Case Report

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Death, a Rare Complication of Cranioplasty in Emergency: A Case Report

Dentistry Section

NANDAKISHORE SAHOO[1], SUDARSHAN BHAT[2], MOHAN RANGAN[3]

ABSTRACT

Cranioplasty is a reconstructive procedure which is performed to restore the calvarial integrity with either a stored autologous bone flap or a custom synthetic prosthesis. It is performed to protect the brain and as a cosmetic procedure. It has been shown to improve patient's functional outcome. This procedure has been performed as early as <14 days postdecompressive craniectomy. Cranioplasty in emergency which is a variant of secondary cranioplasty is rarely indicated after decompressive craniectomy. Complication rate associated with cranioplasty is relatively high. Infection, convulsions, and epidural haematoma are frequent complications of cranioplasty which are not life threatening. Fatal complications associated with this procedure are not well documented and that could be among one of the reason that death, as a complication following cranioplasty is substantially low. Here, a case of unexpected death of a 37-year-old female postcranioplasty which was performed as an emergency procedure is reported. She was a previous case of right mid one-third parasagittal meningioma who developed severe sinking skin flap syndrome after three months of parasagittal craniectomy. The patient was operated under general anaesthesia for reconstruction of the residual calvarial defect. However, the patient developed bacterial meningitis and on the 16th day postcranioplasty procedure, she died of cardiac arrest. The procedure had otherwise been uneventful and it was speculated that infection and cerebral oedema postcranioplasty might have been the cause of death.

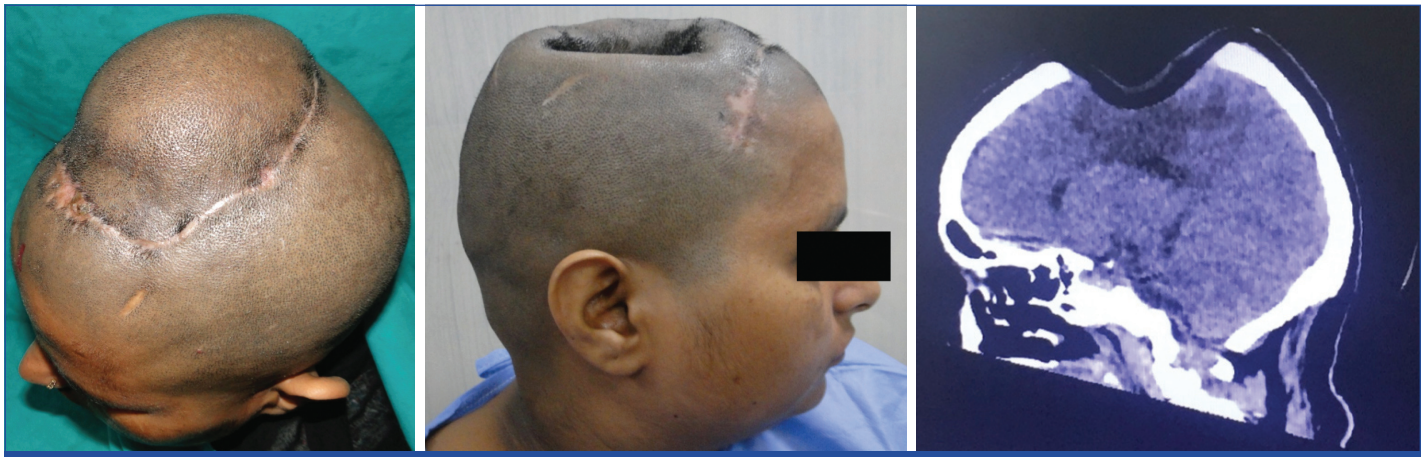
Keywords: Cerebral oedema, Residual calvarial defect, Sinking skin flap

CASE REPORT

A 37-year-old female was referred from the Department of Neurosurgery to the Department of Oral and Maxillofacial Surgery with features of severe sinking skin flap syndrome after three months of parasagittal craniotomy for consideration of emergency cranioplasty. She was a previous case of right mid one-third parasagittal meningioma operated in the month of June 2017 in a tertiary care centre of the Armed Forces Medical Services. She underwent right mid one-third parasagittal craniectomy wherein her superior sagittal sinus was exposed, dura over the tumour was coagulated and Simpson grade I resection of the tumour was carried out. After six weeks of the initial surgery a Non Contrast Computed Tomography (NCCT) of brain showed depression of right cerebral hemisphere, midline shift to left and calcified granuloma of left frontal lobe. After three months of the initial surgery, she developed a large pseudomeningocele with herniated brain matter through the calvarial defect [Table/Fig-1]. She had recurrent seizures, persistent left hemiparesis altered Glasgow Coma Scale (GCS) (E3V4M6-13/15) and was bound to wheelchair. She was managed by the neurosurgical team by Thecoperitoneal (TP) shunting. The shunting was effective, and the bulging of the flap reduced.

However, the patient developed severe sinking skin flap syndrome within 48 hours of TP shunting [Table/Fig-2,3] with symptoms of headache, mental changes and drowsiness. On the 2nd TP shunt day, the shunt was blocked by ligation and patient was observed for improvement but patient continued to be symptomatic. The general condition of the patient further deteriorated with increase in frequency and severity of seizure, loss of orientation and decrease of GCS (E2V3M5-10/15). Hence, the patient was referred to the authors for emergency cranioplasty.

Informed consent was taken and the patient was operated under general anaesthesia for reconstruction of residual calvarial defect. A subgaleal flap was raised to avoid durotomies and epi/subdural bleeding. The defect was exposed and aluminium foil was used as a template to measure the size of the defect and as a guide to mimic the curvature of the skull. A single piece of low profile titanium mesh (manufacturer: Stryker Leibinger) was used to cover the craniotomy defect completely with the edges of the mesh extending

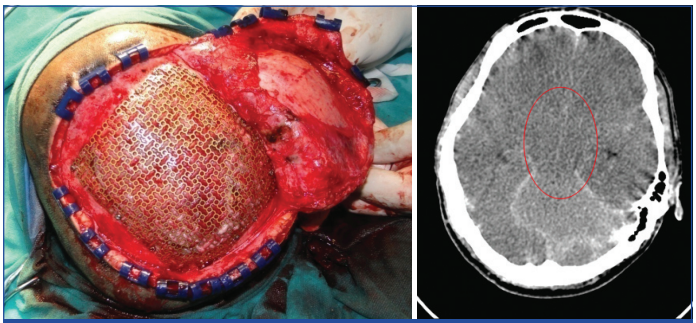


[Table/Fig-1]: Large psuedomeningocele with herniated brain matter through the calvarial defect three months after mid parasagittal craniectomy; **[Table/Fig-2]:** Sinking skin flap syndrome seen 48 hours after Thecoperitoneal (TP) shunting was done to manage psuedomeningocele; **[Table/Fig-3]:** Sinking skin flap syndrome (CT Scan-Sagittal view). (Images from legft to right)

approximately 5 mm on the sound bone [Table/Fig-4]. The cut edges of the mesh were turned down toward the bone so as not to project into the overlying scalp. A 10 monocortical titanium micro screws (1.7x5 mm Stryker Leibinger screws) were used to secure the mesh. Haemostasis was achieved and the wound was closed using a single layer of monofilament suture material. The duration of the surgery was 3 hours and 20 minutes. Postoperatively patient continued to be symptomatic (headache, mental changes and drowsiness).

The intracranial pressure was monitored and the readings were between 10-12 mmHg. The patient was under observation and adequate postoperative care about seizure prophylaxis (levetiracetam 1 gm loading dose followed by 500 mg twice daily) and cardiac stability was taken. The suction drain was removed 48 hours postsurgery.

On the 11th postoperative day, she showed features of meningitis i.e., increased loss of sensorium, fever and neck rigidity. The patient was subjected to NCCT [Table/Fig-5] which showed cerebral oedema with no mass effect and no evidence of subgaleal collection. Cerebrospinal Fluid (CSF) sampling was done through lumbar puncture which revealed increased white blood cell (WBC) count and gram stain was positive and therefore, it was decided to remove the shunt on the 12th postoperative day. The CSF collected during removal of the shunt was sent for culture and the culture showed evidence of bacterial meningitis. The CSF culture showed growth of *Acinetobacter baumannii* which was sensitive to higher antibiotics like tigecycline and colistin. Therefore, the patient was administered with a combination therapy of inj. tigecycline 50 mg twice a day and inj. colistin methane sulphonate 1.25 mg/kg every 6 hour. On the 14th postoperative day the patient developed status epilepticus and was placed on ventilator. Inspite of parenteral antibiotics and other supportive care measures (administration of decongestants and automated external defibrillator) patient continued to have fever and developed features of septic shock. Thereafter, the patient developed multiorgan dysfunction and was started on double ionotropes to maintain her vitals. On the 16th day postcranioplasty procedure, the patient died of cardiac arrest.



[Table/Fig-4]: Intraoperative image showing titanium mesh covering the craniotomy defect fashioned to mimic the curvature of the skull fixed using monocortical titanium microscrews; **[Table/Fig-5]:** (CT scan-Axial view)- Effacement of sulci and gyri, blunting of basal cisterns and kinking of ventricles indicating cerebral oedema. (Images from left to right)

DISCUSSION

Cranioplasty is a reconstructive procedure which is performed to restore the calvarial integrity. It is more than just a cosmetic repair as it has a pivotal role in the rehabilitation process following a patient's neurological injury [1,2]. It may be termed as primary, when performed in conjunction with addressing a trauma or a pathology and secondary, when performed at a later stage. Emergency cranioplasty is a variant of secondary cranioplasty. The incidence of cranioplasty done as an emergency is very less and is mainly indicated in acute cases of syndrome of trephine and sinking brain and scalp syndrome associated with neurological deterioration after decompressive craniectomy [1,3]. The major therapeutic effect of cranioplasty is enhanced cerebral flow and restoration of cerebral dynamics which in turn leads to early neurological recovery of the patient. Infection, convulsions, and epidural haematoma are frequent complications of cranioplasty [4].

Repair of the cranial vault defect can be done by insertion of autologous graft or alloplastic materials. Brain protection and cosmesis are the major indications of cranioplasty. The optimal timing for performing a cranioplasty after craniectomy has been debated. Cranioplasty has been performed as early as 2 weeks to as late as 18 months [5,6]. For several decades it was believed that early cranioplasty was associated with a poor outcome as it involved intervening into a contaminated wound [7]. Whereas several recent clinical reports have established that early cranioplasty provides a better functional and neurological outcome [8,9]. Postoperative haematoma, subdural effusion and herniation through the defect are the common early postoperative complications of craniectomy. Syndrome of trephine, sinking brain and scalp syndrome, hydrocephalus and seizures have also been reported postcraniectomy [9]. Therefore, cranioplasty is performed to overcome these complications. In this case, the patient was treated for right mid one-third parasagittal meningioma by mid one-third parasagittal craniectomy. Three months postcraniectomy, the patient suffered from complications of bulging of flap, headache, seizures, hemiparesis and altered GCS associated with psuedomeningocele. Development of meningocele may be attributed to altered CSF clearance due to damage to the arachnoid structure though exact pathophysiology is not clear. The incidence of postcraniectomy hydrocephalus is high following traumatic brain injury [10].

Since, the general condition of the patient was compromised, the neurosurgeon decided to place a TP shunt to deviate CSF flow. The TP shunting is simpler extracerebral procedure with low risk of shunt infection and malfunction [10]. Though the incidence of over drainage through TP shunt is less than Ventriculo-Peritonial (VP) shunt [10,11]. Within 48 hours of shunting the index patient developed severe sinking flap syndrome with increase in frequency and severity of seizure, loss of orientation and decrease in GCS also known as the syndrome of trephine. This may be attributed to over drainage of CSF. Sinking flap syndrome is generally associated with neurological dysfunction, behavioural changes and severe headaches [9]. This has been attributed to CSF hydrodynamic changes, cerebrovascular/haemodynamic changes, and metabolic changes [3]. Since, the general condition of the patient further deteriorated with increase in frequency and severity of seizure, loss of orientation and decrease of GCS, she was referred for an emergency cranioplasty.

Cranioplasty done in emergency is a variant of secondary cranioplasty which is indicated for its therapeutic effect in cases of syndrome of trephine and severe sinking skin flap syndrome. It is a less practiced procedure and is expected to relieve the symptoms at the earliest. There are a plethora of autogenous and alloplastic materials available for reconstruction of calvarial defects. In emergency, stock titanium mesh implant is suitable as it is readily available. It is sterilised by autoclaving and can be customised on table [5].

Infection, convulsions, epidural haematoma, cerebral oedema, haemorrhage and death are some of the complications of cranioplasty. The overall complication rate, postcranioplasty is estimated to be between 15% to 37% [2,12]. Death, following cranioplasty is a rare complication and the incidence is about 3% [13]. Most patients have complications after cranioplasty but these complications seldom play a role in poor prognosis [12]. In this case, postemergency cranioplasty the patient continued to be symptomatic and NCCT head revealed the features of cerebral oedema (effacement of sulci and giri and blunting of cerebral basal cisterns) [Table/Fig-5]. The incidence of this fatal complication is considerably high and the aetiology of postcranioplasty cerebral oedema remains obscure. Van Roost D et al., first reported a case of devastating cerebral swelling postcranioplasty which they attributed to the negative pressure due

to significant pressure differentials between the intracranial pressure and the atmospheric pressure [14]. This negative pressure theory was further supported by Honeybul S and Sviri GE [13,15]. There have been reports in which cerebral oedema is also referred to as pseudo hypoxia of brain which has provoked death [12]. Similar to present case, none of these cases reported problems during surgery. The neurosurgeon decided to remove the TP shunt on the 12th postoperative day. Culture of CSF showed evidence of bacterial meningitis and the patient was administered with culture specific antibiotics. The source of infection in this case might have been the TP shunt or the cranioplasty. A study has reported that the rate of infection following TP shunt varies from 1% to 9% and that following cranioplasty varies from 7% to 11% [16]. A combination therapy of inj. tigecycline and inj. colistin methane sulphonate was administered to the patient [17,18]. Despite using combination therapy in conjunction to aggressive critical care management the general condition of the patient did not improve and the patient succumbed to the effects of cerebral oedema and bacterial meningitis.

There is limited information with regards to emergency cranioplasty in sunken flap syndrome, therefore it still remains a dilemma for surgeons whether emergency cranioplasty would benefit patients with sunken flap syndrome or not. In this case, there were no other alternative options available, and the procedure was undertaken after due consent.

CONCLUSION(S)

Cranioplasty, is relatively a simple procedure but it may lead to unexpected complications as in this case. Despite adequate postoperative care about paying close attention to seizure prophylaxis, cardiovascular stability and using suction drains at low pressure in order to avoid epidural haematoma, patient developed cerebral oedema and meningitis and succumbed to the illness. In view of these findings, it is of paramount importance to make the patients and their relatives aware of the unexpected complications that could happen during the procedure despite following standard pre and postoperative protocols.

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PARTICULARS OF CONTRIBUTORS:
1. Professor and Head, Department of Oral and Maxillofacial Surgery, Armed Forces Medical College, Pune, Maharashtra, India.
2. Lecturer, Department of Oral and Maxillofacial Surgery, Army Dental Centre R&R, New Delhi, India.
3. Lecturer, Department of Oral and Maxillofacial Surgery, Army Dental Centre R&R, New Delhi, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Sudarshan Bhat,
Lecturer, Department of Oral and Maxillofacial Surgery, Army Dental Centre R&R, New Delhi, India.
E-mail: sudarshan_in@yahoo.com

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