

Case Report

Never say never again: A bone graft infection due to a hornet sting, thirty-nine years after cranioplasty

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Received: 16 February 17 Accepted: 06 June 17 Published: 10 August 17

Abstract

Background: Cranioplasty (CP) is a widespread surgical procedure aimed to restore skull integrity and physiological cerebral hemodynamics, to improve neurological functions and to protect the underlying brain after a life-saving decompressive craniectomy (DC). Nevertheless, CP is still burdened by surgical complications, among which early or late graft infections are the most common outcome-threatening ones.

Case Description: We report the case of 48-year-old man admitted to our neurosurgical unit because of a painful right frontal swelling and 1-week purulent discharge from a cutaneous fistula. He had been undergone frontal CP because of severe traumatic brain injury (TBI) when he was 9-year-old. Since then, his medical history has been being unremarkable without any surgical or infective complication of the graft for 39 years, until he was accidentally stung by a hornet in the frontal region. After the CT scan and laboratory findings had evidenced a probable infection of the graft, the patient was treated by vancomycin and cefepime before he underwent surgical revision of its former CP, with the removal of the graft and the debridement of the surgical field. Subsequent bacteriological tests revealed *Staphylococcus aureus* as causal agent of that infection.

Conclusion: This case illustrates an anecdotal example of very late CP infection, due to an unpredictable accident. Due to lack of consensus on risk factors and on conservative or surgical strategy in case of graft infection, we aimed to share our surgical experience.

Key Words: Cranioplasty, late infection management, risk factors, surgical complications

Access this article online

Website:

www.surgicalneurologyint.com

DOI:

10.4103/sni.sni_68_17

Quick Response Code:

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How to cite this article: Maugeri R, Giammalva RG, Graziano F, Basile L, Gulì C, Giugno A, et al. Never say never again: A bone graft infection due to a hornet sting, thirty-nine years after cranioplasty. *Surg Neurol Int* 2017;8:189.

<http://surgicalneurologyint.com/Never-say-never-again:-A-bone-graft-infection-due-to-a-hornet-sting,-thirty-nine-years-after-cranioplasty/>

INTRODUCTION

Decompressive craniectomy (DC) has become a widespread procedure for treating life-threatening conditions that lead to a higher intracranial pressure (ICP).^[3] DC, either “classic” or “internal” (sinuses cranialization in megasinus subdural empyema patients) is performed in the management of neurological emergencies, such as hemispheric stroke, dural sinus thrombosis, subarachnoid hemorrhage, or peri-cephalic and brain infections.^[15,28,29,32] Otherwise to be noted that craniectomy versus burr hole in subdural empyema is still matter of debate.^[21]

Nevertheless, severe traumatic brain injury (TBI) still remains the most common indication for DC in many clinical series.^[3,14,25] Even if DC is a common neurosurgical procedure that might gain the control of resistant intracranial hypertension, it is often related to many side effects, such as the “trephined syndrome” and mostly the need to repair the bone defect. Thus, once resolved the cerebral swelling and restored the ICP, a cranioplasty (CP) is required in surviving patient in order to restore the physiological cerebral hemodynamics,^[14,15] to protect the brain and to reconstruct bone defect.^[3,11-13] Despite the clear advantages on clinical and neurological conditions of the patient due to CP, this procedure still entail a high morbidity and overall risk of adverse events ranging from 10.9% to 36.5%.^[14,15] Among those, surgical site infection is one of the most common complications described in literature.^[25] Their rate ranges from 5.8% to 16%,^[15,16,33] and in most cases it implicates additional surgical procedures and the revision of the former CP.^[14,26] Although their high incidence, the majority of surgical site infections is seen between 3 months and 10 months after CP,^[2,5,20] instead late infections are extremely rare.^[8,22] We report an exceptional case of graft infection seen 39 years after primary CP in a male patient with an unremarkable clinical history and uneventful graft permanence.

CASE DESCRIPTION

A 48-year-old male was admitted to our neurosurgical unit because of a frontal swelling with purulent discharge from a cutaneous fistula. In his clinical history, he reported a previous frontal TBI when he was 9-year-old. For this reason, he had undergone a frontal craniectomy with subsequent unspecified CP. Since that, his clinical history has been unremarkable for 39 years, without any surgical or infective complications related to the cranial surgical procedure. A week before his admission he had been stung by a hornet while he was working in the countryside, with a progressive swelling of frontal soft tissues, temperature raising, and local pain with inflammatory state. During that week, a progressive

purulent discharge has onset from the site of the sting, so he referred to the emergency department. At the admission, he was awake and aware [Glasgow Coma Scale 15 (GSC 15)], his temperature was 37.8°C and his neurological examination was negative. Initial laboratory studies revealed a pathological increased value of white blood cell (WBC) count ($22 \times 10^9/L$ with 82% neutrophils, 14% lymphocytes, and 7% monocytes) and of serum C-reactive protein level (57 mg/dL with a normal range 0.08–1.5 mg/dL). A head CT scan revealed an irregular subcutaneous fluid collection, adherent to the graft’s anterior face, a peripheral soft tissues edema, and a frontal hypodensity in brain parenchyma contiguous to the graft. The administration of contrast medium revealed a moderate peripheral enhancement of the collection. Because of the radiological evidence of the graft infection with subcutaneous abscess, the patient was then transferred to our neurosurgical unit. A swab test of the purulent discharge from the cutaneous fistula was performed, and, after a multidisciplinary consult, a polychemotherapy was started (1 g of vancomycin and 2 g of cefepime every 12 h). Thirty-six hours after his admission, the patient underwent surgical toilet and removal of the graft. This appeared as a porous acrylic graft, with several fibrotic bands firmly tied with the skin and the underlying synthetic dural substitute. All the samples were harvested for microbiological exam, while the surgical site was washed with iodine solution, peroxide, isotonic saline, and rifampicine. At the end, a spongy layer of dural substitute with two overlaying patch of fibrin sealant (Tachosil©) and some fibrin glue (Vivostat©) was then used to ensure dural seal and to contribute in protecting the brain parenchyma under the bone defect [Figure 1a-d].^[9,10,18,19]

Nor titanium mesh or other allograft was applied on the former craniotomy in order to prevent the rejection,



Figure 1: (a) Fronto-orbital swelling with purulent discharging fistula. (b and c) Intraoperative debridement and exposition of infected bone graft. (d) Closure with the apposition of dural substitute, Tachosil© and Vivostat©

waiting for the resolution of the infection in analogy with other surgical procedures.^[30]

Further bacteriological tests revealed *Staphylococcus aureus* as causal agent of that graft infection. Due to this, the patient was discharged 7 days after surgical procedure under an antibiotic therapy with linezolid and ceftazidime and he is still in follow-up for further CP after the infection resolution and inflammatory markers normalization.

DISCUSSION

CP is a common surgical procedure requisite to restore skull integrity in such cases where it has been compromised. Moreover, it has been demonstrated that CP can be effective in preventing seizures or cerebral atrophy after a DC so avoiding the “trephined syndrome,”^[27] in improving neurological functions due to the re-establishment of normal cerebral hemodynamics, and in restoring the skull shape.^[14,15] Although, necessary for the above-mentioned reasons, CP is a surgical procedure as common as dangerous, carrying a significant rate of complications.^[3,4,6,7,17] It has been estimated that this rate ranges from 15% to 36.5% and further surgical procedures may be necessary up to 76% of the cases.^[14]

CP failure may be attributable to autologous bone flap resorption (when used) or mostly to graft infections,^[3,11] which negatively conditions postoperative prognosis, requiring antibiotic therapy, graft removal, and further surgical procedures. Graft infection rate ranges from 5.8% to 16%,^[15,16,33] and in most cases, it implicates additional surgical procedures with the revision of the former CP.^[14,26]

Even if CP is a relatively old procedure, there still are no solid evidences on the risk factors for graft infections. The first and most controversial issue is the time interval between DC and CP. Many clinical series state that early CP ensures better outcomes, whereas many others advocate late surgery to prevent graft infections, avoiding the risk of performing surgical procedure on a contaminated wound.^[1,3,14,15,23,27] Other risk factors described in literature take account of the use of allograft instead of autologous bone flap, the size of skull defect,^[14,15,25,31,33] and also the etiology of primary DC; Kim *et al.* reported a higher rate of graft infections in those case of CP performed after DC in severe TBI than in non-TBI ones.^[14] Other possible predictors of CP failure include: Motor deficit, Glasgow Outcome Score (GOS) <4, anemia, recent systemic infection,^[14,24] previous surgical site infection, or wound dehiscence.^[25] Identifying those risk factors involved in CP procedure in order to reduce the rate of infection remain a challenge for the surgeon; but unfortunately, despite the good clinical and surgical practice not every

risk could be avoided and some factors may be even unpredictable, especially forward in time. To the best of our knowledge, the present case of 48-year-old male patient represents the most tardive CP infection reported in literature. Formerly, Gürbüz *et al.* reported a case of purulent discharge from the surgical site of a patient 22 years after he had undergone DC and subsequent CP because of temporal TBI.^[8] CP infections usually onset within 10 months after the surgical procedure, but the unpredictable happens: The long-term outcome of an accurate surgical procedure is sometimes irremediably influenced by the fate. In particular, in our reported case the correct engraftment and the 39-years long uneventful permanence of the synthetic CP had been foiled by an accidental hornet sting, which led to the graft infection. This complication might be related to the introduction of a foreign body, such as the hornet stinger and delivering bacteria into its surface.^[27] The most of CP infections has been attributed to the colonization by the bacterial skin flora, in particular, *Propionibacteria*, *Klebsiella*, *Enterobacter*, *Pseudomonas Aeruginosa* also if *Staphylococcus aureus* results as the most common causal agent with a prevalence of 75%.^[3,11,15,25,34] The execution of a swab test of the purulent discharge and the harvest of some intraoperative specimens are strictly recommended for further bacteriological studies, even if in a small percentage of the whole cases they can be inconclusive, showing no bacterial overgrowth.^[26] In our case, the bacterial culture was positive for *Staphylococcus aureus*, both on the swab test and on the intraoperative specimens, in accordance with the literature data.^[15] Due to variety of bacterial agents in such this kind of infection, our patient begun a medical therapy with vancomycin and cefepime before he underwent surgical operation and microbiological exam of intraoperative specimens. There is still not a consensus on the timing and the surgical strategy in treating infected CP.^[26] In this challenging scenario, as it is still not possible to find either definite risk factors of earlier CP infections or reliable predictors of the later ones, which often are even unpredictable, a graft infection has to be considered as a diagnostic option, in case of a congruity between patient's clinical presentation, laboratory findings, and neuroimaging, also after several decades.

CONCLUSION

CP is still necessary in surviving patients after life-saving DC. Despite its widespread employment and the wide variety of techniques and material, postoperative complications still threat patients' clinical outcome. Among these, graft infections are the most common, even many years after the CP have been performed, and they are often due to skin bacterial flora. Nowadays, there is still no consensus on reliable risk factors for early graft infections, moreover late ones are often unpredictable

and totally accidental, as we reported. Due to lack of similar cases of very late CP infections in literature, we aimed to share our surgical experience about the most tardive one that we treated, highlighting its extraordinary fortuity.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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