Successful treatment with hyperbaric oxygen therapy for severe brain edema characterized by radiological appearance of pseudosubarachnoid hemorrhage in a child

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Abstract. Pseudosubarachnoid hemorrhage (PSAH) is a rare neuroradiological finding, particularly in pediatric patients. The appearance of PSAH is commonly associated with poor clinical outcome due to refractory cerebral edema. Recent clinical trials have favored hyperbaric oxygen therapy (HBOT) as a promising therapeutic strategy for adult patients with severe head injuries. The present report describes a pediatric case of diffuse brain edema characterized by the radiological appearance of PSAH successfully treated with HBOT. An adolescent boy collapsed unconscious following convulsion for 3-5 min with fever and headache for 2 days. A brain computed tomography (CT) scan provided an image compatible with subarachnoid hemorrhage (SAH). Lumbar puncture was conducted on admission to hospital and showed no evidence of SAH. The CT scan was again considered and eventually interpreted as PSAH. The patient received drug treatment including acyclovir and mannitol, but the condition deteriorated rapidly. HBOT was administered at 72 h post admission and the condition was clearly improved following the initial therapy. The patient was discharged with 20 sessions of HBOT and recovered completely after 1 year. The appearance of PSAH indicates severe cerebral edema refractory to treatment with conventional internal medicine. HBOT maybe an effective therapeutic strategy for this condition.

Introduction

Subarachnoid hemorrhage (SAH) is one of the most serious acute neurologic events. Computed tomography (CT) of the brain is the first diagnostic imaging method performed in patients suspected of having a SAH. However, in rare circumstance, a similar appearance may occur in CT imaging in the absence of blood in the subarachnoid space, which has been designated as ‘pseudo-subarachnoid hemorrhage, PSAH’ (1). Although the exact mechanisms remain unknown, severe diffuse brain edema might be associated with PSAH, and it is extremely important to increase the recognition of PSAH and to distinguish it from true SAH.

PSAH has been observed in various severe clinical events, including acute poisoning (2), cardiopulmonary arrest without trauma (3,4) and sudden unconscious and generalized convulsions (5). In these cases, it was reported that the prognoses of patients were all poor and the common characteristics of pathophysiology were internal medicine-refractory diffuse cerebral edema due to anoxic encephalopathy. Recent clinical trials have favored hyperbaric oxygen therapy (HBOT) as a promising therapeutic strategy for adult patients with severe head injury (6,7); however, clinical data on the safety and efficacy of HBOT in the pediatric field, with the exception of acute carbon monoxide poisoning (8), remains scarce. The present case report describes a pediatric case of diffuse brain edema characterized by a radiological appearance of PSAH successfully treated with HBOT.

Case report

A 10-year-old previously healthy boy was admitted to the Yuhuangding Hospital (Yantai, China) with a 2-day history of a fever and headache. The patient experienced a 3-5-min long, generalized tonic-clonic convulsion and became unconscious 2 h prior to admission. On admission, he had a temperature of 39°C and was comatose. Brain stem reflexes were preserved and deep tendon reflexes were very brisk, with a Glasgow coma scale (GCS) score of 6 (E1V1M4). Brain CT (Fig. 1A) revealed increased attenuation of the subarachnoid spaces in basal cisterns, as well as signs of diffuse cerebral edema and was interpreted as SAH by a radiologist. Lumbar
puncture was performed following treatment with mannitol (0.6 g/kg body weight infusion over 30 min). Cerebrospinal fluid (CSF) examination revealed the following results: Leukocytes, 180×10^3/μl, with lymphocytic predominance; no erythrocytes; normal protein, chloride and glucose levels; and negative Gram stain. The lumbar puncture showed no evidence of SAH while supporting viral encephalitis. CT results were again considered and eventually interpreted as PASH. The patient was treated with acyclovir (10 mg/kg body weight repeated every 8 h for 10 days) and mannitol (0.6 g/kg body weight repeated every 4-6 h for 72 h).

On day 2 of hospitalization, the mental status of the patient was continuing to worsen, and intubation was required for airway protection. The subsequent brain CT (Fig. 1B) continued to show signs of diffuse cerebral edema and the attenuation values within the cisterns ranged from 22 to 46 Hounsfield units (Hu), with a mean value 33 Hu. HBOT was administered at 72 h post admission. The protocol consisted of 100% oxygen for 60 min at 2.0 absolute atmospheres. Following the initial therapy, the GCS score of the patient improved from 6 to 11 (E4V2M5), and he was successfully extubated on day 4 of hospitalization.

On day 6 of hospitalization, following 3 days of HBOT, the patient was able to eat, occasionally say words, follow commands and display purposeful and spontaneous movements of his arms and legs. Repeat brain CT (Fig. 1C) demonstrated that the basal cisterns had reopened and the abnormal hyperdensity had disappeared. After 20 sessions of HBOT, the patient was discharged with significant improvements in ambulation, balance, speech and cognition. At the 1-year follow-up, the patient had recovered completely and had resumed school without apparent neurological sequelae. The present study was conducted in accordance with the Declaration of Helsinki, and with approval from the Ethics Committee of Yuhuangding Hospital. Written informed consent was obtained from participant's guardians.

Discussion

A PSAH finding is a CT pseudo lesion that exhibits characteristics similar to those of SAH, with a hyperattenuated appearance of the cisterns and cerebral sulci relative to the brain parenchyma (3). Although the exact mechanisms causing the appearance of SAH remain unclear, cerebral edema may be associated with PSAH (9). The dural sinuses are compressed by severe brain edema, which compromises the venous drainage from the brain and results in the cerebral veins becoming engorged; they thus stand out against the edematous low attenuated cerebral parenchyma, mimicking an SAH (10). Senthilkumaran et al proposed the application of the attenuation values (in Hu) to assist emergency physicians in distinguishing PSAH from SAH: In basal cisterns of PSAH the attenuation coefficient was reported to be 21-44 Hu, with a mean of 29-33Hu (11). The present patient had an attenuation coefficient in the basal cisterns of 22-46 Hu (mean, 33Hu), which is consistent with a diagnosis of PSAH on CT imaging.

Previous reports have indicated that patients with PSAH have a poor prognosis, and that he main risk factors for poor outcome were increased intracranial pressure (ICP), higher lactate levels, a longer duration of secondary anoxia due to cerebral edema, and lack of effective treatment using internal medicine (9,10). As early as 1982, Sukoff and Ragatz reported that hyperbaric oxygenation was able to treat acute cerebral edema effectively by reducing intracranial pressure and concomitantly increasing cerebral oxygenation (12). In adults, studies have revealed that HBOT can decrease cerebral edema, maintain blood-brain barrier integrity, improve regional oxygen metabolism, and lower ICP and dialysate lactate levels (6,13). HBOT refers to the inhalation of 100% oxygen inside a hyperbaric chamber pressurized to >1 atmosphere. The mechanisms of HBOT include increasing the partial pressure of oxygen within the blood and the subsequent improvement of mitochondrial metabolism and tissue oxygenation (14).

To the best of our knowledge, the present case report describes the first pediatric case of acute nontraumatic cerebral edema successfully treated with HBOT. However, the optimal regimen for treating acute brain edema, and the efficacy and safety of HBOT in the pediatric field, particularly in neonates and infants remains elusive.
References