

Case Report

Unexpected Severe Cerebral Edema after Cranioplasty : Case Report and Literature Review

Gwang Soo Lee, M.D., Sukh Que Park, M.D., Ph.D., Rasun Kim, M.D., Sung Jin Cho, M.D., Ph.D.

Department of Neurosurgery, Soonchunhyang University Seoul Hospital, Soonchunhyang University College of Medicine, Seoul, Korea

This report details a case of unexpected, severe post-operative cerebral edema following cranioplasty. We discuss the possible pathological mechanisms of this complication. A 50-year-old female was admitted to our department with sudden onset of stuporous consciousness. A brain computed tomography (CT) revealed a subarachnoid hemorrhage with intracranial hemorrhage and subdural hematoma. Emergency decompressive craniectomy and aneurysmal neck clipping were performed. Following recovery, the decision was made to proceed with an autologous cranioplasty. The cranioplasty procedure was free of complications. An epidural drain was placed and connected to a suction system during skin closure to avoid epidural blood accumulation. However, following the procedure, the patient had a seizure in the recovery room. An emergency brain CT scan revealed widespread cerebral edema, and the catheter drain was clamped. The increased intracranial pressure and cerebral edema were controlled with osmotic diuretics, corticosteroids, and antiepileptic drugs. The edema slowly subsided, but new low-density areas were noted in the brain on follow-up CT 1 week later. We speculated that placing the epidural drain on active suction may have caused an acute decrease in intracranial pressure and subsequent rapid expansion of the brain, which impaired autoregulation and led to reperfusion injury.

Key Words : Cranioplasty · Cerebral edema · Complication · Decompressive craniectomy · Subgaleal drain.

INTRODUCTION

Cranioplasty complications are well documented and include infection, bone resorption, intracerebral hemorrhage, and infarction^{2,3,4,6,12}. This report details a case of unexpected, severe postoperative cerebral edema following autologous cranioplasty. There are few reported cases of cerebral edema after cranioplasty^{5,11,14}. We discuss the potential pathological mechanisms of this complication.

CASE REPORT

A 50-year-old female was admitted to our department with sudden-onset stuporous consciousness. A brain computed tomography (CT) scan revealed a subarachnoid hemorrhage with a right temporal lobe intracranial hemorrhage and a right subdural hematoma. Following the CT, angiography demonstrated an aneurysmal sac at the right posterior communicating artery (Fig. 1). Emergency decompressive craniectomy (DC) and an-

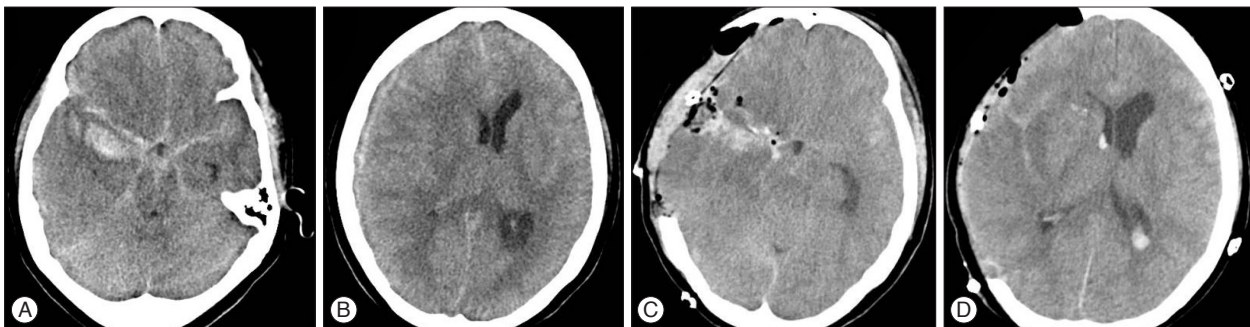


Fig. 1. Brain computed tomography scan on admission showing a subarachnoid hemorrhage, intracerebral hemorrhage, and subdural hemorrhage (A and B). Emergency decompressive craniectomy was performed (C and D).

• Received : June 20, 2014 • Revised : September 22, 2014 • Accepted : September 26, 2014

• Address for reprints : Sukh Que Park, M.D., Ph.D.

Department of Neurosurgery, Soonchunhyang University Seoul Hospital, Soonchunhyang University College of Medicine, 59 Daesagwan-gil, Yongsan-gu, Seoul 140-743, Korea
Tel : +82-2-709-9269, Fax : +82-2-792-5976, E-mail : drcolor@schmc.ac.kr

• This is an Open Access article distributed under the terms of the Creative Commons Attribution Non-Commercial License (<http://creativecommons.org/licenses/by-nc/3.0>) which permits unrestricted non-commercial use, distribution, and reproduction in any medium, provided the original work is properly cited.

eurysmal neck clipping were performed. Following recovery there was some sinking of the scalp flap, and the decision was made to proceed with an autologous cranioplasty 61 days after the initial hemorrhage. There were no complications during the cranioplasty procedure, and there was no evidence of cardiovascular instability or cerebral swelling. Epidural and subdural drains were placed and connected to a suction system during skin closure. Following skin closure the drains were connected to a drainage bag to avoid subgaleal and epidural blood accumulation. However, the patient had a seizure in the recovery room. Emergency brain CT revealed widespread cerebral edema (Fig. 2). The patient extended her extremities in response to painful stimuli with no evidence of awareness. Therefore, the decision was made to clamp the drain catheters. The increased intracranial pressure (ICP) and cerebral edema were controlled with osmotic diuretics, corticosteroids, and antiepileptic drugs. The edema slowly subsided, but new low-density areas were noted on follow-up brain CT 1 week after the cranioplasty (Fig. 2).

DISCUSSION

Patients who survive DC need to undergo a second proce-

dures, a cranioplasty. Complications following cranioplasty, including epidural or subdural hematoma, wound healing disturbance, subdural hematoma, cerebrospinal fluid fistula, infection, and bone graft resorption, are a significant cause of post-operative morbidity^{2,3,4,6,12}. There are only five reported cases of unexpected cerebral edema after cranioplasty^{5,11,14}. Honeybul⁵ reported three cases of sudden death following massive post-operative cerebral swelling after autologous cranioplasty. All three patients had a bifrontal DC for intractably raised ICP following severe neurotrauma, and developed hydrocephalus necessitating shunt insertion. The author speculated that some degree of autoregulatory impairment and change in brain compliance due to the development of hydrocephalus and subsequent insertion of a ventriculoperitoneal (V-P) shunt may have contributed to the outcome. Santana-Cabrera et al.¹¹ reported a similar case of massive post-operative cerebral edema following cranioplasty. Zebian and Critchley¹⁴ reported the case of a patient with an aneurysmal subarachnoid hemorrhage who underwent unilateral DC. During the cranioplasty there was a negative pressure shift causing the brain to expand rapidly to fill the defect within the skull; this shift may have been further exacerbated by the drain being placed on full suction. Of these reported

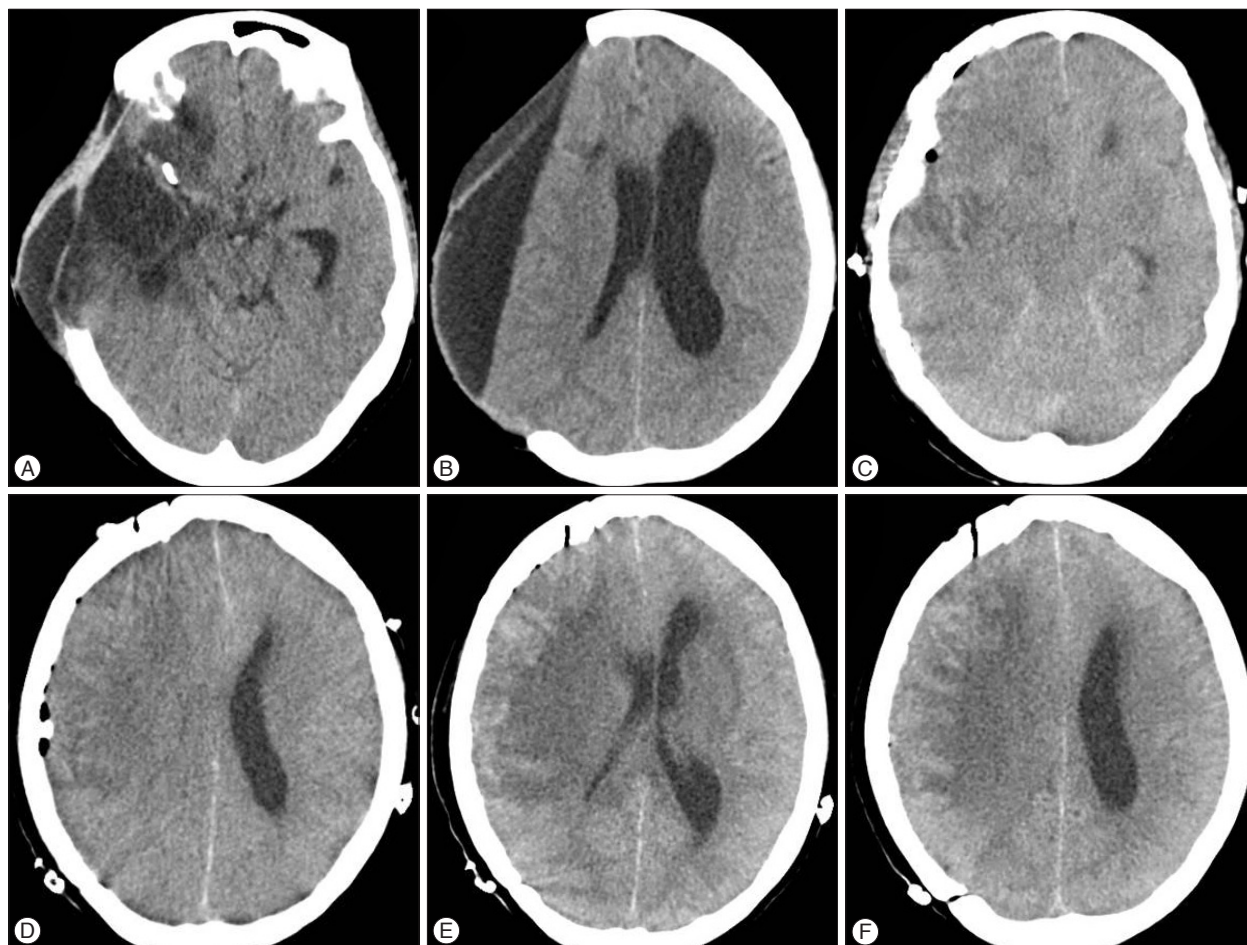


Fig. 2. Brain computed tomography scan prior to cranioplasty (A and B), scan after cranioplasty with massive swelling and a midline shift (C and D), scan 1 week post-operatively with slightly improved cerebral edema (E and F).

cases, four were traumatic brain injury patients and one had cerebral edema following aneurysmal neck clipping. Bifrontal craniectomy was performed in four cases and left-sided unilateral craniectomy in one, while our patient underwent a right-side unilateral craniectomy. In addition, four of the previous cases underwent a V-P shunt operation prior to cranioplasty due to hydrocephalus, but our patient did not have hydrocephalus.

In the presented case, several factors contributed to the development of severe swelling following cranioplasty. The exact pathophysiological mechanism behind severe cerebral edema and sudden death following cranioplasty is poorly understood. Potential contributing factors include some degree of autoregulatory impairment, change in brain compliance due to the development of hydrocephalus, and the acute lowering of ICP. The underlying cause must be related to a failure in the autoregulatory process. The brain, which is subjected to atmospheric pressure prior to cranioplasty, appears to shift toward negative pressure, causing a rapid expansion of the brain to fill the defect within the skull.

Another point to consider is that the acute lowering of ICP may lead to intracranial hemorrhage in various scenarios. Roth et al.¹⁰⁾ reported that continuous suctioning of the subgaleal and epidural space may induce a negative ICP that is sufficient to cause epidural hematomas and hemodynamic collapse. We speculated that placing the epidural and subgaleal drains on active suction in our patient may have caused an acute decrease in ICP leading to subsequent rapid expansion of the brain, which impaired autoregulation and led to reperfusion injury.

A few studies have reported neurological improvement following cranioplasty in craniectomy patients, and they indicated that the mechanism of recovery involves a significant increase in cerebral blood flow and cerebral energy metabolism^{7,8,13)}. Sometimes the brain is unable to endure sudden hyperperfusion after cranioplasty due to complications, such as diminished autoregulation, cerebral blood flow, and cerebral metabolism^{1,9)}. Therefore, physicians should remember that reperfusion injuries can occur following cranioplasty in patients with large skull defects.

CONCLUSION

We speculate that placing the epidural and subgaleal drains on active suction during cranioplasty may have caused an acute

decrease in ICP and subsequent rapid expansion of the brain, which impaired autoregulation and led to reperfusion injury.

• Acknowledgements

This report had been presented at the 53rd Annual Meeting of the Korean Neurosurgical Society.

This work was supported by the Soonchunhyang University research fund.

References

- Cecchi PC, Rizzo P, Campello M, Schwarz A : Haemorrhagic infarction after autologous cranioplasty in a patient with sinking flap syndrome. *Acta Neurochir (Wien)* 150 : 409-410; discussion 411, 2008
- Chang V, Hartzfeld P, Langlois M, Mahmood A, Seyfried D : Outcomes of cranial repair after craniectomy. *J Neurosurg* 112 : 1120-1124, 2010
- De Bonis P, Frassanito P, Mangiola A, Nucci CG, Anile C, Pompucci A : Cranial repair : how complicated is filling a "hole"? *J Neurotrauma* 29 : 1071-1076, 2012
- Gooch MR, Gin GE, Kenning TJ, German JW : Complications of cranioplasty following decompressive craniectomy : analysis of 62 cases. *Neurosurg Focus* 26 : E9, 2009
- Honeybul S : Sudden death following cranioplasty : a complication of decompressive craniectomy for head injury. *Br J Neurosurg* 25 : 343-345, 2011
- Honeybul S, Ho KM : Long-term complications of decompressive craniectomy for head injury. *J Neurotrauma* 28 : 929-935, 2011
- Honeybul S, Janzen C, Kruger K, Ho KM : The impact of cranioplasty on neurological function. *Br J Neurosurg* 27 : 636-641, 2013
- Isago T, Nozaki M, Kikuchi Y, Honda T, Nakazawa H : Sinking skin flap syndrome : a case of improved cerebral blood flow after cranioplasty. *Ann Plast Surg* 53 : 288-292, 2004
- Kwon SM, Cheong JH, Kim JM, Kim CH : Reperfusion injury after autologous cranioplasty in a patient with sinking skin flap syndrome. *J Korean Neurosurg Soc* 51 : 117-119, 2012
- Roth J, Galeano E, Milla S, Hartmannsgruber MW, Weiner HL : Multiple epidural hematomas and hemodynamic collapse caused by a subgaleal drain and suction-induced intracranial hypotension : case report. *Neurosurgery* 68 : E271-E275; discussion E276, 2011
- Santana-Cabrera L, Pérez-Ortiz C, Rodríguez-Escot C, Sánchez-Palacios M : Massive postoperative cerebral swelling following cranioplasty. *Int J Crit Illn Inj Sci* 2 : 107-108, 2012
- Wachter D, Reineke K, Behm T, Rohde V : Cranioplasty after decompressive hemicraniectomy : underestimated surgery-associated complications? *Clin Neurol Neurosurg* 115 : 1293-1297, 2013
- Yoshida K, Furuse M, Izawa A, Iizima N, Kuchiwaki H, Inao S : Dynamics of cerebral blood flow and metabolism in patients with cranioplasty as evaluated by 133Xe CT and 31P magnetic resonance spectroscopy. *J Neurol Neurosurg Psychiatry* 61 : 166-171, 1996
- Zebian B, Critchley G : Sudden death following cranioplasty : a complication of decompressive craniectomy for head injury. *Br J Neurosurg* 25 : 785-786, 2011