

## Combined supra-transorbital keyhole approach for treatment of delayed intraorbital encephalocele: A minimally invasive approach for an unusual complication of decompressive craniectomy

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### Abstract

**Background:** Intraorbital encephalocele is a rare entity characterized by the herniation of cerebral tissue inside the orbital cavity through a defect of the orbital roof. In patients who have experienced head trauma, intraorbital encephalocele is usually secondary to orbital roof fracture.

**Case Description:** We describe here a case of a patient who presented an intraorbital encephalocele 2 years after severe traumatic brain injury, treated by decompressive craniectomy and subsequent autologous cranioplasty, without any evidence of orbital roof fracture. The encephalocele removal and the subsequent orbital roof reconstruction were performed by using a modification of the supraorbital keyhole approach, in which we combine an orbital osteotomy with a supraorbital minicraniotomy to facilitate view and access to both the anterior cranial fossa and orbital compartment and to preserve the already osseointegrated autologous cranioplasty.

**Conclusions:** The peculiarities of this case are the orbital encephalocele without an orbital roof traumatic fracture, and the combined minimally invasive approach used to fix both the encephalocele and the orbital roof defect. Delayed intraorbital encephalocele is probably a complication related to an unintentional opening of the orbit during decompressive craniectomy through which the brain herniated following the restoration of physiological intracranial pressure gradients after the bone flap repositioning. The reconstruction of the orbital roof was performed by using a combined supra-transorbital minimally invasive approach aiming at achieving adequate surgical exposure while preserving the autologous cranioplasty, already osteointegrated. To the best of our knowledge, this approach has not been previously used to address intraorbital encephalocele.

**Key Words:** Decompressive craniectomy, intraorbital encephalocele, minimally invasive surgery, piezosurgery

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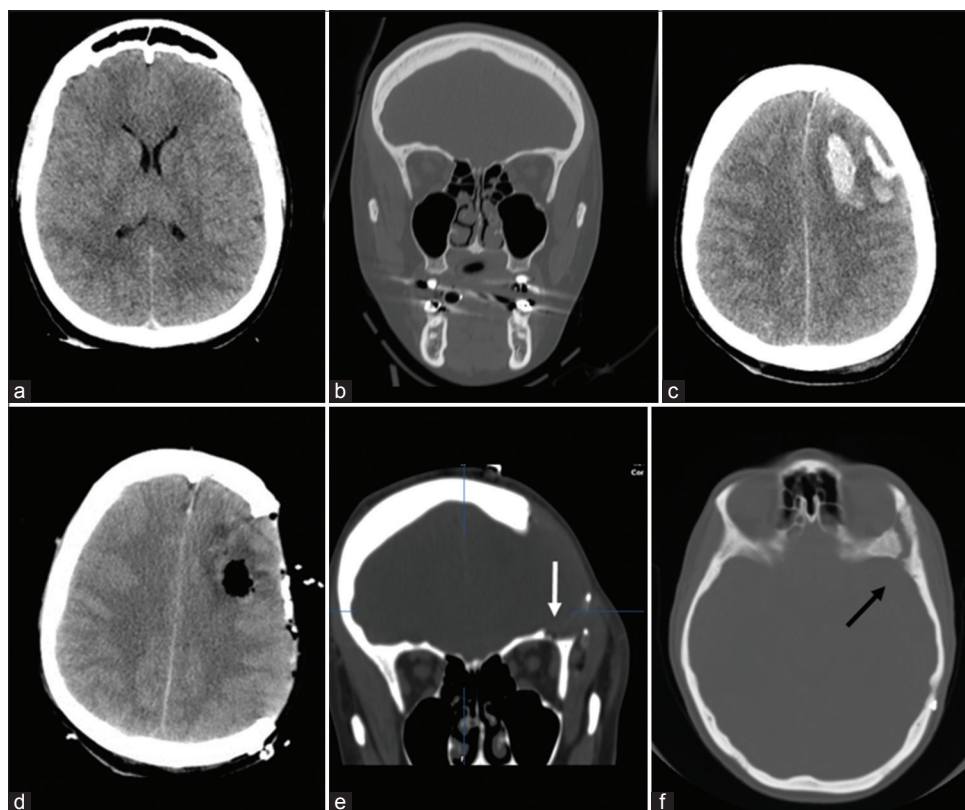
## INTRODUCTION

Intraorbital encephalocele is a rare entity characterized by the herniation of cerebral tissue inside the orbital cavity through a defect of the orbital roof.<sup>[1,3,11]</sup> The most common causes reported in literature are trauma, skull base malformations, and tumors.<sup>[3,4]</sup> In patients who have experienced head trauma, intraorbital encephalocele is usually secondary to orbital roof fracture.<sup>[1-4,8-12]</sup> Early recognition and treatment are very important since the raised intraorbital pressure may irreversibly damage the optic nerve and causes pulsating exophthalmos.<sup>[8]</sup> Excision of the herniated brain tissue, duraplasty and reconstruction of the orbital roof are the key steps of surgery.<sup>[10-12]</sup> In almost all the reported cases, the fronto-basal approach with wide craniotomy is the preferred approach.<sup>[1-4,8-12]</sup> We describe here a case of a patient who presented an intraorbital encephalocele 2 years after severe traumatic brain injury, treated by decompressive craniectomy and subsequent autologous cranioplasty, without any evidence of orbital roof fracture. The encephalocele removal and the subsequent orbital roof reconstruction were performed by using a modification of the supraorbital keyhole

approach, in which we combine an orbital osteotomy with a supraorbital minicraniotomy to facilitate view and access to both the anterior cranial fossa and orbital compartment. The peculiarities of this case are the orbital encephalocele without an orbital roof traumatic fracture, and the combined minimally invasive approach used to fix both the encephalocele and the orbital roof defect.

## CASE REPORT

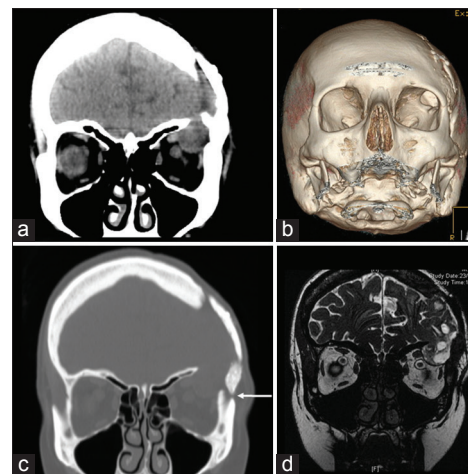
A 40-year-old female was admitted at our institution following a motor vehicle accident. The patient was unresponsive with a Glasgow Coma Scale (GCS) score of 6 (E1V1M4) and was put on mechanical ventilation. The pupils were bilaterally equal and reactive. She had no periorbital hematoma and no proptosis. Visual acuity and extraocular muscle motility of both eyes were not assessed because of the reduced level of consciousness of the patient. The immediate postinjury computed tomography (CT) scan showed only a mild, diffuse brain swelling [Figure 1a]. Thin-slice CT scan with three-dimensional (3D) reconstruction and coronal and sagittal sections did not show skull base fractures, in particular, posttraumatic



**Figure 1:** (a and b) The immediate postinjury thin-slice computed tomography scan with coronal reconstruction showed generalized brain edema without left orbital roof fractures. (c) Computed tomography scan 24 h after trauma revealed a left-sided frontal contusion causing midline shift and increased diffuse brain swelling. (d and e) Postoperative computed tomography scan showing left decompressive craniotomy and evacuation of frontal contusion with a small bony defect in the roof of the orbit (see arrow) probably related to an unintentional opening of the orbit during keyhole burr hole without evidence of encephalocele. (f) Computed tomography scan after cranioplasty confirming the small bone opening on the orbital roof (see arrow)

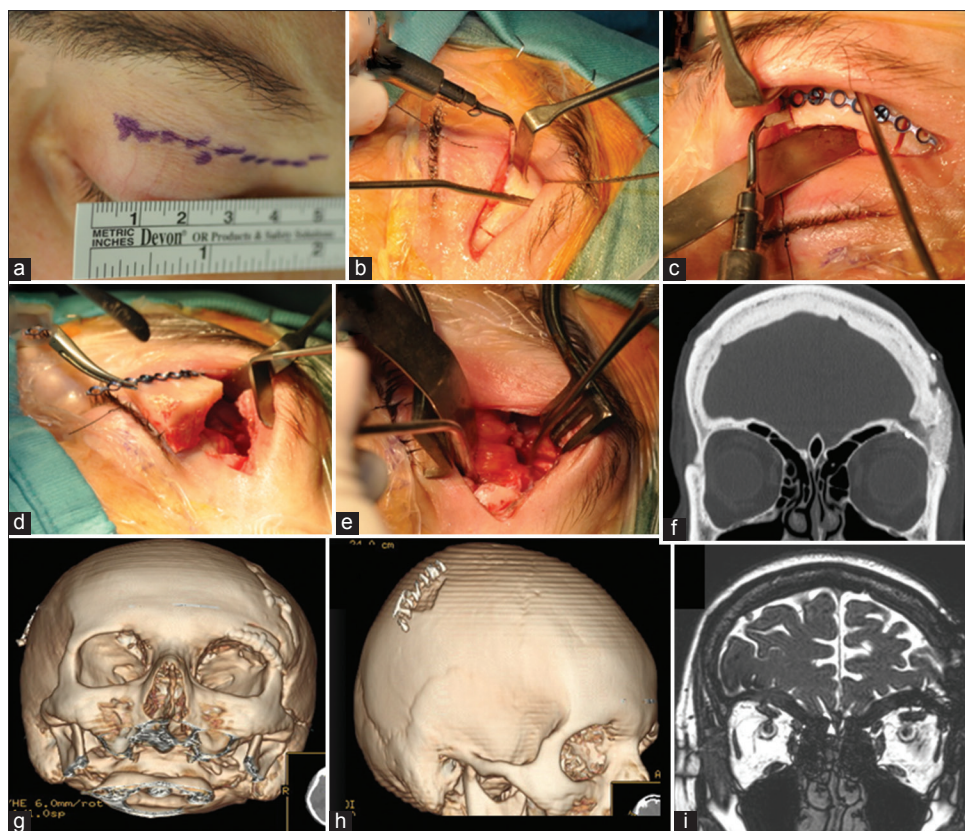
bony defects on the left orbital roof [Figure 1b]. The patient was then transferred to the Intensive Care Unit, and an intracranial pressure (ICP) probe was implanted for ICP monitoring. After 24 h, despite maximal medical treatment, the patient developed intractable ICP with stable values up to 30 mmHg. Hence, she repeated immediately a CT scan that revealed a massive increment of the brain swelling, and a left-sided frontal contusion causing contralateral midline shift [Figure 1c]. She underwent left decompressive craniotomy and evacuation of frontal contusion. Immediately after decompressive surgery, the ICP decreased significantly. The immediate postoperative CT scan confirmed evacuation of left posttraumatic hemorrhage and resolution of the mass effect [Figure 1d]. At a deeper examination, this exam also showed a small bony defect on the orbital roof, undetected until the occurrence of the orbital encephalocele (see the arrow in Figure 1e). During the following days, the ICP remained stable within normal limits. The patient recovered well and was discharged from the hospital on the 25<sup>th</sup> posttraumatic day when she was conscious with a GCS score of 15 and with only a mild right hemiparesis. After 2 months, the patient was submitted to autologous bone flap repositioning. There were no complications during the cranioplasty procedure. Wound healing was uneventful, and postoperative neuroimaging showed accurate fitting of cranioplasty, along with the undetected bony defect of the orbital roof [Figure 1f]. After cranioplasty, the patient presented full neurological recovery, included the hemiparesis, and returned to the previous occupation.

Two years following the last operation, the patient complained left fronto-orbital headache without any others ocular symptoms and/or signs, such as visual disturbance, proptosis, or pulsatile exophthalmos. 3D CT scan demonstrated the complete bone fusion of autologous cranioplasty, and no others intracranial complication. However, a thin-slice orbital CT sections with coronal reconstruction revealed the enlargement of the supero-lateral orbital roof defect with intraorbital encephalocele [Figure 2a-c]. Magnetic resonance imaging (MRI) confirmed the herniation of brain parenchyma into the left orbit through that bony defect [Figure 2d]. Accurate anamnestic investigation excluded a history of congenital anomalies, bone dysplasia, or neurofibromatosis (that could be associated with congenital skull base encephalocele). Hence, the patient underwent a combined left supra-transorbital minimally invasive approach aiming at achieving adequate surgical exposure without compromising the previously implanted autologous cranioplasty, already osteointegrated. A left superior blepharoplasty skin incision was performed to expose the orbital rim. The piezoelectric bone scalpel (piezosurgery) was used to perform a small supraorbital



**Figure 2:** (a-c) Thin-slice orbital computed tomography sections with three-dimensional and coronal reconstruction (2 years after the cranioplasty) revealed the enlargement of the lateral orbital roof bony defect associated with intraorbital encephalocele. The arrow indicates the burr hole, filled by autologous bone dust mixed with a bone substitute, too close to the orbital wall. (d) Magnetic resonance imaging image confirming the herniation of brain matter into the left orbit

keyhole craniotomy including the supero-lateral orbital rim in one piece using the defect of the orbital roof as the posterior edge of the orbitotomy [Figure 3a-e]. This combined minimally invasive approach gave access to both the superior orbital cavity and the intracranial supraorbital anterior skull base without the need for cranioplasty to be removed [Figure 3e]. The periorbital and orbital contents were identified and separated from the herniated cerebral tissue. Herniated brain parenchyma was resected with microsurgical technique revealing the clear margin of dural and bony defect of the orbital roof. The dura was re-approximated with a locked running stitch, and water-tight closure was achieved. Then, the orbital roof was reconstructed with autologous bone, obtained from the outer bone parietal layer contralateral to the cranioplasty through a small linear skin incision and fixed by titanium low-profile miniplate and screws [Figure 3c, d, and f-h]. The thin bone cutting line, performed by piezoelectric scalpel, allowed a perfect realignment of bone stumps [Figure 3c]. The postoperative period was uneventful, and she was discharged from the hospital 5 days after surgery. The esthetic results were excellent and patient was able to achieve complete eye closure within 2 weeks. Postoperative thin-slice 3D CT scan showed the stable reconstruction of the orbital roof and good restoration of roof contour and normal orbital volume, excluding intracranial complications [Figure 3f-h]. MRI images 2 months after surgery confirmed the complete reduction of encephalocele [Figure 3i]. At the last follow-up after 3 years, the patient was in good health with no neurological deficits.



**Figure 3: Abcdefghi:** Intraoperative images and postoperative thin-slice computed tomography scan with three-dimensional reconstruction. (a) Left superior blepharoplasty incision. (b and c) Supra-transorbital keyhole approach with the combination of an orbital osteotomy with a supraorbital minicraniotomy. The use of piezoelectric scalpel allows to realize precise and thin osteotomies, for better future bone healing and better aesthetic result. (c and d) The one-piece bone flap includes the frontal bone, and the orbital rim using the defect of the orbital roof as the posterior edge of the orbitotomy. The one-piece bone flap was then fixed by titanium low-profile miniplate and screws. (e) Intraoperative working area with the exposure of both orbital and intracranial compartments. (f-i) Postoperative thin-slice computed tomography scan with three-dimensional reconstruction and magnetic resonance imaging showing left orbital roof reconstruction with autologous bone, obtained from the split calvarial parietal bone contralateral to the cranioplasty

## DISCUSSION

Orbital encephalocele is a rare entity, and it is characterized by the herniation of part of the brain through a congenital, traumatic, neoplastic, or iatrogenic orbital roof defect.<sup>[1-4,11]</sup> The herniation of brain parenchyma can also occur through natural foramina such as the superior orbital fissure or the optic foramen. Orbital encephaloceles may be congenital or acquired with congenital encephaloceles more likely to occur in early childhood.<sup>[3]</sup> In the adult population, acquired encephaloceles are reported in patients who have experienced head trauma with associated orbital roof fracture.<sup>[3,9,11]</sup> However, even traumatic orbital encephaloceles are a quite rare event. There are <25 reported cases in literature thus far, including both pediatric and adult population.<sup>[1-4,8-12]</sup> In all these cases, there was a radiological or direct intraoperative evidence of an orbital roof fracture associated with the encephalocele. The incidence of orbital roof fractures is higher in children because an impact to the orbital rim before the age of 7 years, cannot be dissipated by the

frontal sinus (not yet completely pneumatized), and the traumatic force is transmitted directly to the orbital roof. In a pediatric series, orbital roof fractures were found in 7%, 1% of patients who suffered head injuries, and 13% of those patients developed intraorbital encephalocele.<sup>[3]</sup> An orbital encephalocele due to trauma may develop acutely or may develop many years later as the arachnoid and tissue gradually herniate, or as the fracture in the orbital wall grows over time.<sup>[1-4,7]</sup> Growing fractures of the orbital roof have been reported several times, especially in pediatric population.<sup>[3]</sup> Gradual herniation of the arachnoid into the fracture, the physiologic growth of the cranium and the brain (in children), continuous pulsation of the CSF, and the absence of bony counter compression are the accepted mechanisms of growth and expansion of the fracture.<sup>[1]</sup> Our patient developed the intraorbital encephalocele without any evidence of orbital roof fracture at the initial thin-slice CT scan with 3D and coronal reconstructions. The first evidence of bony defect in the orbital roof was undetected but present at the immediate CT scan after decompressive craniectomy, probably related to an unintentional opening of the

orbit by the keyhole burr hole. The development of the intraorbital encephalocele could be related to the restoration of a positive pressure gradient between the intracranial compartment, and the orbital compartment after cranioplasty similar to the etiopathological mechanism of the growing fracture in children. To the best of our knowledge, this is the first case of intraorbital encephalocele secondary to decompressive craniectomy and subsequent bone replacement.

The main symptoms and signs of intraorbital encephalocele are diplopia, exophthalmos, orbital edema, subconjunctival hemorrhage, restricted movements of the eye, and loss of vision. Among these, pulsatile exophthalmos is the most reported symptom after traumatic intraorbital encephalocele in the literature.<sup>[8]</sup> Our patient complained only left fronto-orbital headache. Early recognition and treatment are very important since the raised intraorbital pressure may irreversibly damage the optic nerve.<sup>[8,11]</sup> Thin-slice CT scan with 3D reconstruction is the gold standard imaging modality to assess orbital roof fractures or bony defects. However, MRI imaging is essential to detect and evaluate the herniated brain tissue and for differential diagnosis with others intraorbital lesions such as tumors or vascular malformations.<sup>[1-3]</sup>

Excision of the herniated brain tissue, duraplasty, and reconstruction of the orbital roof are the key steps of surgery.<sup>[10,12]</sup> There are two main routes to the orbital roof: The transcranial and the extracranial transorbital route.<sup>[10]</sup> The transcranial approach is commonly performed through a bicoronal incision and a uni- or bi-frontal craniotomy. This approach can also allow to address concomitant intracranial lesions in the same session. The extracranial approaches to the orbit are generally performed through a superior blepharoplasty skin incision or through a preexisting skin laceration. In all but one cases described to date in the literature, the herniated brain parenchyma into the orbit was accessed through a wide frontal craniotomy followed by the removal of fractured bony fragments inside the orbit.<sup>[1-4,8-12]</sup> The main limitation of this approach is the extensive brain exposure and retraction that could result in cerebral parenchyma contusion, infection, and epilepsy.<sup>[10,12]</sup> In our case, to preserve the already osteointegrated autologous cranioplasty, we adopted a modification of the supraorbital keyhole approach, by combining an orbital osteotomy with a supraorbital minicraniotomy. This combined minimally invasive approach was used to facilitate both views and access to the anterior cranial fossa and orbital compartment. The one-piece bone flap includes the frontal bone, and the orbital rim using the defect of the orbital roof as the posterior edge of the orbitotomy. The use of piezoelectric scalpel enhance safety by preventing injuries to nonosseous structures and allows to realize precise, thin osteotomies, so facilitating the subsequent bone healing with better esthetic result.<sup>[5-7]</sup>

## CONCLUSIONS

The peculiarities of this case are the development of intraorbital encephalocele without evidence of traumatic orbital roof fracture and the combined supra-transorbital minimally invasive approach. The delayed intraorbital encephalocele is probably a complication related to an unintentional opening of the orbit during decompressive craniectomy through which the brain herniated following the restoration of physiological ICP gradients after the bone flap repositioning. The reconstruction of the orbital roof was performed by using a combined supra-transorbital minimally invasive approach aiming at achieving adequate surgical exposure while preserving the autologous cranioplasty, already osteointegrated. To the best of our knowledge, this approach has not been previously used to address intraorbital encephalocele.

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## Conflicts of interest

There are no conflicts of interest.

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