Received: 9 May 2024 | Revised: 4 June 2024 | Accepted: 13 June 2024

DOI: 10.1002/ccr3.9115

CASE REPORT

Clinical Case Reports Open Access WILEY

An unusual presentation of orbital encephalocele following a self-inflicted gunshot wound injury: A case report and literature review

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Key Clinical Message

While orbital encephaloceles secondary to orbital roof fractures, in the setting of gunshot wound injuries, are rare, it is important to discuss diagnosis, treatment, and outcomes. This comprehensive manuscript aims to accomplish these objectives.

KEYWORDS

case report, gunshot wound injury, orbital encephalocele, suicide, traumatic encephalocele

1 | INTRODUCTION

Self-inflicted GSWs are a common cause of death in people committing suicide.[1] Most frequently, the entry wound is in contact range and on the temple.[2,3] Most patients who present with facial self-inflicted gunshot wounds—especially in the young and non-penetrative brain injury cases—will survive, although readmission rates after self-inflicted gunshot wounds are frequent.[4,5] Often, early primary reconstruction can be successful for patients, particularly when the entry point of the bullet is in the upper and midface areas.[6] We report a case of self-inflicted GSW injury in an adult leading to an uncommon complication. This case report has been reported in line with the SCARE criteria.[21]

2 | METHODS

A scoping review of PubMed and Google Scholar was performed in May 2023 using the following search terms: orbital encephalocele, gunshot wound injury, traumatic encephalocele, self-inflicted, penetrating injury. We reviewed the literature as far back as 1970, and it was apparent that the first reported case was in 2015. We found four cases from the literature and then reviewed the references from each manuscript, which yielded no additional cases. To the best of our knowledge, these are the only reported cases of traumatic orbital encephaloceles secondary to gunshot wound injuries reported in the literature since 1970.

3 | CASE PRESENTATION

3.1 | Patient information

A 39-year-old man was transferred from an outside hospital (OSH) to our medical center for concern of abscess of the left orbit. He had suffered a self-inflicted gunshot wound to the face, occurring 11 days prior with fractures of his hard palate, left maxillary sinus, nasal bones, left orbital walls, frontal sinus, frontal bone, and a frontal intracranial hematoma all identified on initial imaging. There was no indication of orbital encephalocele at the time of initial imaging. Hospital course at OSH was complicated by hyponatremia, which was thought to be secondary to diabetes insipidus, and resolved after treatment with Desmopressin acetate and hypertonic saline. General surgery was consulted regarding concerns for dysphagia and attempted to place a PEG tube, however, attempts were unsuccessful due to the inability to transilluminate during the procedure. A fiberoptic NG tube was placed instead for nutrition. Subspecialty evaluation was completed, but no interventions were performed for any fracture. The trajectory of the single bullet lacerated the left chin and penetrated the tongue, hard palate, left maxillary sinus, orbit, frontal lobe, and exited through the frontal bone and scalp (Figure 1). The patient has a history of schizoaffective disorder and was otherwise stable, nonverbal, and review of systems was unable to be obtained given the patient's mental state. He was initially intubated and then extubated, off sedation and on prophylactic anticoagulant. Over the 11 days, patient developed worsening orbital exam with exophthalmos, globe displacement and absent vision. Patient developed fever and an elevated white count, therefore CT scan was performed and read as concern for orbital abscess. Patient was transferred to our institution and there was concern for infected encephalocele both within the orbit and through the frontal bone. MRI was performed for confirmation. Psychiatry was consulted for management and stabilization of patients' underlying mental health disorder.

3.2 | Clinical findings

The visual acuity was unable to be obtained due to patient's inability to cooperate with exam, however his left eye exhibited obvious proptosis with inferolateral orbital displacement and a fixed, dilated pupil in addition to vitreous hemorrhage. A fluctuant fluid collection was also present at the frontal bone exit site. An evolving left frontal contusion was evident on a repeat CT of his face compared to his initial imaging as well as a traumatic subarachnoid hemorrhage. A CT of his face with contrast showed an enhancing collection within the left orbit and associated proptosis of the left eye (Figure 2A,B). He also had a comminuted left orbital roof, medial wall, and floor fractures, anterior skull base fractures, left frontal sinus fracture that did not involve the posterior table, and comminuted left frontal bone fracture. An MRI with contrast was performed revealing transcalvarial herniation of left anterior frontal lobe into the orbit and maxillary sinus with enhancement as well as herniation anteriorly and superiorly out of the frontal bone exit site (Figure 3). The patient was taken to the operating room the next day for multidisciplinary intervention with neurosurgery, otolaryngology, and ophthalmology.

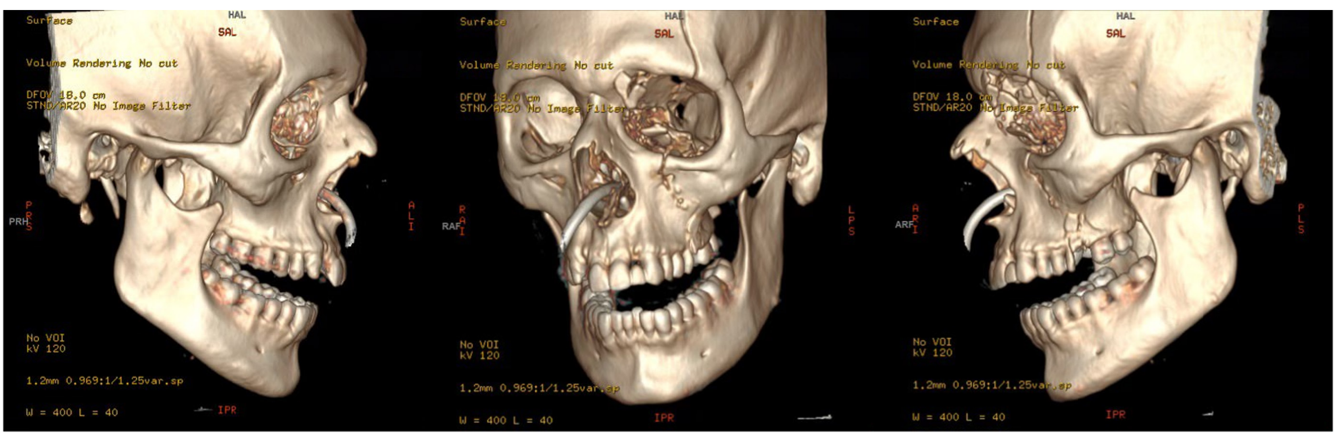

FIGURE 1 3-D reconstruction of his outside CT scan showing gunshot wound trajectory leading to several facial fractures.

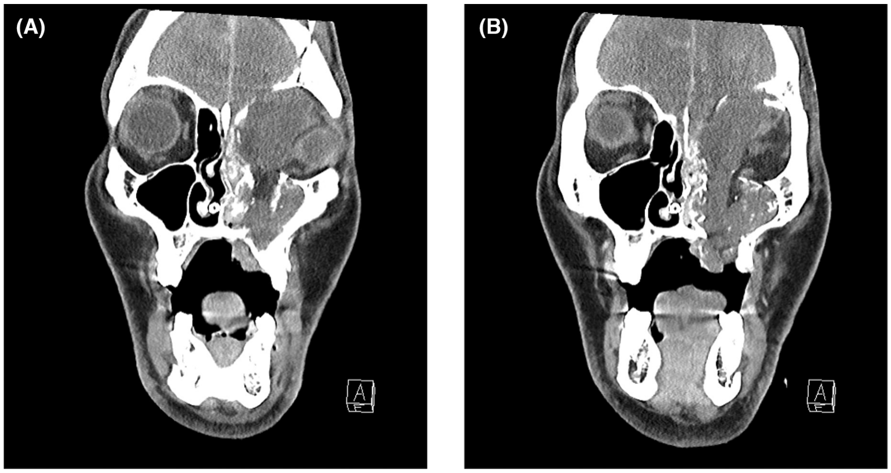

FIGURE 2 (A) CT of his face showing complex left orbital wall, midface, and palate fractures, an intraorbital collection of the left eye with proptosis with (B) extension into the maxillary sinus and palate.

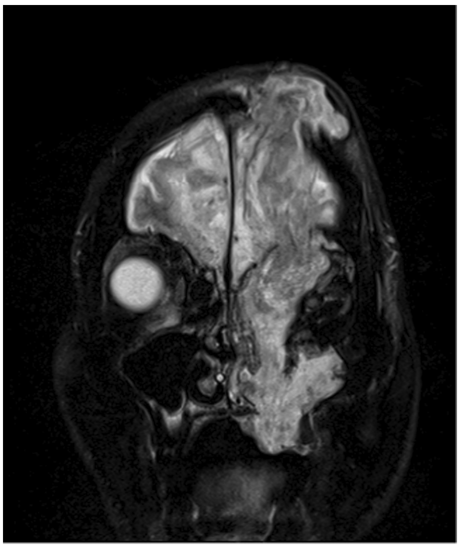

FIGURE 3 MRI showing transcalvarial herniation of left anterior frontal lobe into the orbit and maxillary sinus as well as herniation out of the frontal skull defect.

3.3 | Interventions

Intraoperatively, he was found to have a portion of the anterior frontal lobe herniating into the orbit and the superior maxillary sinus as well as superiorly through the frontal fracture underlying the closed left frontal scalp wound with subdural empyema and necrotic tissue. All apparent prolapsed and necrotic tissue was surgical debrided. Orbitotomy was achieved using the transcranial approach, which involved developing the subperiosteal plane along the superior and lateral orbital rim. A Freer elevator was used to dissect through the arcus marginalis. After preserving the supraorbital neurovascular bundle, two malleables were used to develop the subperiosteal plane from the superior orbital wall down toward the superior orbital fissure anteriorly. Simultaneously, the cranial side was explored in a dual fashion until the defect was entirely identified. All apparent prolapsed tissue was removed. Once the excision was completed, the eye was examined, and the globe was found to be intact with a significant reduction in axial proptosis. Additionally, the globe was found to move freely within the orbit. Intraoperatively, ballottement of the eye showed complete fracture and loss of the medial, superior and inferior walls with a visible transmission of pressure. Through the palate and maxillary sinus, herniated brain was noted in the sinus. The orbital floor was examined, which confirmed a large floor and medial wall defect. The dural defect was repaired and the orbit was decompressed, which allowed the eye to rest in a more relaxed position, and decreased the chance of recurrent infection. This also helped to salvage any remaining visual function by reducing the potential compartment syndrome. We then performed a left side orbital roof and skull base reconstruction using split-thickness calvarial bone grafts to separate the intracranial and orbital compartments (Figure 4). Necrotic palate and sinus tissue were debrided. Long-term antibiotics were provided.

3.4 | Follow-up and outcomes

The patient was stabilized and kept inpatient for nearly 1 month before being discharged to an inpatient rehabilitation facility. Psychology was intimately involved to restart and optimize the patient's mental health medications and confirm no additional suicidal ideations or intents. After the rehab facility, the patient was discharged to a group home. Vision was not recovered in the left eye. He was planned for delayed reconstruction of his palatal defect, though this ultimately healed with only a small asymptomatic oroantral fistula and patient was not interested in further repair of his fistula or periocular procedures to optimize his facial outcomes.

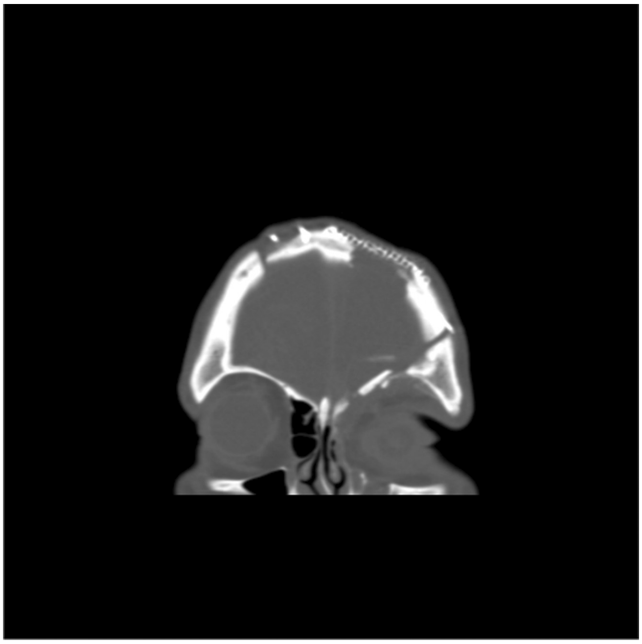

FIGURE 4 Postoperative CT scan showing split-thickness calvarial bone grafts to reconstruct the orbital roof and walls with reduction of the encephalocele.

4 | DISCUSSION

Suicide rates in the United States increased 37% between the years 2000 and 2018, according to the CDC.[17] Though firearms are the most lethal method for people who attempt suicide, 15% of attempts with firearms do not result in death.[18] Of these failed suicide attempts, skull fractures are common. Since the temples are most frequently the associated area of impact, the orbital roofs are at high risk for traumatic injury. Another possible cause of nonfatal injuries is assault with firearms.[19] During 2006–2014, close to 50% of patients presenting to the ED in the U.S. with firearm-related injuries were due to assault.[20] With attempted suicide and assault with firearms becoming more common, it is particularly important to understand complications of GSW injuries and how they are managed. Our study aimed to characterize orbital roof fractures secondary to GSW injuries that resulted in orbital encephaloceles since 1970, as summarized in Table 1. Although numbers are low, we found that gunshot injuries were due to assault in 50% of reported cases, with self-inflicted and accidental comprising the remaining cases. The literature was reviewed as far back as 1970 to include as many cases as possible, allowing over 50 years of reported data.

Orbital roof fractures are usually asymptomatic and frequently occur secondary to facial trauma from motor vehicle collisions (MVC).[14] These fractures can also present in the setting of penetrating injuries, such as gunshot wounds. Orbital encephaloceles, or herniation of the brain and meninges into the orbit, are rare complications of orbital roof fractures that have only been described in a limited number of cases.[7–10,16] In a recent review of orbital roof fractures complicated by meningoencephaloceles, 9 out of 20 reported cases since the year 2000 had MVC as the mechanism of injury.[11] Including our patient, there have been five reported cases since 1970 of GSW injuries resulting in traumatic orbital encephalocele. An association between mechanism of injury and the likelihood of developing a traumatic orbital encephalocele secondary to orbital roof fractures may exist, however, further investigation is needed.

Orbital encephaloceles are more common in younger populations than in adults. Children under the age of 7 years have a higher incidence of orbital roof fractures because traumatic force is transmitted to the orbital roof rather than the frontal sinus. Following 7 years of age, pneumatization of the frontal sinuses is complete, and trauma on the superior orbital rim can be dissipated by the frontal sinus.[25] This creates a diagnostic challenge as many practitioners are not used to diagnosing orbital encephaloceles in adults, and therefore may be less likely to suspect or include them in their differential diagnoses.

Patients with facial trauma routinely receive a noncontrast CT of the head. CT is particularly useful in identifying the bony defect in patients with orbital roof fractures.[12] Identifying post-traumatic orbital encephaloceles on CT can be more complex because concurrent orbital or intracranial hematomas may present similarly to herniated brain tissue.[7] Similarly, diagnosis can be complicated by the presence of periorbital edema and ecchymosis.[23] Direct X-ray of the skull may be performed, but usually is only helpful in identifying large depressed fractures.[24] Although all patients with orbital encephaloceles in Table 1 were diagnosed with CT, our case involved an orbital encephalocele that was missed on initial CT. Upon further work up involving MRI of the brain and orbits with contrast due to a high index of suspicion, the diagnosis was correctly made. In a review of six patients with traumatic orbital encephalocele, CT scans successfully identified the presence of orbital encephalocele in only four out of the six patients. However, when MRI was used for diagnosis, all three patients who underwent this imaging modality had the orbital encephalocele accurately diagnosed.[16] Further, MRI is the preferred imaging modality for visualizing orbital encephalocele and its proximity to vascular structures, as well as for detailed tissue characterization.[13] These findings demonstrate how additional imaging with MRI is an important next step if the CT is unrevealing, especially in the presence of physical symptoms, such as exophthalmos.

Uncomplicated orbital roof fractures do not typically require surgery. Indications for surgical repair of an orbital roof fracture include exophthalmos, loss of

TABLE 1 Summary of traumatic orbital encephaloceles in the setting of GSW injuries published since year 1970.

Authors	Year	Age/ Gender	Mechanism	Diagnosis	Presentation	Imaging	Treatment	Outcomes
Pawar et al[8]	2015	26/M	Suicide attempt	Left orbital floor fracture, left superior orbital rim fracture, left dural injury with frontal lobe herniation.	-Facial swelling -Periorbital edema -Ecchymosis	CT demonstrated left orbital injury, comminuted fractures of left orbital roof, superior and inferior orbital rims, and orbital floor. Left frontal lobe injury, left frontal sinus fractures, ZMC fracture.	-Left frontal craniotomy. -Split thickness calvarial bone graft.	-Subsequent procedure to remove displaced bone fragment and place a dermal fat graft for additional volume augmentation. - No focal neurological motor deficits. - Numbness over V1 of the trigeminal nerve was noted.
Wei et al[7]	2016	3/M	Accidental	Right orbital encephalocele, intracranial bleeding, hydrocephalus, large skull base bony defects.	-Afferent pupillary defect -Proptosis -Exophthalmos	CT demonstrated extensive skull defect extending from right superior orbit to occiput.	-Right craniectomy. -Hygroma evacuation. -Duraplasty -Ventriculoperitoneal shunt placement.	-Subsequent frontoparietal reconstructive cranioplasty with EVD placement. -At 5 month follow up, visual acuity in the right eye was 20/80. Exotropia, amblyopia, and ptosis. -Development of seizures required EVD revision.
Czyz et al[9]	2021	20/M	Assault	Bilateral orbital floor and roof fractures, left sided herniation of frontal lobe tissue through orbital roof defect and CSF accumulation.	-Edema -Ecchymosis -Proptosis	CT demonstrated displaced fractures of middle cranial fossa, sphenoid of left greater wing, and bilateral frontal sinuses with intracranial and orbital extension of bony fragments and brain tissue.	-Canthal cut down. -Bifrontal craniotomy with exoneration of the frontal sinuses.	-At 3 month follow up, there was resolution of most previous abnormal findings. An afferent pupillary defect was noted OS. -Patient had trace left sided periorbital ecchymosis, including globe proptosis.
Callahan et al[10]	2021	29/F	Assault	Orbital roof fracture with herniation of dura mater within orbit.	-Exophthalmos -Proptosis -Limited supraduction -Periorbital edema	CT demonstrated a blow-in fracture of orbital roof with loss of orbital volume, herniation of the dura mater within orbit, and intact supraorbital rim.	-Upper blepharoplasty. -Durepair Regeneration Matrix. -Titanium mesh adapted and secured with screws.	-At 2 month follow up, vision and extraocular movements were normal. -Good adaptation of the plate and restoration of orbital volume.

visual acuity, and other signs of intraorbital injury or CSF leak.[15] Another indication for emergent surgical repair is compression of the optic nerve by bony spicule on imaging.[26] Importantly, surgery can be delayed up to 7–10 days to allow time for swelling to be reduced.[27] There are different treatment options for the repair including split-thickness calvarial bone grafts and titanium mesh, with both summarized in Table 1. Further, there are different ways to access the orbital roofs. The transconjunctival approach involves making an upper blepharoplasty incision along the patient's eyelid, which was utilized for the 29-year-old female in Table 1. The transcranial approach involves performing a craniotomy, which was used for all other patients. The type of approach utilized depends on the specific requirements of the individual case and outcomes appear to be similar between both approaches. Frontal bone fractures present commonly in craniomaxillofacial trauma patients and our patient had a left frontal sinus fracture that did not involve the posterior table. When the frontal sinus is fractured, options for treatment are dependent on the involvement of the anterior and posterior walls, as well as involvement of the frontonasal duct. Conservative treatment can be used when the fractures are limited to the anterior wall of the frontal sinus and the frontonasal duct is not involved. Displaced fractures require surgical reduction and osteosynthesis.[22]

Patients typically experience good recovery following surgical repair, with three out of four cases in Table 1 demonstrating full return of vision in the affected eye. The patient whose visual acuity did not normalize was only 3 years old, which suggests that age could have been a contributing factor. The patient from our institution also did not recover vision in the affected eye, which may have been related to the delay in diagnosis. Three of the four cases presented in Table 1 had preservation of extrinsic ocular motility. The 20-year-old male demonstrated a persistent afferent pupillary defect (APD). While short-term outcomes have been reported, the literature is limited regarding long-term outcomes of surgical repair of post-traumatic orbital encephaloceles, particularly in adult patients. One case report highlights the long-term effects of surgical reconstruction in a 5-year-old female with post-traumatic orbital encephalocele due to a fall accident. Results demonstrated 8 years after surgical repair of the orbital roof with titanium mesh, the child developed proptosis, ptosis, and decreased visual acuity in the affected eye, which the researchers hypothesize may have been caused by chronic irritation of the bony cortex by the titanium mesh.[25] While traumatic orbital encephaloceles secondary to orbital roof fractures are rare, there exists a need for greater reporting of long-term effects following surgical repair.

Psychological assessments are important, especially when dealing with patients who have access to firearms. Suicide risk assessments—which are commonly performed by psychiatrists, family medicine physicians, and other primary care physicians—encompass the evaluation of the patient's mental health, risk factors, and underlying motivations. A key part of the risk assessment involves asking patients about ownership and access to firearms. All physicians, including surgeons, should be familiar with the suicide risk assessment so that potential suicide have a higher chance of being avoided or prevented. Intervention strategies include psychiatry consults, particularly for patients that are admitted to the hospital after self-inflicted injuries. Early identification can alert practitioners to exacerbating factors and guide the implementation of tailored interventions. A holistic approach may encompass crisis intervention, psychiatric consultations, and psycho-therapeutic support, with the aim of mitigating risk of recurrent self-harm, facilitating emotional recovery, and enhancing overall patient well-being. Suicide hotlines also provide a societal line of defense against these destructive impulses, for example, the 988 Suicide and Crisis Lifeline provides 24/7 support for United States citizens and has a variety of language options.

In our case, we faced the challenge of dealing with a self-inflicted GSW patient presenting in a delayed fashion. The orbital and intracranial hematoma, lack of patient responsiveness, lack of adequate imaging, and the initial nonvisualized herniated brain tissue suggested the presence of an abscess, particularly in the context of fever and elevated white blood cell count. The referring hospital noted progressive proptosis and ocular deviation over his admission without additional evaluation, ophthalmologic evaluation, or additional specialist examination. Early evaluation with MRI in patients with similar presentations may be beneficial for improving patient outcomes. Our study highlights the importance of GSW as the mechanism of injury for orbital roof fractures causing traumatic orbital encephaloceles. Future studies should compare the mechanism of injury and the likelihood of orbital encephalocele secondary to orbital roof fractures.

5 | CONCLUSION

This case demonstrates the complexity of accurately diagnosing post-traumatic orbital encephalocele on CT and the need for further imaging such as an MRI of the orbits and brain for definitive diagnosis. This is a unique complication of self-inflicted GSW injury, with only four cases reported in the literature since 1970, which should be considered by physicians to potentially lead to more favorable outcomes for patients with early intervention.

Additionally, it is important to approach a complex case like this with a multidisciplinary team involving neurosurgeons, otolaryngologists, oral and maxillofacial surgeons, ophthalmologists, and anesthesiologists.

AUTHOR CONTRIBUTIONS

Marc Levine: Conceptualization; writing – original draft. **Olivier F. Noel:** Conceptualization; writing – original draft. **Shivam Patel:** Writing – original draft. **Haejoe Park:** Writing – original draft. **Christopher L. Weller:** Writing – original draft. **Jessyka G. Lighthall:** Conceptualization; supervision; writing – original draft; writing – review and editing.

FUNDING INFORMATION

None declared.

CONFLICT OF INTEREST STATEMENT

No conflict of interest was declared by the authors.

DATA AVAILABILITY STATEMENT

Data sharing not applicable to this article as no datasets were generated or analysed during the current study.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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How to cite this article: Levine M, Noel OF, Patel S, Park H, Weller CL, Lighthall JG. An unusual presentation of orbital encephalocele following a self-inflicted gunshot wound injury: A case report and literature review. *Clin Case Rep*. 2024;12:e9115. doi:10.1002/ccr3.9115