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Case report

Spontaneous cranial bone regeneration following craniectomy for traumatic brain injury in a pregnant woman: A case report

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ABSTRACT

Introduction and importance: Spontaneous bone formation following craniectomy is an extremely rare in adult. As in the medical literature, this is the first case report on total spontaneous ossification following craniectomy in a pregnant woman.

Case presentation: In this paper, we reported a 20-year-old female currently in the 30th week of her pregnancy suffered from head trauma following motorcycle accident. On admission to our hospital, her GCS score was 3 points. She was treated with emergency extradural hematoma evacuation with craniectomy and Caesarean section with uterine artery ligation. 3 weeks post-operation, the patient and her daughter were discharged from the hospital. At follow-up, spontaneous cranial bone generation was observed.

Clinical discussion: The presentation, diagnosis and strategy of treatments were discussed.

Conclusion: Diagnostic imaging in traumatic pregnant patient is often postponed for the concern of fetus exposure to radiation. Traumatic pregnant patient with possible head trauma should be transferred to a center with expertise in neurotrauma and obstetrical care. Spontaneous cranial bone regeneration following craniectomy in adult is rare. Surgery techniques and hormones in pregnancy contribute to bone formation.

1. Introduction and importance

Spontaneous bone formation following craniectomy is an extremely rare in adult. As in the medical literature, this is the first case report on total spontaneous ossification following craniectomy in a pregnant woman.

2. Case presentation

2.1. History and examination

A 20-year-old female, currently on the 30th week of her pregnancy (gravida 2, para 1), suffered from a motorbike accident. She was alert and had some headache following the accident. She was then transferred to a local hospital. As she was pregnant, the patient did not get a CT scan at the local hospital, and she was transferred to our hospital. Her husband told us that halfway to our hospital, her headache got more severe, and then she went into coma. It was 4 h from the accident to when she was admitted to our hospital. Past medical history revealed no drug use. No family history was observed.

On admission, the patient presented with coma. Her GCS score was 3 points [1], both her pupils were totally dilated, she showed neither eye, verbal nor motor response to stimuli, no pupillary reflex, no respiratory reflex [2]. Her vital signs were relatively steady, as her heart rate was 130 bpm, her blood pressure was 110/70 mmHg without vasopressors, her respiratory rate was 20 breaths per minute, her oxygen saturation was 90–91%. She was treated by ABCDE approach [3] with emergency tracheal intubation immediately [4]. A head CT scan demonstrated a large extradural hematoma in the right tempo-parietal-occipital region (Figs. 1A and 2A). No other major injury was found. She was indicated urgent surgery with obstetrics consultation simultaneously with affirmation from all of the patient's family members. It was 30 min from her admission to the operation.

Abbreviations: 3D, 3-dimensional image; CT, Computed topography; GCS, Glasgow coma scale.

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2.2. Operation

In the operation room, after the anesthesia team done, two teams operated simultaneously. The neurosurgery team performed an extradural hematoma evacuation via temporal parietal craniectomy. We performed a U-shaped skin incision. Then, 5 layers of the scalp, consisting of the Skin, the subcutaneous tissue, the galea Aponeurosis, the Loose connective tissue, and the Periosteum was elevated in a single flap [5]. A burr hole was quickly drilled to relieve the pressure. Then, a temporal parietal craniectomy was performed. The size of the bone flap was approximately 10 cm × 10 cm in size.

A large extradural hematoma was observed, the bleeding meningeal vessels were coagulated. The dura was unopened. We decided to store the bone flap. We closed the scalp in two-layer fashion. The inner layer consists of pericranium to the subcutaneous was closed using interrupted absorbable sutures. The outer layer consists of the skin and the subcutaneous was closed using interrupted non-absorbable sutures. The procedure was performed by Dr. D.T and his team.

The obstetrics team performed a Caesarean section and uterine arteries ligation for hemorrhage control. Total time of surgery was 30 min.

2.3. Post-operation

Post-operation, the patient remained in the intensive care unit for 3 weeks. 3 – day post-operation, neurologic examination revealed mild left hemiparesis with muscle strength of 4/5. 3 – days post-operation CT scan demonstrated no hematoma, no bleeding in craniectomy site (Figs. 1B, 2B, and 3A). The baby was treated in pediatric intensive care unit for 3 weeks with surfactant to achieve fetal lung maturation. After 3 weeks, both the patient and her baby were dismissed from the hospital. The patient GCS score at dismission was 15 points; she had no headache, mild left hemiparesis. The baby was alert, and he could breathe without assistance.

7 weeks after the operation, the patient came back for a check-up. The patient GCS score was 15 points, her left hemiparesis subsided. She denied neither neurological deficit nor seizures. On examination, the surgical site was surprisingly firm and stable. The CT scan showed thin bone ossification in the craniectomy region (Figs. 1C, 2C, and 3B). She was indicated cranial reconstruction by autologous bone graft at 7-week after the first operation. In the cranioplasty operation, we found the newly remodeled bone was steady, it spread roughly throughout the craniectomy region, and it tied closely to the underlining dura and it was continuous with the surrounding skull edge in some areas (Fig. 4). We decided not to replace the bone flap. No further procedure was done, and we closed the surgical site. The procedure was performed by Dr. D.T and his team. The patient was treated with antibiotics for 5 days and then dismissed from the hospital.

23 weeks post-operation, the patient had a follow-up. Her GCS score was 15 points. On examination, no neurological deficit was observed. The craniectomy site was solid and uninterrupted. The CT scan demonstrated spontaneous cranial bone regeneration which spread nearly throughout the craniectomy site (Figs. 1D, 2D, and 3C).

This paper has been reported in line with the SCARE 2020 criteria [6].

3. Clinical discussion

3.1. Craniectomy decision

In the first time we operated on the patient, we decided to store the

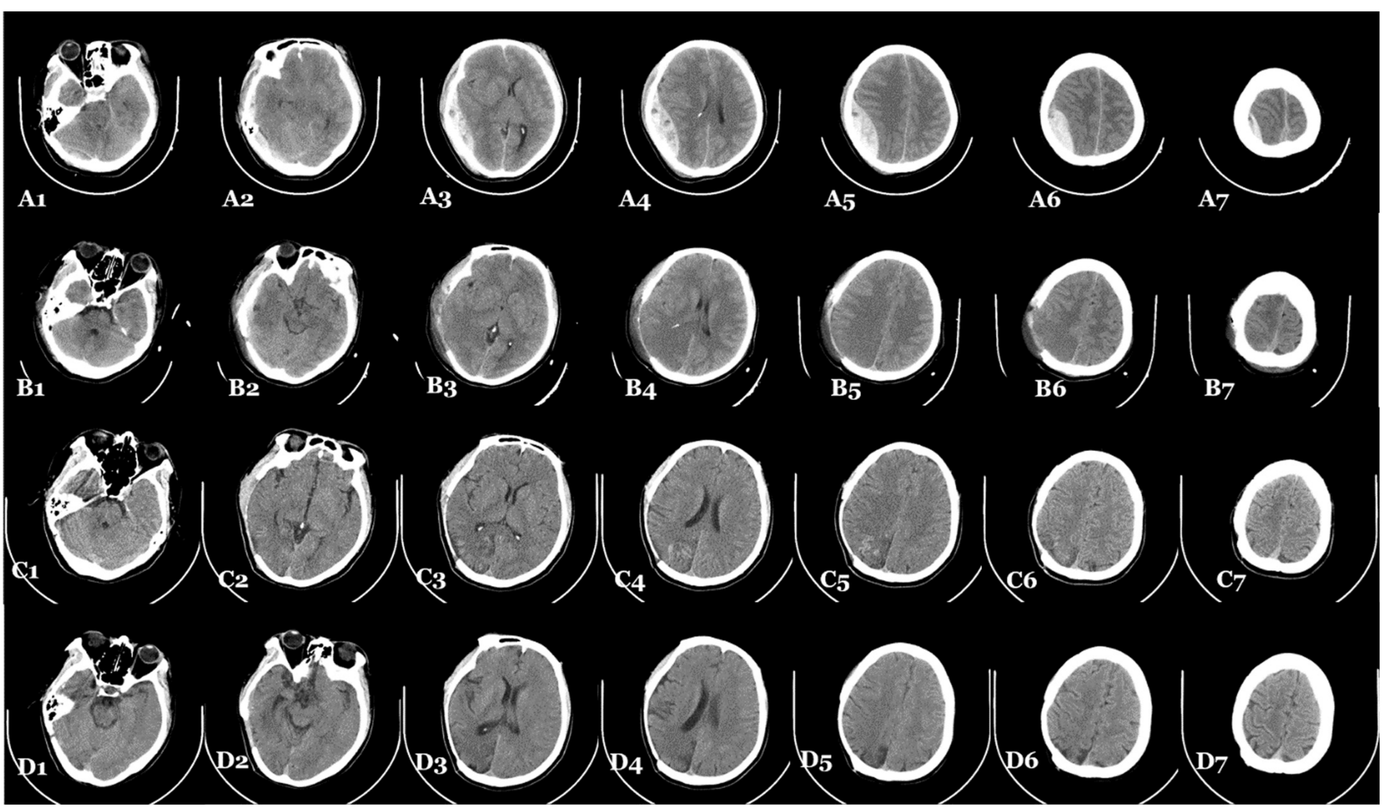

Fig. 1. Brain window.
A: 1–7: The pre-operative CT scan showed a large extradural hematoma in the parietal occipital and posterior fossa region.
B: 1–7: The 3 days post-operative CT scan showed no mass effect with bone flap removed.
C: 1–7: The 7 weeks post-operative follow-up CT scan showed bone remodeling in the craniectomy region.
D: 1–7: The 23 weeks post-operative follow-up CT scan showed ossification in the craniectomy region.

bone flap for the following reasons. We were currently trying to save a 3-point Glasgow Coma Scale pregnant patient, who entered on two emergency operations simultaneously. The major reason was to prevent further blood loss and anesthetic complications. Both operation teams would work as quickly as possible, so the anesthesia team could provide intensive care. The minor reason was that we wanted to exclude delayed cerebral edema following the operation [7]. Although an intact normal dura does not expand much, in our experiences, an intact dura without the bone flap would be better than an intact dura with the bone flap on.

In the second operation, we decided not to replace the original skull flap. In our observation at the operation room, removing the newly regrowth bone would provide risk of blood loss and dura tear, which may complicate the situation and require further surgery. Although the newly regrowth bone was thinner than a completely normal skull, it was stable to protect the inner cerebral structure. In the follow-up, we could observe that the newly regrowth bone increased in size and calcification.

3.2. Spontaneous bone formation

In our observations, we found 4 articles focused on spontaneous bone formation after craniectomy in humans. One of them described the phenomenon occurred in a 29-year-old woman [8]. The other three cases were children [9–11]. Our case described a woman had spontaneous ossification following craniectomy in her pregnancy period, which we believed to be the first report on this issue.

3.3. Spontaneous bone formation >< surgery technique

Spontaneous bone formation occurred as the result of the three possible layers: periosteum, dura mater, and adjacent skull diploe. Skull fracture could lead to the stimulation of latent osteogenic cells in periosteum and the surrounding skull diploe [12]. Dura and periosteum add in osteogenesis by contact [13]. Some articles suggest that skull diploe is the dominant source of osteoblast, and dura contact was more productive than pericranium [14,15]. Besides, vascularization makes a crucial contribution to osteogenic processes [16].

In our case, the new bone in this case mainly grew up from the dura and some from the edges of the craniotomy (Fig. 4). We respected all the three layers and vascularization processes (Table 1). We performed a U-shaped skin incision, which allowed good vascularization of the scalp flap. All the 5 layers of the scalp: skin, the subcutaneous layer, galea, loose Areolar layer, the temporal muscle along with the periosteum layer were dissected in single flap. After drilling a burr hole, we used suction to evacuate some hematoma to relieve further pressure. A tempo-parietal craniectomy was performed with a large amount of saline solution to avoid high temperature. After evacuation of the extradural hematoma, the bleeding meningeal artery was coagulated. Along the edges of craniectomy, numerous tack-up stitches from the dura to the surrounding pericranium and galea were used. The dura remained intact.

3.4. Spontaneous bone formation >< pregnancy, uses of calcium and vitamin D3

In pregnancy, there is an increase in level of steroid hormones. Progesterone rises in the first semester followed by increase in estrogen and prolactin in the second and third trimesters [17]. Estrogen is proved to facilitate bone formation through cytokine levels [18]. Furthermore, increase in cardiac output and stroke volumes in pregnancy assists in bone healing by providing hormones and cellular factors at the

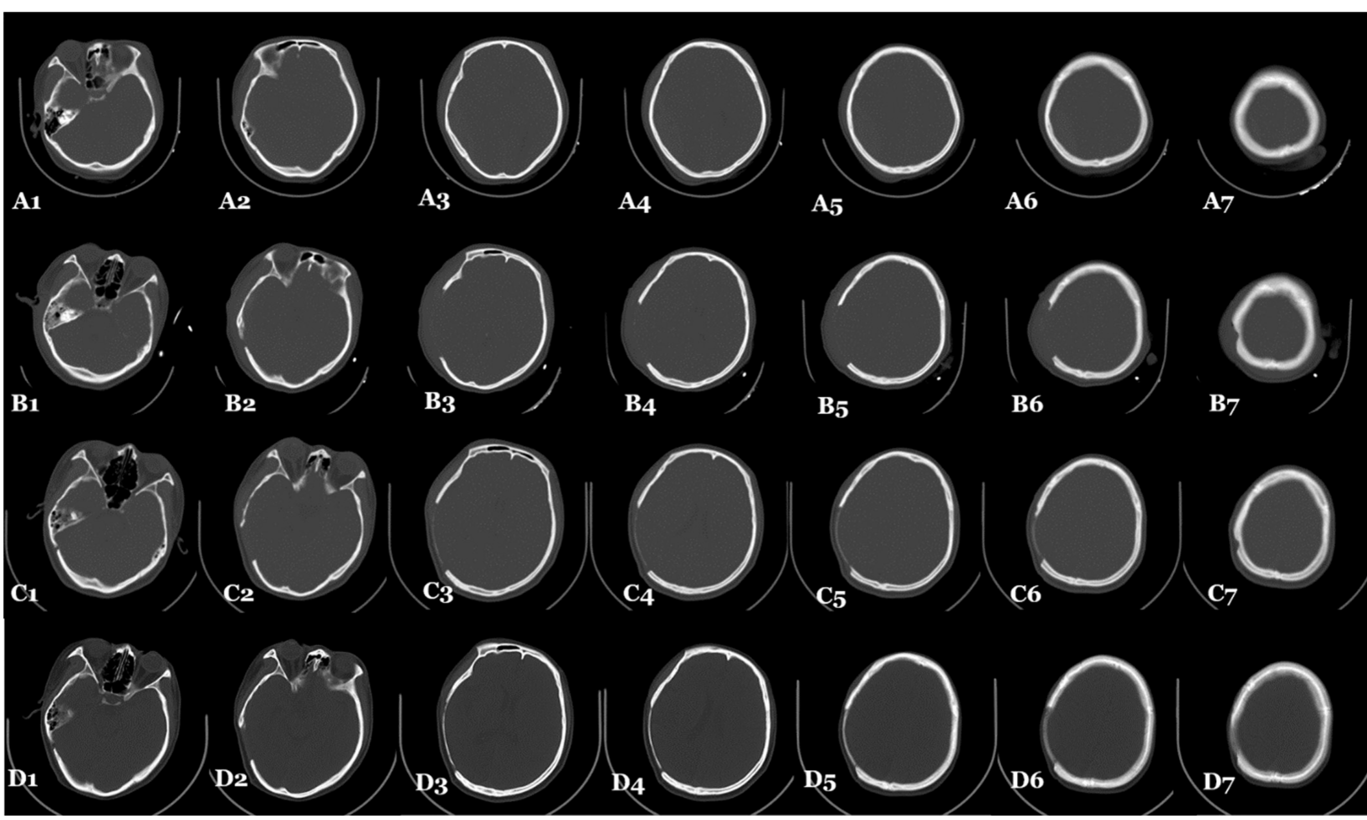

Fig. 2. Bone window.
A: 1–7: The preoperative CT scan.
B: 1–7: The 3 days post-operative CT scan showed no mass effect with bone flap removed.
C: 1–7: The 7 weeks post-operative follow-up CT scan showed bone remodeling in the craniectomy region.
D: 1–7: The 23 weeks post-operative follow-up CT scan showed ossification in the craniectomy region.

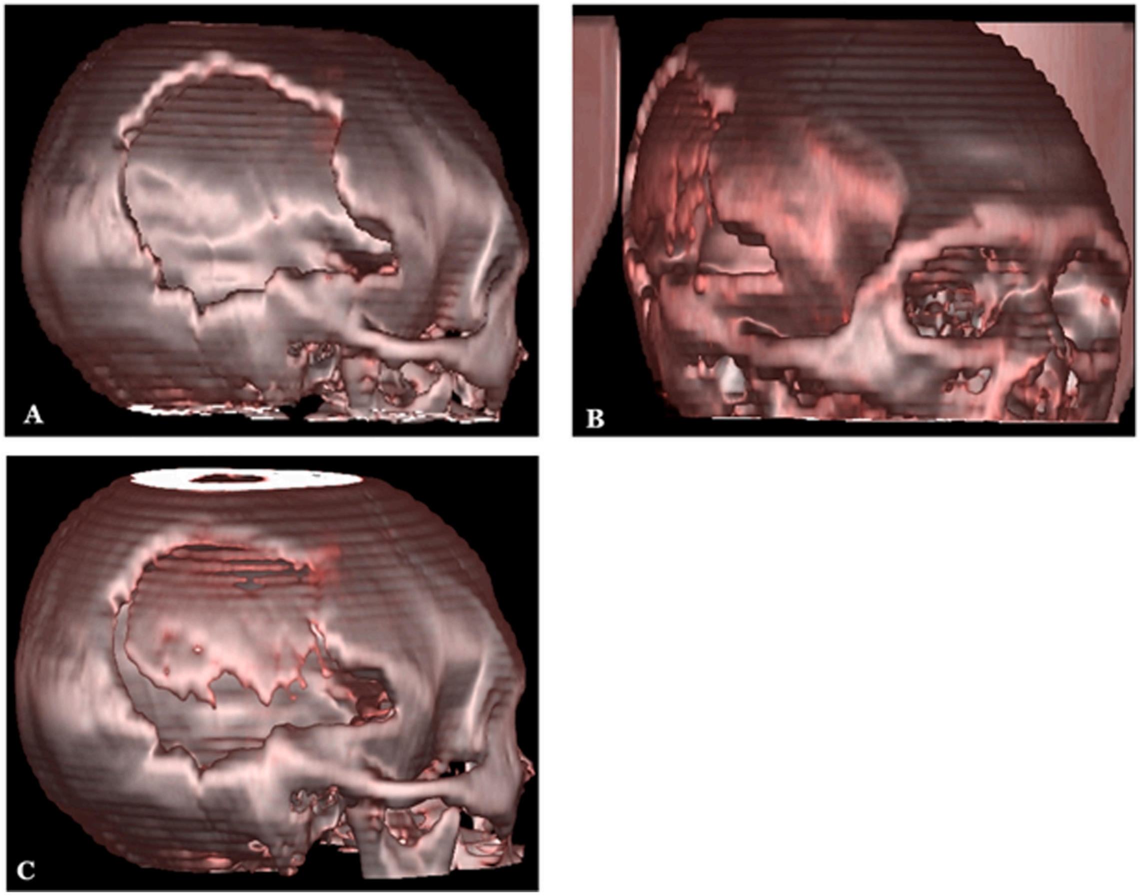

Fig. 3. The 3D reconstruction image.
A: The 3D image 3-day post-operative shows a craniectomy region.
B: The 3D image 7 weeks post-operative shows bone remodeling in the craniectomy region.
C: The 3D image 23 weeks post-operative showed ossification in the craniectomy region.

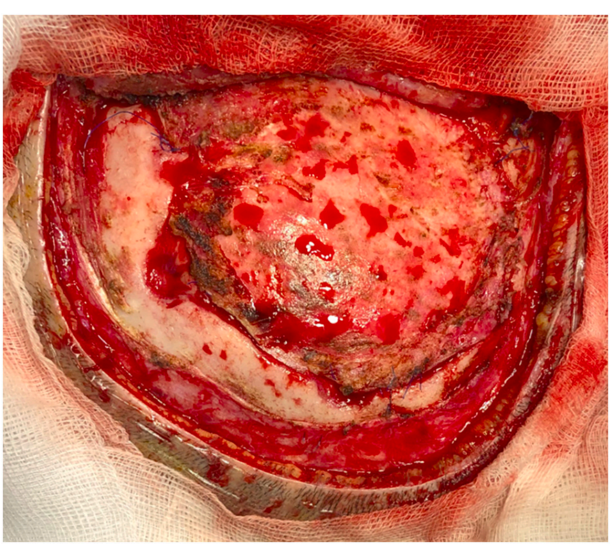

Fig. 4. Intra cranioplasty operation image shows bone formation at the craniectomy site.

craniectomy site [19]. Our patient also took oral vitamin D3 and calcium (200UI Cholecalciferol and 125 mg Calcium everyday) during her pregnancy, which contributes to bone formation [20].

Table 1
Surgery technique and effects on spontaneous bone formation.

Procedure	Justification of procedure
U-shaped skin incision	Respect vascularization
Single scalp flap	Respect the periosteum
Single burr hole	Respect diploe
Craniectomy with a large amount of saline solution to avoid high temperature	Respect diploe
Peripheral tack-up stitches using surrounding pericranium and galea instead of drill holes around the perimeter of the craniotomy	Respect diploe
Coagulation of the bleeding meningeal artery, no dura opening	Respect dura

4. Conclusions

Diagnostic imaging in traumatic pregnant patient is often postponed for the concern of fetus exposure to radiation. Traumatic pregnant patient with possible head trauma should be transferred to a center with expertise in neurotrauma and obstetrical care. Spontaneous cranial bone regeneration following craniectomy in adult is rare. Surgery techniques and hormonal changes in pregnancy may contribute to this phenomenon. In case of extensive spontaneous bone regeneration, cranioplasty is unnecessary.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Research registration (for case reports detailing a new surgical technique or new equipment/technology)

Not applied. This was not a first time a new surgical technique or new equipment/technology was used.

Ethical approval

The study was approved by the Research Ethics Committee of Hanoi Medical University. The procedures used in this study adhere to the tenets of the Declarations of Helsinki.

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CRediT authorship contribution statement

Dat Tran: Conceptualization, Methodology, Investigation, Writing - review & editing, Supervision.

Hung Thanh Chu: Conceptualization, Methodology, Investigation, Writing - original draft, Writing - review & editing, Visualization.

Tam Duc Le: Conceptualization, Methodology, Investigation, Writing - original draft, Writing - review & editing, Visualization.

Tuan Anh Le: Visualization, Writing - original draft, Writing - review & editing.

Ha Dai Duong: Conceptualization, Resources, Supervision.

He Van Dong: Conceptualization, Resources, Supervision.

All authors contributed to the interpretation of the results, discussed the results. All authors read and approved the final manuscript to submit.

Consent to participate

Written informed consent was obtained from the patient and her family members for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Consent to publication

Not applicable.

Availability of data and material

Data is available upon reasonable request and with permission of Viet Duc Hospital. No patient or author details are included in the figures.

Declaration of competing interest

The authors declared no conflict of interest.

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References

[1] F.C. Reith, R. Van den Brande, A. Synnot, R. Gruen, A.I. Maas, The reliability of the Glasgow Coma Scale: a systematic review, Intensive Care Med. 42 (1) (Jan 2016) 3–15, https://doi.org/10.1007/s00134-015-4124-3.

[2] Widdicombe MT, Hanacek J, J. The expiration reflex from the trachea and bronchi. (2008-02-01 2008);doi:https://doi.org/10.1183/09031936.00063507.

[3] T. Thim, N.H. Krarup, E.L. Grove, C.V. Rohde, B. Løfgren, Initial assessment and treatment with the Airway, Breathing, Circulation, Disability, Exposure (ABCDE) approach, Int. J. Gen. Med. 5 (2012) 117–121, https://doi.org/10.2147/ijgm.S28478.

[4] V. Jain, R. Chari, S. Maslovitz, et al., Guidelines for the management of a pregnant trauma patient, J. Obstet. Gynaecol. Can. 37 (6) (Jun 2015) 553–574, https://doi.org/10.1016/s1701-2163(15)30232-2.

[5] Themes UFO. Reconstruction of the Scalp. 2016-03-05 2016.

[6] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, The SCARE 2020 guideline: updating consensus Surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (Dec 2020) 226–230, https://doi.org/10.1016/j.ijsu.2020.10.034.

[7] G.L. Prasad, Steroids for delayed cerebral edema after traumatic brain injury, Surg. Neurol. Int. 12 (2021) 46, https://doi.org/10.25259/sni_756_2020.

[8] González-Bonet Lg, Spontaneous Cranial Bone Regeneration After a Craniectomy in an Adult, World Neurosurg. (Mar 2021) 147, https://doi.org/10.1016/j.wneu.2020.12.056.

[9] B.D. Thombre, A.R. Prabhuraj, Spontaneous bone formation in a large craniectomy defect, Childs Nerv. Syst. 34 (8) (Aug 2018) 1449–1450, https://doi.org/10.1007/s00381-018-3863-1.

[10] J. Mathew, A. Chacko, Spontaneous re-ossification of a large calvarial defect in an older child, Turk. Neurosurg. 18 (4) (Oct 2008) 407–408.

[11] D.A. Hoover, A. Mahmood, Ossification of autologous pericranium used in duraplasty. Case report, J. Neurosurg. 95 (2) (Aug 2001) 350–352, https://doi.org/10.3171/jns.2001.95.2.0350.

[12] S. Debnath, A.R. Yallowitz, J. McCormick, et al., Discovery of a periosteal stem cell mediating intramembranous bone formation, Nature. 562 (7725) (Oct 2018) 133–139, https://doi.org/10.1038/s41586-018-0554-8.

[13] A.K. Gosain, S.A. Gosain, W.M. Sweeney, L.S. Song, M.T.J. Amarante, Regulation of osteogenesis and survival within bone grafts to the calvaria: the effect of the dura versus the pericranium, Plast. Reconstr. Surg. 128 (1) (Jul 2011) 85–94, https://doi.org/10.1097/PRS.0b013e31821740cc.

[14] C.H. Hämmerle, J. Schmid, N.P. Lang, A.J. Olah, Temporal dynamics of healing in rabbit cranial defects using guided bone regeneration, J. Oral Maxillofac. Surg. 53 (2) (Feb 1995) 167–174, https://doi.org/10.1016/0278-2391(95)90396-8.

[15] A.K. Gosain, T.D. Santoro, L.S. Song, C.C. Capel, P.V. Sudhakar, H.S. Matloub, Osteogenesis in calvarial defects: contribution of the dura, the pericranium, and the surrounding bone in adult versus infant animals, Plast. Reconstr. Surg. 112 (2) (Aug 2003) 515–527, https://doi.org/10.1097/01.Prs.0000070728.56716.51.

[16] C. Szpalski, J. Barr, M. Wetterau, P.B. Saadeh, S.M. Warren, Cranial bone defects: current and future strategies, Neurosurg. Focus. 29 (6) (Dec 2010), E8, https://doi.org/10.3171/2010.9.Focus10201.

[17] Hua Chen, H, a QJ, et al. Pituitary apoplexy leading to cerebral infarction: a systematic review, Eur. Neurol. 83 (2) (2021) 121–130, https://doi.org/10.1159/000507190.

[18] H.K. Väänänen, P.L. Härkönen, Estrogen and bone metabolism, Maturitas. 23 (Suppl) (May 1996) S65–S69, https://doi.org/10.1016/0378-5122(96)01015-8.

[19] L.M. Kohlhepp, G. Hollerich, L. Vo, et al., Physiological changes during pregnancy, Anaesthesist 67 (5) (May 2018) 383–396 (Physiologische Veränderungen in der Schwangerschaft), https://doi.org/10.1007/s00101-018-0437-2.

[20] U. Schulze-Späte, T. Dietrich, C. Wu, K. Wang, H. Hasturk, S. Dibart, Systemic vitamin D supplementation and local bone formation after maxillary sinus augmentation - a randomized, double-blind, placebo-controlled clinical investigation, Clin. Oral Implants Res. 27 (6) (Jun 2016) 701–706, https://doi.org/10.1111/clr.12641.