cases need to be accumulated.

Although CSF leakage is considered the main cause of PHBS, its pathology is not fully understood. This report highlights the importance of recognizing it as a possible complication of burr hole drainage without obvious CSF leakage.

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Conflicts of Interest Disclosure

All authors declare no conflicts of interest.

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