

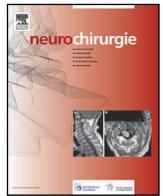


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Craniosynostosis: State of the Art 2019

Virtual 3D planning of osteotomies for craniosynostoses and complex craniofacial malformations



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ABSTRACT

Introduction and objectives. – Computer-assisted surgery has been more and more widely used in craniofacial surgery in recent years. It is useful in many situations: stereolithographic models, surgical simulations of osteotomies and bone repositioning, and cutting guides and customized implants. The present paper argues that computer-assisted surgery is particularly useful in complex cases such as rare malformations, or to address the sequelae of previous surgeries. The various advantages of the technique are emphasized from a surgical and from a teaching standpoint.

Materials and methods. – Forty cases of various computer-assisted surgeries were analyzed, allowing a comprehensive review of outcomes in cases such as craniosynostosis, complex craniosynostosis, hypertelorism, craniosynostosis sequelae and cranio-facial and orbital trauma.

Results. – Results were promising in all of the cases reviewed, except in a few cases for which computer-assisted surgery with cutting guides may not be necessary. In these specific cases, the pedagogical input is nevertheless interesting for residents and students.

Conclusion. – Computer-assisted surgery is revolutionizing the surgical approach to complex craniofacial malformations, as well as easing management of less complex ones. It is likely that in the years to come this technique will supersede previous ones. However, using this technique implies being willing to rely on a non-human device. We need to consider computer-assisted surgery as a tool that can change surgical practices. The surgeon can rely on it, yet nothing will replace his/her eye and experience. It is the combination of both this experience and the appropriate use of computer-assisted surgery that, ultimately, leads to successful surgery.

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1. Introduction

Today, computer-assisted surgery (CAS) in maxillo-facial contexts is mainly involved in 3 cases: orthognathic surgery [1–3], zygoma osteotomies and repositioning [4–6] and fibula free flaps [7–9]. Its application in craniofacial surgery, more specifically in craniosynostosis, is very recent and few teams have been using it so far [10–13]. In this article, we are going to focus on craniofacial malformations. After five years' use of CAS in about forty patients, it is about time to take a closer look at the various clinical situations that have been addressed successfully with this technique. The purpose of this article is to provide some insight on the condi-

tions under which CAS can, and even more should be, be selected as the best option, and on the multiple situations that can be now addressed thanks to CAS and have been highlighted in recent years. In this perspective, three contexts of use are introduced: first, stereolithographic models; second, surgical simulation of osteotomies and bone repositioning; and third, cutting guides and customized implants. The presentation of all of these cases can improve understanding of the reliability and usefulness of CAS.

2. Stereolithographic models

The first field of application of CAS was stereolithographic models [14–16], obtained via 3D printing. Briefly, reconstruction uses a DICOM CT scan. First, the Mimics 19.0[®] software (Materialise, Leuven, Belgium) transfers the data from DICOM into a 3D format. Then the 3D information is printed into a polyamide device. It is possible to obtain a very reliable outcome, which enables very accurate

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Fig. 1. Stereolithographic model of a Pfeiffer syndrome with clover-leaf skull.

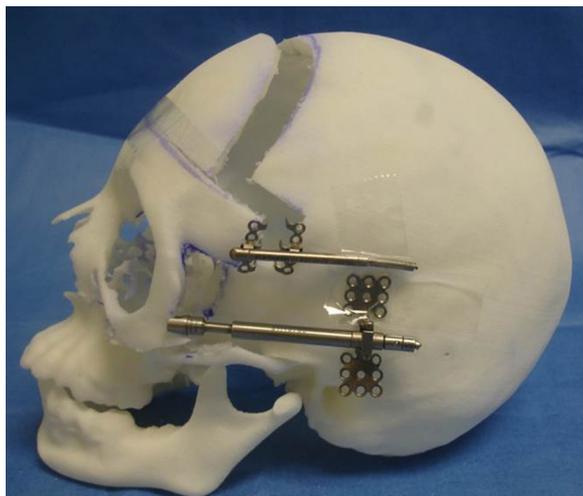


Fig. 2. Simulation of osteotomies in an orbito-frontal monobloc advancement and simulation of internal distractor positioning.

analysis of the malformation [14] (Fig. 1). With these stereolithographic models [17,18], the osteotomies (Fig. 2) to be performed can be simulated. One other major advantage is the autonomy of the surgeon in the process of surgical planning, because there is no need for external help at this stage from clinical engineers.

Stereolithographic models also provide a good opportunity for teaching residents complex cases of craniofacial osteotomy. However, CAS has progressed so much in recent years that its use for printing stereolithographic models may no longer be the best option. Other techniques are replacing these models more satisfactorily: surgical simulations, and the cutting guides and customized implants that derive from them. These techniques address some of the major drawbacks of stereolithographic models: lack of

measurement precision during the surgery simulation phase, difficulties related to cutting the polyamide, which can result in heating of the surgical handpiece, and the impossibility of a second chance if the polyamide has been badly cut, which can occur every now and then.

3. Craniosynostoses

CAS is suitable for most cases of craniosynostosis. Of course, it is not used for sagittal synostoses with an H craniectomy technique, but it can be helpful for other techniques [11]. CAS can be appropriate in case of craniosynostosis with bandeau remodelling, in case of oxycephaly or to address the sequelae of craniosynostosis in the elderly [19].

The main interest of CAS in primary craniosynostosis surgery with orbital bandeau removal and new forehead construction is the possibility of pre-determining the best place for removing the new forehead on the skull, and the best shape.

Obviously, CAS is not mandatory for craniosynostosis correction. It is an additional tool, which allows very precise calculation of the optimal curves and positioning of the future forehead, which is unique in terms of both precision and timesaving. The calculation process takes into account all of the theoretical possibilities on the skull, and finally recommends the best option.

In the following sections, two cases of craniosynostosis surgery are detailed, both of which involved CAS.

3.1. Plagiocephaly

During the presurgical step, the surgeon worked hand in hand with the clinical engineers, using CMF Planner® 3.0 software (Materialise, Leuven, Belgium). This stage simulates the surgical procedure, based on virtual 3D model specifically designed for each patient. The clinical engineers design the various virtual osteotomies and the repositioning following the surgeon's directions. The surgeon can then visualize the osteotomies (cutting angles, resection sizes), bone repositioning and final expected result.

In case of plagiocephaly, the supraorbital bandeau is designed and the cutting plan is decided upon. Then, a simulation of the retruded forehead removal is performed. The most appropriate place for removing the future forehead is then defined (Fig. 3). After all these preliminaries, it is time for simulation of the future positioning of the osteotomies (stage 1). The software defines the new position of both the bandeau and the new forehead (Fig. 4).

Once the surgical simulation has been performed, the next stage (stage 2) is to build the cutting and repositioning guides. This stage uses 3-matic®-11.0 software (Materialise, Leuven, Belgium). Again, with the help of the clinical engineers, the first task is to calculate a very precise cutting guide that will be used by the surgeon as the first step during surgery (Fig. 5). Second, the software aggregates the various skull parts that are to be removed following the instructions of stage 1, thus creating a repositioning guide that takes into account the shapes of new bone parts as well as their respective new positions. A repositioning guide acts as an external structure that holds all the parts of the new skull together like a jigsaw puzzle (Fig. 6). It is fixed onto the remaining parts of the original skull. This stage is the most important one, as it secures the precision of the final outcome.

In the specific case of plagiocephaly, the next stage (stage 3) is in the operating room. The first task to be performed by the surgeon is to position the cutting guide. The exact contours of the cutting areas are marked with the burr. After marking, the cutting guide is removed. The bandeau, the retruded forehead and the new forehead can then be sectioned according to the marking (Fig. 5). Then

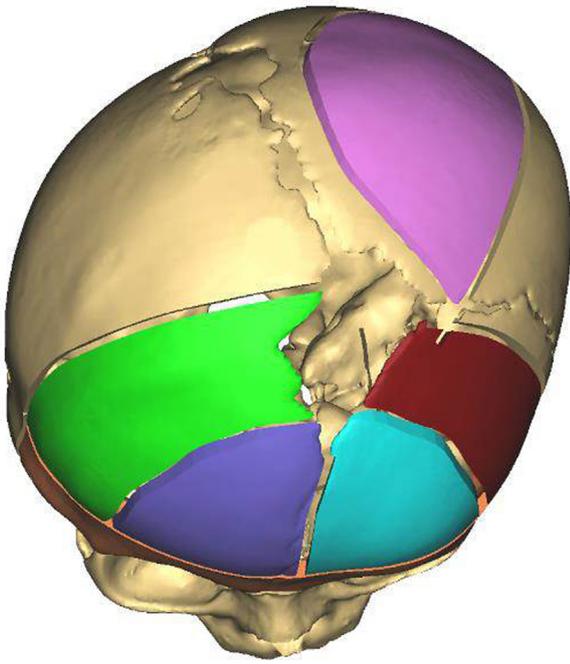


Fig. 3. Top view of the bandeau, forehead and new forehead osteotomies simulations for a plagiocephaly.

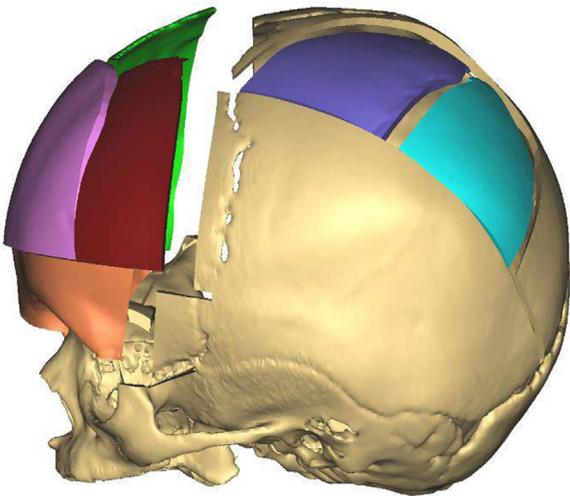


Fig. 4. Simulation of the new position of the bandeau and the new forehead.

the bandeau and the new and old forehead can be remodelled to conform to the repositioning guide. For instance, the retruded forehead can be cut into two pieces that will in turn be placed at the back of the skull from where the new forehead was taken. There need to be two pieces from the old forehead to overcome the issue of the repositioning angles at the back of the cranium.

At this stage, the various skull parts are placed within the repositioning guide, which we call a “repositioning spider” (Fig. 6). The new forehead is fixed to the bandeau with Maxon® 2.0 stitches. Thanks to the repositioning guide, the retruded side can be advanced for a predetermined distance (2 cm). Then, the bandeau can be fixed to the skull with absorbable plates. Then, the whole “spider guide” is positioned on the remaining parts of the original skull. To be sure that the repositioning is correct, it needs to fit perfectly (Fig. 6); if not, the surgeon needs to look for a new positioning angle that will fit and ensure that everything is in the right place. Finally, the spider guide can be removed and the scalp closed.



Fig. 5. Surgical top view of the cutting guide for marking the contour of the future osteotomies.

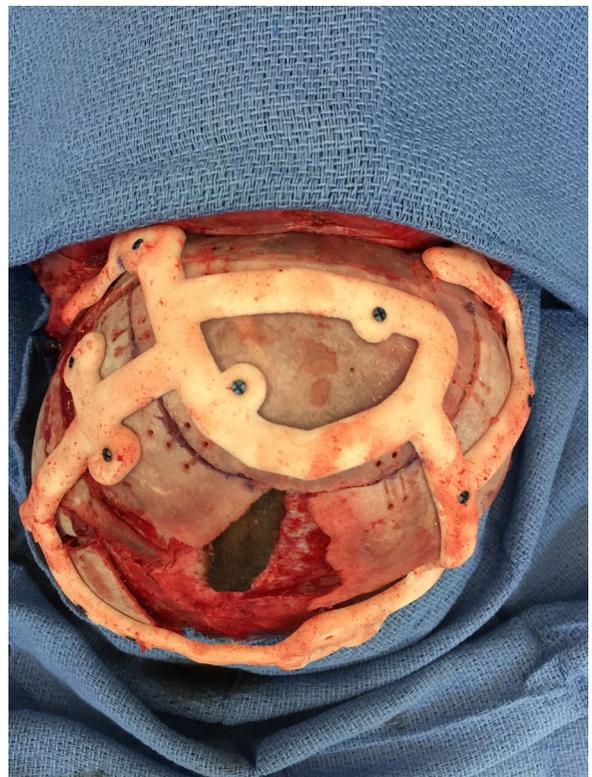


Fig. 6. Top views of repositioning guide on the skull when fully reassembled.

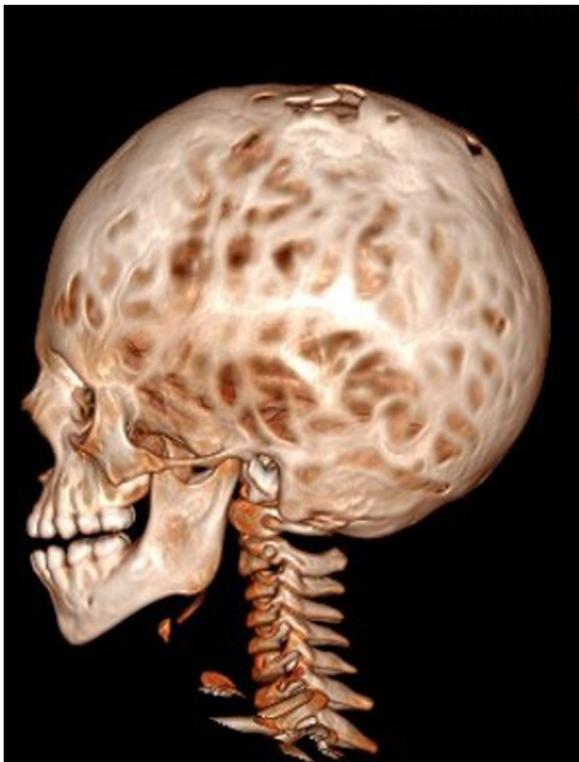


Fig. 7. Lateral view of 3D CT-scan of a scaphocephaly that has progressed to closure of all the sutures: severe oxycephaly.

The advantage of this whole technique is to allow assembly of all of the pieces of the new skull on the operating table, thanks to the spider guide (Fig. 6a). The important result of this is that the surgeon does not need to focus on the implantation of the various parts, as they are all held together like a 3D jigsaw: there is no need to evaluate the distances and perspectives during surgery. The repositioning of the spider guide is also a major improvement in that all the pieces are repositioned at once at the exact place decided ahead of surgery (Fig. 6b), instead of the previous technique that needed all the pieces to be positioned one by one, with loss of time incurred by the systematic evaluation of the appropriateness of each placement. The fact that the spider guide is well positioned with all the pieces in place allows an easier fixation, as all the parts are held properly and there is no longer any need for forceps. The surgeon's hands are free for the most important tasks.

3.2. Oxycephaly

In oxycephaly, many operating techniques are available. The following case describes a 9-month-old boy who had already been through surgery for scaphocephaly. Unfortunately, the craniosynostosis progressed into closure of all sutures (Fig. 7). We decided to address this severe oxycephaly by reverse frontal cranioplasty and enlargement of the parietal flaps. Following the technique described above, a cutting guide was developed and the contours of the forehead, parietal flaps and three bone grafts were marked (Fig. 8). First, the bandeau remained in place while the forehead was removed and reversed with a small advancement. Then, the parietal flaps were cut and the medium space between them was enlarged so as to create two spaces of 2 cm width each. These two spaces were intended to fuse. Between the forehead and the parietal zone, a long bone arch was removed then cut in three pieces that were turned through 90° to be used as three cleats to link the forehead and parietal areas (Fig. 9). As described before, all of these pieces were assembled within the repositioning guide, outside the

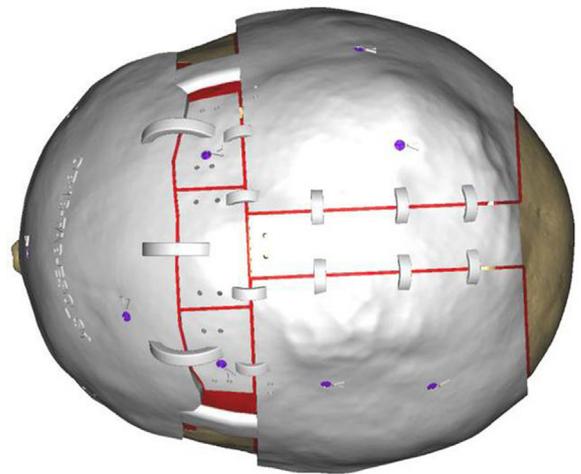


Fig. 8. Oxycephaly: cutting guide.

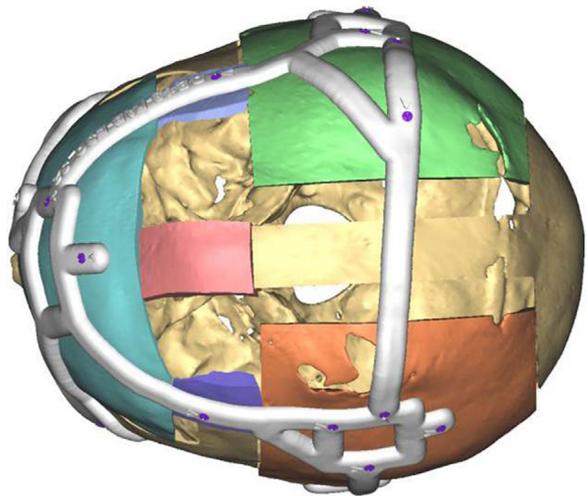


Fig. 9. Lateral view of the repositioning guide: frontal reversion and advancement, enlargement of parietal flaps.

operating field (Fig. 10a), and then put back in place for fixation (Fig. 10b). Finally, the repositioning guide was removed and the scalp was closed. Postoperative CT at 3 months showed compliance between preoperative plan and postoperative outcome (Fig. 11).

3.3. Craniosynostosis sequelae

Primary craniosynostosis surgery is widely developed and addressed in the literature [20–22]. It is acknowledged that this type of surgery can induce sequelae [23] for which computer-assisted surgery can provide a new hope with a different kind of surgical process. More specifically, for patients in their teens or even older and whose bone plasticity no longer allows optimal flexibility, CAS is an appropriate alternative [24]. The case introduced here is that of a 14-year-old patient with right plagiocephaly who had already been through a surgery when she was younger, with unsatisfactory results. She needed a new correction, to achieve frontal symmetry. The plan was to remove the retruded forehead and replace it with a new one. The Mimic® software calculated the optimum place for new bone harvesting. We ended up switching bones, with the one from the back flap devoted to the new forehead while the old forehead was placed at the back of the calvarial vault. A cutting guide was then designed for the retruded forehead

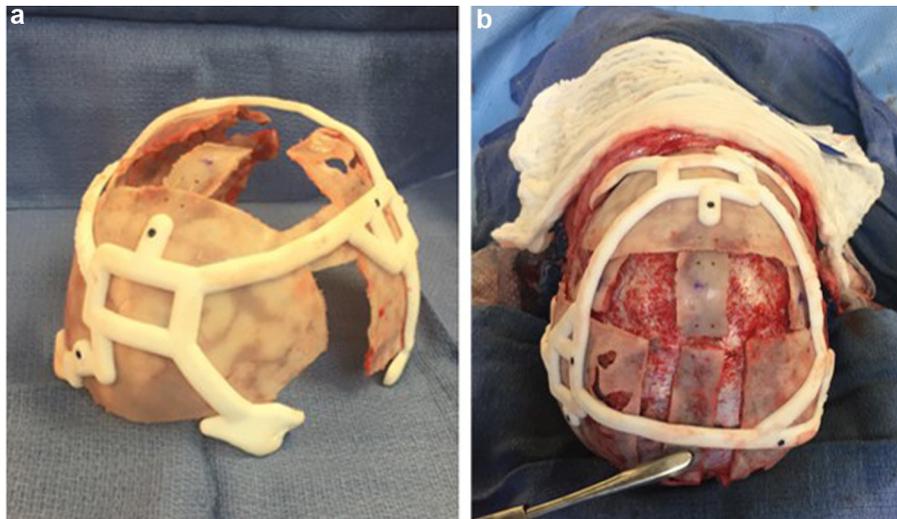


Fig. 10. a and b: top and lateral views of the spider guide while assembling all pieces on the table and back in place on the patient.



Fig. 11. Lateral view of the postoperative 3D CT-scan of the oxycephaly.

(Fig. 12a) and to allow harvesting of the exact shape for the new forehead (Fig. 12b).

4. Complex craniosynostoses and rare craniofacial malformations

In the previous sections, the procedure of computer-assisted surgery was described for simple craniosynostoses, matching the outcomes observed in previous research [25,26]. It seems that complex craniosynostoses can also benefit from this technique. Three different situations of complex craniofacial malformations are introduced below, for which CAS has shown interesting results.

4.1. Hypertelorism

The first case is of wide symmetric hypertelorism in a fronto-nasal dysplasia with bilateral cleft lip. The patient was 10 years old when we first met him. We decided to perform box osteotomies, and designed five cutting guides (Fig. 13). The contours of the polyamide guides were marked with a burr before being removed. The titanium guides were slightly different and allowed simple marking of the future osteotomies. Series of holes were also made to mark future stitch positioning. They allowed the appropriate

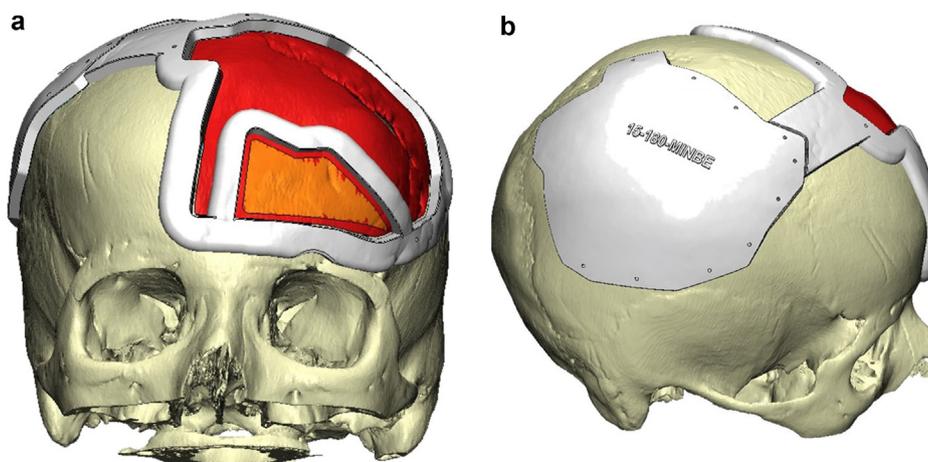


Fig. 12. a and b: cutting guide for retruded forehead removal and cutting guide for new forehead harvesting.

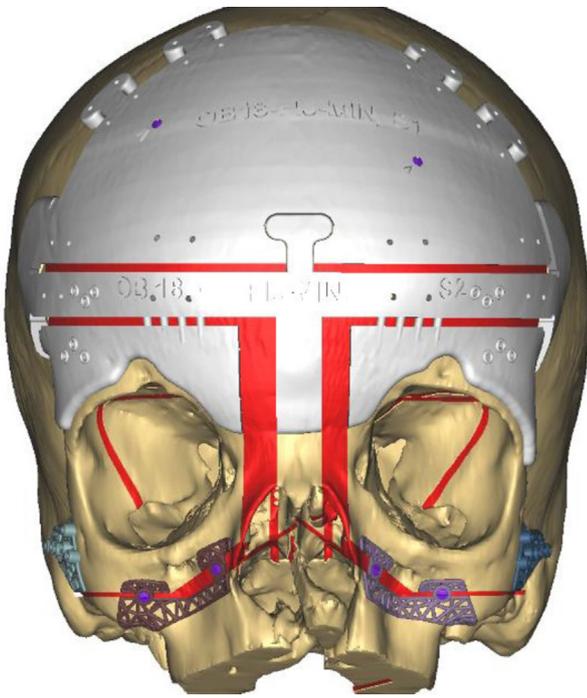


Fig. 13. Surgical simulation of box osteotomies and design of cutting guides.

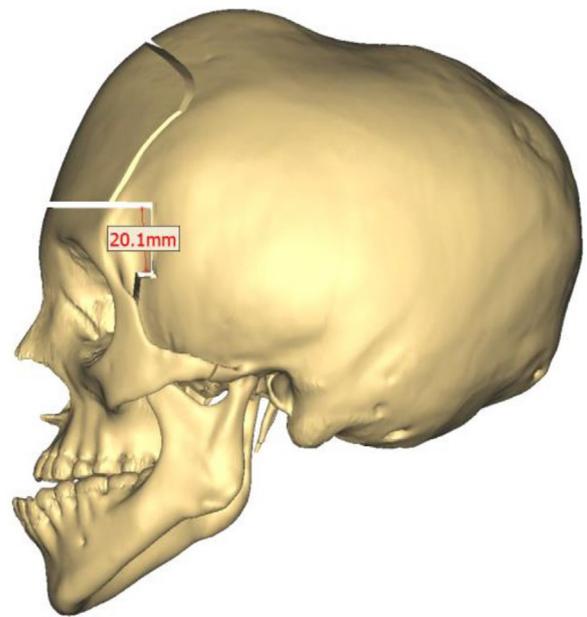


Fig. 15. 3D simulation of the patient's craniofacial morphology: Crouzon syndrome.

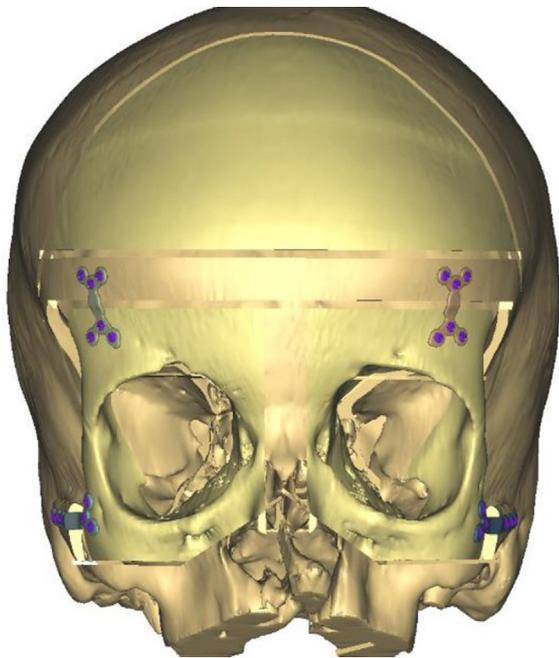


Fig. 14. Repositioning of osteotomies and expected final result with customized plates.

positioning of the osteotomy via the customized plates (Fig. 14). These customized plates have two advantages:

- precise positioning of the osteotomies and;
- osteotomy fixation.

The nasal dorsum was rebuilt by calvarial bone graft.

In the specific case of facial bipartition, computer-assisted simulation can also be used with very satisfying outcomes [27].

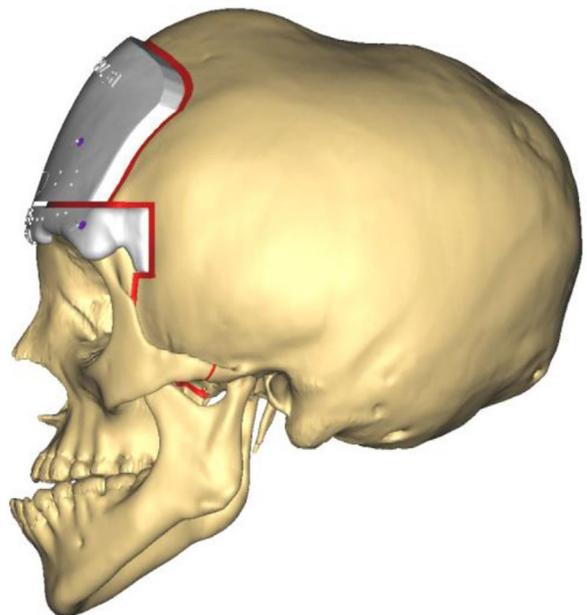


Fig. 16. 3D simulation of the monobloc and design of the cutting guides.

4.2. Crouzon disease

A 40-year-old patient was referred for previously untreated Crouzon disease (Fig. 15). Despite her age, we decided to perform fronto-facial monobloc advancement with internal distraction in order to increase the cranial and orbital volume and to correct the class III malocclusion at the same time. Again, computer-assisted surgery allowed precise preparation of the monobloc (Fig. 16). However, in this particular case, it was unnecessary to resort to this technology: the advancement could have been calculated on telero-diography or CT scan. Finally, a cutting guide was manufactured to raise the frontal flap and the upper third of the osteotomy (Fig. 17). The major advantage of CAS in this case was for residents' training, for them to fully understand this complex facial osteotomy.

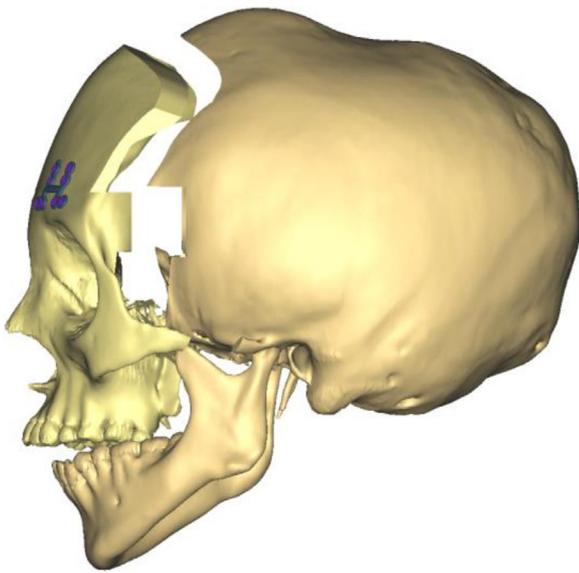


Fig. 17. Simulation of the monobloc outcome.

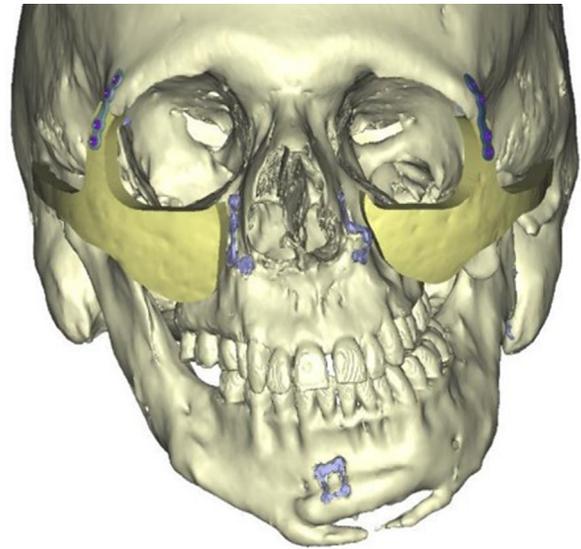


Fig. 19. 3D simulation of two ideal zygomas.

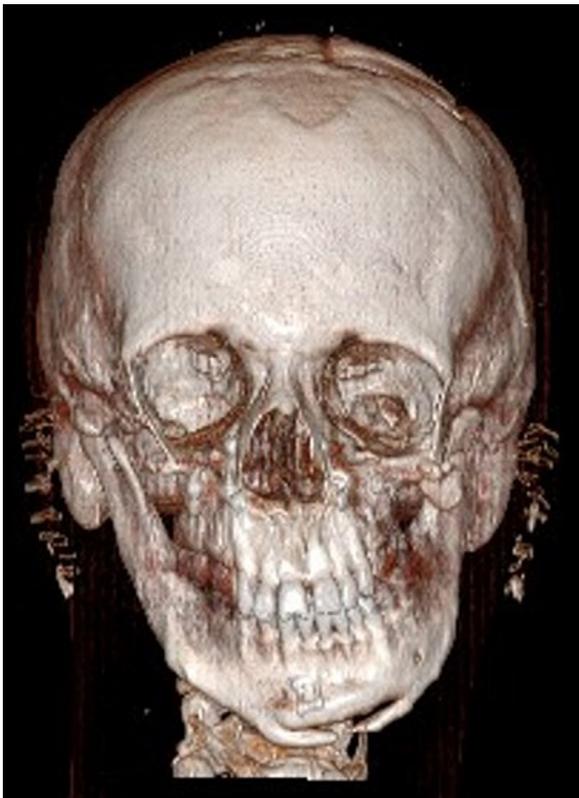


Fig. 18. CT scan of the Treacher-Collins syndrome with major sequelae of previous surgeries.

4.3. Treacher Collins

Treacher Collins syndrome has previously been addressed with recent techniques such as customized PEEK and alloplastic implants or cranial bone grafts [28,29]. The third case is a 25 year-old patient with Treacher Collins syndrome. He had undergone several in his youth, but had major sequelae (Fig. 18). It was decided to concentrate on the zygoma issue [30]. The computer-assisted procedure calculated the ideal curve to locate two anatomic zygomas that would perfectly fit once in their new place. Fig. 19 displays

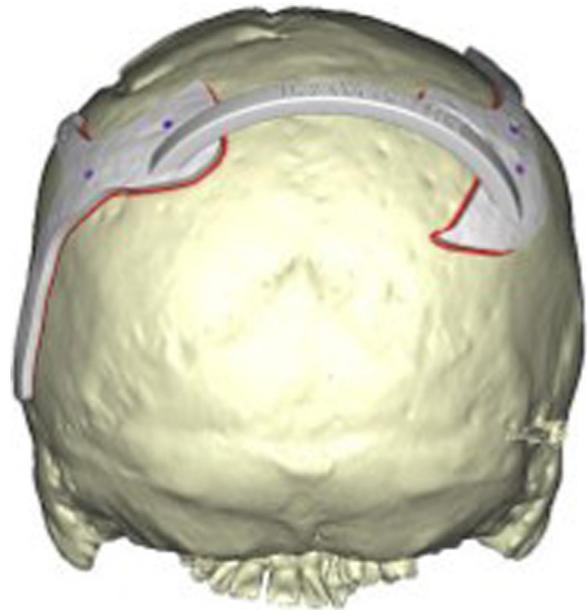


Fig. 20. Cutting guides design for the harvesting of the bicortical zygomas.

the two ideal zygomas on the 3D simulation and Fig. 20 shows the cutting guide design for harvesting the two bicortical zygomas. During surgery, as planned, two zygomas were extracted from the calvarium, and perfectly matched those of the 3D simulation (Fig. 21). One-year postoperative CT confirmed symmetry and absence of bone resorption (Fig. 22).

5. Discussion

In the light of the present cases, several important implications of the use of computer-assisted surgery emerge. The major contribution of the technique is precise forecasting, enabling more accurate preparation for surgery a more explicit presentation of the expected outcome to the patient. Secondly, during the presurgical phase, much time is devoted to the explanations needed by the clinical engineers to run the Mimic 19.0® software (Materialise, Leuven, Belgium) to calculate the exact places of bone extraction and implantation; however, once the clinical engineers have been



Fig. 21. Zygoma extraction: perfect match with the 3D simulation.



Fig. 22. Postoperative CT scan at 1 year: symmetry and absence of bone resorption.

trained in a specific type of case, they are able to run the software much faster for subsequent cases. During surgery, much time is saved by the precise planning of the surgical steps to be performed, which is a major advantage for both the surgical team and the patient. The surgeon can rely on the forecast, and the cutting guides allow faster and more accurate cutting whenever bone removal is required. Bone positioning is also both faster and more precise, as the new bone parts fit exactly in their new place. This combination of timesaving and precision is a major benefit: patients spend less time in the operating room, and hence experience less fatigue due to anesthesia. Reconstruction is more anatomical, as it is adapted to the individual patient's morphology; however, it remains crucial for the surgical team to pay attention to the soft tissues that also need to be addressed. A further benefit is the possibility for the scrub staff to prepare most of the instrumentation ahead of surgery, with the trays ready on the table next to the surgical team, streamlining the whole procedure, which both saves time and is useful for residents who are being trained, as they can see the whole procedure in advance. From a more pragmatic and economic standpoint, computer-assisted surgery reduces operating room occupation rates and hence the overall cost of surgery.

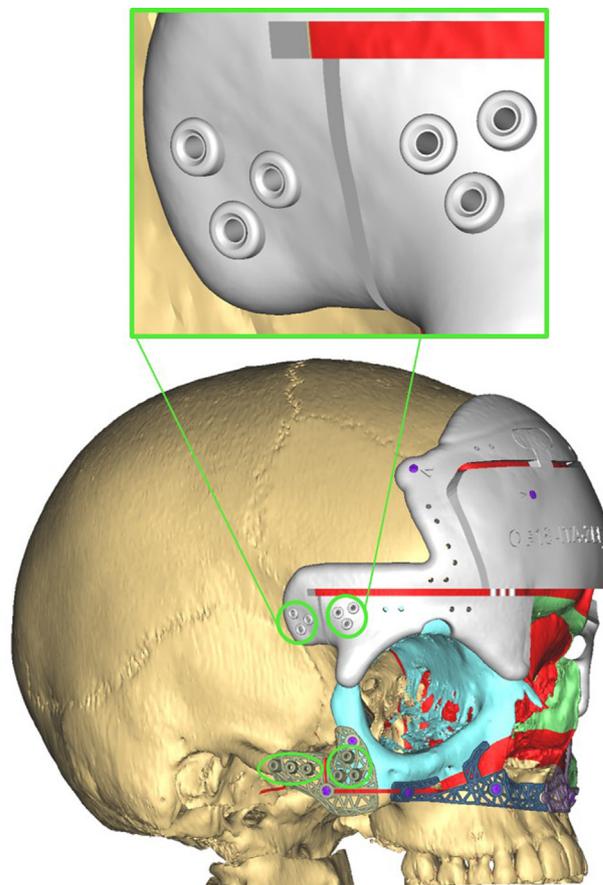


Fig. 23. Prospective image of a complex hypertelorism surgery.

By definition, simulation is a flexible tool with a wide variety of uses. Its limitations are that it addresses only bone structure and cannot provide soft tissue simulation. Another limitation concerns the accuracy of the CT scan to ensure correct rendering of anatomy in the 3D model: infra-millimetric slices should be used. While simulation is flexible, once the surgical plan is defined and the devices have been produced, it does not allow any further flexibility: if any change is made preoperatively, the devices cannot be used to their full potential. Moreover, simulation relies on data taken at given moment of time; any change in anatomy between scanning and surgery increases the risk of non-conformity. Great care must be taken when dealing with young patients still in growth. This is an important factor to take account of in planning surgery, as simulation, modelling and production of devices takes at least 2 weeks.

Computer-assisted surgery also provides an opportunity for new ways of training residents. In addition to the usual photographs and CT scans displayed on the wall or on a computer screen in the operating room, all of the images that are generated by the Mimic 19.0[®] software (Materialise, Leuven, Belgium) can also be hung on the walls, giving both surgeon and residents an accurate prospective view of the surgical steps that are going to be implemented. More precisely, the cutting-guide, the titanium plates that are going to be used to reassemble small bone parts and the spider guide that may be used to reassemble large bone parts can be visualized in colors that elucidate the successive stages, especially in complex cases such as the one presented in Fig. 23. Another important advantage of these images is that the various details that need specific attention are highlighted. For instance, in the image displayed in Fig. 24, the detail of the upper right side of the forehead bone is enhanced, to clarify what needs to be done in shaping the new forehead; that

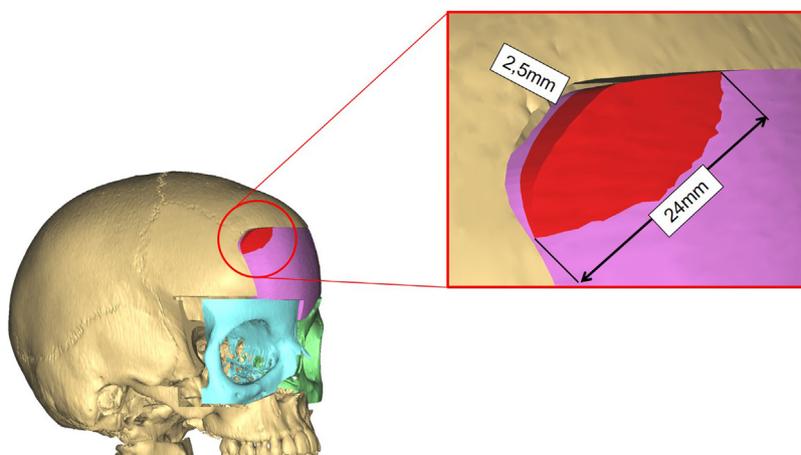


Fig. 24. Details of a prospective image of complex surgery.

specific area cannot be cut with the help of the cutting guide, as the bone shape that does not fit exactly, so it is emphasized that the surgeon needs to do this freehand. In this particular case, the technical contribution of the Mimic 19.0[®] software (Materialise, Leuven, Belgium) is all the more useful as it highlights the complex stages which require more attention, while easing the simpler ones.

In asymmetric hypertelorism, 3D positioning of the osteotomies is difficult. The pre-drilled holes and the customized titanium repositioning plates are of great help in that respect. Also, when it is time to assemble all the parts together, the spider guide enhances overall coherence. If the surgeon had to reassemble all the parts freehand, it would take much longer and the final result might not be as precise. Surgeons implementing this type of surgery most often consider the outcome very impressive in terms of compliance with the initial plan.

Overall, computer-assisted surgery offers infinite possibilities. Once a discussion with the clinical engineers is initiated, everything becomes possible: customized implants, all sorts of complex cases, variations in the osteotomies, etc.

The cost of the device depends on the complexity of each case, but ranges between 2000 and 4000 euros in complex cases.

6. Conclusion

To conclude, computer-assisted surgery is revolutionizing the surgical approach to complex craniofacial malformations, as well as easing less complex cases. However, using this technique implies relying on a device that is not human. Computer-assisted surgery is a tool that can change surgical practice; the surgeon can rely on it, yet nothing can replace his or her eye and experience. It is the combination of this experience and the appropriate use of computer-assisted surgery that, ultimately, leads to successful surgery.

Disclosure of interest

The authors declare that they have no competing interest.

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