




The Silent Threat: Unraveling the Rare Catastrophic Complication after Elective Cranioplasty

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Abstract

Although cranioplasty seems to be a simple procedure, fatal complication of development of diffuse severe cerebral edema following cranioplasty although unusual have been reported in a few cases. The mechanism for this occurrence is still speculative. A 38-year-old male patient presented with a history of having undergone left decompressive hemicraniectomy following a road traffic accident with traumatic left intracranial internal carotid artery dissection leading to anterior cerebral artery and middle cerebral artery territory infarct and endovascular embolization of traumatic type 1 left caroticocavernous fistula. The preoperative computed tomography scan of the brain revealed left frontotemperoparietal craniectomy defect with sunken flap and diffuse encephalomalacia with gliosis of the entire left cerebral hemisphere. Immediately following an uneventful cranioplasty surgery with titanium mesh, the patient developed severe hypotension and dilated fixed pupils. Postoperative imaging revealed diffuse severe cerebral edema in bilateral hemispheres with a significant midline shift toward the ipsilateral (left) side, that is, toward the side of cranioplasty. The patient immediately underwent removal of the titanium mesh, and despite all efforts, the patient had a fatal outcome on postoperative day 5. Although this type of fatal complication of diffuse severe cerebral edema is rare in postcranioplasty patients, neurosurgeons must be aware of this complication and close monitoring postprocedure is important, especially in patients with a large craniectomy defect and sunken skin flap.

Keywords

- ▶ complication
- ▶ cranioplasty
- ▶ edema

Introduction

Cranioplasty is one of the common neurosurgical procedures that involves the repair of a skull defect with a graft (either autologous bone or synthetic materials such as titanium, polymethyl methacrylate, polyetheretherketone, etc.). The wide use of decompressive hemicraniectomy surgery in patients with medically refractory intracranial hypertension secondary to trauma or cerebral infarction has resulted in the increased use of cranioplasty to repair these skull defects.

Although cranioplasty seems to be a simple procedure, the immediate postoperative complication rate is around

34%.¹ Fatal complications following cranioplasty although unusual have been reported in a few cases. Here, we report an unusual complication of severe cerebral edema immediately following cranioplasty, as the mechanism for this occurrence is still speculative.

Case Report

A 38-year-old male patient presented with a history of having undergone left decompressive hemicraniectomy at his hometown, in July 2023, following a road traffic accident with traumatic left intracranial internal carotid artery

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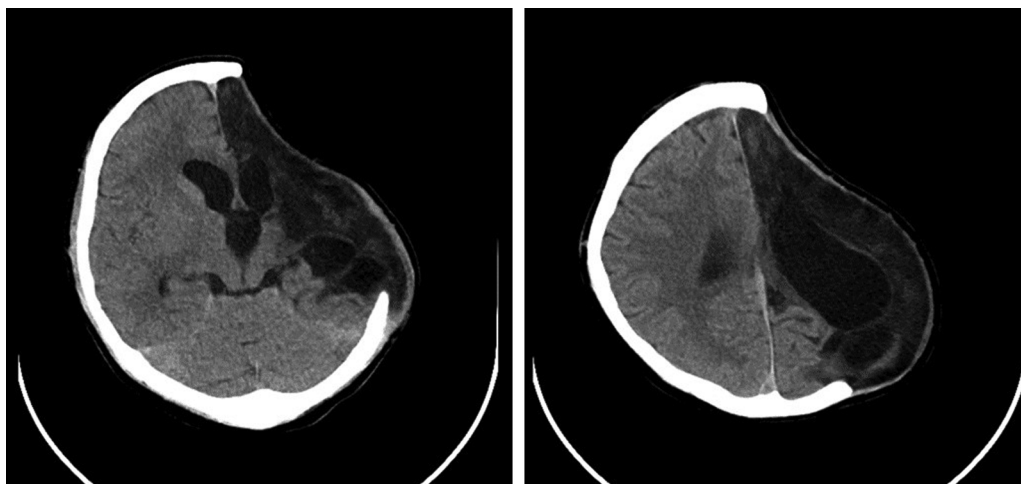


Fig. 1 Preoperative computed tomography scan (brain).

dissection leading to anterior cerebral artery and middle cerebral artery territory infarct. A few months later, he was diagnosed with type 1 direct caroticocavernous fistula on left side for which embolization using detachable coils was done. His Glasgow Coma Scale (GCS) was E4V2M6. There was no further deterioration in vision, and the redness of the eye that was present before embolization had completely

resolved. Computed tomography (CT) scan (→**Fig. 1**) revealed left craniectomy defect with a sunken flap and diffuse encephalomalacia with gliosis and ex vacuo dilatation of the left lateral ventricle.

The patient then underwent a customized titanium mold cranioplasty at our hospital. Intraoperatively, the brain was sunken. The surgery was uneventful with minimal blood loss.

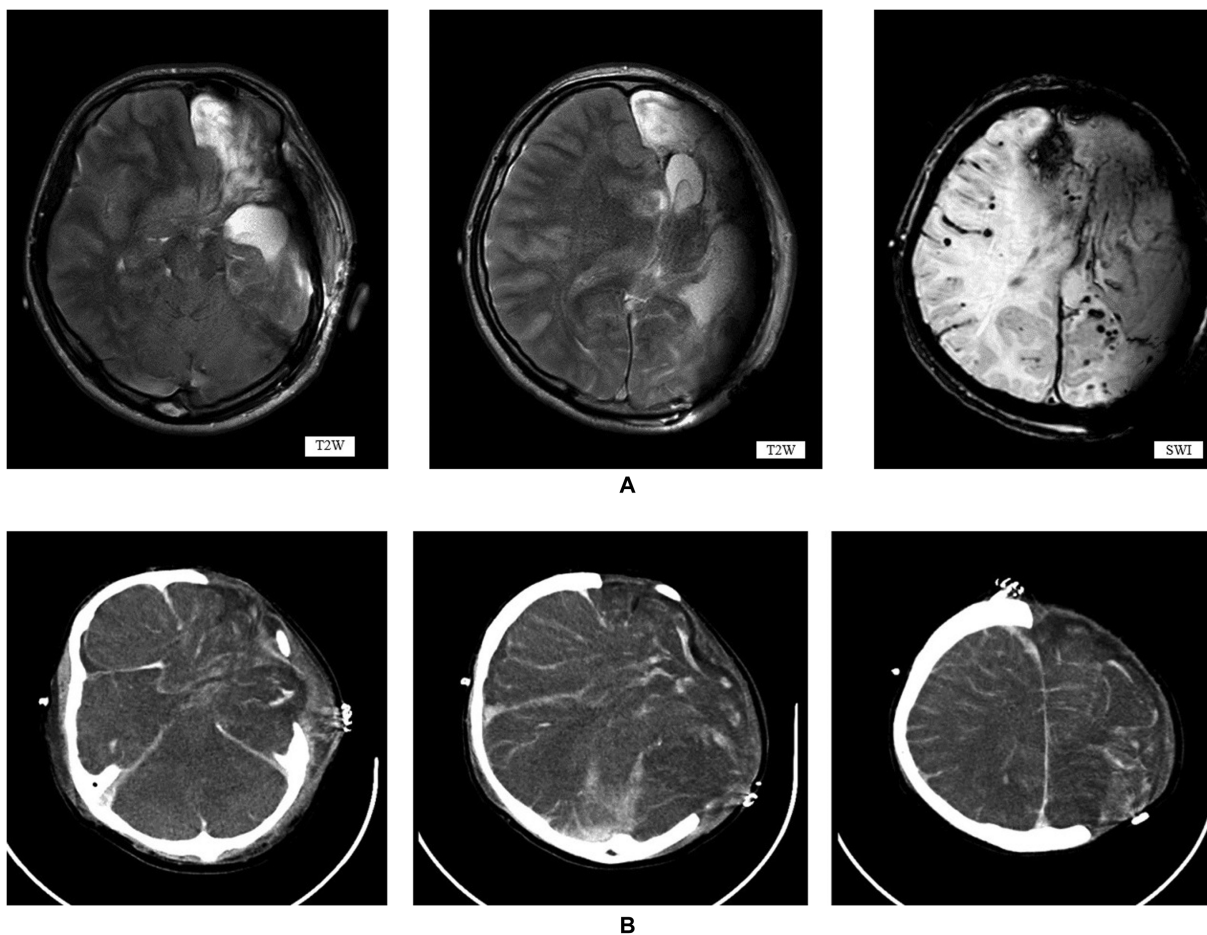


Fig. 2 (A) Immediate postcranioplasty magnetic resonance imaging scan (brain). (B) Postoperative day 3 computed tomography scan (brain).

However, the patient did not wake from anesthesia and had sudden hypotension (70/30 mm Hg) within a few minutes in the operation theatre while waiting to extubate. The hypotension lasted for a few seconds and stabilized with inotropes, with close titration, as the blood pressure was fluctuating to minor adjustments in inotropes dose. Arterial blood gases and electrolytes were within normal limits. Screening echocardiogram ruled out cardiac cause for hypotension. During resuscitation, the patient pupils dilated bilaterally from 2 to 4 mm over 20 minutes.

The patient was immediately shifted from the operation theatre for emergent imaging, and screening magnetic resonance imaging scan (→**Fig. 2A**) revealed diffuse severe cerebral edema in bilateral hemispheres with effaced basal cisterns, microhemorrhages, and poor gray–white matter differentiation in the contralateral (right) side with significant midline shift toward the ipsilateral (left) side, that is, toward the side of cranioplasty. There was no evidence of any diffusion restriction. So, the patient was immediately taken up for the removal of titanium mesh.

Postoperatively, there were significant hemodynamic disturbances. The operative flap was tense and bulging. GCS remained E1VtM1, and pupils were bilateral 6 mm dilated and fixed.

A repeat imaging with CT brain (→**Fig. 2B**) was performed on postoperative day 3 that showed diffuse cerebral edema with effaced CSF spaces and ventricles. Despite all possible efforts and intervention, the patient had a fatal outcome on postoperative day 5.

Discussion

Cranioplasty is one of the common procedures performed in neurosurgery for a skull defect. It can be associated with various complications that include infection, bone resorption, convulsions, and hematomas. Another complication that has been reported only in a few patients is the development of diffuse and severe cerebral edema immediately after cranioplasty. Although there have been several hypotheses for this unusual complication, they seem to be speculative and do not provide a satisfactory explanation.

The onset of the event, that is, the development of cerebral edema, is following implantation of the bone flap which is then followed by a series of changes that leads to a catastrophic outcome in most of the cases.

The probable causes for this catastrophic event in our patient could be:

- (1) The sudden negative pressure difference, due to the removal of atmospheric pressure following cranioplasty, could have induced the sudden onset of cerebral edema. Roost et al² in 2003 first reported a devastating cerebral swelling in a patient due to negative pressure from cranioplasty.
- (2) The chronically atrophied brain before surgery could have had impaired self-regulatory capacity, and in this state, the brain could not tolerate negative pressure gradients and lead to cerebral edema.

- (3) It has been reported that the brain parenchyma and basal cisterns communicate via the paravascular spaces, called Virchow–Robin spaces.^{3,4} Based on this, it could be speculated that following the initial onset of catastrophic events, the CSF could have shunted from the cisterns to the brain parenchyma through the paravascular spaces which in turn led to increased intracerebral pressure leading to cerebral edema and also impairment of the glymphatic system.
- (4) It has been identified that there is a link between TBI and the accumulation of metabolic wastes and misfolded proteins leading to long-term secondary brain damage⁵ due to the impairment of the glymphatic fluid circulation.⁶ But the intriguing feature is the rapid onset of cerebral edema, which cannot be solely attributed to the impairment of the glymphatic system.
- (5) Another possibility is that the blood–brain barrier is already disrupted, secondarily to traumatic brain injury. Following cranioplasty and the sudden change in the atmospheric pressure, there could have been an increase in cerebral blood flow, which is normally seen in postcranioplasty patients, especially in the microvascular beds which led to elevated capillary pressure and capillary leakage in an already disrupted blood–brain barrier leading to severe cerebral edema.

A subgaleal drain without suction was placed in our patient, and the theory of use of subgaleal drains postoperatively causing intracranial hypotension is not that convincing in our case unless the patient had significant postoperative CSF leakage, as these kinds of patients are exposed to long periods of intracranial hypotension secondary to decompressive hemispherectomy.⁷

An immediate imaging of the brain is suggested in postcranioplasty patients who do not recover from anesthesia or if there is inadequate spontaneous breathing or generalized seizures⁸ or a low/drop in GCS postoperatively.

Conclusion

Massive cerebral edema after cranioplasty is a rare but catastrophic complication. The above-mentioned hypotheses as reported in the literature and as speculated by us will require further validation in large numbers of cranioplasty patients, to identify these high-risk patients developing cerebral edema postcranioplasty.

Conflict of Interest

None declared.

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