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TITLE: Sinking bone flap due to overdrainage of a ventriculoperitoneal shunt. A case report and review of the literature.

ABSTRACT

The widespread use of decompressive craniectomy and subsequent cranioplasty has led to a better understanding of its complications. However, cases of a sunken bone flap have hardly ever been described. We present the eighth case reported up to date and perform a review of the literature of this sporadic complication.

A 40-year-old Caucasian male suffered a traumatic brain injury that required a decompressive craniectomy. One month after initial trauma autologous cranioplasty was performed. A ventriculoperitoneal shunt was also placed. Neurological status progressively improved but his therapist noted cognitive status decline 8 months later. Follow-up computed tomography showed a progressive sinking bone flap. The patient underwent bone flap removal and a custom-made calcium phosphate-based implant was inserted, leading to symptoms resolution.

Bone resorption has been described as the main cause of sinking bone flap following cranioplasty. This entity may manifest with symptoms of overdrainage in patients with cerebrospinal fluid shunt devices.

KEYWORDS

cerebrospinal fluid; decompressive craniectomy; overdrainage; sinking; traumatic brain injury; ventriculoperitoneal shunt.

TITULO: Hundimiento de colgajo óseo por sobredrenaje de una derivación ventriculoperitoneal. Caso clínico y revisión de la literatura

RESUMEN

El uso extendido de la craniectomía descompresiva y la consiguiente craneoplastia ha propiciado un mejor conocimiento de sus complicaciones. Sin embargo, esporádicamente se han descrito casos de hundimiento del colgajo óseo. Describimos el octavo caso descrito hasta la fecha y realizamos una revisión de la literatura de esta infrecuente complicación.

Un varón de 40 años sufrió un traumatismo craneoencefálico que requirió craniectomía descompresiva. Un mes después se sometió a la reposición de su colgajo óseo, junto con la implantación de una derivación ventriculoperitoneal. Presentó mejoría neurológica progresiva que se frenó y empeoró 8 meses después. La tomografía computerizada de control mostró hundimiento progresivo del colgajo óseo. El paciente se sometió a la retirada del colgajo óseo y craneoplastia con implante a medida, con resolución de los síntomas.

La resorción ósea se ha descrito como la principal causa del hundimiento del colgajo óseo tras craneoplastia. Sin embargo, esta entidad puede manifestarse como síntomas de sobredrenaje en pacientes con derivación de líquido cefalorraquídeo.

PALABRAS CLAVE

líquido cefalorraquídeo; craniectomía descompresiva; sobredrenaje; hundimiento; traumatismo craneoencefálico; derivación ventriculoperitoneal.

MAIN TEXT

Introduction

The widespread use of decompressive craniectomy has led to rise the frequency of cranioplasty. It not only protects the brain and restores cosmesis, but also improves neurologic function. The overall complication rate for cranioplasty after decompressive craniectomy is cited to be in the range from 10% to 40%.¹ These complications include infection, (which is the most important one), bone flap resorption, intracranial haemorrhage, extra-axial fluid collections and hydrocephalus, as well as seizures.¹⁻³

A still rarer complication, hardly ever being described, and the motivation behind writing this paper, is the presence of a sinking bone flap, giving rise to neurological symptoms. Also known as the “sinking bone flap syndrome”, only seven cases have been reported up to date in the literature.⁴⁻⁹ The aim of this study, then, is to report the eighth case of this clinical entity and to carry out a review of the literature. This sporadic complication may manifest clinically as overdrainage in patients with implanted cerebrospinal fluid (CSF) shunt devices.

Case Report

A 40-year-old Caucasian male suffered severe traumatic brain injury (TBI) while participating in a runners meeting. Initial physical exam showed a Glasgow Coma Scale score of 10, mild right paresis and dysphasia. A cranial computed tomography (CT) scan showed acute subdural hematoma, subarachnoid hemorrhage and several cranial vault fractures. Endotracheal intubation was necessary once he was moved to the intensive care unit due to deterioration in the level of consciousness. A second CT scan showed bifrontal parenchymal contusions (total volume under 10 cc; Figure 1A) as well as brain swelling and an external ventricular drainage was inserted to monitor intracranial pressure (ICP). Initial values of 40 mmHg were managed according to the Brain Trauma Foundation guidelines.¹⁰ However, ICP values did not drop under 25 and a decompressive procedure was considered necessary eight hours after admission. Cerebral contusions were not significant due to their volume, so a bifrontal craniectomy was discarded. Then, a left-sided unilateral decompressive craniectomy was performed due to the presence of a left acute subdural hematoma which could be evacuated during the same procedure.

The patient maintained raised ICP (20 mmHg) during the first days following the surgery, whilst progression of the parenchymal contusions was evidenced in follow-up CT scans. The external ventricular drainage was closed thirteen days after TBI, once ICP was well controlled. A CT scan was performed 48h later (prior to catheter removal) and a right subdural hygroma with mass effect was then seen, with associated brain herniation through the bone defect. A CSF external brain tamponade was also present under the craniectomy site (Figure 1B). Follow-up magnetic resonance imaging was obtained to confirm progressive worsening of the CSF hygromas (Figures 1C, 1D) despite the clinical improvement of the patient, who presented cognitive disorder, right hemiparesis, and motor dysphasia. One month after initial TBI, an autologous cranioplasty was performed and the implant was fixed with four titanium miniplates and 4mm screws (Figure 1E). A cerebrospinal fluid circulation disorder was considered according to radiological findings which were progressively worsening (ventriculomegaly, right hygroma, interhemispheric hygroma, left hygroma). Since an external hydrocephalus was suspected, a programmable ventriculo-peritoneal shunt was also placed. The patient's neurological status progressively improved and he was discharged home two months after admission.

He followed a specific rehabilitation program with excellent initial results. However, his therapist subsequently noticed cognitive status decline 8 months later, consisting in a reduced ability to find vocabulary in phonemic and semantic verbal fluency tests, as well as disorganized language with lack of grammar resources. Besides that, regular non-orthostatic left supraorbital headache also appeared, but it was refractory to amitriptyline and carbamazepine. Follow-up CT scans showed progressive sinking of the bone flap, in spite of increasing the CSF shunt valve opening pressure (Figures 1F, 1G, 1H). Finally, he also showed three 5-minute-long episodes of motor dysphasia, each, in turn, associated with sudden and acute worsening of said headache.

He underwent bone flap removal and a patient-specific calcium phosphate-based implant was inserted. CSF shunt valve was initially programmed at 200 mmH₂O until the subdural collection was resolved. A lower opening pressure was then required for better outcome. The supraorbital headache and dysphasia episodes resolved, and the patient recovered his previous cognitive status. A final follow-up CT scan also evidenced radiological improvement (Figure 2).

Discussion

Decompressive craniectomy is employed when intracranial hypertension is uncontrollable with medical management. It has been shown that the effects of increased ICP can be mitigated by this technique. The initial studies looking at preventive decompressive craniectomy yielded dispiriting results, however, more recent series on the procedure have pointed towards improved outcomes compared to conservative management. The bone flap may be frozen or stored in the abdominal subcutaneous tissue until ICP normalizes and the patient can undergo subsequent cranioplasty. Even though it is a technically easy procedure, overall complication rate is not insignificant.¹

When it comes to postoperative infection, there are several risk factors associated, such as operation time exceeding 90 minutes, older patients and female sex, to name a few.² Although views on the matter abound, and the literature is fraught with debate, there appears to be a significant difference between early and late cranioplasty when it comes to the incidence of post-operative subdural effusion, favoring early cranioplasty (taking the definition of early cranioplasty to be before 3 months after the bone was first removed). Furthermore, there is evidence that suggests that early cranioplasty permits dissection of tissue planes with less damage to the dura and brain parenchyma, the basis of this being that there is less scar tissue. Finally, when contrasted with late cranioplasty, early cranioplasty has other advantages, such as better neurological outcomes and a reduction of overall costs, secondary to a decreased length of stay.¹¹

A phenomenon known as external brain tamponade occurs when subgaleal fluid accumulates and exerts pressure on the brain parenchyma across the craniectomy defect. The factors giving rise to external brain tamponade haven't been fully elucidated. Proposed etiologic candidates such as ICP spikes with coughing, a 'ball-valve' effect across the dural repair, and a pressure gradient favoring subgaleal fluid accumulation have all been considered.¹² Even though CSF dynamics frequently restore following cranioplasty, the rate of hydrocephalus ranges from 8.7 to 25%.¹ In the case hereby reported a programmable ventriculoperitoneal shunt was inserted since by that time, clinical evaluation was difficult (the patient did not walk due to hemiparesis, he showed urinary incontinence from the beginning after catheter removal and motor dysphasia interfere in cognitive evaluation) and radiological findings were progressively worsening.

In addition to all the complications cited above, the aim of this article was to bring to attention a very rare complication that can arise after cranioplasty, that one must be cognisant of, and that is the symptomatic bone flap syndrome. This clinical entity is characterized by the appearance of neurological symptoms, (these symptoms vary in their nature, depending on the location of the craniectomy) that arise after a depressed bone flap exerts pressure on the brain parenchyma. These symptoms disappear after the pathophysiological problem is addressed. A review of the literature yields only seven cases of symptomatic bone flap (Table 1). All patients showed neurological deficit when diagnosed and most of them referred headache. The majority of these cases manifested in a delayed fashion, at least one year between the surgical procedure and the onset of symptoms. Only one case was symptomatic at two months.⁶

The case hereby described provides some novelty in the sense that it is the first to our knowledge that had a CSF shunt implanted and manifested as ventriculoperitoneal shunt overdrainage. This may have influenced in the relatively early appearance of the complication. Most patients with shunt overdrainage refer positional headache and it is of note that nearly half of cases showed positional symptoms related to bone flap sinking syndrome.^{7,9} However, the case hereby reported presented with atypical supraorbital headache. Besides that, bone flap sinking did not improve when opening pressure of the valve was raised.

In such cases, other factors must be considered too. Bone flap fixation technique must be reviewed, even though it was standard in the present case and postoperative CT scan showed correct position of the bone flap with regard to the cranial vault. Bone resorption has also been postulated as the main etiological factor of a sinking bone flap following cranioplasty in the few cases published to date. Moreover, ventriculo-peritoneal shunt has been identified as a risk factor of aseptic bone resorption.¹³ Even though it did not seem to be relevant in the case hereby reported according to CT scans, a patient-specific implant was designed in order to avoid the same complication if refixation of the own bone flap was achieved.

Multifactorial etiology of sinking bone flap syndrome must be considered, moreover when the patient bears a CSF shunt device. All patients showed significant improvement of the symptoms following flap refixation or replacement, so the benign course is the most probable outcome.

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TABLES

Table 1. Sunken bone flap cases reported in the literature

Author (year)	Duration from initial surgery to sunken flap	Onset symptoms	Treatment Outcome
Rengachary et al. ⁴ (1979)	1 year	Dizziness, gait disturbance, memory impairment Flap movement noticeable with diurnal postural variation	-Refixation + methylmethacrylate -Prompt resolution of symptoms
Anei et al. ⁵ (2010)	15 years	Contralateral hemiparesis	-Refixation -Symptoms improved
del Mar Carmona Abellán et al. ⁶ (2012)	2 months	Hemiparesis, speech disorder, depressed sensorium	-Bone refixation -Significant improvement
Di Rienzo et al. ⁷ (2013)	1 st patient - 1 year	Headache on getting up followed by confusion, difficulty in walking progressing to third nerve palsy and hemiplegia. Symptoms aggravated by erect position and relieved on lying down. Flap movement noticeable with postural variation	-Flap replaced by hydroxyapatite implant. -Complete recovery
	2 nd patient - 4 years	Fatigue, confusion, worsening of hemiparesis, diplopia and ptosis on assuming erect posture and hemiparesis with regression on lying down	-Flap replaced by hydroxyapatite implant. -Complete recovery
Reddy et al. ⁸ (2013)	6 years	Progressive hemiplegia	-Refixation
Krishnan et al. ⁹ (2018)	1 year	Headache, heaviness and weakness of contralateral limbs with change in posture. Flap movement noticeable with postural variation	-Bone refixation -Complete recovery
Present case (2022)	8 months	Reduced verbal fluency, motor dysphasia, 3 seizure episodes, supraorbital cephalalgia	-Flap replaced by calcium phosphate-based implant + CSF shunt reprogrammed -Significant improvement

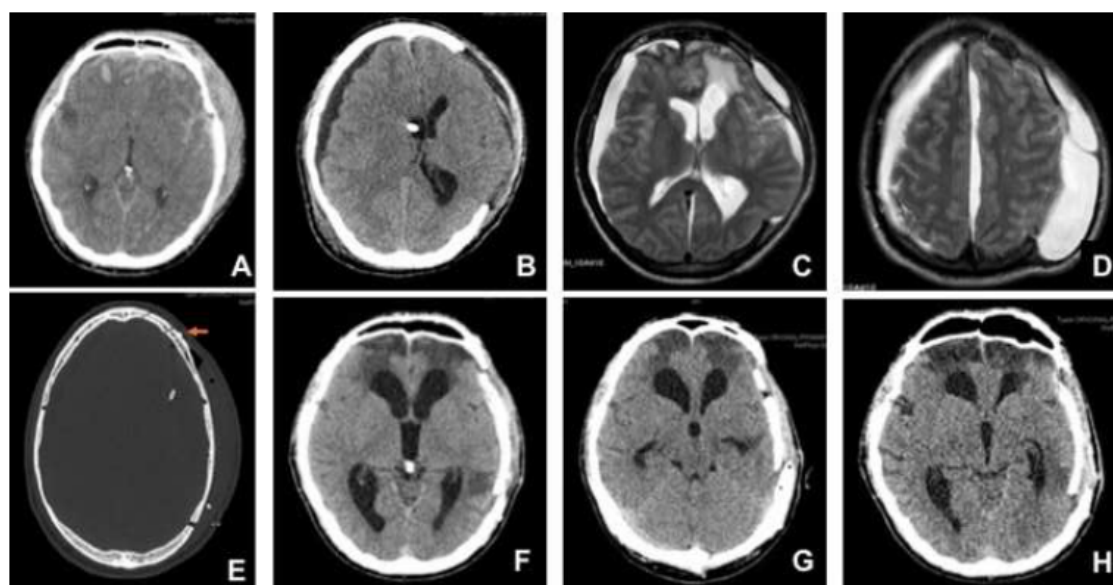


Figure 1. (A) CT scan performed five hours after admission showing small bifrontal contusions, subarachnoid hemorrhage, a small left subdural hematoma and diffuse edema. (B) Follow-up CT scan performed 2 weeks postoperatively and after ventricular catheter closure prior to its removal. A right subdural hygroma with mass effect is evidenced, and a left subcutaneous collection can be appreciated too. (C, D) Follow-up MRI performed 1 month postoperatively, showing right and interhemispheric hygromas, as well as increased size of the left CSF hygroma. (E) Postoperative CT scan performed 24 hours after bone flap replacement. 4-mm screws and miniplates were used to fix the implant (arrow) (F) Postoperative CT scan 2 months after bone flap replacement. (G) 6-month follow-up CT scan, showing slight sinking bone flap in absence of symptoms. (H) 1-year follow-up CT scan, showing significant sinking bone flap when symptoms were evident.



Figure 2. (A, B, C) Preoperative CT scan showing significant sinking of the bone flap. Shunt catheter can be appreciated. (D, E) Postoperative AP and lateral plain x-ray showing cranioplasty with good cosmetic result. (F) Postoperative CT scan showing the implant correctly lined up with the cranial vault, as well as reduced ventricular size when comparing to preoperative image.