Case Report

Unexpected complications immediately after cranioplasty

Akihito Kato,[1] Hiroyuki Morishima,[2] and Goro Nagashima[1,2]

[1]Center of Emergency and Disaster Medicine, St. Marianna University School of Medicine, Kawasaki Municipal Tama Hospital; and [2]Department of Neurosurgery, St. Marianna University School of Medicine, Kawasaki Municipal Tama Hospital, Kawasaki, Japan

Case: An 84-year-old man with subarachnoid hemorrhage underwent craniotomy and clipping with external decompression. Perfusion magnetic resonance imaging showed subclinical sinking skin flap syndrome, and he underwent cranioplasty on postoperative day 58. No problems occurred during the operation, but cerebral edema and hemorrhage were recognized on immediate postoperative computed tomography. Edema continued to progress, but edema and bleeding eventually improved without additional surgery.

Outcome: Neurological symptoms improved to presurgical baseline and stabilized.

Conclusion: Causes of cerebral edema and hemorrhage immediately after cranioplasty include reperfusion, reduction of automatic adjustment function, sinking skin flap syndrome, negative pressure due to s.c. drain, venous stasis, vascular damage following restoration of midline shift, and allergic reaction. Although cranioplasty itself is a relatively minor surgery, the fatality rate for this complication is high. Most complications are due to infection, convulsions, or epidural hematoma, but brain edema and hemorrhage also occur. It is necessary to elucidate the etiology before corrective surgery.

Key words: cerebral edema, cerebral hemorrhage, reperfusion, sinking skin flap syndrome, venous stasis

INTRODUCTION

CRANIOPLASTY IS UNDERTAKEN after decompressive craniectomy to improve the brain herniation or increased intracranial pressure caused by brain swelling as the result of severe traumatic brain injury or severe acute stroke. When the intracranial pressure is normalized, cranioplasty is carried out using autologous that was frozen or stored in the body, or artificial bone such as titanium or hydroxyapatite for the purpose of cerebral protection and cosmetic treatment, sinking skin flap syndrome treatment. Infection, convulsions, and epidural hematoma are common complications immediately following cranioplasty. Cerebral edema and hemorrhage have also been reported. The etiology of post-cranioplasty edema and hemorrhage remains unknown, and the mortality of this complication is high. We experienced a case of cerebral edema and hemorrhage immediately following cranioplasty. Here we consider the etiology of post-cranioplasty edema and hemorrhage.

Corresponding: Akihito Kato, MD, PhD, Center of Emergency and Disaster Medicine, St. Marianna University School of Medicine, Kawasaki Municipal Tama Hospital, 1-30-37, Shukugawara, Tama-ku, Kawasaki, Kanagawa 214-8525, Japan. E-mail: akihito@marianna-u.ac.jp.

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CASE

AN 84-YEAR-OLD MAN experienced left internal carotid artery/posterior communicating artery aneurysm rupture and subarachnoid hemorrhage (WFNS, grade V; Fisher, group 3). He underwent craniotomy and clipping on the day of admission, followed by left external decompression (Fig. 1A–D). He additionally underwent percutaneous revascularization on day 8 for cerebral vasospasm and tracheotomy on day 17 for inability to wean from the ventilator. Glasgow Coma Scale improved to E4VTM5. However, the cranial skin surface recessed, causing midline shift (Fig. 1E). The patient successfully underwent cranioplasty

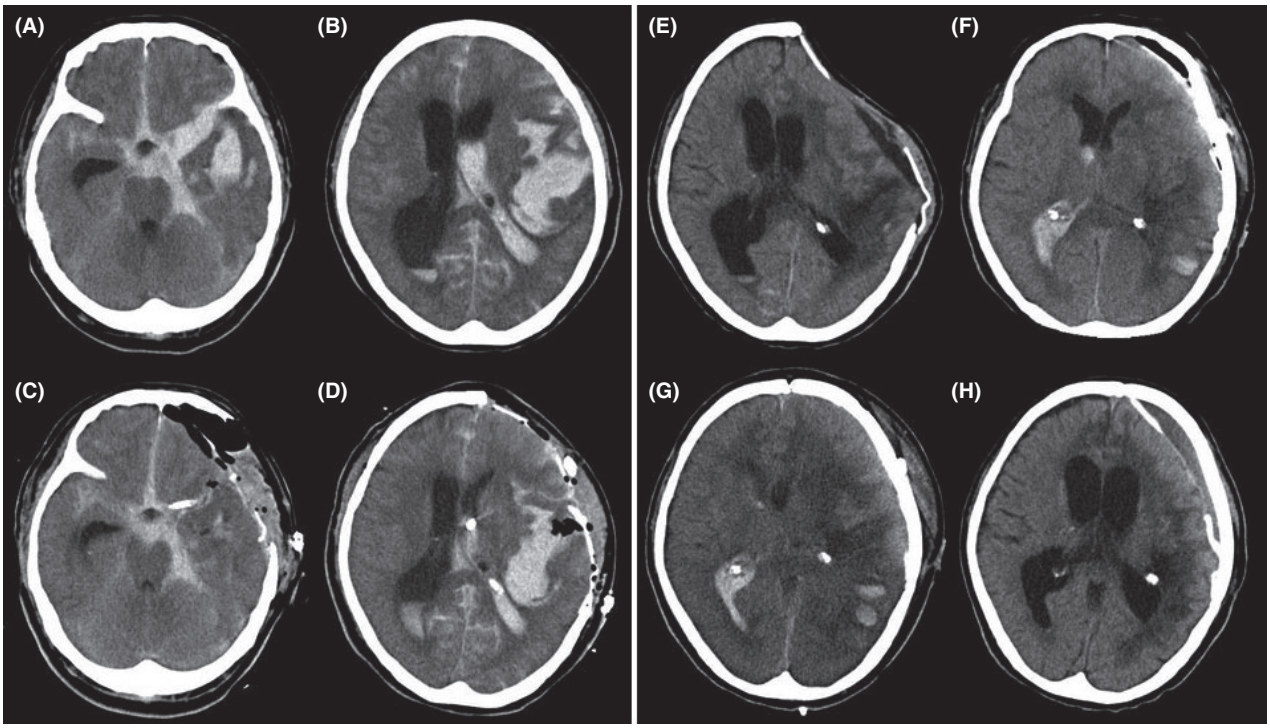

Fig. 1. Computed tomography of an 84-year-old man with subarachnoid hemorrhage. (A, B) Head CT showing subarachnoid hemorrhage with cerebral hematoma. (C, D) Head CT after surgical clipping and decompressive craniectomy. (E) Head CT showing slight midline shift and moderate anterior depression before cranioplasty. (F) Head CT showing intracerebral hemorrhage, intraventricular hemorrhage, and brain edema, with slight midline shift immediately after craniotomy. (G) Head CT showing progression of brain edema; midline shift was worse 6 h after cranioplasty. (H) Head CT showing improvement in brain edema and midline shift on day 34 following cranioplasty.

with artificial hydroxyapatite bone on postoperative day 58. An s.c. drain was placed on the exterior surface to facilitate negative pressure. Immediate postoperative computed tomography (CT) showed right intraventricular hemorrhage, left trigone bleeding, and left-sided cerebral edema (Fig. 1F). Magnetic resonance (MR) angiography showed no recurrence of clipped aneurysm nor any new blood vessel abnormalities, including cerebral vasospasm, on postoperative day 65 (7 days after cranioplasty). These findings progressed on 6-h postoperative CT, causing right midline shift (Fig. 1G). Given our suspicion that the bleeding was due to overperfusion, the patient underwent propofol sedation and strict blood pressure control. We administered glycerol and mannitol and clamped the s.c. drain to decrease negative pressure. The bleeding ceased, the edema improved, and his neurological symptoms returned to presurgical baseline (E4VTM5) (Fig. 1H). Ventricular expansion was observed and a ventriculoperitoneal shunt was placed on postoperative day 128. The patient was transferred to rehabilitation on day 176.

Perfusion magnetic resonance imaging (MR System Achieva 1.5T release 3.2; Philips Medical Systems, The Netherlands) was obtained on postoperative days 44 (14 days before cranioplasty), 65 (7 days after cranioplasty), and 159 (101 days after cranioplasty). Following cranioplasty, the ipsilateral anterior and middle cerebral artery cortical territories showed increased cerebral blood flow and volume compared with the healthy contralateral territories, but ultimately stabilized (Fig. 2).

DISCUSSION

DECOMPRESSION CRANIECTOMY IS carried out to reduce intracranial pressure following severe head trauma or stroke. As intracranial pressure stabilizes, autologous bone is frozen or stored within the body; subsequent cranioplasty is carried out for protection and cosmesis using autologous or artificial titanium or hydroxyapatite bone. Infection, convulsions, and epidural hematoma are frequent complications of cranioplasty. However, there are only 18

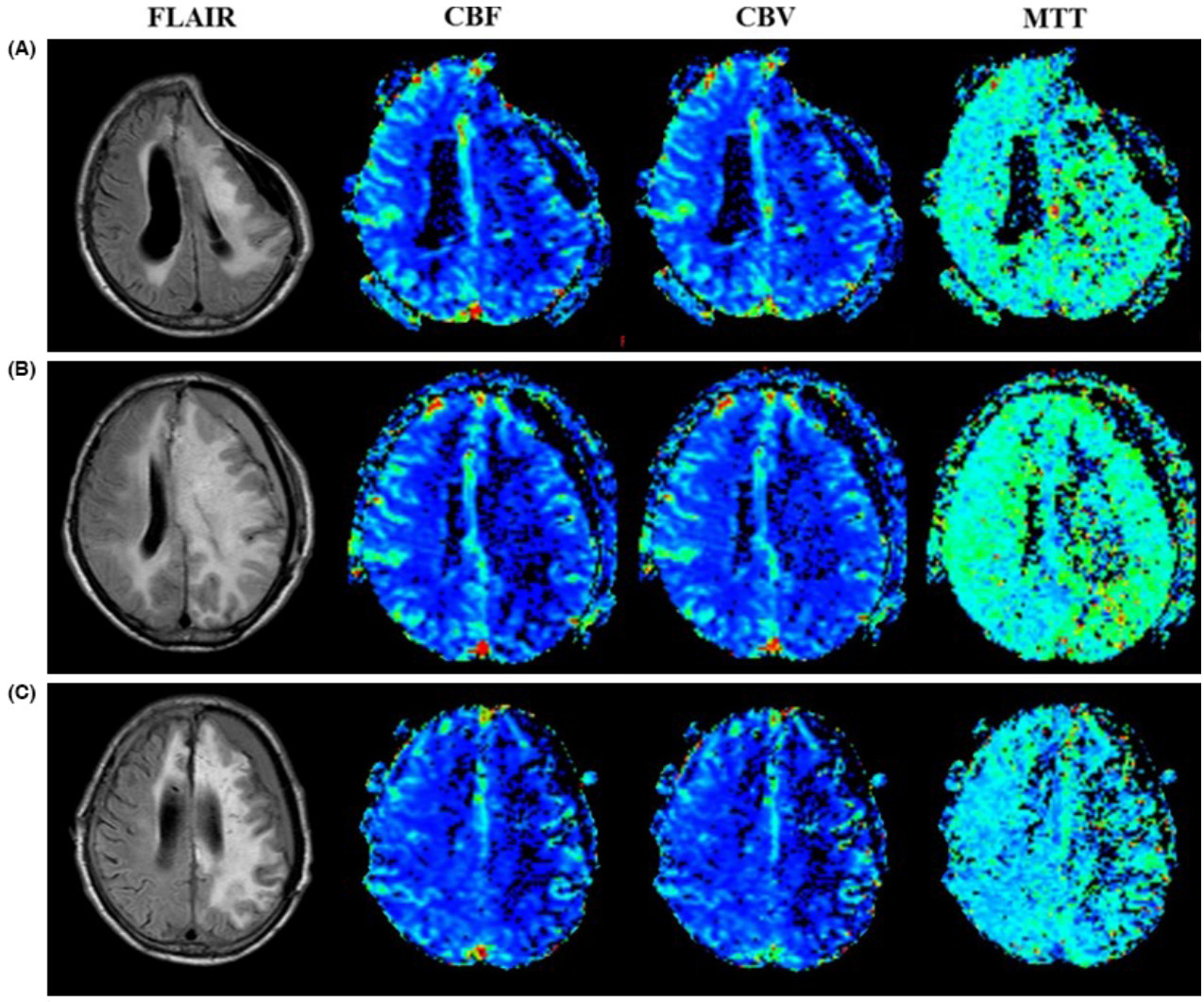

Fig. 2. Magnetic resonance images of fluid attenuated inversion recovery (FLAIR), cerebral blood flow (CBF), cerebral blood volume (CBV), and mean transit time (MTT) in the cerebral cortex in an 84-year-old man with subarachnoid hemorrhage. (A) Fourteen days before cranioplasty. (B) Seven days after cranioplasty. (C) One hundred and one days after cranioplasty.

published reports of cerebral edema and hemorrhage immediately after cranioplasty, including one case in which brain edema was recognized after hematoma evacuation. Similar to our case, none of these cases reported problems during surgery (Table 1). The etiology of post-cranioplasty edema and hemorrhage remains unknown.

Defective bone often recesses before cranioplasty, leading to midline shift. Sarov *et al.*[1] described this condition as asymptomatic sinking skin flap syndrome (SSFS). In cases with midline shift or shunt placement, intracranial pressure is decreased. In our review of published work, we found nine cases of midline shift prior to surgery (47.4%), 10 cases of prior shunt surgery (52.6%), and 15 cases of prior midline shift or shunt surgery (78.9%) (Table 1). Yoshida *et al.*[2] reported increased cerebral blood flow and metabolism in SSFS. Mangubat *et al.*[3] suggested 3-D CT angiography/CT venography as the cause of brain edema and hemorrhage following cranioplasty; using MR angiography/MR venography, Chitale *et al.*[4] failed to detect any embolic source, such as a venous thrombosis. After failing to detect dissection or occlusion, they suggested that it was caused by venous stasis, vascular injury, or reperfusion. Sviri[5] reported four similar cases. In one case, autopsy failed to demonstrate arterial or venous occlusion but

Table 1. Clinical and radiological characteristics of patients with edema or cerebral hemorrhage after cranioplasty

References	Age, years / sex	Cause	Site of initial injury	Midline shift before CP	Shunt	GOS before CP	Time to CP, months	Postoperative seizure	Type of implant	CT findings	Salvage operation	Outcome
Van Roost, 2003[11]	32/M	TBI	Left	NR	+	NR	NR	+	NR	CI	–	Dead
Cecchi, 2008[12]	77/M	CI	Right	+	–	SD	2	–	Autologous	Hemorrhagic infarct	–	Dead
Eom, 2010[13]	63/M	CI	Right	+	–	MD	10	–	Autologous	Edema, multiple hemorrhage	–	Dead
Honeybul, 2011[14]	22/M	TBI	Bifrontal	NR	+ Ligation	SD	2.5	+	Autologous	Edema	NR	Dead
	16/M	TBI	Bifrontal	NR	+ Ligation	PVS	2.3	–	Autologous	Edema	NR	Dead
	16/M	TBI	Bifrontal	NR	+ Ligation	SD	2.6	–	Autologous	Edema	NR	Dead
Zebian, 2011[7]	40/F	CI	Left	+	+	PVS	24	+	Titanium	Edema, brainstem hemorrhage	–	Dead
Kwon, 2012[6]	63/M	SAH	Right	+	–	NR	2	+	NR	ICH, IVH	–	NR
Santana-Cabrera, 2012[8]	17/M	TBI	Bifrontal	–	–	GR	2	+	NR	Edema	–	Dead
Hettige, 2012[10]	64/F	MN	Bifrontal	NR	+	GR	NR	–	Titanium	Edema	+	Dead
Chitale, 2013[4]	64/M	CI	Right	–	–	NR	12	+	PEEK	Edema	+	Dead
Evins, 2013[15]	46/M	CI	Right	NR	+	MD	0	–	PMMA	Edema	–	Dead
Mangubat, 2015[3]	14/F	TBI	Left	+	+	SD	2.5	–	PEEK	Edema, ICH	–	Dead
Sviri, 2015[5]	22/M	TBI	Right	–	–	MD	9	–	Autologous	Edema	+	Dead
	14/M	TBI	Left	+	+	SD	10	–	Autologous	Edema	+	Dead
	28/M	TBI	Right	+	+	MD	17	–	PMMA	Edema	+	Dead
	24/M	TBI	Left	–	–	SD	3	–	Autologous	Edema	+	Dead
Lee, 2015[16]	50/F	SAH	Right	+	–	NR	2	+	Autologous	Edema	NR	NR
This study	84/M	SAH	Left	+	–	PVS	2	–	HA	Edema, ICH, IVH	–	PVS

CI, cerebral infarction; CP, cranioplasty; CT, computed tomography; F, female; GOS, Glasgow Outcome Scale; GR, good recovery; HA, hydroxyapatite; ICH, intracerebral hemorrhage; IVH, intraventricular hemorrhage; M, male; MD, moderately disabled; MN, meningioma; NR, not reported; PEEK, polyetheretherketone; PMMA, polymethylmethacrylate; PVS, persistent vegetative state; SAH, subarachnoid hemorrhage; SD, severely disabled; TBI, traumatic brain injury.

confirmed brain necrosis. Kwon *et al.*[6] described one case in which the cause was reported as reperfusion due to surgery-related pressure changes. Our case included preoperative midline shift. Ipsilateral cerebral blood flow and volume was increased from before cranioplasty (Fig. 2). The postsurgical improvements suggest reperfusion as the cause of cerebral edema and hemorrhage.

Kwon *et al.*[6] reported impaired autoregulation due to decreased cerebral blood flow and metabolism when bone is deficient. Cranioplasty frees the brain from atmospheric pressure and causes brain expansion. Reduced cerebrovascular resistance increases perfusion pressure, leading to a sudden increase in blood flow. Automatic adjustment dysfunction has also been reported as the cause of brain edema.[7,8] Further damage can induce encephalomalacia-like transformation, and overperfusion can cause pressure changes that outpace the brain's capacity for elastic response.[6] In our case, the long-term bone-deficient state caused automatic adjustment dysfunction; inability to accommodate reperfusion induced further edema.

Santana-Cabrera *et al.*[8] reported cerebrovascular injury and hemorrhage due to microvasculature displacement following rapid flow restoration. Because this case showed preoperative midline shift, it is conceivable that microvasculature was displaced, causing damage after restorative surgery.

Negative pressure due to s.c. drain has also been reported to cause cerebral edema.[5,7] Negative pressure on cerebrospinal fluid changes intracranial pressure, leading to venous stasis. Resulting brain edema has been reported after normal cranial surgery.[9] An s.c. drain was placed in our case, thereby increasing negative pressure. The change from atmospheric pressure and negative pressure causes brain expansion, which contributes to venous stasis.

Hettige *et al.*[10] reported brain edema due to artificial bone material allergy. Although we did not test for allergies, we did not recognize any characteristic allergic changes, such as erythema.

There may be some overlap among all of these factors in the emergence of unexpected complications immediately following cranioplasty. A definitive cause often cannot be determined, but awareness of these factors may increase survival.

CONCLUSION

WE OBSERVED BRAIN edema and hemorrhage shortly after cranioplasty, attributed to automatic adjustment dysfunction following a long-term bone-deficient state, in conjunction with damaged brain tissue unable to withstand reperfusion. Venous stasis developed due to inhibition of venous return, contributing to edema expansion. Midline shift was recognized preoperatively. Our case showed asymptomatic SSFS concurrent with diminished intracranial pressure. Sudden release from atmospheric pressure and drain-related negative pressure following cranioplasty caused reperfusion-related changes in intracranial pressure, edema, and hemorrhage. Microvascular damage following rapid restoration may have contributed.

Although cranioplasty is a relatively minor operation, it is important to remember the various risk factors for unexpected complications. Immediate action may lead to increased survival.

CONFLICT OF INTEREST

NONE DECLARED.

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