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Strategy for Surgical Excision and Primary Reconstruction of Giant Frontal sinus Osteoma

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Abstract:	Background Huge craniofacial tumors represent a real challenge to surgeons from several aspects; total excision of tumor, preservation and restoration of physiological functions of sensitive organs in the region, acceptable cosmetic outcome, and avoidance of complications. Tumors larger than 30 mm in diameter are considered giant tumors. The objectives of surgery in such tumors are to overcome those challenges. Case Description In this article we present a 22-year-old female patient with a huge frontal sinus osteoma, probably the largest reported in the literature so far, measuring 10cm × 8cm × 8cm, and causing recurrent peri-orbital cellulitis and severe disfigurement of the face. It took us 3 months of multidisciplinary preoperative planning for surgical excision of the tumor and 1ry reconstruction using custom-made PEEK prosthesis. Surgery went smoothly, the postoperative hospital stay was short (22 days), and there were no postoperative complications. Histology confirmed cancellous (sponge) osteoma. The outcome was excellent and satisfactory for both patient and the surgeons. Conclusions Giant osteoma can be safely removed by craniotomy, planning and simulation of surgery on 3-D skull model, and preparing custom-made prosthesis, preferably titanium, for primary reconstruction is the key for successful treatment of such challenging tumors with good outcome.
Suggested Reviewers:	

Cover Letter

Dr. Edward C. Benzel

Editor in Chief

World Neurosurgery Journal

Dear Dr Edward C. Benzel

I would like to submit this manuscript to the World Neurosurgery Journal.

I do confirm that this manuscript is not submitted to other journals or has been published before and we have no disclosures.

It is probably the largest frontal sinus osteoma reported in the literature so far. In this case report, the evolution of tumor over 15 years is documented with photographs, and role of infection theory in the evolution of this type of tumor is well observed in this case.

We present the elaborate preoperative planning and detailed description of the surgical steps, and 5-year follow up photographs of patient.

Main Author

Sherif Elwatidy

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Case Report Manuscript

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Abstract

Background: Huge craniofacial tumors represent a real challenge to surgeons from several aspects; total excision of tumor, preservation and restoration of physiological functions of sensitive organs in the region, acceptable cosmetic outcome, and avoidance of complications. Tumors larger than 30 mm in diameter are considered giant tumors. The objectives of surgery in such tumors are to overcome those challenges.

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Keywords: Giant Osteoma, craniofacial tumors, frontal sinus, craniotomy, custom-made prosthesis, reconstruction, complications.

Background

Osteomas are benign bone tumors that constitute 1% of all bone tumors, they are the most common benign tumors of the paranasal sinuses, usually found in the frontal sinus (47-80 %) [(1-5)] and less often in the ethmoid sinuses. It is usually seen in the second and third decades of life with a male predominance of 2:1. In the frontal sinus, they usually (37%) originate near the frontal sinus ostium and the rest from the roof, floor, inter-frontal septum, and anterior or posterior walls. In the fronto-ethmoidal localization it is most commonly located near the nasolacrimal duct. Frontal sinus osteomas are usually asymptomatic, in most cases they are detected accidentally in 3% of computed tomography (CT) scans and 1% of radiographs of the sinuses done for some other reasons. However, larger tumors can present with mass effect or complications which could be ophthalmological, sinus related or intracranial.

Ophthalmological complications are rare and consist of proptosis, diplopia, ptosis [6,7,8]. Very rarely visual loss and epiphora can occur due to compression of lacrimal sac by the osteoma. Sinus related complications of frontal osteoma include frontal sinusitis, mucocoele, and the vacuum 'sinus 'syndrome due to extension of osteoma into the anterior cranial fossa through the posterior wall of the frontal sinus or the cribriform plate, and can lead to pneumocephalus, meningitis, or cerebral abscess [4,7,9].

Histologically, 3 different types of osteoma are described, cortical variant, sponge variant and mixed type (both cortical and sponge). Three different theories explain the etiology of osteomas; traumatic, infection, and embryological theories [1,2,4,10-14].

Surgical excision of frontal sinus osteoma could either be done by endoscopic or open surgery depending on its size, location, and surgical expertise. However, giant osteomas, like our case, need open surgery where we did bifrontal craniotomy. The steps of surgery are described in details below.

Cases Description:

This is a case of a 22-year-old female, Known to have psychomotor impairment. She presented to the ophthalmology emergency room with recurrent periorbital cellulitis, the first attack was in 2010, **(Fig 1- A, B, C)**. She was treated conservatively with antibiotics and she responded well to the treatment. During the period 2010-2015, the patient developed relatively fast growing bony swelling that caused severe disfigurement of the face. Physical examination showed a midline swelling in the forehead and upper face, growing more to the left side. The mass is painless, hard in consistency, immobile, about 10 cm in diameter and has lobulated surface. The overlying skin is healthy and mobile with dilated veins. Eye examination showed, downward and lateral displacement of the left eye with proptosis, restriction of eye movements, and increased inter-pupillary distance. It was difficult to assess visual acuity and fields of vision as the patient was uncooperative. The rest of neurological examination was unremarkable.

Full radiologic examinations included **(Fig 2- A,B,C,D)**; plain x-ray of the skull, CT scan with 3-D reconstruction, magnetic resonance imaging (MRI) with magnetic resonance arteriography (MRA) and magnetic resonance venography (MRV), and cerebral angiogram. They revealed avascular large (10*8*8 cm) lobulated bony mass centered in the left fronto-ethmoid region with central enhancement**.** The antero-superior aspect of the mass has showed mucosal enhancement. There is also an inferior extension in the nasal cavity and ethmoid air cells. There is severe compression of the left orbit with exophthalmos and stretching of the optic nerve. Large intracranial component causing significant compression of the left frontal lobe and mild brain edema, compression of the superior sagittal sinus, however the tumor was avascular, and causing only compression and displacement of the anterior cerebral arteries. Endoscopic biopsy showed osteoma. Cerebral angiogram showed non filling of the left internal carotid artery (ICA) as it exits the cavernous sinus, Fig 2-G (arrow), as the patient was asymptomatic, no intervention was considered for it.

She was scheduled for bifrontal craniotomy excision of the tumor and primary craniofacial reconstruction using synthetic custom-made bone. The surgery was performed in combination with plastic surgery, ophthalmology and ENT teams. Surgery went smoothly and in harmony between different teams, it went as planned in stages; craniotomy stage, isolation of tumor from dura and orbital fascia, excision of tumor, and finally reconstruction. The stages are described below in details.

Preoperative planning

After completing investigations, CT scan (special protocol) is done to build 3-D model of the skull (actual size) including the tumor, **(Fig 3-A)**. We marked the cut lines on the 3-D model, then another 3-D model without the tumor was created to identify the resultant defect and prepare the custom-made prosthesis. Prototype prosthesis was created and approved before making the final prosthesis from PEEK (Poly Ether Ether Ketone)**, (Fig 3- B, C).**

Surgical Technique

Multidisciplinary meeting with parents of the patient to explain the surgery, possible complications, and take consent for surgery.

1. Skin flap and complete exposure of tumor**;** under general anesthesia, supine position, head fixed in 3-point clamp, Neuronavigation is used, bilateral temporary eye closure (tarsoraphy). Preparation of the head and face including upper lip and nose. Bicoronal skin incision, from tragus to tragus running 5 cm behind the tumor margin. Skin, periosteum and temporalis fascia were reflected as one flap exposing both orbits and nasal bones completely with preservation periorbital fascia intact. Both anterior and posterior walls of the frontal sinus were found defective, only thin bone trabeculae remained, and the sinus cavity was full of thick mucus (removed and sent for microbiology), **(Fig 4-A).**
2. Craniotomy**;** multiple burr holes surrounding the tumor, strip (donut) craniotomy flap around the tumor was elevated, dura dissected from the tumor all around and from the skull base using the microscope. All efforts were made to keep dura intact and do water tight repair of any accidental dural tears**.**
3. Tumor removal; tumor was lobulated, made of very hard bone, and difficult to remove with bone nippler, so oscillating saw and pneumatic drill were used to break the tumor into smaller pieces. As anticipated, tumor removal resulted in a huge cavity with large bone defects in the anterior and posterior walls of the frontal air sinus (forehead bone), medial walls of both orbits, floor of the anterior cranial fossa and ethmoid air sinuses down to the nasal floor, **(Fig 4-B)**.
4. Reconstruction; large vascularized pericranial and temporalis fascia flap was prepared and sutured to the dura of the anterior cranial fossa to isolate the intracranial cavity from the nasal cavity and paranasal sinuses. The custom bone was trimmed to fit nicely into the bony defect. The ophthalmology team tested the integrity of the lacrimal system and anchored the medial canthal ligaments to the prosthesis. The prosthesis and cranial vault bone were fixed with titanium plate and screws, and finally wound was closed in layers with drain, **(Fig 4-C, D).**

The post-operative course was uneventful with fast recovery, postoperative CT scan showed total tumor removal, no intracranial insults or bleed, and good reconstruction **(Fig 5-A, B, C).** Five years ' postoperative photos for the patient is shown in **(Fig 6-A, B).**

Discussion:

Frontal sinus osteoma are usually discovered incidentally in CT scans and x rays done for some other reasons. Small osteoma are treated endoscopically, while giant ones (3 cm or bigger) might need combined or craniofacial surgical removal (Howard Lynsch approach or even craniotomy) [15,17,18,19,20].

Cheng et al 2013 published 3 cases of giant osteoma of frontal and ethmoid sinuses and reviewed the literature on giant osteomas from 1975 to 2011. He collected 45 patients with giant osteomas from 41 articles including his 3 cases. He reported more males than females , (28 males and 17 females), the male to female ratio was 1.6:1, and the patient's age ranged from 11 to 70 years of age at initial presentation, with a mean age of 39.5 years. He did not comment on the size of tumors [12].

Our patient had a giant tumor (10cm×8cm×8cm) which is the largest frontal sinus osteoma reported in the literature so far (1-20). The patient presented with severe disfigurement, proptosis, lateral displacement of the eyes, squint, and with large intracranial extension and significant mass effect on the brain and compression of the frontal lobes. The tumor caused erosion of the anterior and posterior walls of the frontal air sinus that resulted in recurrent periorbital cellulitis with a significant risk of extension of infection into the cranial cavity due to bone erosion. Infection is one of the theories involved the etiology of osteoma, in our patient the tumor has rapidly grown to huge size in a relatively short period of time (about 5 years) most likely due to recurrent infection, which supports the infection theory in the etiology of osteoma [1, 2,7,12,19,20].

Surgical excision of giant frontal sinus osteoma entails a real challenge and carries serious morbidity, specifically meningitis, subdural empyema, brain abscess. Other postoperative complications include; nasal and sinus complications (epistaxis, septal perforation, chronic sinusitis, and mucocoele), ophthalmologic (epiphora, strabismus, diplopia, ptosis, infection and even loss of vision), and poor cosmetic outcome.

The bicoronal incision and strip (donut) craniotomy surrounding the tumor gives a good approach to the frontal sinus, the ethmoid and the eyeballs. It allows isolation of the tumor from the dura of the anterior cranial fossa particularly at the floor and allows also water tight repair of any dural tears, as well as dissection and preservation of intact peri-orbital fascia. In addition, it allows harvesting large vascularized pericranial flap used to isolate the cranial cavity from the nasal cavity as shown in fig 5-B (red arrow). The corner stone for avoidance of postoperative complications is preservation and restoration of natural barriers between the intracranial cavity and the nasal cavity and paranasal sinuses. This can only be achieved by (1) microscopic water tight dural repair of any dural tears, (2) vascularized pericranial and temporalis fascia flap sutured all around to the dura and enforced with tissue glue, (3) reconstruction of

the orbital walls and orbital structures (lacrimal apparatus and canthal ligaments), and ensure no compression or kink of the optic nerves, (4) proper reconstruction of the cranial and fascial bones using synthetic custom-made prosthesis preferably titanium, and ensure adequate healthy skin coverage.

Conclusions: Adequate preoperative planning of steps of surgery, simulation of surgery on 3-D skull model, and preparing custom-made prosthesis preferably Titanium for primary reconstruction made the surgery run smoothly, postoperative course uneventful, and achieve excellent cosmetic outcome.

References

1. Izci Y: Management of the large cranial osteoma: experience with 13 adult patients. Acta Neurochir (Wien) 147: 1151-1155, 2005.
2. Erdogan N, Demir U, Songu M, Ozenler NK, Uluç E and Dirim B: A prospective study of paranasal sinus osteomas in 1889 cases: changing patterns of localization. Laryngoscope 119: 2355-2359, 2009.
3. Bignami M, Dallan I, Terranova P, Battaglia P, Miceli S and Castelnuovo P: Frontal sinus osteomas: the window of endonasal endoscopic approach. Rhinology 45: 315-320, 2007.
4. Olumide AA, Fajemisin AA and Adeloye A: Osteoma of the ethmofrontal sinus. Case report. J Neurosurg 42: 343-345, 1975.
5. Malcolm WM and Haskell N: Transcoronal removal of an atypical orbitoethmoid osteoma. Plast Reconstr Surg 72: 874-877, 1983.
6. Mugliston TA and Stafford N: A cranio-facial approach to large osteomas of the fronto-ethmoidal region. J Laryngol Otol 99: 979-983, 1985.
7. Lunardi P, Missori P, Di Lorenzo N and Fortuna A: Giant intracranial mucocele secondary to osteoma of the frontal sinuses: report of two cases and review of the literature. Surg Neurol 39: 46-48, 1993.
8. Shady JA, Bland LI, Kazee AM and Pilcher WH: Osteoma of the frontoethmoidal sinus with secondary brain abscess and intracranial mucocele: case report. Neurosurgery 34: 920-923, 1994.
9. Rappaport JM and Attia EL: Pneumocephalus in frontal sinus osteoma: a case report. J Otolaryngol 23: 430-436, 1994.
10. Mendelsohn DB, Hertzanu Y and Friedman R: Frontal osteoma with spontaneous subdural and intracerebral pneumatocele. J Laryngol Otol 98: 543-545, 1984.
11. Brunori A, de Santis S, Bruni P, Delitala A, Giuffre R and Chiappetta F: Life threatening intracranial complications of frontal sinus osteoma: report of two cases. Acta Neurochir (Wien) 138: 1426-1430, 1996.

12. Cheng K-J, Wang S-Q, Lin L. Giant osteomas of the ethmoid and frontal sinuses: Clinical characteristics and review of the literature. Oncology letters. 2013;5(5):1724-30.

13. Summers LE, Mascott CR, Tompkins JR and Richardson DE: Frontal sinus osteoma associated with cerebral abscess formation: a case report. Surg Neurol 55: 235-239, 2001.

14. Johnson D and Tan L: Intraparenchymal tension pneumatocele complicating frontal sinus osteoma: case report. Neurosurgery 50: 878-880, 2002.

15. Osma U, Yaldiz M, Tekin M and Topcu I: Giant ethmoid osteoma with orbital extension presenting with epiphora. Rhinology 41: 122-124, 2003.

16. Park MC, Goldman MA, Donahue JE, Tung GA, Goel R and Sampath P: Endonasal ethmoidectomy and bifrontal craniotomy with craniofacial approach for resection of frontoethmoidal osteoma causing tension pneumocephalus. Skull Base 18: 67-72, 2008.

17. Yiotakis I, Eleftheriadou A, Giotakis E, Manolopoulos L, Ferekidou E and Kandiloros D: Resection of giant ethmoid osteoma with orbital and skull base extension followed by duraplasty. World J Surg Oncol 6: 110, 2008.

18. Adeleye AO: A giant complex fronto-ethmoidal ivory osteoma: surgical technique in a resource-limited practice. Surg Neurol Int 1: 97, 2010.

19. Humeniuk AM, Stryjewska-MG, Janik MA, Kolebacz B. Giant fronto-ethmoidal osteoma–selection of an optimal surgical procedure. Brazilian journal of otorhinolaryngology; 84 (2): 232-239, 2018.

20. R Vishwakarma, ST. Joseph, KB Patel, A Sharma. Giant Frontal Osteoma: Case Report with Review of Literature. Indian J Otolaryngol Head Neck Surg; 63(Suppl 1): S122–S126, 2011.

Legends

Fig1: Evolution of the tumor

Fig 1-A, photograph for the patient in 2005, no tumor detected

Fig 1-B, photograph for the patient in 2010 during the first attack of periorbital cellulitis, notice broadening of the forehead and inter-orbital distance

Fig 1-C, photograph for the patient in 2015 during the last attack of periorbital cellulitis, notice the tomor has grown to huge size in 5 years with severe disfigurement.

Fig 2, radiologic investigations

Fig 2-A, sagittal reconstruction of computerized tomography (CT) scan showing the lobulated tumor filling the anterior cranial fossa and extends down into the nasal cavity and paranasal sinuses almost reaching the hard palate.

Fig 2-B, 3-D reconstruction of CT scan showing the lobulated tumor in the frontal sinus causing erosion of its anterior wall, as well as medial wall of the left orbit and nasal bones.

Fig 2-C, Cerebral angiogram (left common carotid artery injection) showing avascular tumor, occluded left internal carotid artery (arrow).

Fig 2-D, Magnetic resonance imaging (MRI) scan, T2 axial images, showing huge tumor displacing the left orbit and compressing the frontal lobes.

Fig 3, preoperative planning

Fig 3-A, 3-D skull model with tumor showing the cut line in normal bone around the tumor (Black marker).

Fig 3-B, 3-D skull model without the tumor showing the anticipated skull defect after tumor removal.

Fig 3-C, intra-operative image of the custom made prosthesis (made of poly ether ether ketone – PEEK).

Fig 4, Surgical steps

Fig 4-A, exposure of the tumor after bicoronal fascio-cutaneous flap, notice lobulated osteoma in the frontal sinus with thick mucus secretions.

Fig 4-B, remaining bone trabeculae of the anterior wall of the frontal sinus.

Fig 4-C, huge defect in the floor of the anterior cranial fossa, left orbit (green arrow), Right orbit (black arrow), nasal cavity (blue arrow), Dura (yellow arrow).

Fig 4-D, reconstruction of craniofacial defect using custom made prosthesis

Fig 4-E, final intraoperative image after skin closure.

Fig 5, postoperative CT scan

Fig 5-A, axial CT images showing total excision of the tumor, notice the contour of the frontal region of the skull.

Fig 5-B, sagittal reconstruction CT image, notice the pericranial flap (red arrow) isolating the cranial cavity from nasal cavity.

Fig 5-C, 3-D reconstruction image showing the custom made PEEK prosthesis.

Fig 6, 5-year postoperative photograph of the patient; front projection (fig 6-A), and lateral projection (fig 6-B)

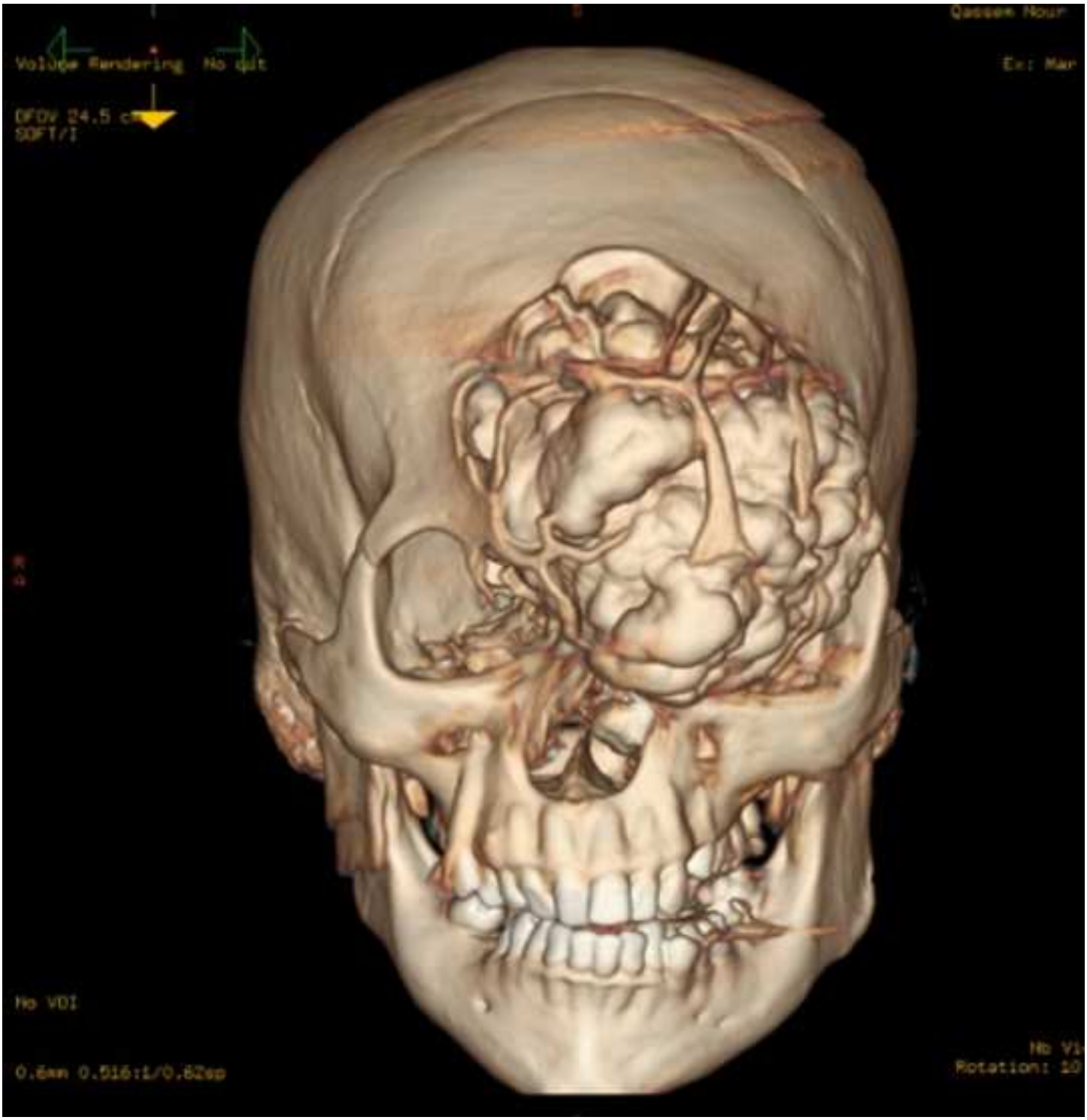

Figure 2 C

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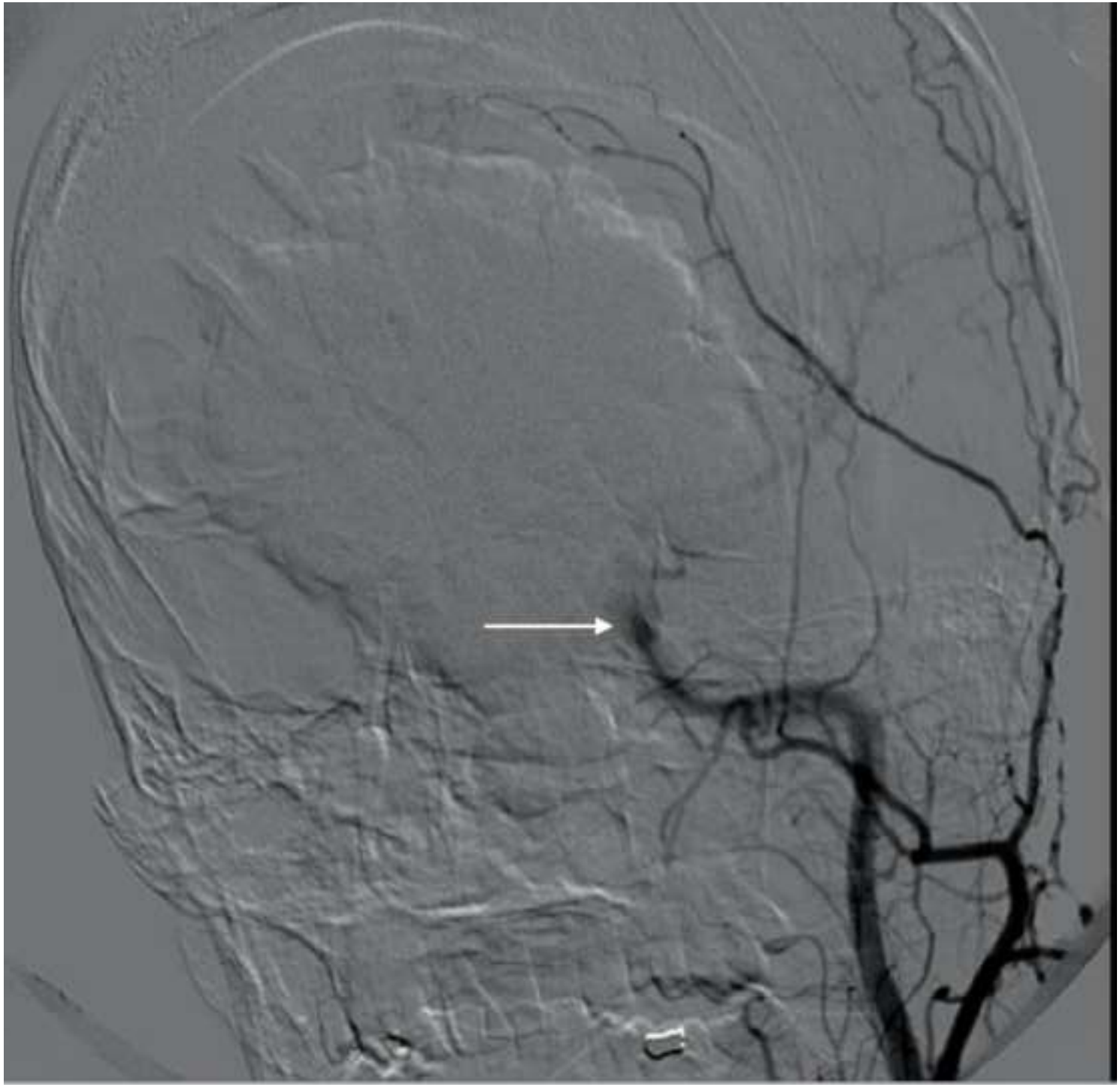

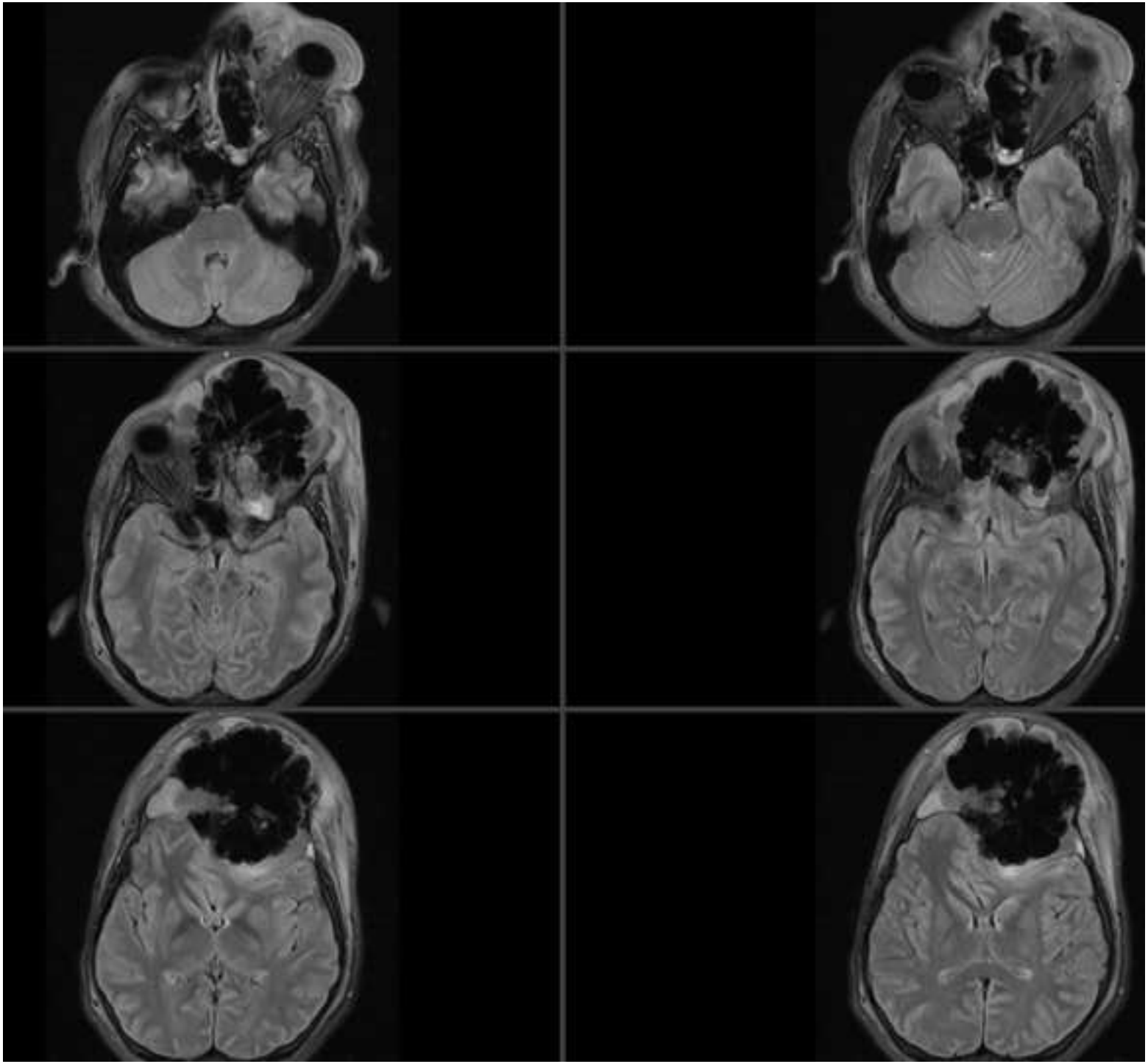

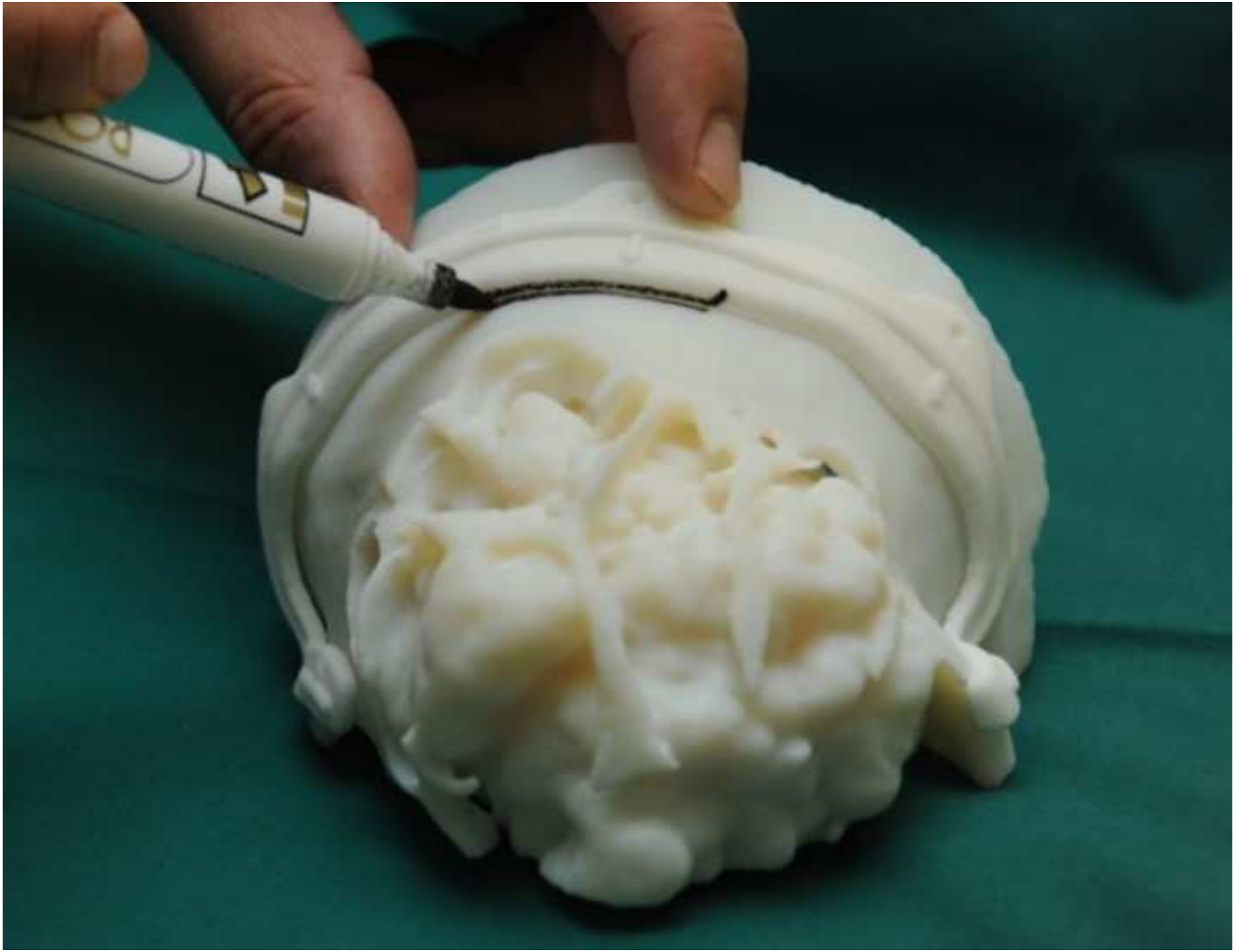

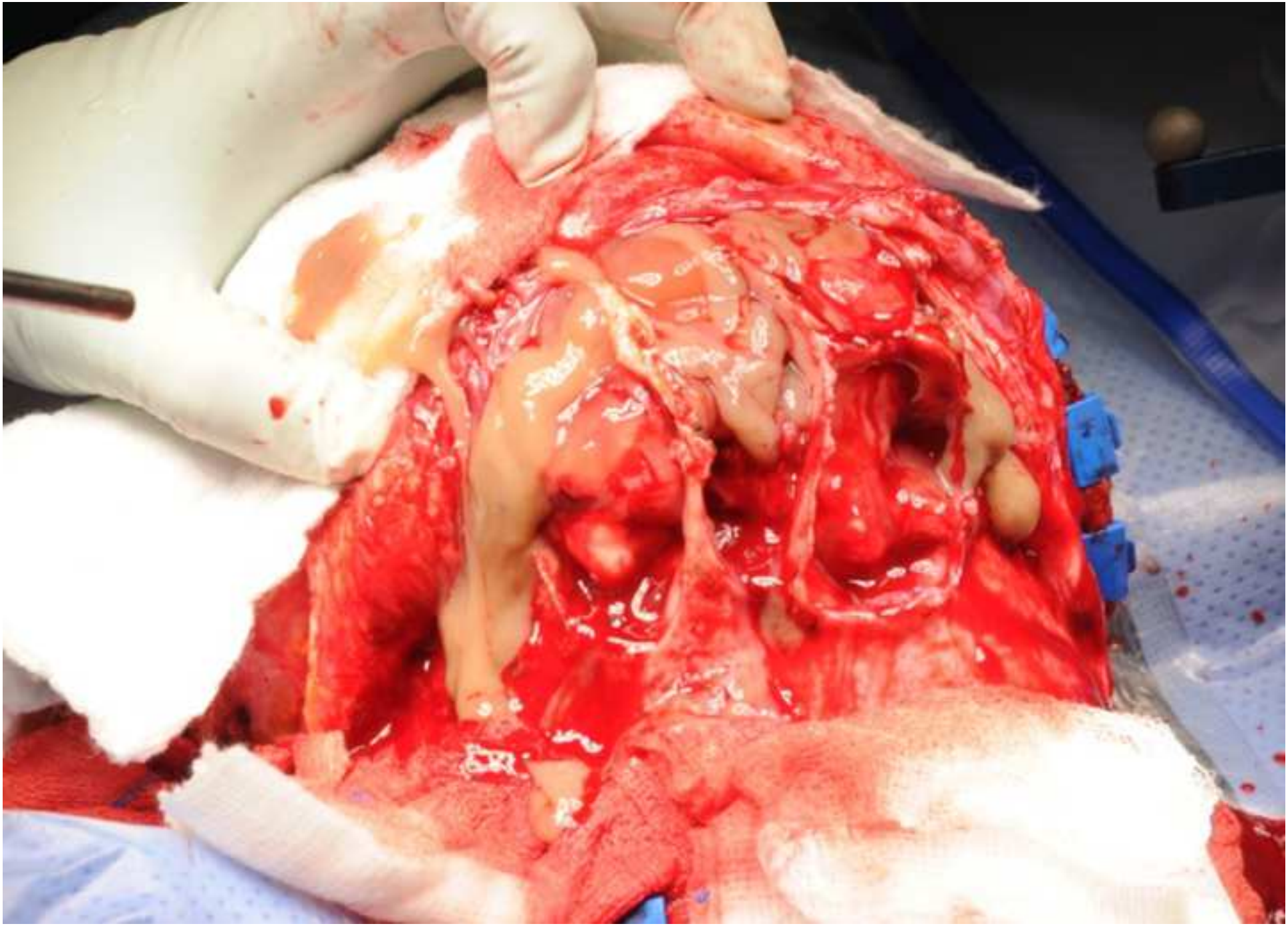

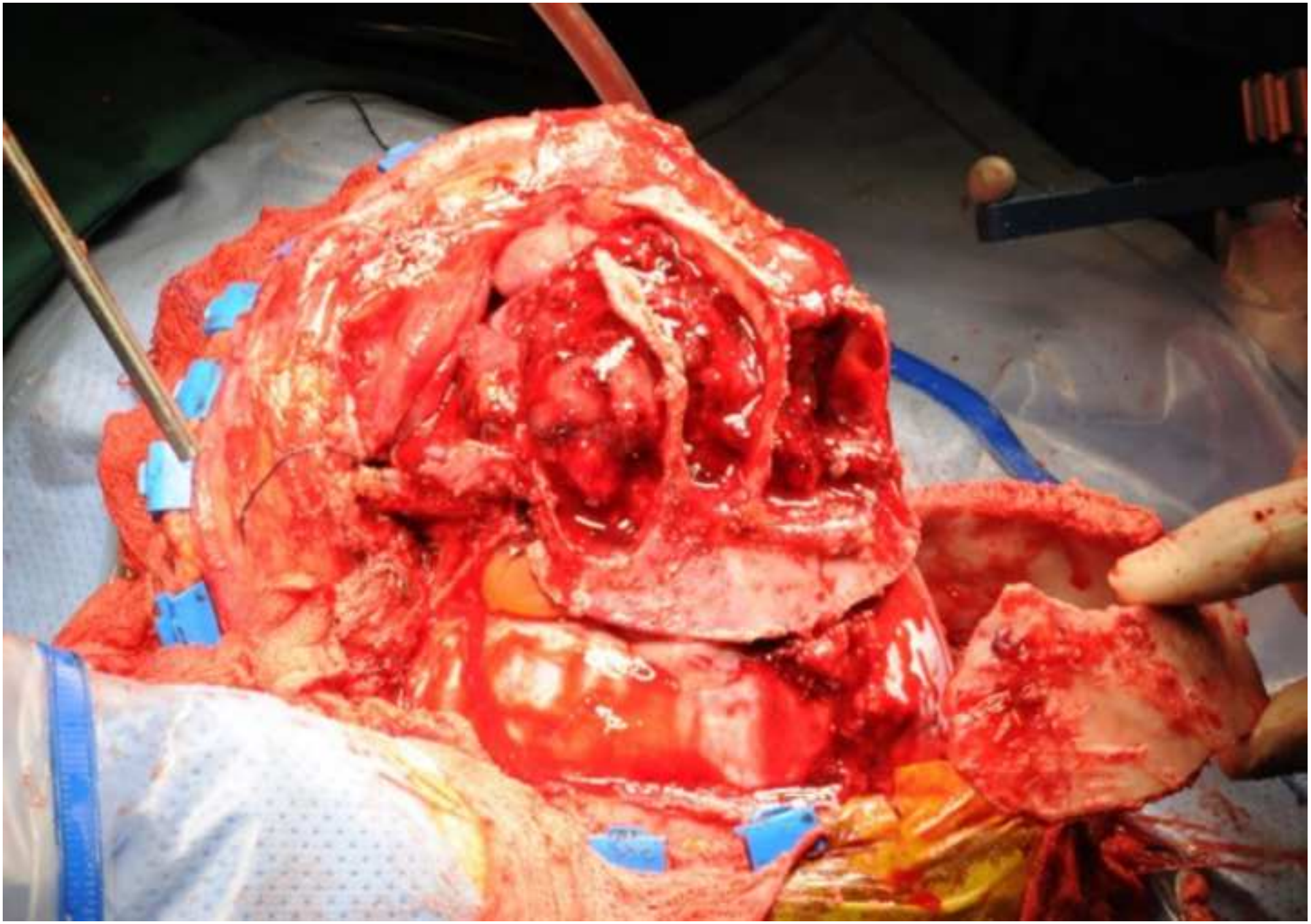

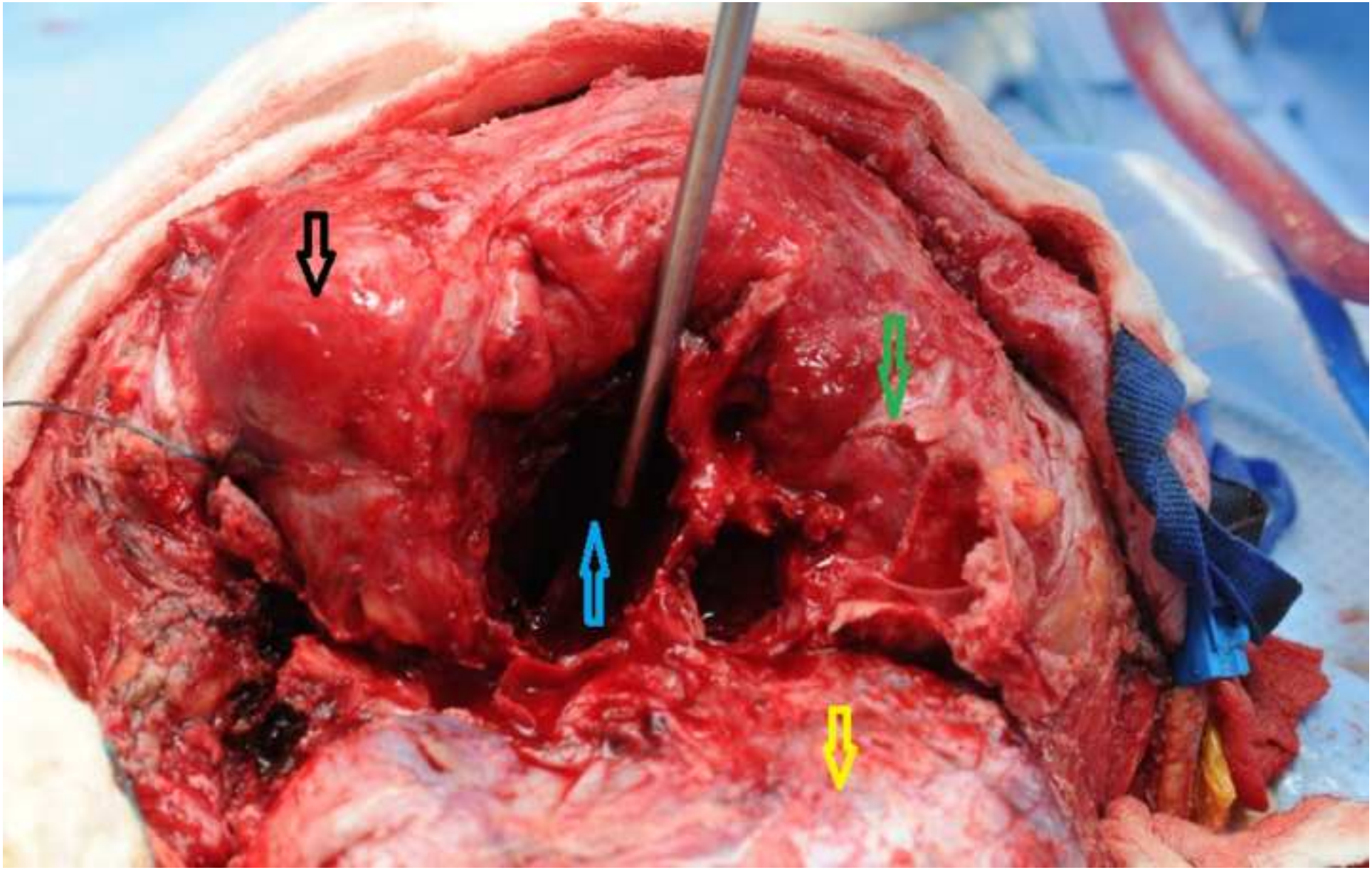

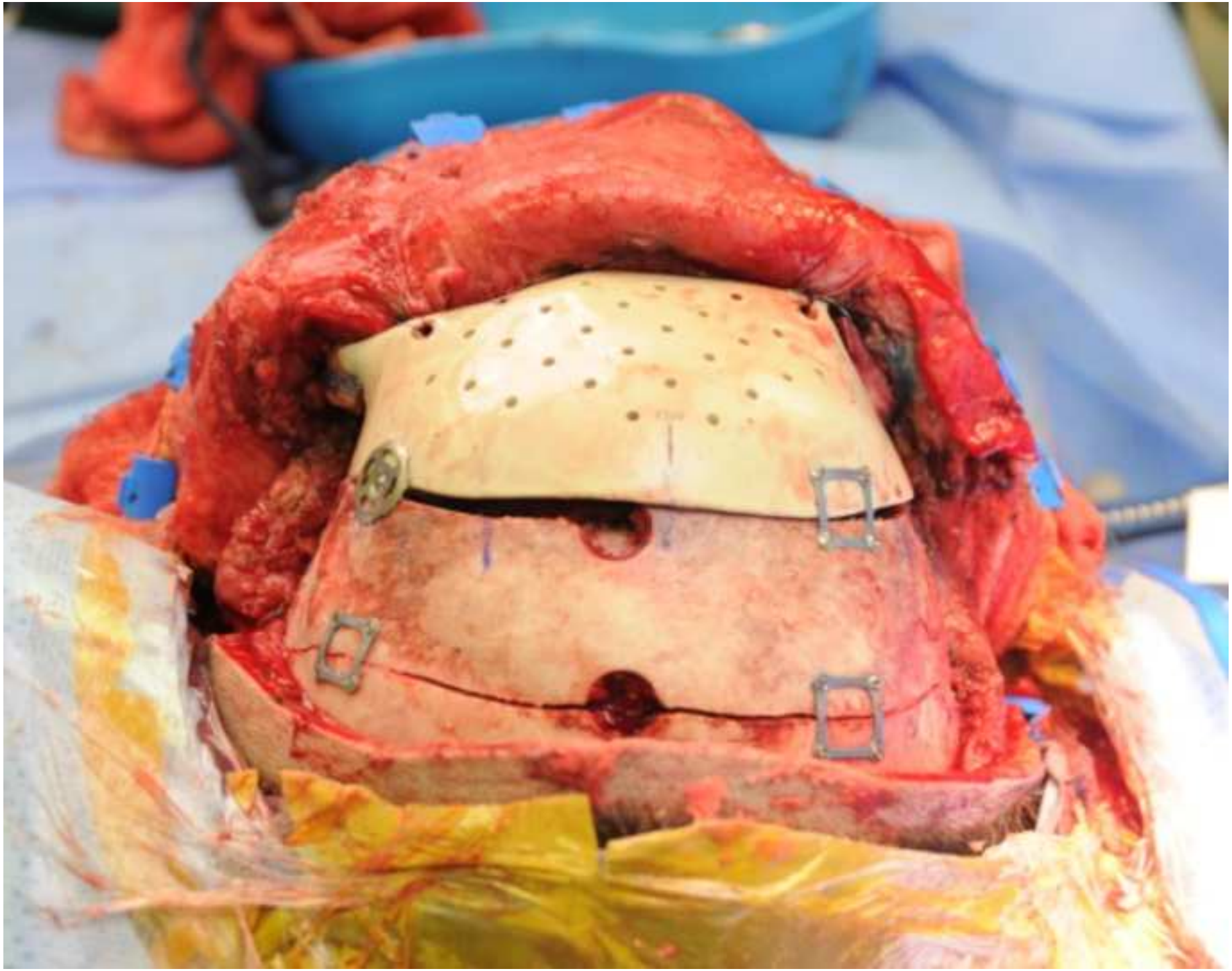

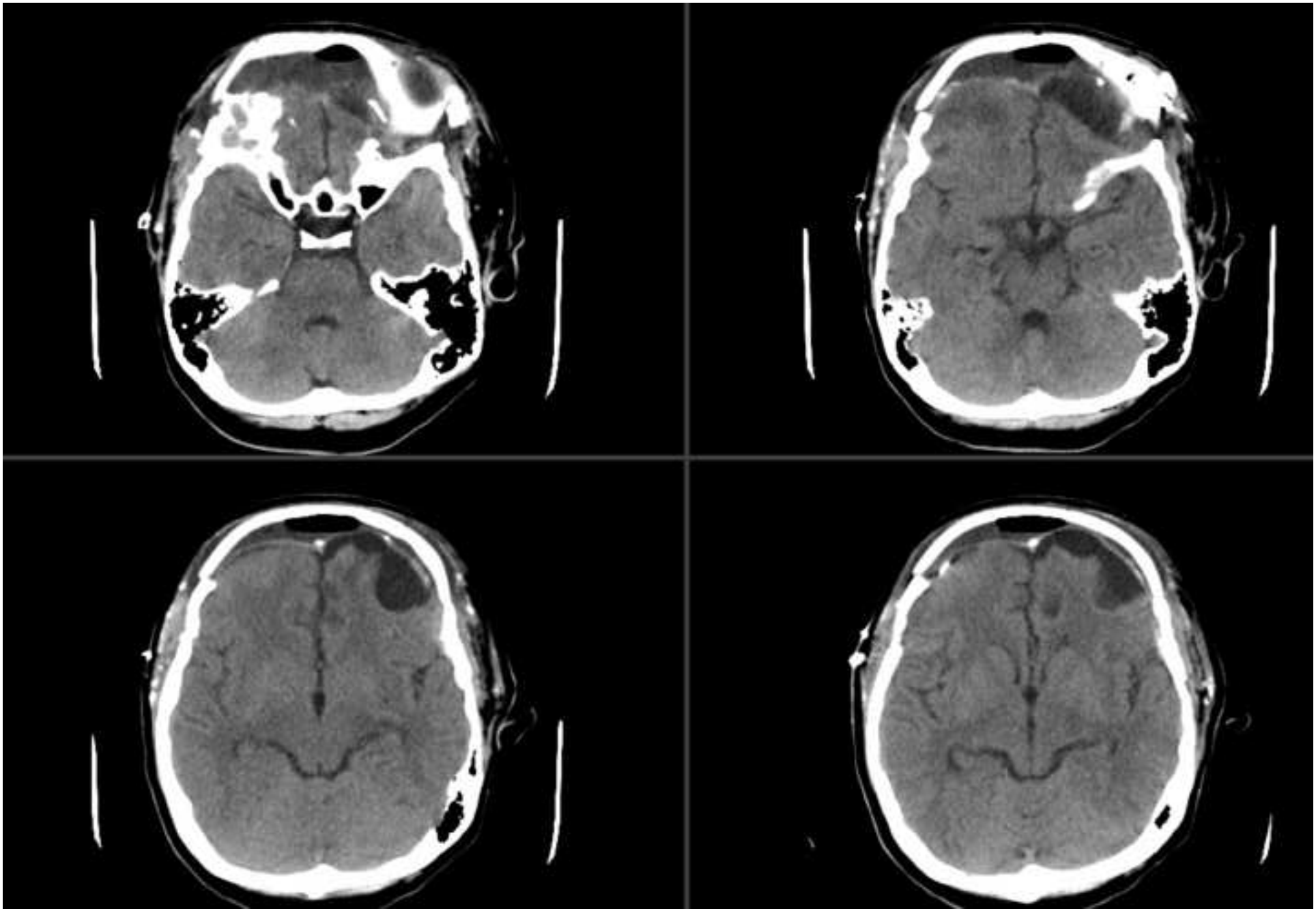

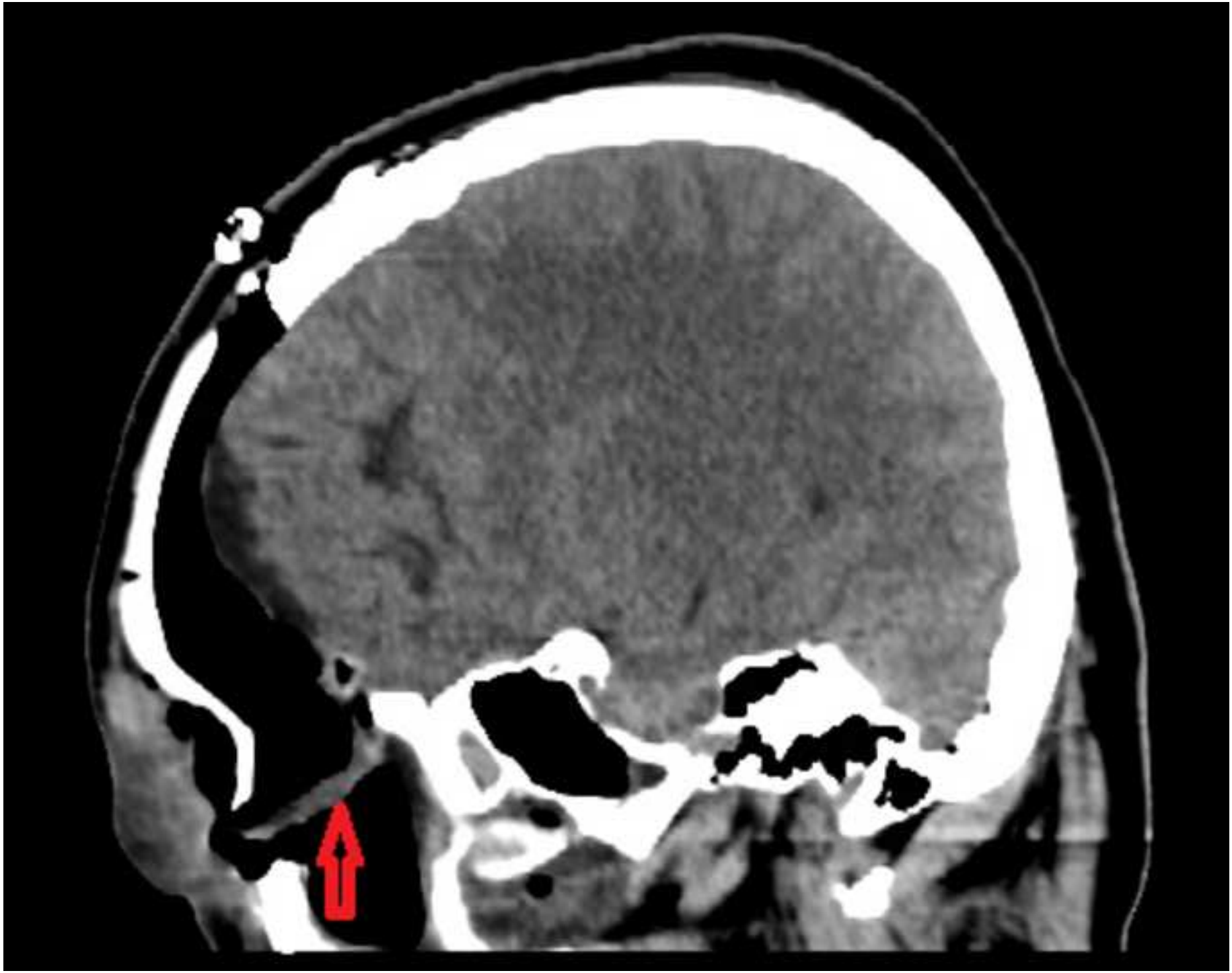

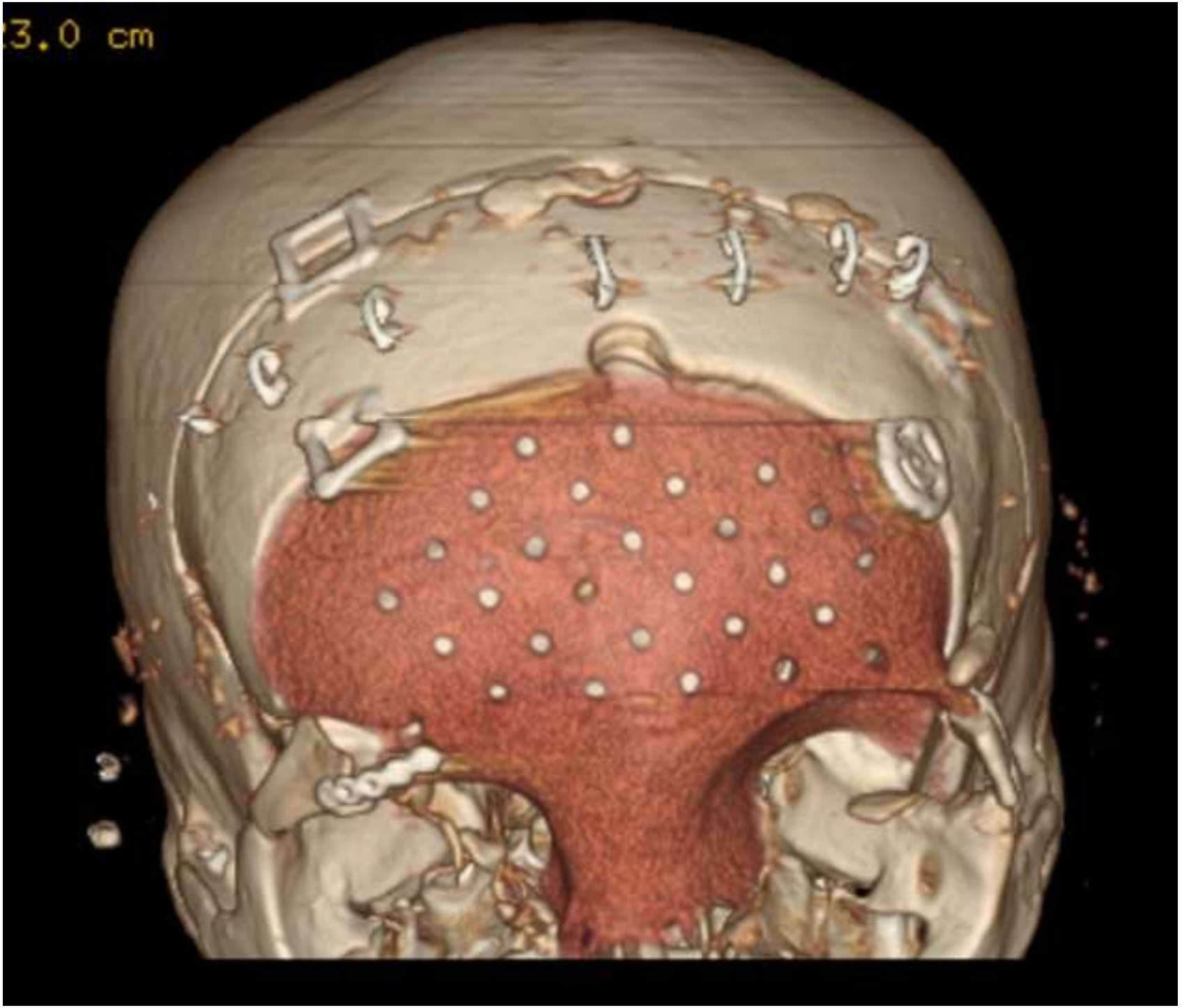

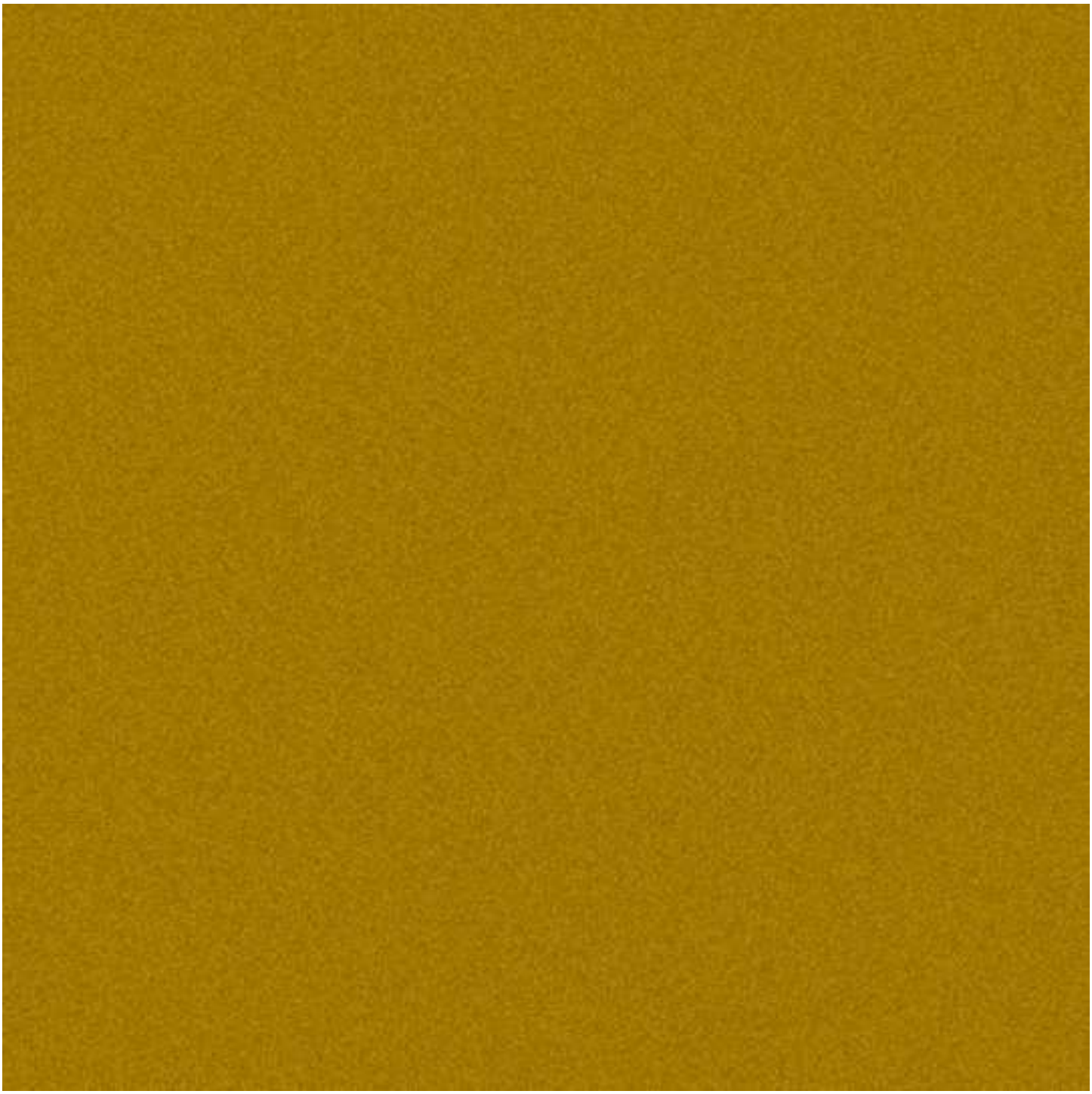

Disclosure

The Authors have no disclosures or conflicts of interest to declare