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Craniofacial/Pediatric

The Role of Virtual Surgical Planning in Surgery for Complex Craniosynostosis

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Background: Virtual surgical planning (VSP) decreases reliance on intraoperative subjective assessment of aesthetic and functional outcomes in craniofacial surgery. Here, we describe our experience of using VSP for complex craniosynostosis surgery to inform preoperative decision making and optimize postoperative outcomes.
Methods: Chart review was performed for children treated with craniosynostosis at our institution from 2015 to 2021. Eight VSP maneuvers were defined and assigned to each patient when applicable: (1) complex cranioplasty: combined autologous and synthetic; (2) autologous cranioplasty; (3) synthetic cranioplasty; (4) vector analysis and distractor placement; (5) complex osteotomies; (6) multilayered intraoperative plans; (7) volume analysis; and (8) communication with parents. Outcomes between VSP and non-VSP cohorts were compared.
Results: Of 166 total cases, 32 were considered complex, defined by multisutural craniosynostosis, syndromic craniosynostosis, or revision status. Of these complex cases, 20 underwent VSP and 12 did not. There was no difference in mean operative time between the VSP and non-VSP groups (541 versus 532 min, $P = 0.82$) or in unexpected return to operating room (10.5% versus 8.3%, $P = 0.84$). VSP was most often used to communicate the surgical plan with parents (90%) and plan complex osteotomies (85%).
Conclusions: In this cohort, VSP was most often used to communicate the surgical plan with families and plan complex osteotomies. Our results indicate that VSP may improve intraoperative efficiency and safety for complex craniosynostosis surgery. This tool can be considered a useful adjunct to plan and guide intraoperative decisions in complex cases, reducing variability and guiding parental expectations. *(Plast Reconstr Surg Glob Open 2024; 11:e5524; doi: 10.1097/GOX.0000000000005524; Published online 10 January 2024.)*

INTRODUCTION

Virtual surgical planning (VSP) has emerged as a tool to simulate osteotomy and hardware placement and to decrease reliance on intraoperative subjective assessment in predicting both aesthetic and functional outcomes in craniofacial surgery. When utilized in conjunction with computer-assisted design and manufacturing (CAD/CAM), VSP has been shown to decrease intraoperative time and create a more efficient surgical workflow across an array of surgical procedures, most notably orthognathic surgery. To date, the literature surrounding this topic as it relates to cranial reconstruction is primarily limited to use of VSP in cases of single-suture craniosynostosis despite increased understanding that more complex cases may incur the greatest benefit. Herein, we describe our institutional experience of performing VSP in cases of complex craniosynostosis and repair of calvarial abnormalities over a 6-year period.

THE ROLE OF VSP IN CRANIOSYNOSTOSIS MANAGEMENT

Craniosynostosis is a congenital abnormality characterized by premature ossification of one or more calvarial

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sutures, which typically manifests within the first few weeks of infancy and results in progressive, dysmorphic skull growth. It is associated with a number of syndromic conditions with multisutural involvement and mid-face dysmorphology, necessitating multiple staged procedures to address both intra- and extracranial manifestations. In these complex cases, optimal surgical management is dependent on factors such as age, extent of sutural involvement, skull thickness, venous anomalies, skull base morphology, intracranial volume, and associated intracranial pathology such as hydrocephalus and tonsillar ectopia. Historically, the craniofacial surgeon relied heavily on subjective assessment and personal experience in deciding proper osteotomy and hardware placement and planned reconstruction to achieve optimal functional and aesthetic outcomes. VSP has recently emerged as a tool to introduce more objective metrics in the preoperative planning of craniofacial procedures and can result in a substantial improvement in selection of procedure, timing of procedure, and overall outcome in cases of complex craniosynostosis.

The literature to date has demonstrated that cranial vault remodeling in single-suture craniosynostosis as aided by VSP and computer-assisted design and manufacturing (CAD/CAM) has been associated with greater conformity of the reconstructed bandeau compared with nontemplate-guided cranial vault remodeling as assessed by postoperative computed tomography (CT).[1] This is attributed to the decreased need for intraoperative reassessments of distances and perspectives by the surgeon due to reliance on prefabricated bandeau templates.[2,3] Numerous studies have also demonstrated decreased procedure time for craniosynostosis repair aided by VSP.[1,4–7] For example, Ganesh et al found significant reductions in both procedure time and estimated blood loss in patients undergoing template-guided anterior cranial vault remodeling with fronto-orbital advancement compared with nontemplate-guided retrospective controls.[5] It has been argued that savings in procedure time afforded by VSP and CAD/CAM increase overall downstream cost-effectiveness in craniofacial practices and cultivate a more efficient workflow compared with more traditional practices.[2,8–10]

Moreover, VSP provides an excellent visual tool for patients and their families to discuss planned procedures. The images generated from this technology can be used to explain upcoming procedures and to graphically indicate how these complex cases will be performed. This serves to bridge a common communication gap between patients/parents and the surgeon and ultimately results in greater understanding of the surgical plan.

TRENDS IN THE UTILIZATION OF VSP AND ROLE IN THE MANAGEMENT OF COMPLEX CRANIOSYNOSTOSIS

As noted by Kalmar et al, use of VSP for pediatric craniofacial procedures has increased over the past decade.[9] Between 2011 and 2018, the authors found an increase in VSP utilization from 2.0% in 2011 to 18.6% in 2018 in their craniofacial registry. The most frequent application of VSP was for orthognathic surgery. Surprisingly, they found no utilization of VSP for craniosynostosis repair throughout the study period at their center.

Other authors have reported increased utilization of VSP in patients requiring repeated intervention and late presentation of synostosis.[11,12] Andrew et al found significant reductions in surgical duration for cranial vault remodeling in single-suture synostosis when utilizing VSP compared with non-VSP-treated patients. This time-saving effect was further amplified in multisuture synostosis patients, indicating even greater benefit for more complex cases.[4] However, despite the demonstrated potential in multisuture synostosis, VSP has been more frequently used for patients with single-suture craniosynostosis. Reports of the use of VSP for multisutural, syndromic, or other cases of complex synostosis remain limited to case reports or small case series.[2,13,14] In addition, there have been no reports made which focus on comparing directly the utility of VSP in cases of complex craniosynostosis to a similar cohort of cases where VSP was not used. As digital techniques for cranial modeling continue to advance and become more readily available, increased utilization of VSP for cases of skeletal facial reconstruction and complex craniosynostosis repair is anticipated.[6,15,16] It is in this setting that we will discuss our experience with the use of VSP for cases of complex craniosynostosis while also presenting our comparative analysis to our cohort of complex cases completed without the use of VSP.

Takeaways

Question: What are some ways in which VSP can be used to improve clinical outcomes in complex craniosynostosis?

Findings: VSP improves outcomes in complex craniosynostosis.

Meaning: VSP is a powerful adjunct to improve patient care and clinical outcomes in cases of complex craniosynostosis.

PATIENTS AND METHODS

A retrospective review was performed of all children who underwent a craniofacial procedure by the senior surgeons (T.A.I., M.S., C.H.) at Weill Cornell Medical Center/Komansky Children's Hospital and Columbia University Medical Center/Morgan Stanley Children's Hospital of New York from January 2016 to September 2021.

Periprocedural characteristics and outcomes were collected including age at surgery, date of surgery, diagnosis, type of surgical procedure performed, utilization of VSP, syndromic patient status, primary versus revision surgery, and complex versus simple reconstruction. Complex cases were defined as multisutural, syndromic, or secondary/revision cases. For all patients, total surgical time and length of hospitalization were also recorded. Children who underwent concomitant orthognathic or mid-facial surgery were excluded.

For all patients who underwent VSP, the specific application of VSP in each case was recorded by the attending physician performing the case as described by the following eight defined categories: (1) complex cranioplasty: combined autologous and synthetic; (2) autologous cranioplasty; (3) synthetic cranioplasty; (4) vector analysis and distractor placement; (5) complex osteotomies; (6) multilayered intraoperative plans; (7) volume analysis; and (8) communication with parents. All applicable maneuvers for a given case were recorded.

Statistical Analyses

The distribution of patient and procedural characteristics was evaluated between VSP and non-VSP cohorts. Unpaired *t* tests were used to compare outcomes between the two groups. Statistical significance was defined as a *P* value less than 0.05.

RESULTS

In this multi-institutional, retrospective chart review, 166 children were identified as having undergone craniofacial surgery between January 2016 and September 2021. Of these 166 children, 32 patients underwent surgery for craniosynostosis with cases defined as "complex" or involving multiple sutures, secondary procedures, or revision status.

Of the 32 patients, 20 underwent VSP and 12 did not. Patients who underwent VSP were significantly older than those in the non-VSP cohort at age of surgery (mean age 87.25 versus 12.92 mo, $P = 0.0011$) (Table 1). Cases were stratified by complexity on a four-point scale whereby two points were allotted for revision or secondary procedures, one point was allotted for syndromic patients, and one point for multisuture involvement.

The VSP group had a higher average complexity score compared with the non-VSP group (2.25 versus 1.75), although this difference was not statistically significant ($P = 0.2563$). There was no difference in mean time of surgery between the VSP and non-VSP groups with a mean operative time of 541 minutes for the VSP group compared with 532 minutes for the non-VSP group ($P = 0.82$). Mean length of stay was not significantly different between the two groups at 5.3 days for the VSP group compared with 3.83 for the non-VSP group ($P = 0.35$). There was no difference in unexpected return to the operating room rate across the VSP versus non-VSP groups (10.5% versus 8.3%, $P = 0.84$) and no difference in estimated blood loss between the two groups at 393 mL in the VSP group compared with 235 mL in the non-VSP group ($P = 0.2412$). (**See table, Supplemental Digital Content 1**, which displays patient demographics across cohorts, **http://links.lww.com/PRSGO/D6**.)

Furthermore, there was no difference in the amount of blood transfused across the two groups ($P = 0.413$). Of the patients requiring transfusion, there was no difference in amount of blood transfused per kilogram of patient weight ($P = 0.95$). Each case was reviewed, and all relevant VSP maneuvers were assigned. (**See table, Supplemental Digital Content 2**, which displays VSP maneuvers, **http://links.lww.com/PRSGO/D7**.) Of the patients who underwent VSP for cranial reconstruction, an average of three different maneuvers or applications were used (range 2–5 maneuvers). The most common application of VSP was used to communicate the surgical plan with parents (used in 18 of 20 patients, 90%). The second most common application was to plan complex osteotomies, and this occurred in 17 of 20 patients (85%). VSP was used the least to develop a multilayered operative plan (two of 20 cases or 10%). (**See table, Supplemental Digital Content 3**, which displays VSP maneuvers utilized, **http://links.lww.com/PRSGO/D8**.) Exemplary cases are presented to highlight the use of each maneuver.

Table 1. Patient Characteristics

	VSP		No VSP	
	No.	%	No.	%
Total	20	62.5	12	37.5
Age at surgery				
Mean age of cohort (mo)	87.25		12.92	
0–6	1	5	2	17.6
6–12	3	15	4	33.3
12–24	2	10	6	50
24+	14	70	0	0
Sex				
Male	9	45	7	58
Female	11	55	5	42
Multisuture involvement	14	70	8	67
Secondary procedure	3	15	1	8.3
Revision surgery	9	45%	3	25

CASE DISCUSSION

Case 1: Complex Osteotomies and Volume Analysis

The patient was a 5-year-old boy presenting with a history of developmental delay, frequent falls, and tonsillar ectopia. Preoperative workup revealed papilledema and imaging confirmed fusion of his sagittal and metopic sutures. He was diagnosed with craniocerebral disproportion. Given this presentation, cranial vault remodeling was recommended. Virtual surgery planning was performed to determine the extent of surgery needed and guide the choice of procedure. Initially, VSP was used to map out two different plans (plans A and B). Plan A involved complex osteotomies of the mid-vault while also incorporating a modified fronto-orbital advancement for correction of his frontal bossing, Plan B involved a mid and posterior vault remodeling. Volume analysis indicated that plan B would provide a greater total volume change primarily through posterior vault expansion (Fig. 1). The patient ultimately underwent a total cranial vault reconstruction which involved the elements of both plans A and B, thereby allowing for full cranial recontouring while also maximizing volume expansion. VSP with volume analysis was key in guiding this decision-making. The patient's papilledema resolved postoperatively and postoperative imaging showed complete resolution of tonsillar ectopia. Postoperative follow-up 18 months later showed an excellent cosmetic result.

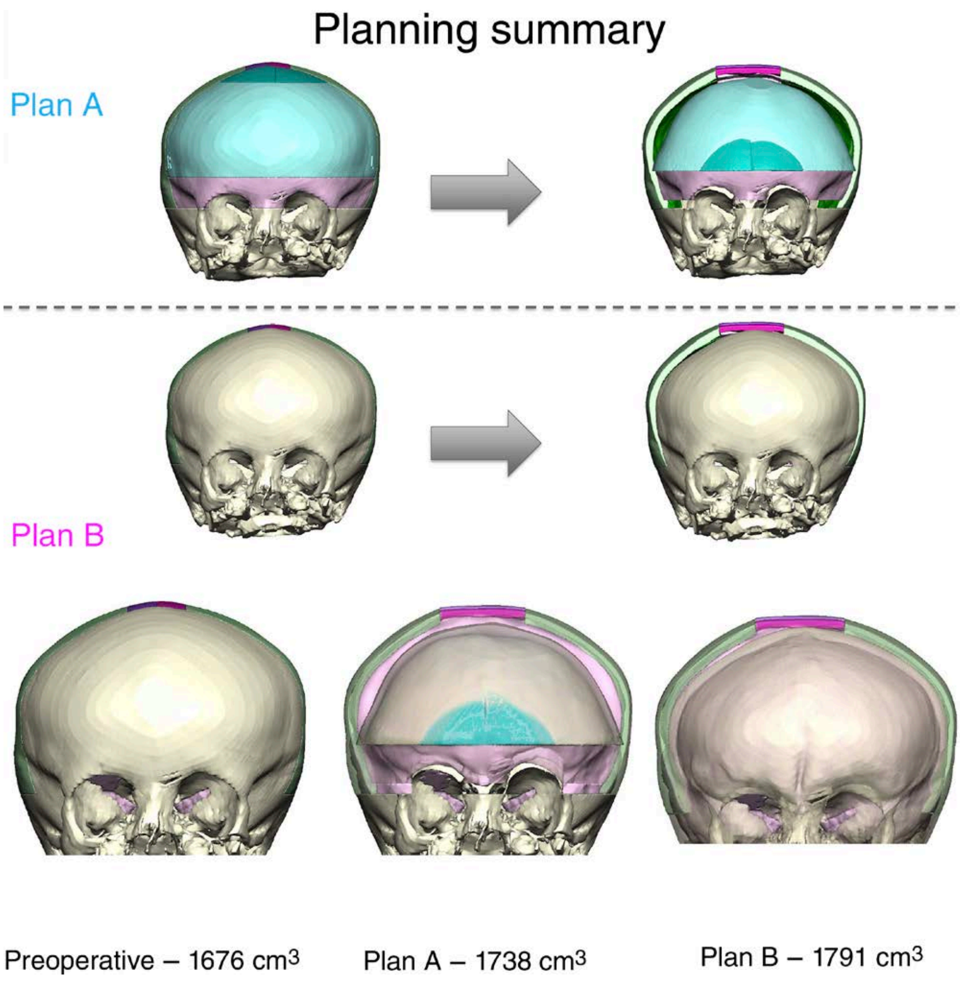

Fig. 1. Virtual surgical plan delineating complex osteotomies and volumetric analysis.

Case 2: Complex Osteotomies, Vector Analysis, and Distractor Placement

The patient was initially seen on day of life 3. She was noted to have bilateral choanal atresia and turribrachycephaly, proptosis, complex syndactyly of both hands and feet (mitten hands), and mid-face hypoplasia. Postnatal imaging demonstrated bicoronal craniosynostosis. Genetic workup confirmed a diagnosis of Apert syndrome with FGFR-2 mutation. Cranial vault remodeling was recommended; however, hyperdilation of the metopic and sagittal suture precluded traditional fronto-orbital advancement at that time (Fig. 2). Given this, VSP was used to plan an extended posterior vault distraction osteogenesis, along with a staged correction of turribrachycephaly. Use of VSP was critical in determining not only the final positioning of the distraction transport segment but also the appropriate distraction vector needed to provide a reduction in the maximum posterior height of contour while maximizing cranial volume. In addition, VSP workup allowed for predictive modeling of the most secure fixation sites for the distraction devices while also providing CAD/CAM-fabricated cutting guides to facilitate intraoperative craniotomy (Fig. 3). After approximately 5 months of consolidation, the patient underwent the second stage of her cranial vault reconstruction. Repeated CT scan at this time showed adequate bony ingrowth along the prior hyperdilated cranial sutures, resulting in ample frontal bone stock. VSP was again utilized to plan a modified fronto-orbital advancement with further correction of residual mid-vault brachycephaly, along with removal of bilateral cranial vault distraction devices. At this time, VSP was critical in planning the necessary extended complex osteotomies, which incorporated the prior areas of distraction regenerate. In addition, age-matched controls were used to plan the necessary residual turribrachycephaly correction. Predictive modeling was again utilized to plan fixation (Fig. 4). At 4 years of follow-up, the patient has not yet required any additional surgical procedures and remains without a VP shunt and without any progression of hydrocephalus.

Case 3: Synthetic Cranioplasty

The patient was a 15-year-old adolescent boy with a history of metopic craniosynostosis. He had undergone initial correction as an infant at an outside hospital. He noted progressive fronto-facial deformity with notable hypoplasia of the forehead and the bilateral superior-lateral brows. The CT scan indicated relapse of position of the orbital bandeau and restriction of grow of the bilateral frontal

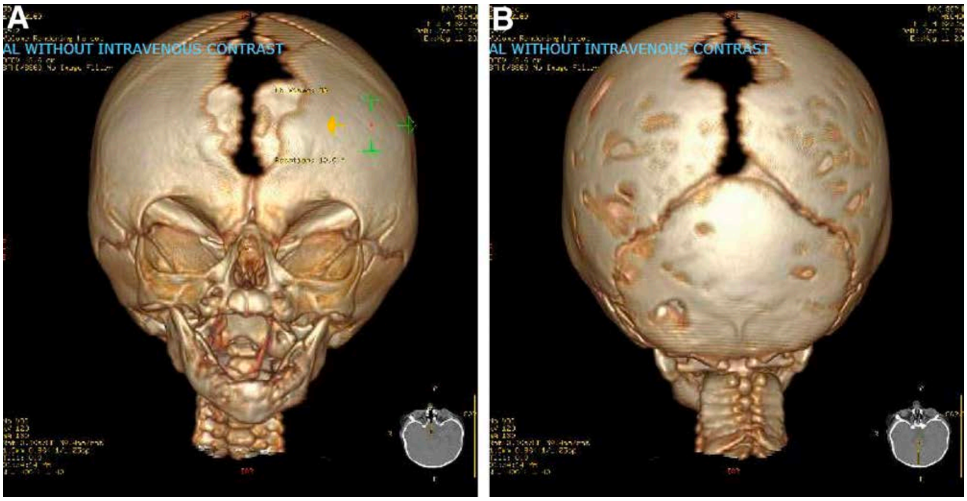

Fig. 2. Three-dimensional reconstruction images from patient CT scan demonstrating hyperdilation of metopic and sagittal sutures. A, Frontal view. B, Rear view.

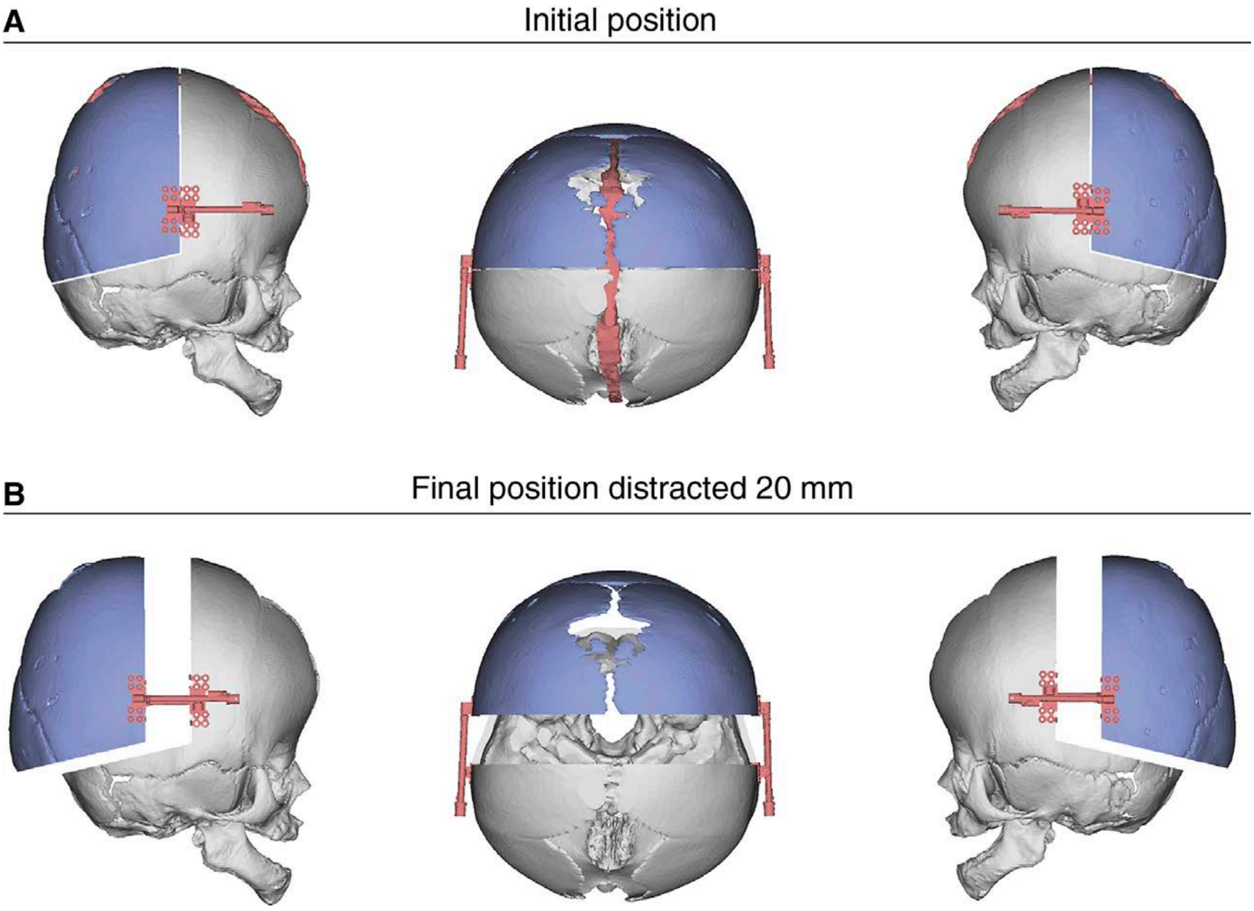

Fig. 3. Virtual surgical plan demonstrating placement of cranial distractors and subsequent movement. A, Initial position. B, Final position.

bone complex (Fig. 5). Revision cranial remodeling was recommended with patient-specific customized synthetic cranioplasty. VSP was utilized to create an implant to optimize the forehead projection, contour, and width while also correcting lateral brow hypoplasia. In addition, the implant was layered to correct for some soft tissue deficiency. Once again, predictive modeling allowed for ideal placement of bony fixation, with avoidance of underlying

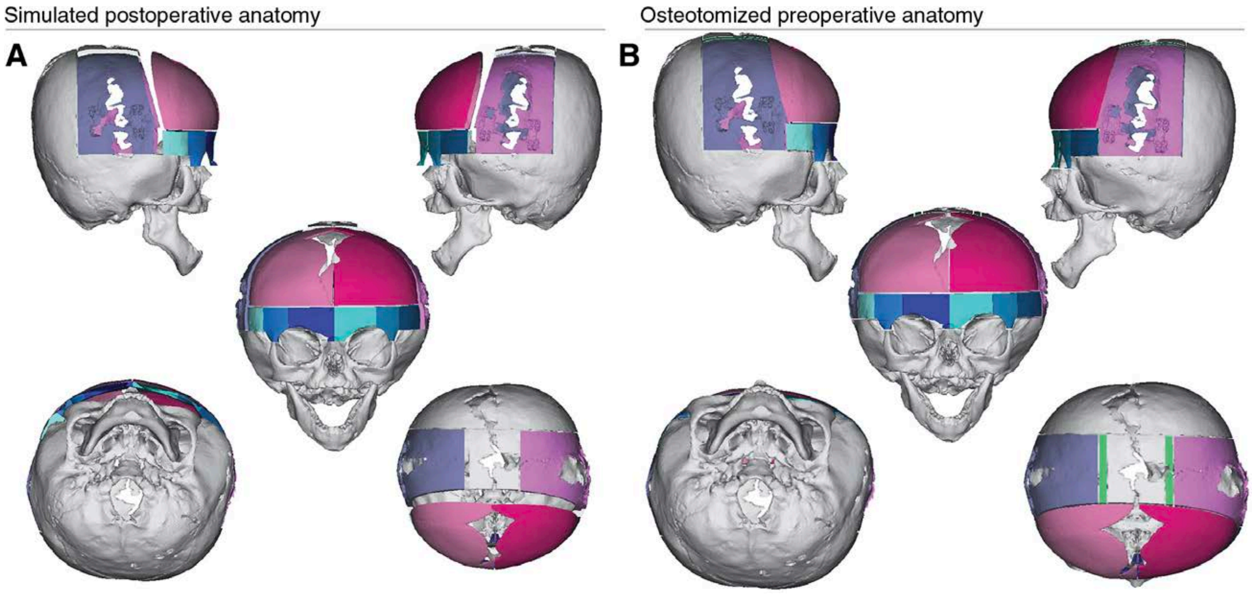

Fig. 4. Virtual surgical plan illustrating fixation of complex osteotomized segments. A, Simulated postoperative anatomy. B, Osteotomized preoperative anatomy.

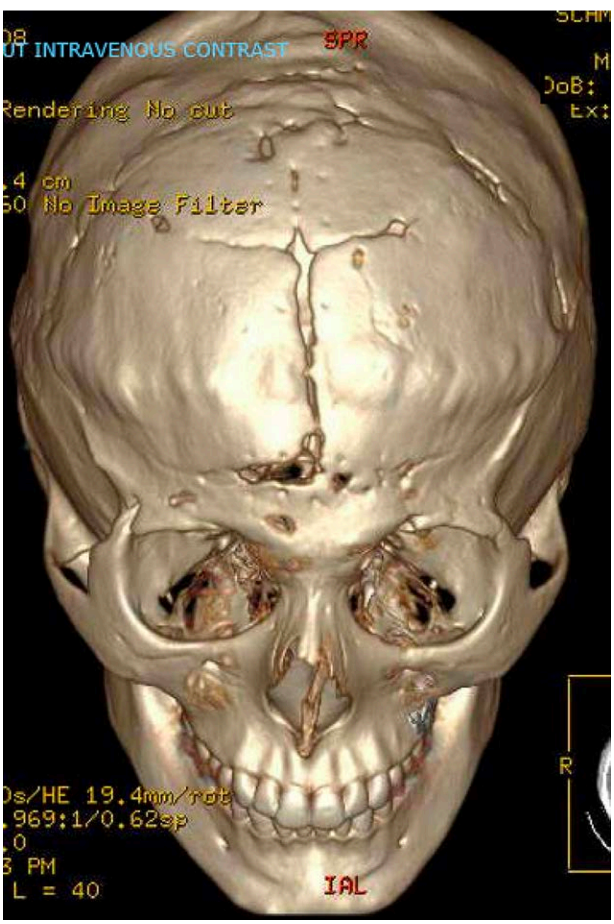

Fig. 5. Three-dimensional reconstruction images from patient CT scan demonstrating preoperative restriction of frontal bone complex.

bone gaps (Fig. 6). Follow-up at 2 years demonstrated maintenance of correction.

Case 4: Autologous Cranioplasty with Volume Analysis, Complex Osteotomies

The patient presented at 13 months with concern for abnormal head shape. Examination demonstrated severe right posterior occipital flattening, and brachycephaly, right forehead hypoplasia, right lateral orbital protrusion, and vertical orbital dystopia. Imaging confirmed multisuture craniosynostosis (right lambdoid, sagittal, and right coronal) and the noted cranial abnormalities (Fig. 7). Several surgical approaches were considered, including cranial vault distraction, staged anterior and posterior vault remodeling versus single-stage total cranial vault remodeling. VSP was used to simulate several plans to determine the most efficient and effective way to correct the patients severe cranial dysmorphology. The VSP was critical in creating a treatment plan for total cranial vault remodeling that allowed for correction of the posterior vault hypoplasia, vertex saddle deformity, compensatory occipital and frontal bossing, and superior orbital rim hypoplasia. Age-matched normative skulls were used to guide the new cranial width and height proportions, whereas a volume analysis confirmed that the reconstruction allowed for a significant increase in total cranial volume. Importantly, given how many bony pieces were being moved in this case, CAD/CAM technology was used to create cutting guides, positioning guides, and postoperative models to assist in accurate cranial recontouring (Fig. 8). The patient has been followed closely, and at 18 months postoperative, he demonstrates complete correction, without need for further revisions.

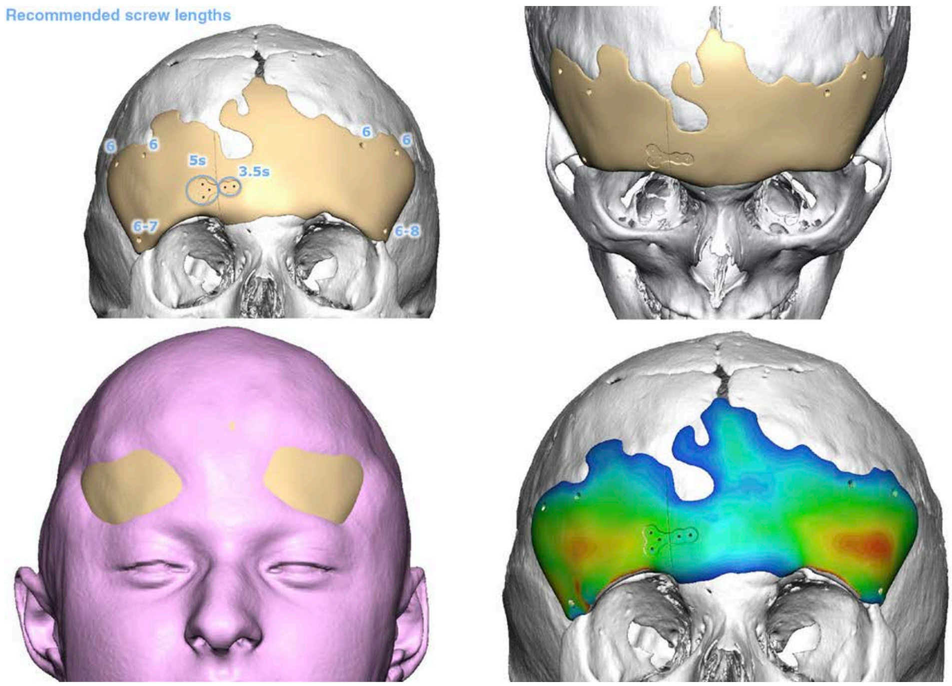

Fig. 6. Virtual surgical plan demonstrating improved forehead projection, contour, and width.

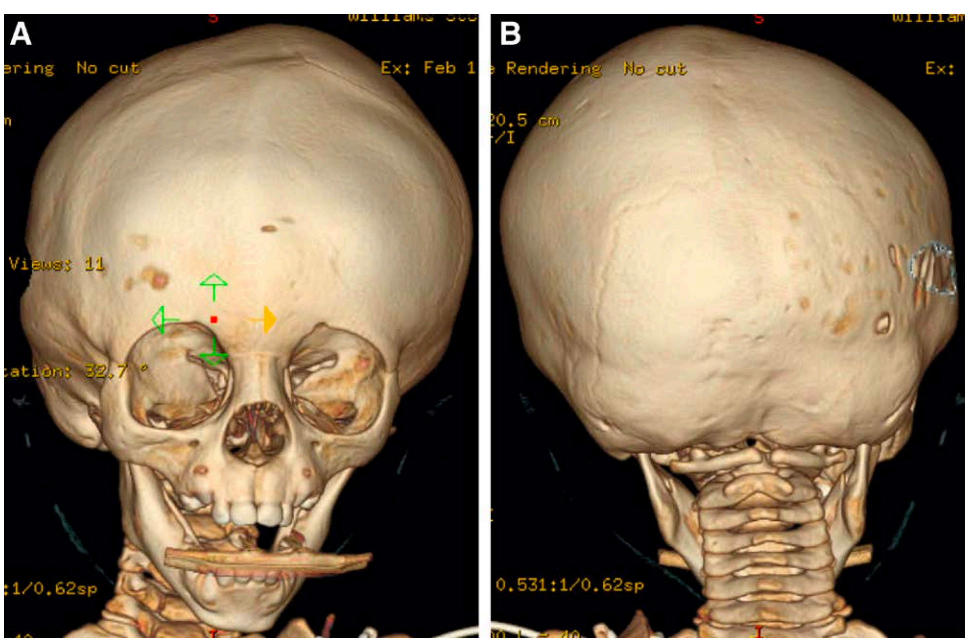

Fig. 7. Three-dimensional reconstruction images from patient CT scan illustrating multisuture craniosynostosis. A, Frontal view. B, Rear view.

Case 5: Complex Cranioplasty (Combined Synthetic and Autologous, Complex Osteotomies, Multilayered Plans

The patient was an 8-year-old girl with DiGeorge syndrome (22q11.2 deletion syndrome), VSD status postrepair, submucosal cleft and VPI, bifid uvula, and associated developmental delays with sagittal synostosis. She was noted to have craniosynostosis at a young age at an outside hospital; however, treatment was deferred. On presentation, she was found to have scaphocephaly with ridging of the sagittal suture, bitemporal restriction, and frontal compensation consistent with her diagnosis of sagittal synostosis. VSP was again used to create a surgical plan for the patient. In this instance, given the patients age, it was no longer technically feasible to expand the bony vertex to the desired width. Therefore, VSP was used to create a complex cranioplasty plan for her which combined

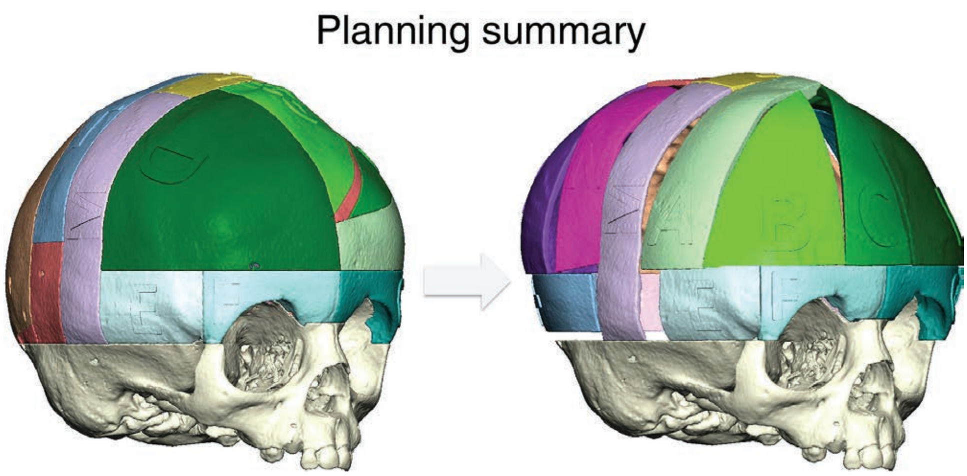

Fig. 8. Virtual surgical plan delineating new position of osteotomized segments after cranial recontouring.

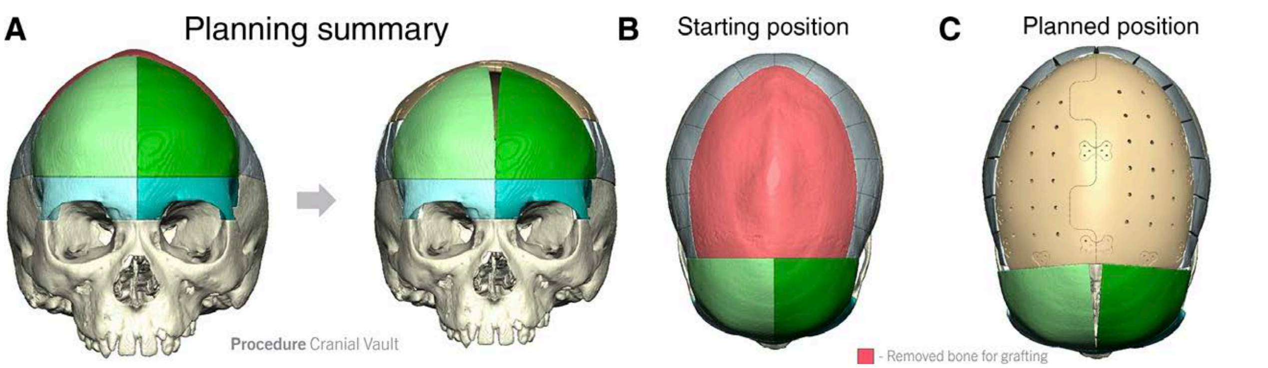

Fig. 9. Virtual surgical plan demonstrating postoperative position of cranial segments and implant in a complex cranioplasty with combined synthetic and autologous reconstruction. A, Planning summary. B, Starting position. C, Planned position.

elements of both an autologous and synthetic cranioplasty. To do this, VSP was used to first create a craniectomy defect, then to simulate complex osteotomies. A synthetic cranioplasty was then designed to fit into this virtually created neo-anatomic position (Fig. 9). We anticipated that there may be some variability in the degree to which the biparietal area was also able to be expanded; therefore, a multilayered intraoperative plan was created. To accommodate for multiple potential widths, the implant was designed with an interlocking sliding design that allowed for some adjustment of the final width intraoperatively.

DISCUSSION

The implementation of VSP is an interdisciplinary process requiring the input from craniofacial practitioners, namely pediatric neurosurgeons and plastic surgeons, and clinical software engineers.[17] Contour data are first obtained from CT scans of the patient's head which are packaged as DICOM images and transformed into three-dimensional (3D) renderings via various software platforms that can be virtually manipulated by the software engineers. These renderings include clear delineation of critical structures such as dural sinuses and shunt catheters and provide projections of potential alterations in critical structures such as the torcula to determine maximal safe distraction and placement of osteotomy cuts and craniofacial hardware. Anthropometric data, describing patient's head shape, such as biparietal diameter, fronto-occipital diameter, and cranial length are precisely tabulated and conformed to accurately plan osteotomy placement, advancement of the supraorbital bandeau, and alignment of bone segments.[18,19] When used in conjunction with CAD/CAM, stereolithographic templates and precise cutting guides can be created to dictate osteotomy placement and bone segmentation ahead of the procedure date. Typically, CT data of age-matched controls are overlapped with the patient's skull dysmorphology to guide intraoperative manipulation for targeted volumes and long-term aesthetic outcomes.[20–22] Practitioners can also generate normative contours from clinical assessments of normal child's head shape to avoid unnecessary exposure to ionizing radiation in healthy children.[5]

Although there is a growing body of literature which has demonstrated the utility of VSP for multiple craniofacial procedures, including cranial vault remodeling for single-suture craniosynostosis, reports such as ours greatly expand on the scope of this powerful tool. To the extent that prior reports have discussed the utility of VSP for complex craniosynostosis, it has been limited to small case series, without comparative cohorts. In the described cases, we demonstrate that VSP allowed for greater safety and efficiency for the most complex cases of cranial vault remodeling for patients with complex craniosynostosis (defined as multisuture, revision, syndromic or recurrent status). The results of our analysis, comparing cohorts of patients in whom VSP was and was not used, indicate that VSP was critical in minimizing time in the operating room by allowing for the majority of operative decision-making to happen before arriving in the operating room. Surgical time for the VSP and non-VSP groups was comparable, which is significant in that patients in the VSP cohort were older and had a higher complexity score compared with the non-VSP cohort. In aggregate, these two factors would have certainly lengthened the case time of this more complex population; however, we found that the case times were indeed similar.

In this report, we have described eight different VSP maneuvers, which we used in varying combinations during our planning sessions. Case discussions have been presented as a way to highlight how each of these maneuvers can be utilized to execute complex cranial vault reconstructions. The case discussions also document the manner in which VSP was a critical element in allowing for the proper execution of each treatment plan. Both cases 1 and 4 highlight the manner in which VSP can be utilized to guide the choice of procedure while also utilizing a volume analysis which ensures that the final reconstruction significantly addresses intracranial volume deficiency. Cases 2, 4, and 5 demonstrate the critical role that VSP plays in idealizing distractor placement and vector. Furthermore, the VSP also allowed for the innovative design of complex osteotomies which were used to achieve normative cranial contour. Cases 3 and 5 exemplify the irreplaceable role that VSP plays in creating customized implants that can address multiple aspects of cranial deficiency and secondary deformity often seen in complex craniosynostosis cases. Finally, it should be noted that, to the authors' knowledge, this is the first report to document the use of VSP in complex combined cranioplasty and layered intraoperative plans.

Our analysis clearly indicates that VSP was used most often to plan complex osteotomies and also to discuss the proposed plans with patients and their parents. We included this eighth and final maneuver as a way to highlight the ability that VSP gives the surgeon to communicate the information gleaned during the planning sessions to patients and parents who are often left struggling to understand exactly how the surgery will be executed. This was the most frequently utilized maneuver, which underscores the importance that this tool has in facilitating surgeon-parental communication.

Moreover, VSP serves a pedagogical role for both craniofacial trainees and families who are often encouraged to join preoperative interdisciplinary VSP meetings.[23,24] Virtual surgical simulation also allows for greater appreciation of regional anatomy and anticipation of incongruous interdigitation of osseous borders that may occur with manipulation.[25] Both the use of templates and the ability to perform preoperative simulation can aid less experienced surgeons who benefit from decreased reliance on intraoperative subjective assessment.[26–28] Given these multitiered applications, VSP is poised to increase in the number and diversity of applications for the care of craniosynostosis patients in the coming years.

Finally, it should be noted that VSP itself will likely become more and more sophisticated over time. This is likely to address some of the limitations of the technology which include how the plan relays information about the predictive response of overlying soft tissue and to changes in underlying bony anatomy. Adding additional anatomic detail to models, including soft tissue guides, and the potential incorporation of other virtual platforms to further facilitate intraoperative execution of prior planned procedures are just some of the ways in which this technology could potentially evolve over time.

CONCLUSIONS

VSP provides a comprehensive tool which can increase the intraoperative safety and efficiency of complex craniosynostosis surgery. We describe eight VSP maneuvers which may be useful in cases of complex cranial reconstruction. We expect that VSP will become an increasingly ubiquitous tool in the armamentarium of craniofacial surgeons to further optimize the safety and efficiency of surgery for complex craniosynostosis.

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DISCLOSURE

The authors have no financial interest to declare in relation to the content of this article.

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