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Case Report

Managing the "big black brain" in low resource setting: A case report of early outcome after hinge craniotomy

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ABSTRACT

Background: The big black brain (BBB) phenomenon is described as an infant's response to an acute subdural hematoma (SDH). It is characterized by hypodensity and swelling of the supratentorial compartment as a whole. Numerous factors may contribute to the formation of the BBB. Due to its high morbidity and mortality, the management of BBB is still debatable. In this report, we describe a 2-month-old boy who had bilateral hemispheric hypodensity and underwent hinge craniotomy.

Case Description: The patient was referred to our hospital with decreased consciousness. The patient had a history of seizures and cardiopulmonary arrest. There is no history of trauma. The computed tomography revealed a subacute SDH on the left parietal and occipital lobe along with hypodensity in both hemispheres with preservation of posterior fossa, consistent with hemispheric hypodensity. We performed a hinge craniotomy for the emergency procedure and evacuated only the hemisphere with the bleeding side. The patient cried spontaneously 24 hours after the procedure and was discharged six days later.

Conclusion: Early outcomes of hinge craniotomy as an alternative procedure for treating the BBB were positive. However, long-term outcomes, particularly the infant's development, should be monitored.

Keywords: Big black brain, Hinge craniotomy, Infants, Outcome

INTRODUCTION

The big black brain (BBB) phenomenon is described as a response to acute subdural hematoma (SDH) in infants.[6] This term was first reported in 1993 and was associated with hypodensity and swelling of the entire supratentorial compartment on a computed tomography (CT) scan.[5,15] It is believed that BBB represents a decompensation of hemispheric cerebral demand as a result of SDH.[1] Numerous factors may contribute to the formation of the BBB. Due to its high morbidity and mortality, the management of BBB is still contested.[6,7]

Hinge craniotomy is a technique that permits a degree of decompression while retaining the bone flap *in situ*, in a "floating" or "hinged" fashion, and maintains cerebral protection while reducing postoperative complications and eliminating the need for subsequent cranioplasty procedures.[12,21] Since decompressive craniectomy carries a high risk of complications, particularly

in infants, hinge craniotomy can be a useful alternative to reduce intracranial pressure (ICP).[23] Numerous studies have also reported infants undergoing hinge craniotomy.[17,23] However, not all studies reported the status of functional outcomes at the conclusion of follow-up.[12] Here, we describe a 2-month-old boy who underwent hinge craniotomy to treat the big black brain and had a positive early outcome.

CASE DESCRIPTION

A 2-month-old boy was referred to our hospital with decreasing consciousness. His mother had noticed that the patient had "ordinary cold symptoms" 5 days prior that were escalating gradually. The child had a mild fever and was not doing well at first, but her mother noted that the patient had become paler than before and had a seizure on the 3rd day. The seizure lasted around 2 min on the left side of the body and ended without any intervention. The patient was subsequently taken to a primary healthcare service nearby. Anemia (hemoglobin) 6.1 g/dL, hematocrit 15.8%, leukocyte 14.000/L, and thrombocyte 450.000/L were discovered during the initial laboratory evaluation. There was no history of trauma, and according to the mother, the child was in a healthy condition before. The patient was referred to our hospital after receiving a loading dose of phenytoin.

The paramedic reported that the patient had stopped breathing en route to our hospital's emergency department. The patient was unconscious and had apnea, with a heart rate of 80 beats/min, according to the initial assessment. Intubation and stabilization were started as an emergency treatment. Following intubation, the vital signs were as follows: blood pressure 124/72 mmHg, heart rate 78 beats/min, and oxygen saturation on a ventilator of 97%. There was no sign of trauma to the head or any other area of the body. The main fontanelle, on the other hand, was bulging and tight, with a sluggish-symmetric pupil (2 mm). Following painful stimuli, the child was decorticated with minimum flexion. A non-contrast head CT scan revealed a subacute SDH on the left parietal and occipital lobe along with hypodensity in both hemispheres with preservation of posterior fossa, consistent with hemispheric hypodensity (the "big black brain") [Figure 1]. Additional laboratory testing revealed a somewhat extended hemorrhagic screening test, with a prothrombin time of 15.4 s (control 12.5 s), an activated partial thrombin time of 34.2 s (control 27 s), an international normalized ratio of 1, and a thrombin time of 26.2 s. (control 22.1 s). There was also metabolic acidosis (pH 7.348, pCO_2 24.9, pO_2 178, HCO_3 13.8, base excess -12.1, and oxygen saturation 99.7%).

To treat the anemia and prolonged hemorrhagic screening test, packed red cells and fresh frozen plasma transfusions were started within the first 48 h. We performed a hinge craniotomy and subdural blood clot removal after stabilization [Figure 2]. A chronic SDH, along with severe brain edema, was found during the operation. We left the bone "floating" without fixation to facilitate brain expansion.

Following the operation, the patient's condition significantly improved. The patient had spontaneous breathing for the first 24 hours after the operation and was then extubated. The patient cried spontaneously the next day, and the nasogastric tube was withdrawn on the 5th postoperative day. The patient was released from the hospital on the 6th day. The patient was active two months after discharge and had no seizures. However, two months following the operation, a control CT scan indicated substantial brain atrophy in both hemispheres [Figure 3].

DISCUSSION

BBB was first described by Duhaime *et al.* as a hemispheric hypodense associated with subdural hemorrhage.[5] According to a study, a blood flow disturbance at the level of the peripheral arteries to microcirculation may account for a potential BBB mechanism.[16] This phenomenon is unique to infant head trauma and was rarely observed in adults.[19] Brain oxygen supply-demand mismatch and systemic insults that impair oxygen delivery may contribute to the formation

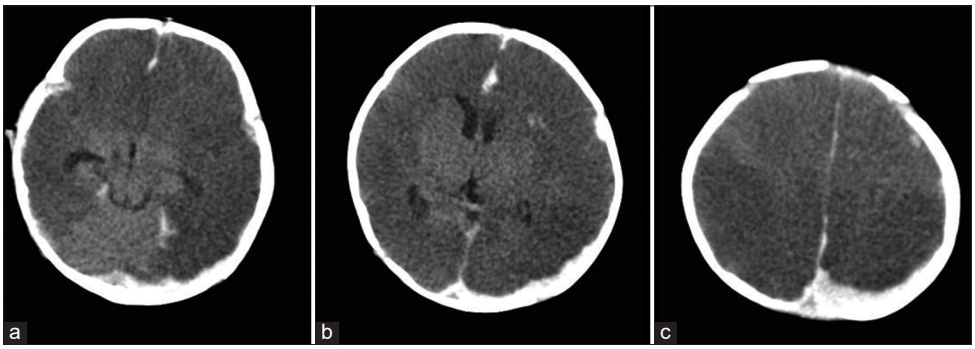

Figure 1: Non-contrast computed tomography scan. (a) Bilateral hemispheric hypodensity with preservation of posterior fossa was seen, consistent with a "big black brain" (b) Preservation of central nuclei was also seen, along with a left hemispheric crescentic lesion, suggestive of a subdural hematoma. (c) The hypodense lesion was also seen up to the vertex region.

of BBB.[7] Recent animal studies also revealed that apnea, hypoventilation, and seizures are part of the pathophysiological cascade of the BBB.[2] It is believed that SDH is also associated with shaken baby syndrome.[14] Study shows that the bridging veins are more susceptible to tearing during shaking in infants, who had bigger cerebrospinal fluid space than in adults.[3] In this study, we did not find any signs or histories of trauma, so it might be the baby-shaken syndrome.

Our patient presented with a history of cardiopulmonary arrest (CPA) and seizures. A study outlined four pathological conditions in which CPA arrest can occur and be manifested in infants.[18] Diffuse brain injury and injury to the respiratory center are unquestionably attributable to CPA in abusive head trauma in infants.[18,19] Another study added three patterns of extensive low-density area, which is typical of BBB, and their pathophysiology based on when it occurs, where it occurs, and the presence of midline shift.[19] It has been demonstrated that early surgical evacuation of blood near the cerebral cortex is not protective against the development of BBB.[25] Momose *et al.* identified five predictors for cerebral infarction following acute SDH: Seizure, consciousness disturbance at admission, absence of skull fracture, hematoma thickness, and midline shift, more than 5 mm more than 3 mm on CT scan, respectively.[16] At present, CT scans are used to detect hemorrhages, particularly in settings with limited resources.[13] Nonetheless, a study revealed the importance of magnetic resonance imaging follow-up in infants with non-accidental trauma (NAT) for improved brain injury characterization, prognosis, and long-term management, particularly in the "big black brain" phenomenon.[1] In settings with limited resources, a CT scan is, therefore, still the initial method of imaging the BBB.

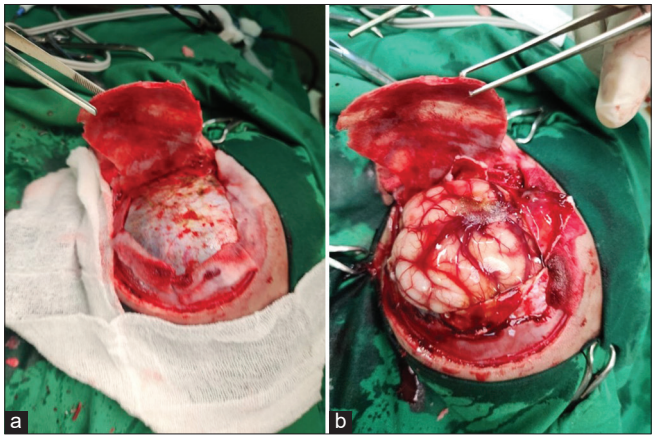

Figure 2: Hinge craniotomy. (a) After a "three-sided craniotomy," periosteal on the temporal base was preserved. A craniectomy on the temporal base was performed to improve the decompression size. A blue dura mater was seen. (b) After subdural blood clot removal, the brain was bulging with engorgement of the cortical vein. The bone was then left at the initial site without any fixation to maintain a "hinged" position.

Foster *et al.* reported intracranial hypertension and ICP monitoring intensity.[7] Due to limited resources, we did not include angiography and ICP measurements in this study. Despite this, we were confident that the angiography was consistent with the BBB due to the absence of occlusion. Moreover, ICP monitoring was not a common practice, particularly in our setting. In addition, we considered this patient's limited access to ventilators and lack of intensive care unit (ICU) availability. Therefore, we must overcome our limitations to treat the patient. We did not perform a craniotomy due to the numerous reported complications.[11] We also believed that the subsequent cranioplasty would be exceedingly difficult to perform, as it had a high incidence of complications.[20] We propose emergency evacuation of the bleeding side as an alternate procedure to reduce patient costs and ICU requirements in low-resource settings. In this study, patients cried spontaneously and showed no signs of CPA 24 h after undergoing surgery. The patient was discharged 6 days after surgery.

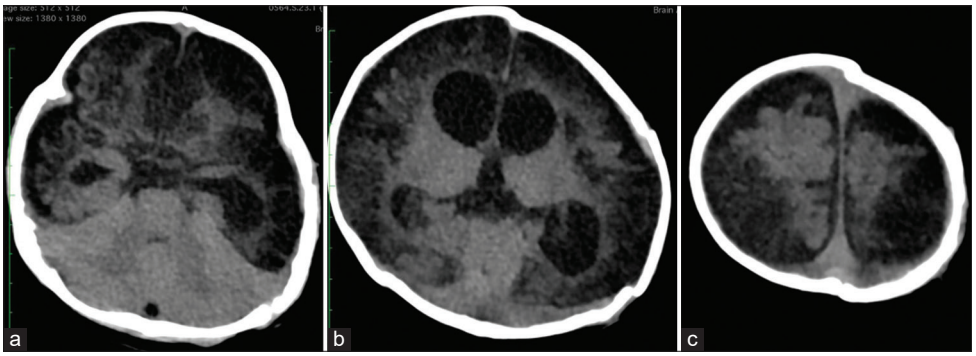

Figure 3: Post-operative non-contrast head computed tomography scan two months following surgery. (a) There was an extensive hypodense lesion on the cortical region, as well as dilatation of the temporal horn (b) The extensive hypodense lesion on the cortical was also seen along with dilatation of the lateral ventricle. "Hinged" bone could be found on the left side. (c) The cortical hypodense lesion was also identified up to the vertex region, but relatively less than the preoperative finding.

A retrospective study found that the hinge technique effectively prevents procedure-related morbidity as well as the need for a second incision to explant the bone flap and surgery to restore cranial contour.[10,21] Layard Horsfall *et al*. also suggested that hinge craniotomy has potential clinical utility, particularly in low-resource settings and when neurosurgeons are distributed unequally, as was the case in Indonesia.[4,12,22] According to a review study, hinge craniotomy is effective for controlling ICP. It is indicated by a decrease in ICP and a corresponding decrease in midline shift.[12] In addition, Kenning *et al*. suggested that adequate cerebral decompression was achieved with hinge craniotomy.[8,9] In this study, the hinge craniotomy was effective in controlling the ICP, and the clinical presentation gradually improved; however, three months after the procedure, the left hemisphere showed atrophy. In accordance with our findings, a study reported a year after emergency craniotomy, a diffuse atrophy of one cerebral hemisphere, but the patient aged normally without paresis or developmental delay.[24] In addition, long-term follow-up should be required, particularly in cases of motoric or developmental delay.

CONCLUSION

The management of the big black brain is still debatable, especially in infants. Early outcomes of hinge craniotomy were reported to be favorable in this case report; however, long-term outcomes, particularly the development of the infant, should be monitored.

Ethical approval

Not applicable

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Conflicts of interest

There are no conflicts of interest.

Use of artificial intelligence (AI)-assisted technology for manuscript preparation

The authors confirm that there was no use of artificial intelligence (AI)-assisted technology for assisting in the writing or editing of the manuscript and no images were manipulated using AI.

REFERENCES

1. Beriwal N, Misko AL, Duhaime AC. Big black brain phenomenon: Understanding clinicoradiological dissociation in non-accidental traumatic brain injury in children. Cureus 2020;12:e8011.
2. Costine-Bartell BA, McGuone D, Price G, Crawford E, Keeley KL, Munoz-Pareja J, *et al.* Development of a model of hemispheric hypodensity ("Big Black Brain"). J Neurotrauma 2019;36:815-33.
3. Cox LA. The shaken baby syndrome: Diagnosis using CT and MRI. Radiol Technol 1996;67:513-20.
4. Dewan MC, Rattani A, Fieggen G, Arraez MA, Servadei F, Boop FA, *et al.* Global neurosurgery: The current capacity and deficit in the provision of essential neurosurgical care. Executive Summary of the Global Neurosurgery Initiative at the Program in Global Surgery and Social Change. J Neurosurg 2018;130:1055-64.
5. Duhaime AC, Bilaniuk L, Zimmerman R. The "big black brain": Radiographic changes after severe inflicted head injury in infancy. J Neurotrauma 1993;10:S59.
6. Duhaime AC, Durham S. Traumatic brain injury in infants: The phenomenon of subdural hemorrhage with hemispheric hypodensity ("Big Black Brain"). Prog Brain Res 2007;161:293-302.
7. Foster KA, Recker MJ, Lee PS, Bell MJ, Tyler-Kabara EC. Factors associated with hemispheric hypodensity after subdural hematoma following abusive head trauma in children. J Neurotrauma 2014;31:1625-31.
8. Kenning TJ, Gandhi RH, German JW. A comparison of hinge craniotomy and decompressive craniectomy for the treatment of malignant intracranial hypertension: Early clinical and radiographic analysis. Neurosurg Focus 2009;26:E6.
9. Kenning TJ, Gooch MR, Gandhi RH, Shaikh MP, Boulos AS, German JW. Cranial decompression for the treatment of malignant intracranial hypertension after ischemic cerebral infarction: Decompressive craniectomy and hinge craniotomy. J Neurosurg 2012;116:1289-98.
10. Ko K, Segan S. *In situ* hinge craniectomy. Neurosurgery 2007;60:255-8.
11. Lassen B, Helseth E, Egge A, Due-Tønnessen BJ, Rønning P, Meling TR. Surgical mortality and selected complications in 273 consecutive craniotomies for intracranial tumors in pediatric patients. Neurosurgery 2012;70:936-43.
12. Layard Horsfall H, Mohan M, Devi BI, Adeleye AO, Shukla DP, Bhat D, *et al.* Hinge/floating craniotomy as an alternative technique for cerebral decompression: A scoping review. Neurosurg Rev 2020;43:1493-507.
13. Le TH, Gean AD. Neuroimaging of traumatic brain injury. Mt Sinai J Med 2009;76:145-62.
14. Lee Y, Lee KS, Hwang DH, Lee IJ, Kim HB, Lee JY. MR imaging of shaken baby syndrome manifested as chronic subdural hematoma. Korean J Radiol 2001;2:171-4.
15. Luyet FM, Feldman KW, Knox BL. The big black brain: Subdural hemorrhage with hemispheric swelling and low attenuation. J Child Adolesc Trauma 2018;11:241-7.
16. Momose H, Sorimachi T, Aoki R, Atsumi H, Matsumae M. Cerebral infarction following acute subdural hematoma in infants and young children: Predictors and significance

of FLAIR vessel hyperintensity. Neurol Med Chir (Tokyo) 2015;55:510-8.
17. Okuma Y, Yasuhara T, Kin I, Daido S, Date I. Inverted gull-wing hinge decompressive craniotomy for infantile acute subdural hematoma: A case report. Brain Circ 2023;9:35-8.
18. Paek D, Kwon DI. A review on four different paths to respiratory arrest from brain injury in children; implications for child abuse. J Forensic Leg Med 2020;71:101938.
19. Park YS. Complex pathophysiology of abusive head trauma with poor neurological outcome in infants. J Korean Neurosurg Soc 2022;65:385-96.
20. Rocque BG, Agee BS, Thompson EM, Piedra M, Baird LC, Selden NR, *et al.* Complications following pediatric cranioplasty after decompressive craniectomy: A multicenter retrospective study. J Neurosurg Pediatr 2018;22:225-32.
21. Schmidt JH 3rd, Reyes BJ, Fischer R, Flaherty SK. Use of hinge craniotomy for cerebral decompression. Technical note. J Neurosurg 2007;107:678-82.
22. Wicaksono AS, Tamba DA, Sudiharto P, Basuki E, Pramusinto H, Hartanto RA, *et al.* Neurosurgery residency program in Yogyakarta, Indonesia: Improving neurosurgical care distribution to reduce inequality. Neurosurg Focus 2020;48:E5.
23. Yokota H, Sugimoto T, Nishiguchi M, Hashimoto H. Greenstick fracture-hinge decompressive craniotomy in infants: Illustrative case and literature review of techniques for decompressive craniotomy without bone removal. Childs Nerv Syst 2019;35:1491-7.
24. Yoshimura J, Morikawa M, Kumagai S, Saito T, Takahashi H, Fujiwara H, *et al.* Surgical case of accidental infantile acute subdural hematoma caused by household minor head trauma: Hyperperfusion during postoperative hemispheric hypodensity, namely "big black brain". No Shinkei Geka 2020;48:841-7.
25. Zhao J, Hylin MJ, Kobori N, Hood KN, Moore AN, Dash PK. Post-injury administration of galantamine reduces traumatic brain injury pathology and improves outcome. J Neurotrauma 2018;35:362-74.

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