

“Sunken brain and scalp flap” syndrome following decompressive “extra-craniectomy”

Abdul Rashid Bhat M Ch, Altaf R Kirmani M Ch, Furqan Nizami MS,
Ashish Kumar MS, Mohammed Afzal Wani M Ch

Department of Neurosurgery, Sher-i-Kashmir Institute of Medical sciences, Srinagar, Kashmir

Abstract: The sinking brain and scalp syndrome associated with neurological deterioration after decompressive craniectomy in traumatic brain edema is an uncommon condition. The recovery of neurological and imaging deficits following cranioplasty is well known. Although lumbar puncture and ventriculo-peritoneal shunts have been labelled precipitating factors to induce sinking brain syndrome but occasionally “extra” decompressive craniectomy alone may be sufficient to cause it. Clinically the non-pulsatile depressed brain and scalp flap with a marked concavity in an obtunded patient and radiological features like midline shift, paradoxical herniation and ‘kidney-bean’ shaped brain (Axial CT) are remarkable. It is more like a dehydrated infant with non-pulsatile depressed fontanelles. The cranioplasty has a dramatic effect on the overall recovery of the patient. We present a patient of traumatic brain injury who developed the “non-pulsatile sunken brain syndrome” without VP shunt, lumbar puncture or a external ventricular drainage and recovered completely after cranioplasty. The extra component of craniectomy is likely cause.

Keywords: cranioplasty; extra-craniectomy; low GCS; non-pulsatile sunken brain

INTRODUCTION

The procedure of decompressive craniectomy is widely used in neurosurgical field for the relief of intractable intracranial hypertension in patients with severe traumatic brain injury, acute stroke, and severe brain edema after intracranial procedure^{1,2}. The syndrome of the sinking skin flap was introduced to explain neurological deterioration after decompressive craniectomy³. This phenomenon may result from CSF hypovolemia, atmospheric pressure gradient that may be aggravated by CSF diversion, dehydration, and position change^{1,4}. A patient of sinking brain and skin flap syndrome is managed by the Neurosurgical Unit of Kashmir and is presented in detail.

CASE REPORT

A 45 year adult male was received by the accident/emergency staff, at SKIMS, Kashmir with a history of road traffic accident three hours back without any roadside or in-transportation resuscitation, in a condition of unattended airway, neck movements and urinary

bladder. After complete resuscitation, an immediate and quick general physical and neurological examination revealed that patient had a GCS (Glasgow Coma Scale) score of 7/15 and all signs of raised intracranial pressure (heart rate 56, blood pressure 190/110 mmHG, irregular and decreased respiratory rate) with lateralization (left hemiparesis and right papillary dilatation and non-reaction) to the right side. An immediate plain CT scan brain (Fig 1) depicted bitemporal lobe contusion (larger on right side) and right temporal acute subdural hematoma with severe brain edema and midline shift towards left side. The patient was prepared and a right temporo-parietal craniotomy followed by evacuation of acute subdural hematoma and contusion with a duroplasty was carried out to add space to the underlying

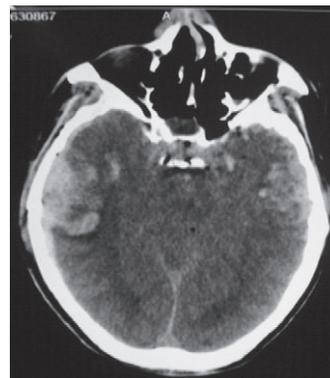


Fig 1 : Plain CT scan brain showing large right and small left temporal lobe contusion with brain edema and obliterated cisterns

Address for Correspondence:

Dr. Abdul Rashid Bhat

Additional Professor

Department of Neurosurgery, (c/o B-4 Faculty Quarters)

Sher-i-Kashmir Institute of Medical Sciences

Soura, Srinagar, J&K, India

Email: seven_rashid@rediffmail.com / huwa9adam@gmail.com

edematous brain. The calvarial bone flap was buried subcutaneously in the abdomen. Post-operatively patient was electively ventilated for 48 hours and was put on antibiotics, anticonvulsants and decongestant drugs. He showed a good response in all signs and a plain check CT scan brain (Fig 2) showed evidence of skull bone defect, regression of brain edema, resumption of midline and resolving left sided contusion. The patient improved neurologically to a GCS score of 14/15 within two weeks. However the buried bone flap got infected in a subcutaneous parietal wall abscess and was discarded from reuse. The patient was discharged home with a GCS score 15/15 and mild residual left hemiparesis after 24 days of hospital stay. About six weeks later, he presented with seizures, vomiting, headache and alteration in consciousness for the last nine days. On examination patient was hemodynamically stable, had sunken eyes, dry tongue. Neurological examination revealed an irritable and arousable patient, and a GCS score of 13/15. Local examination showed an indrawn and sucked-in right temporo-parietal scalp flap, over the craniectomy defect, at an acute angle to the skull margins of the defect creating a non-pulsatile gorge-like pit, and the skin was unpinchable (Fig 3). Plain CT brain showed cranial cavity in the form of a kidney-bean shape like a

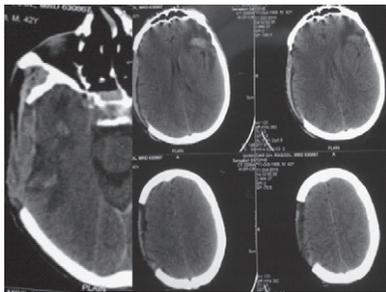


Fig 2: Postoperative plain CT scan brain with evident decompressive “Extra”-craniectomy extending to parietal region, evacuated contusion & acute SDH and relaxed cisterns



Fig 3: Clinical photograph showing non-pulsatile sunken brain and skin flap 6 weeks postoperatively revealing a pit-like craniectomy defect.

concavo-convex surface (Fig 4) with concave surface facing skull defect as if held in position by vacuum. The patient was rehydrated well and prepared for cranioplasty. A defect-sized flap of methymethacrylate powder and solvent in due proportion was made and applied to the cranial defect. Patient had uneventful postoperative period and dramatic recovery of GCS score deficit, headache, irritability and vomiting was recorded. A post-cranioplasty check CT scan brain proved the imaging recovery of the brain and cranial defect (Fig 5). There were no seizures during the hospital stay, and he was discharged six days later.

DISCUSSION

In the absence of any lumbar puncture, ventriculo-peritoneal shunt, external ventricular drainage or brain atrophy on the opposite side, the patient at SKIMS Kashmir had non-pulsatile sunken scalp and brain following decompressive craniectomy with an ‘extra’ component of parietal craniectomy. The non-pulsatile concave and sunken brain and un-pinachable scalp is a sign of vacuum sucked overlying brain and scalp either vertically down or horizontally across. This is like

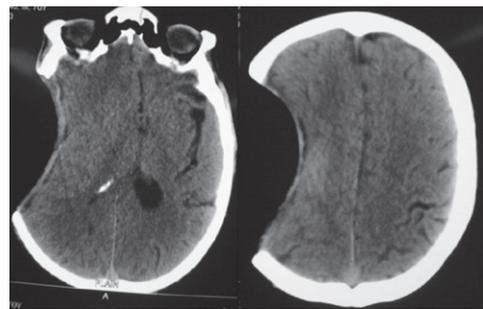


Fig 4: Plain CT scan 6 weeks postoperatively revealing right cerebral sinking, beginning at the margins of the “Extra” – craniectomy defect till central depth and looking like a kidney-bean.

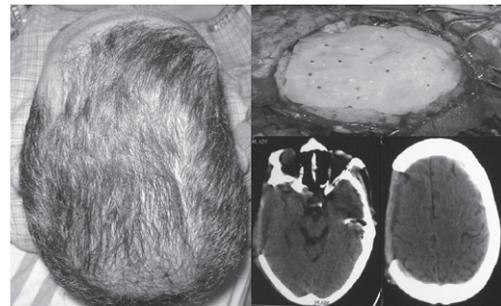


Fig 5: Plain post-cranioplasty CT scan brain, Clinical and intra-operative photographs showing evidence of cranioplasty, reversal of sinking brain and resumption of midline and cisterns back to normal position.

depressed and non-pulsatile anterior fontanellae of a dehydrated infant where lower central venous pressure leads to negative venous pressure, vacuum formation and absence of pulsations in superior sagittal sinus. This vacuum effect leads in turn to depression of the roof of superior sagittal sinus as well as the overlying skin of the fontanellae. By the law of physics, the vacuum or negative pressure is created due to loss of volume and pressure in an air/water tight closed space like cranial cavity (containing fixed volume of brain, blood and CSF). For the suction of right brain across and towards the left, creation of vacuum on the left side is required in the form of external ventricular drainage, brain atrophy (post-injury) and lumbar puncture to produce local intracranial hypotension and hypovolemia. Conversely, high atmospheric air pressure may also cause depression and shift of the scalp and brain to opposite side but in the absence of vacuum and presence of volume/pressure on the opposite side, the sunken scalp and brain shall continue to be pulsatile irrespective of concavity and depression.

Since the first reporting of the sinking skin flap syndrome following decompressive craniectomy by Yamamura in 1977, the few reports about this entity and postulations related to its causation are found in the literature^{1-3,5-8}. Many investigators have sought to explain the pathophysiology of this phenomenon. Langfitt 1968 had theorized that the atmospheric pressure is directly transmitted to the intracranial cavity, causing inward shifting of the scalp over craniectomy site⁹. According to this theory, George et al showed in a series of angiographies that there was a correlation between the restoration of the midline shift and clinical improvement following cranioplasty¹⁰. Recently, several authors proposed that a negative gradient between atmospheric and intracranial pressure, which is aggravated by changes in the CSF compartment following CSF hypovolemia, to be the mechanism of neurological deterioration after craniectomy. The CSF drainage in a patient suffering from hydrocephalus or meningitis exacerbates this effect by creating a pressure gradient through craniectomy site. The prolonged dehydration and up-right position may also precipitate this phenomenon^{7,8}. Han et al postulated that multiple calcification around superior sagittal sinus caused hydrocephalus requiring a V-P shunt which increased negative pressure gradient between atmospheric and intracranial pressure and caused neurological deterioration such as the syndrome of the sinking skin flap¹¹. The post-traumatic hydrocephalus

occasionally develops in patients who undergo decompressive craniectomy after head injury, accounting for 10.1% to 28.6% hydrocephalus in decompressive craniotomies^{12,13}. At SKIMS Neurosurgical Centre it is observed that many neonates with congenital hydrocephalus and open fontanellae, develop cranial bony overlapping due to decrease in intracranial pressure and hypovolemia after VP shunt insertion and CSF over drainage. The mechanism of sinking brain syndrome in patients with hydrocephalus on VP shunt post-decompressive craniectomy is similar to the bony overlapping in neonates. Another observation at SKIMS is, during sub-occipital craniectomy in sitting position in the presence of lower central venous pressure and hypovolemia, the dura/arachnoid overlying cerebellum is depressed and sticks to the surface of non-pulsatile cerebellum. The mechanism of vacuum development is similar in the sinking brain syndrome.

Although the obvious causal relationship between decompressive craniectomy and hydrocephalus remains unclear, the disturbance of CSF flow around the cerebral convexities may be the possible mechanism. In the context of hydrocephalus after decompressive craniectomy, it is difficult for neurosurgeon to decide whether CSF diversion can be safely performed. Accordingly, it is important to recognize the presence of craniectomy with extreme caution to perform a continuous CSF diversion¹. The goal of treatment in patient with the syndrome of the sinking skin flap is restoration of the pressure exerted by depression of craniectomy site. Several authors suggested that severe CSF hypovolemia after craniotomy may produce a dramatic herniation syndrome that is completely reversed by the Trendelenberg position^{4,6}. Also, it was reported that intrathecal saline infusion reverses impending transtentorial herniation in patient with decline of mental status due to intracranial hypotension¹⁴. However conservative management of the syndrome of the sinking skin flap with neurological deterioration is largely ineffective, and cranioplasty is the cornerstone of surgical treatment. Researchers suggested that cranioplasty improves the neurological deficits by an increase in local intracranial pressure and correction of abnormal CSF dynamics¹⁵. Another report suggested that the cranioplasty may affect postural blood flow regulation, cerebrovascular reserve capacity and cerebral glucose metabolism¹⁶. However, the cranioplasty for severe sinking at the skull defect may result in the dysfunction of the underlying brain, risk of fluid collection and

hematoma formation in the subdural space due to the large dead space. The safe and effective surgical methods to expand the scalp depression and to eliminate the dead space (in the context of V-P shunt) are temporary occlusion or removal of shunt device before cranioplasty¹. Additionally, some reporters used a temporary increase of pressure valve to restore the scalp depression and midline shift¹¹.

The notable finding in the patient at SKIMS Kashmir was parietal 'extra-craniectomy' and opposite side temporal contusion leading to possible loss of volume and pressure over a period of time. The parietal component of 'extra' craniectomy/craniotomy in temporal lobe contusion, the duroplasty and burying the craniotomy-bone flap temporarily in the abdomen is understood to increase the cranial-cavity space to wave-off the effects of raised intracranial pressure later on. The cranium-uncovered and bare but normal brain in case of 'extra' craniectomy sinks along with the overlying scalp due to changes in the CSF pressure dynamics is not understood yet. How does the defect causes CSF pressure changes in the subarachnoid spaces on the convexity of the cerebral cortex, underlying parenchyma and ventricles needs pathophysiological evaluation.

CONCLUSION

The non-pulsatile sunken skin flap and brain syndrome is a complication of decompressive craniotomy/craniectomy in the presence of intracranial hypovolemia and hypotension. Although this syndrome may also occur by the decompressive craniectomy over the normal brain which is 'extra-decompression' than is warranted for the treatment of traumatic brain edema but its mechanism is not known. Further clinical and laboratory studies are needed to analyze the pathophysiology. However every case of decompressive craniectomy is not liable to develop sunken skin flap and brain syndrome.

REFERENCES

- Liao CC, Kao MC. Cranioplasty for patients with severe depressed skull bone defect after cerebrospinal fluid shunting. *J Clin Neurosci* 2002; 9: 553-5.
- Schiffer J, Gur R, Nisim U, Pollak L. Symptomatic patients after craniectomy. *Surg Neurol* 1997; 47: 231-7.
- Yamamura A, Sato M, Meguro K, Nakamura T, Uemura K, Makino H. Cranioplasty following decompressive craniectomy. Analysis of 300 cases. *No Shinkei Geka* 1977;5: 345-53.
- Kelley GR, Johnson PL. Sinking brain syndrome: craniotomy can precipitate brainstem herniation in CSF hypovolemia. *Neurology* 2004; 62: 157-9.
- Cho H, Kim CH, Kim JH, Kim JM. Paradoxical herniation after decompressive craniectomy for acute subdural hematoma. *J Korean Neurosurg Soc* 2006; 40: 51-3.
- Komotar RJ, Mocco J, Ransom ER, et al. Herniation secondary to critical postcraniotomy cerebrospinal fluid hypovolemia. *Neurosurgery* 2005; 57: 286-92.
- Oyelese AA, Steinberg GK, Huhn SL, Wijman CA. Paradoxical herniation secondary to lumbar puncture after decompressive craniectomy for a large space-occupying hemisphere stroke : case report. *Neurosurgery* 2005;57:594.
- Schwab S, Erbugth F, Aschoff A, Orberk E, Spranger M, Hacke W. Paradoxical herniation after decompressive trephining [in German]. *Nervenarzt* 1998; 69: 896-900.
- Langfitt TW. Increased intracranial pressure. *Clin Neurosurg* 1968; 16: 436-71.
- George AE, Morantz RA, Abad RM, Rovit RL, Chase N. Neuroradiology of the posthemicraniectomy patient with special emphasis on the radiology of unilateral atrophy. *Radiology* 1974; 111: 627-31.
- Han PY, Kim JH, Kang HI, Kim JS. "Syndrome of the Sinking Skin-Flap" secondary to the ventriculoperitoneal shunt after Craniectomy. *J Korean Neurosurg Soc* 2008;43: 51-3.
- Phuenpathom N, Ratanalert S, Saeheng S. Post-traumatic hydrocephalus: experience in 17 consecutive cases. *J Med Assoc Thai* 1999; 82: 46-53.
- Polin RS, Shaffrey ME, Bogaev CA, et al. Decompressive bifrontal craniectomy in the treatment of severe refractory posttraumatic cerebral edema. *Neurosurgery* 1997;41: 84-94.
- Binder DK, Dillon WP, Fishman RA, Schmidt MH. Intrathecal saline infusion in the treatment of obtundation associated with spontaneous intracranial hypotension: technical case report. *Neurosurgery* 2002; 51: 830-37.
- Segal DH, Oppenheim JS, Murovic JA. Neurosurgical recovery after cranioplasty. *Neurosurgery* 1994; 34: 729-31.
- Winkler PA, Stummer W, Linke R, Krishnan KG, Tatsch K. Influence of craniaoplasty on postural blood flow regulation, cerebrovascular reserve capacity, and cerebral glucose metabolism. *J Neurosurg* 2000; 93: 53-61.