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Role of Fresh Cadaver Practice and Jesus Prayers on Daily Base in Innovations of a Difficult Neurosurgical Operation (Craniopagus Separation)

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Abstract

Purpose: Challenging cases in neurosurgery require experience, which is gained by operating on a number of similarly difficult cases. However, even in large population centers, there are extremely rare cases, such as craniopagus twins. In these instances, other case-specific solutions are required which were improved in the course of fresh cadaver practice on daily base. Methods: During a 20-month preparation period by fresh cadavers the surgical strategy was developed step by step, comprising five neurosurgical ideas, facilitated by deep Jesus prayers as a spiritual source. Possible causes of postoperative complications that occured on 33d postoperative day were also analyzed. Results: During surgery, these nuances proved to be useful, which is also reflected by the postoperative clinical results. One of the twins advanced to a GOS score of 5three months after the surgery. The other twin, despite nonsurgical septic complications, continued to progress well, but on postoperative day 33 for seemingly unknown reasons, she suffered a severe cerebral hemorrhage, which significantly delayed her rehabilitation. A thorough revision of the whole process much later revealed a pitfall that could have a role in this complication. Conclusions: The challenging neurosurgical separation of the craniopagus twins was carried out successfully with the help of five novel neurosurgical refinements, which were found during 300 fresh cadaver practices. Mistakes can serve as a lesson. We hope that the procedures developed and the knowledge gained can be applied in the future.

Keywords

Craniopagus, Conjoined Twins, Fresh Cadaver Practice

1. Introduction

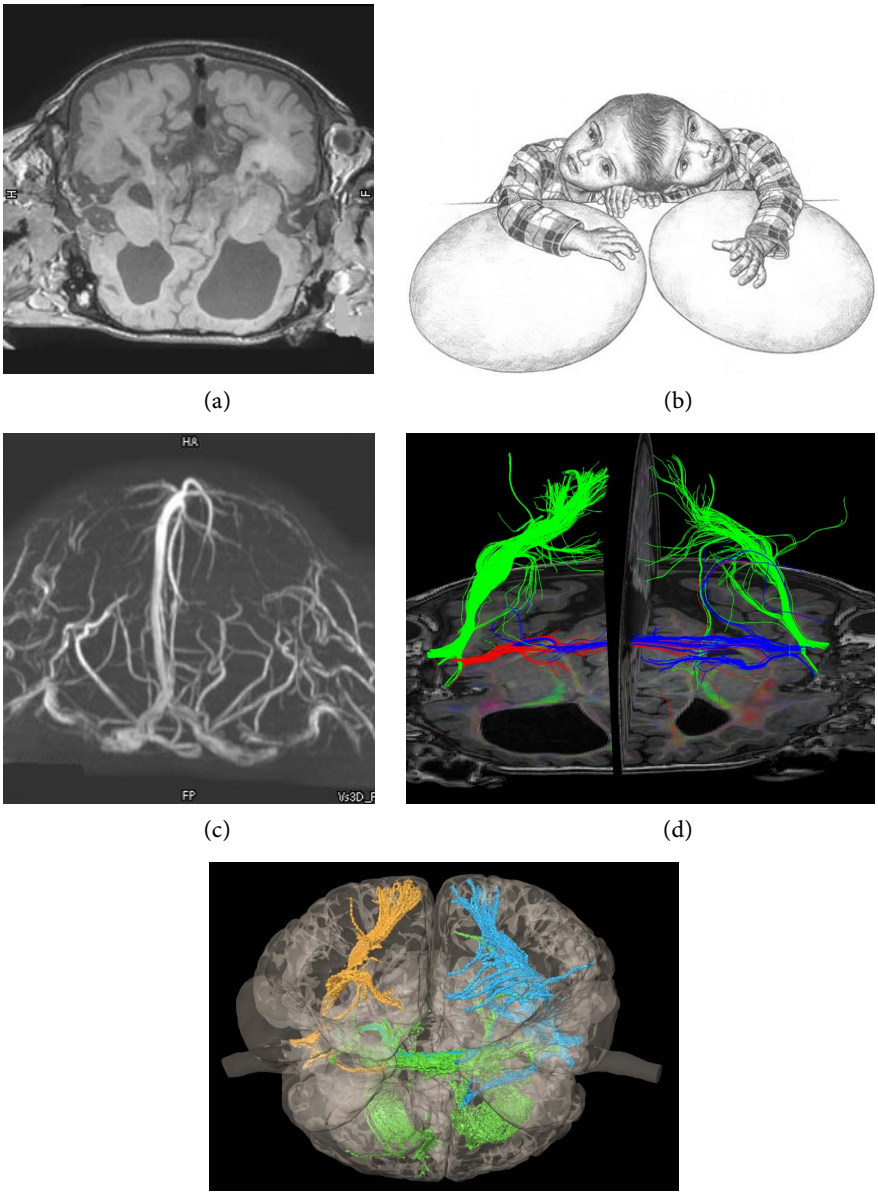
After publishing a previous short video article [1] we were encouraged by reviewers to explain in more detail the preparation for the major surgery, the technical innovations used and to analyse in more detail the possible causes and lessons learned from the severe late bleeding complication. In this article, we do just that, so that better results can be achieved in future similar cases.

The challenges posed by rare pathologies require knowledge and experience, which can only be acquired through the study and operation of a number of similar cases. However, there are some cases that are extremely rare even in large population centers, [2] [3] [4] such as the separation of craniopagus twins. In these instances, other unique, case-specific operative solutions are required, which can be developed step by step organically. The Jesus Prayer written in the title was the greatest help and spiritual strength during our preparation (300 autopsy exercises) and during the 24 hours of surgery. It was a real experience that the scientific ideas that we are talking about in the article were born during Jesus prayers and also the serious professional decisions during surgery. This was the truth, and it is precisely in science that we must not deny the facts, because science, including psychology, is also searching for the truth, for the movements of the soul and its role in intellectual, even scientific, decisions.

2. Case Description

A case of 16-month-old, total vertical craniopagus twins was presented in November 2017 to our group of volunteer medical specialist doctors from Hungary in Dakha. An article on the separation of these specific Bangladeshi craniopagus twins was published within some months after separation [5], with special emphasis on the plastic surgical, international organisational and legal aspects of the case. However, the article lacked a detailed description of the 26-hour-long neurosurgical portion of the final separating surgery itself, which was the most challenging part of the operative series and did not provide information about 20 months long preoperative neurosurgical preparation either. Analysis of severe complications was also missed in the article due to the relatively short follow-up. A short video article was published later [1] as a supplement to the 24-hour neurosurgical intervention but this could not be complete due to publication restrictions. We have now clarified the lessons learned to publish the results of our analytical work.

Our case showed a total vertical, type I conjointment (**Figure 1(a)**, **Figure 1(b)**). The different venous structures were confirmed by MRA venography. The



(a) (b) (c) (d) (e)

Figure 1. (a) MRA picture about the common venous structure; (b) MR picture about the interdigitated part of the brains; (c) Drawing about the twins before operation; (d) Interthalamic neural pathway; (e) Tractography.

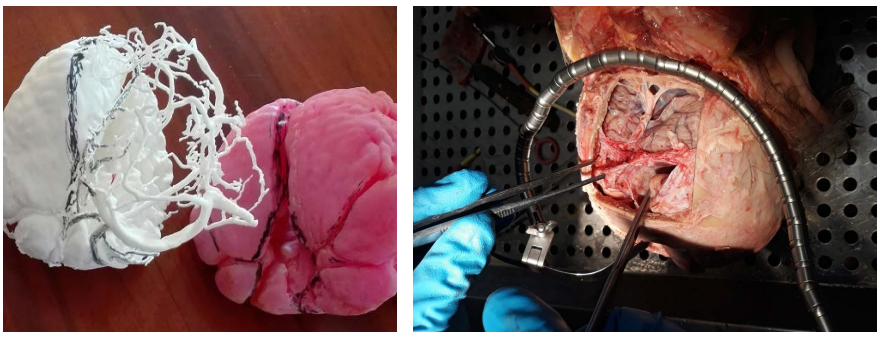
twins shared a common superior and inferior sagittal and a circumferential sinus (**Figure 1(c)**). Multiple venous bypassing seemed indispensable which is technically even more demanding, and implies very high surgical risks, such as in the case of Laleh and Ladan [6]. The next feature, that required consideration is the relationship of the two brains to one another. The MRI showed a close attachment of the two brains dominantly at their parietal and occipital lobes: on one side, a flattened cortical surface was expected to be found at these lobes of corresponding hemispheres. However, on the other side, a much more pronounced interdigitation was found between the hemispheres. Moreover, an'interthalamic

bridge' was also found for both twins (**Figure 1(d)**, **Figure 1(e)**). The dissection and relatively atraumatic separation of these common brain surfaces—the regions of pronounced interdigitation in particular—also posed an enormous surgical challenge, namely, to remain relatively speedy to reduce the risk of infection, and at the same time to remain as atraumatic during dissection of the adherent surfaces as possible for functional reasons. In addition, a gradual slow opening of the hemispheres *i.e.* the skull—halves during the separation of these interdigitated surfaces would also promote this particularly long surgical phase. During further surgical planning, after the circumferential separation of the two skulls and partial separation of the two brains on one side, a 180-degree turn of the twins—at least in theory—seemed necessary. At this point, we encountered yet another issue to consider how to hold the two still-connected hemispheres in the skull halves together during and after the 180-degree rotation of the bodies [7].

We tried to cover tha gap of our knowledge and we presented a way to increase knowledge and experience directed at this specific case by identifying novel neurosurgical steps that were necessary to overcome the above technical challenges. We also tried to explore the possible causes of late postoperative complications.

3. Methods

3D-printed copies helped to identify the crucial surgical steps and to assess their risks (**Figure 2(a)**). We developed a surgical strategy comprising neurosurgical steps during a 20-month preparation period. The steps of patient positioning, the turning of the twins in an operational setup using a doll model were also rehearsed by the surgical team several times before the final separation. The critical surgical steps were practiced on fresh cadavers, and doll models and then successfully applied during the actual separation surgery. (**Figure 2(b)**). During the preoperative fresh cadaveric studies, we continuously focused on findingmicrosurgical and microanatomical situations that will be similar to what we can expect in the course of the final surgery (interdigitations, confluence of venous sinuses in regular cadavers).



(a) (b)

Figure 2. (a) Fresh cadaver practice to modelize the living situations at confluens sinus; (b) 3 D model for practicing and understanding the brain and vascular structure.

4. Results

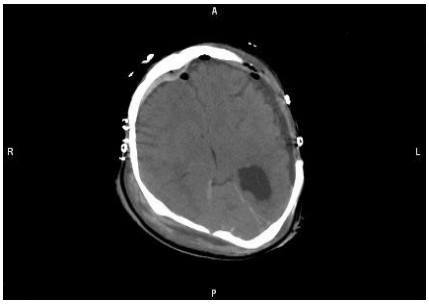
The final separation of the 3-year-old conjoined twins took place 22 months after an initial presentation on August 1 and 2, 2019, in Dhaka, Bangladesh, Combined Military Hospital. Details of the 26-hour-long neurourgical part and the details of post-up course (CT scans) have not been published in the written article so far either. Promising CT images were obtained straight after surgery, which demonstrated cortical oedema and a not extensive ischemic lesion. (**Figure 3(a)**, **Figure 3(b)**). The most frequent anticipated surgical complications (meningitis, wound infection, and cerebritis), did not occur throughout the entire postoperative period. In Twin B however, serious intercurrent infections (bacterial or viral septic conditions, pneumonia, pancreatitis) occurred, causing a serious life-threatening condition. In Twin A similar infection disappeared and resolved within two weeks. This condition regressed more slowly in Twin B, although on postoperative day 30 she came close to being extubated, woke up, and was moving her limbs on both sides, in spite of right hemispheric venous haemorrhage. Three days later, on day 33, twin B experienced a severe intracerebral hemorrhage in the left hemisphere. (**Figure 3(c)**, **Figure 3(d)**) Life-saving partial removal of the hematoma was performed immediately.

No such alterations were detected on the CT scan of Twin A (**Figure 3(e)**). The artifact caused by Onyx made it difficult to interpret the CT findings accurately. Twin A, who had better clinical status from the beginning, experienced a good recovery, with a GOS (Glasgow outcome scale) of 5 at 3 months. and her rehabilitation is ongoing. Due to the skin deficiency, her cranioplasty was performed 15 months after the separation, but 15 months later it should have been removed again because the skin deficiency occurred again. Twin B still shows a GOS score of 3 due to the severe left-hemispheric cerebral hemorrhage; her rehabilitation is ongoing as well, and her cranioplasty was performed 4 months after separation. She is able to stand for now with help for a few minutes, has emotional reactions, smiles at her family members, and resonates with their mood. Her attention can be drawn, but her awareness is far from that of the level of an average four-year-old child. The fact that her psycho-motor skills remain at a GOS score of 3 raises the question of whether she has reached a cognitive plateau, given the massive extent of the brain tissue destruction detected on CT (**Figure 3(f)**) but neuroplasticity in this age can develop her functions. (**Figure 4**)

5. Discussion

1) The first thought was that multiple sequential balloon-occlusion tests could be used to precisely assess the collateral venous cerebral blood flow. The procedure was carried out stepwise. Endovascular occlusion of the common sinuses was successful. [1] (**Figure 5**). [7] [8].

2) We focused on modeling the critical surgical steps namely, dissection and separation of the interdigitating surfaces (**Figure 1(b)**) which were expected to occur during surgery. This modeled microsurgical separation of interdigitations,



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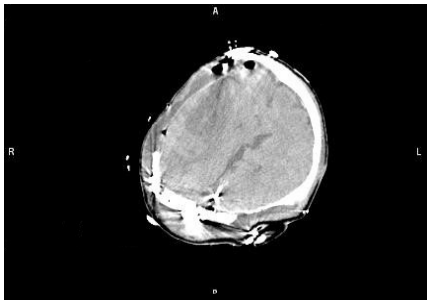

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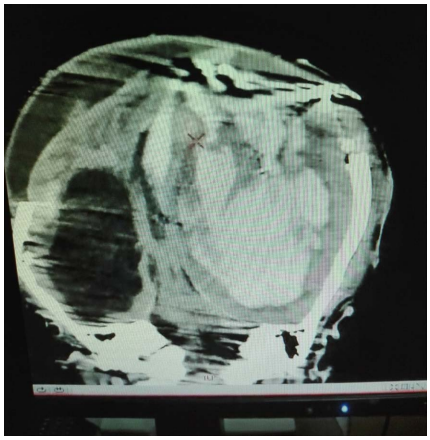

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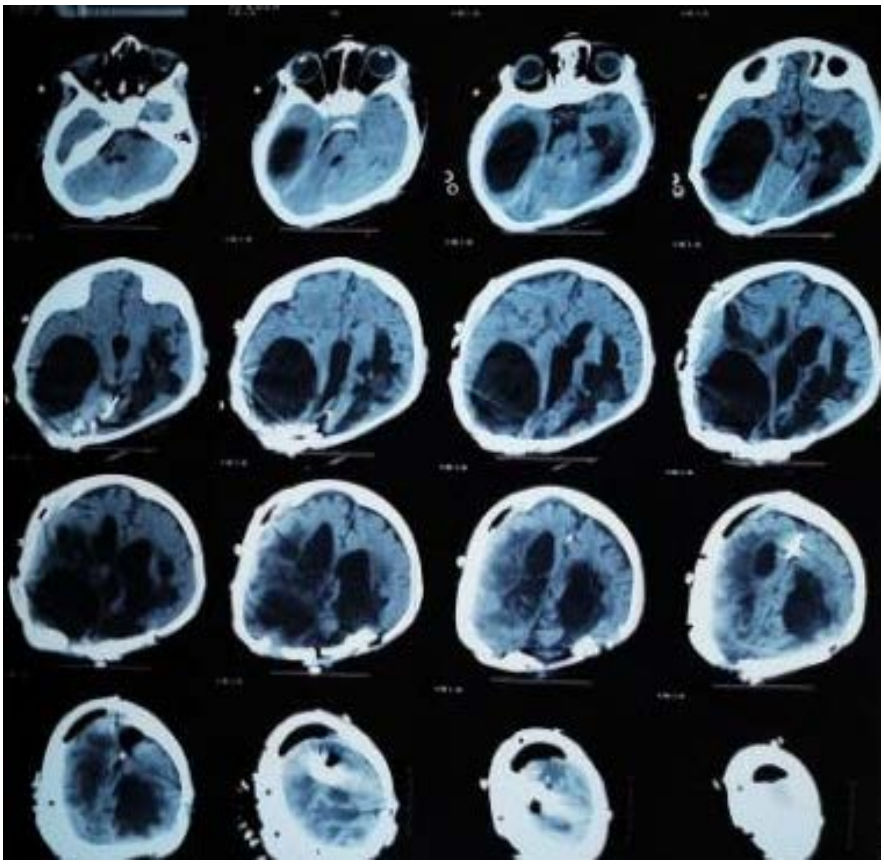

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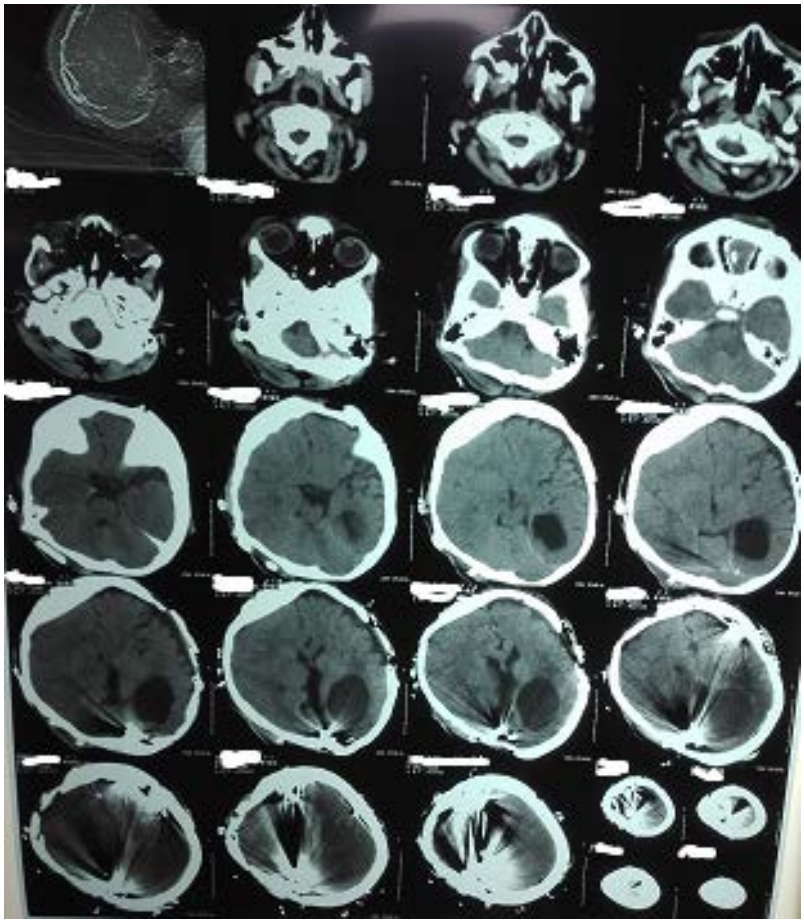


(f)

Figure 3. (a) Post op. 24 hours CT of Twin A. Distended posterior ventricular horn visible as in preop.images; (b) Post op. 24 hours CT of Twin B. Signs of venous infarction visible on right side hemisphere. The left hemisphere was intact. Distended right ventricular posterior horn was also verified on other slides as preoperatively; (c) CT Twin B. on 33. post op day after hemorrhage in the left hemisphere. (axial view); (d) Twin B. on 33. post op day after hemorrhage in the left hemisphere. (coronal view); (e) CT Twin B 8 months after the enormous bleeding occurred on 33. post op. day in the left hemisphere. The right distended occipital ventricle was visible as on preop MRI; (f) CT Twin A. 15 months post op. CT. Cranioplasty was performed. Patient status GOS 5.

Figure 4. 3 months after the surgery.

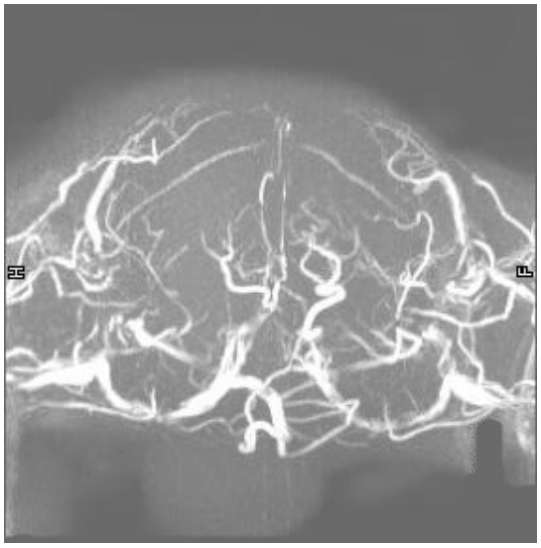


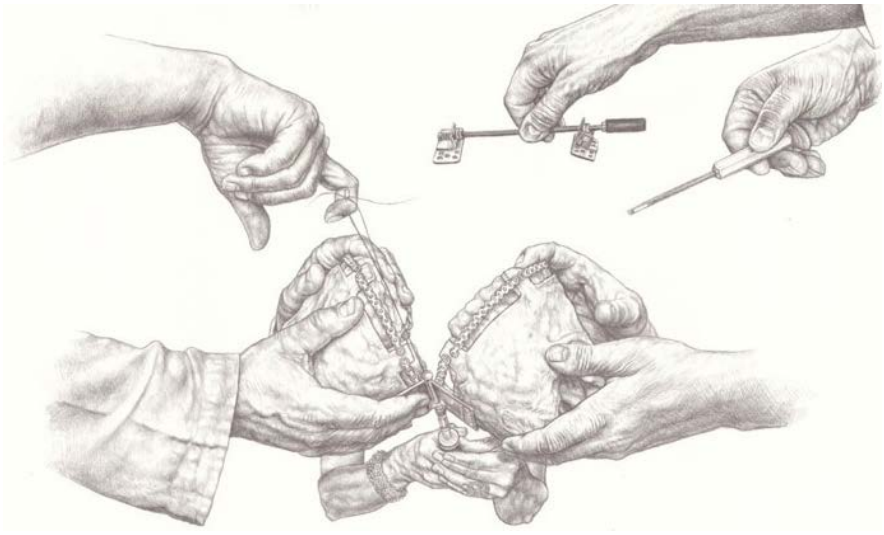
Figure 5. MRA after the endovascular occlusion of common veins.

which were expected to occur during surgery, was practiced on fresh cadavers more than 300 times. These daily exercises were composed of two significant steps: during each cadaveric dissection, we looked for sulci on the surface of the brain where the gyri were particularly interdigitated. At the same time, we performed Sylvian fissure split more than 300 times, during which we were looking for such anatomically abnormal Sylvian fissures, where the gyri of frontal and temporal lobes were interdigitated into one another. Nevertheless, after the rotation of the twins [1] we noticed an unexpected adhesion loss in the area of the interdigitating gyres that was also detected in the preop MRI. (**Figure 1(b)**) demonstrates an unexpected adhesion after inverting the twins.

This phenomenon was facilitated by CSF loss during the surgery. The difficulties of separation of this part were demonstrated in the video article. [1]

3) Fresh cadavers also provided excellent practice in performing venous bypasses (the detached vein of Labbe replantation to sinus). Similarly, at sites of the confluence of sinuses, initial steps of the expected microsurgical detachment of venous walls and sinuses were practiced during cadaveric exercises. (**Figure 2(b)**) At the final separation, after detaching the remaining common occipital venous complex [1], we did not experience abnormal bridging vein expansions. Considering, that a previous endovascular separation of the venous system was accomplished many months prior to this final surgery, at this point, additional, venous bypass did not seem to be necessary.

4) The challenge of holding the two still-connected hemispheres in the skull-halves together after the 180-degree rotation of their bodies, was addressed by our novel "hinge technique" (**Figure 6(a)**, **Figure 6(b)**). Several options were considered when planning the intraoperative fixation during the 180-degree turn of the twins. The hinge—technique provided a sufficient degree of motion between the two skull-halves.



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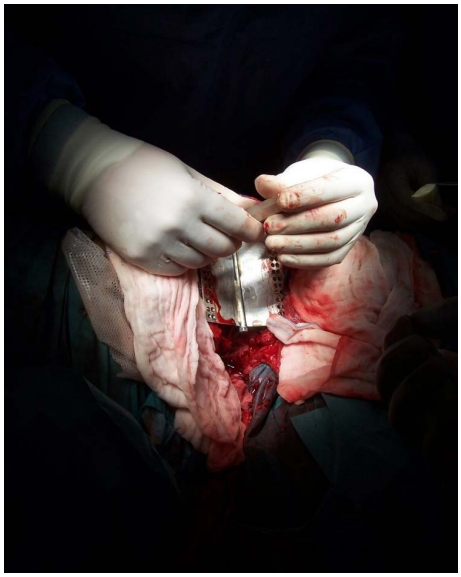

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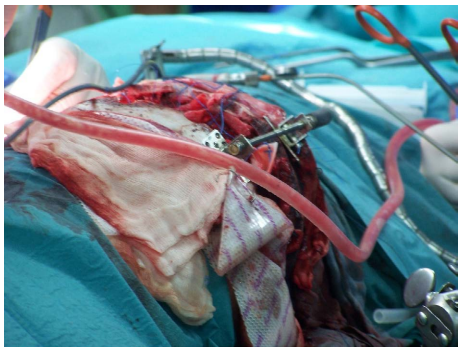


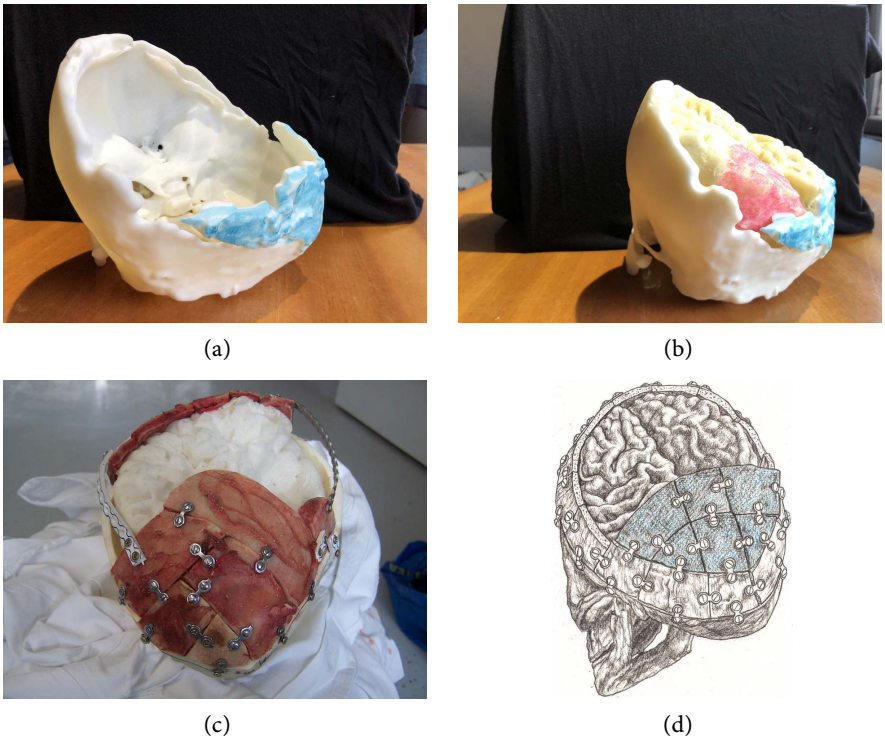
(c)

Figure 6. (a) Drawing of the hinge and the distractor. Moments before the separation as shown in the drawing. The preparation of surgical drawings was of particular help, to be able to visualize the surgical steps; (b) Intraoperative photo of the hinge; (c) Intraoperative photo of distractor.

5) The idea of the hinge together with the custom-made distraction—device proved to be ideal because it enabled a gradual opening of the operative field of interest during the critical microsurgical separation of the corresponding interdigitated cerebral hemispheres. Continuous opening of the two hemispheres was performed, millimeter by millimeter, by utilizing a novel custom-manufactured distraction device (**Figure 6(a)**, **Figure 6(c)**).

We also analyzed the complications that occurred on the 33 postoperative days.

Based on our experience, we hypothesize that the suspected cause of the severe left hemispheric hemorrhage (**Figure 3(c)**, **Figure 3(d)**) was a chronically low platelet count due to, surgically unrelated, tropic viral and bacterial septic complications. Thrombocytopaenia, however, might not be the only reason for the haemorrhage, because the platelet count was within the lower limit by the time the CT revealed the haemorrhage on 33 post op day. Another additional possible ethiological factor of this late complication was the recurring or constant shear—stress and tactile microtrauma of the brain due to lack of sufficient reconstruction of the posterior part of the skull (**Figure 7(a)**, **Figure 7(b)**) Intracerebral haemorrhage is a well-known complication after decompressive craniectomies [9]. We have also experienced such complications after huge bilateral fronto-temporo parieto-occipital decompressive craniectomy, especially on



(a) (b) (c) (d)

Figure 7. (a) Preop model on 3D doll about the planned cranioplasty in twins; (b) Preop the planned cranioplasty in model (drawing) about twins; (c) 3D model about the real situation in twin B after separation. The reconstruction of the posterior part of the skull probably was not enough sufficient, against tactile recurrent microtraumatizations in supine position. I could be the main cause of hemorrhage; (d) 3D model with the brain hemorrhage.

the parieto-occipital region. The reconstruction of the posterior part of the skull was also an important issue during the surgical planning. During the fresh cadaver and 3D model exercises we prepared ourselves for a single-session partial cranioplasty using autologues skull (**Figure 7(c)**, **Figure 7(d)**). During the final separation, this cranioplasty could not be achieved perfectly because of unsatisfying interdisciplinary coordination within the hungarian team, mostly due to misunderstandings and fatigue. This part of the operation was not performed by participants who practiced the most during the cadaver-, 3D-, and doll-model exercises. We did not recognize the threat of insufficient bone replacement in the immediate postoperative stage. This was not in focus because of the ongoing severe septic condition. Later on, in twins, a protection helmet was used until the cranioplatsty.

6. Conclusion

The continous fresh cadaver practices were the base of above mentioned scientific innovation. The spiritual power originated from Jesus prayers and facilitated the birth of creative scientific innovations. The great lesson of the case is that it is not enough to have a good professional idea facilitated by deep prayers if the interdisciplinary surgical planning is not comprehensive enough. After the initially very successful-looking surgery, more thought should have been made about possible late complications. Our possible theory by an early cranioplasty the late cerebral haemorrhoage might have been avoided. Compared to the craniopagus separation operations of the last 50 years, the operation was successful. Avoiding the shortcomings identified in the analysis might have led to an even better result. In the almost four years that have elapsed so far, the B twin has not improved a lot. Further progress is not excluded, given the neuroregenerative capacity of the children. We hope that in the future this action, alongside the neurosurgical innovations in the separation of craniopagus twins discussed in this article, can continue to find application.

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Ethical Approval

Under Bangladeshi law, there was no requirement to request a statement of approval from the IRB/Ethics Committee. The consent for the separation surgery of 3-year-old children was signed by the parents after detailed information. The parents provided permission to publish images.

Ethical Considerations

To perform such an intervention before each phase, a medical team reviewed the planned actions with the parents who were supported by native Bangladeshi doctors present in Hungary. They provided translation and explanation and could lead and accept a detailed consent form. We also took into account the religious beliefs of the parents and the bioethical aspects of the operation. Preliminary internal medicine expert examinations also suggested that the twins theoretically could live separately, because they did not have cross-dependent organ functions.

Conflicts of Interest

The authors declare no conflicts of interest regarding the publication of this paper.

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