Craniosynostosis has an estimated incidence of 1 in 2500 births. The sagittal suture is the most commonly involved in single-suture synostosis. Restenosis after initial surgery for sagittal synostosis has been reported to range from 2% to 16% in case series. Reoperation in the setting of restenosis is more challenging than first-time surgery; it is often complicated by delayed intracranial hypertension and typically requires calvarial vault reconstruction (CVR) through the healed surgical bed to cut the skull, realign bone segments, and reconstruct the calvaria with appropriate expanded dimensions.

Computer-assisted design and manufacturing (CAD/CAM) is a technique that uses 3D imaging to render a virtual patient-specific anatomical model that can be used to plan and execute operative maneuvers or customize devices and implants. This allows the precise formulation of operative plans in anticipation of surgical intervention. With the advent of additive manufacturing in the form of 3D printing, these virtual plans are materialized into anatomical replicas, cutting guides, or customized splints, prostheses, and implants using a variety of materials. The use of CAD/CAM for preoperative planning and intraoperative guidance has been reported in orthopedic, maxillofacial, plastic, craniofacial spine, and neurovascular surgery, among others. Described benefits include decreased operative times, increased potential for reproducible surgical outcomes that are accurate to intended results, enhanced patient understanding of surgical pathology and treatment plan, facilitated learning for surgical residents, and streamlined surgical instrument requirements.

The use of CAD/CAM for the cranial reconstruction in children with craniosynostosis has been reported in small numbers, mostly in the plastic surgery community. This technology allows for patient-specific reconstructions referencing standardized metrics, aiming for efficiency of execution and reproducibility of surgical outcomes. Despite its potential benefits, this modality has received less attention by the neurosurgical commu-
nity. Here, we present a technical note for discussion in the pediatric neurosurgery community. This is a case of a 15-month-old girl with restenosis of the sagittal suture requiring repeat surgery planned and executed using CAD and 3D printed intraoperative cutting guides, and it illustrates the use of this new technology.

Case Description

This 15-month-old girl was born at 39 weeks’ gestation without complication. She initially presented developmentally on track at 2 months of age with scaphocephaly (head circumference 91% percentile; cephalic ratio 0.776) and a palpable sagittal ridge, with no signs of elevated intracranial pressure. 3D CT scanning of the head confirmed a diagnosis of single-suture sagittal synostosis. The patient underwent an uneventful endoscopic sagittal synostectomy (4 cm in width) with bilateral wedge-shaped barrel stave osteotomies paired behind the coronal sutures and paired anterior to the lambdoid sutures. Her postoperative course included a custom helmet orthotic as an adjunct to cranial reshaping. Her cephalic ratio improved to 0.822, which was sustained for the next 9 months.

At 12 months postoperatively, the patient was noted to exhibit visible and palpable bony overgrowth along the synostectomy defect, recapitulation of biparietal narrowing, and frontal bossing, with a notable decrease in her head circumference percentile. Her development was age appropriate, and she had no evidence of elevated intracranial pressure on neurological and ophthalmological examinations. Repeat 3D CT revealed restenosis of the sagittal suture and scaphocephaly, and cranial vault remodeling was recommended to expand the intracranial volume. Due to our center’s experience with and accessibility to virtual surgical planning (VSP) and 3-dimensionally printed aides, these were used for the preoperative planning and intraoperative assistance of her calvarial reconstruction.

The 3D CT was used to plan osteotomies with VSP reconstruction (3D Systems Healthcare) (Fig. 1 upper). These planned cuts were virtually manipulated by superior and lateral expansion of bone segments flanking the center cut bone strip to match the dimensions of a normative 2-year-old female cranium (Fig. 1 lower). Using this schematic, a model of the patient’s skull was produced with planned osteotomy overlay. This was then used to render a sterilized cutting guide and normative 2-year-old female cranial template with outlines for the planned expansion. The model, cutting guide, and template were created using additive manufacturing (3D printing). Intraoperatively, the sterile manufactured cutting guide was used to mark the location of planned osteotomies on the skull (Fig. 2). Osteotomies were performed, positioned within the expanded outlines on the normative cranial template, and attached.

FIG. 1. Preoperative VSP. Upper: 3D head CT scans with virtually planned osteotomies. Lower: Planned expansion to the dimensions of a normative 2-year-old female cranium (superimposed in red). Note lateral (left), where anterior is facing down, and superior (center) distractions with overall outward expansion (right). Figure is available in color online only.
using resorbable plates and screws (Fig. 3). The expanded top of the calvaria was placed and secured to the native bone (Fig. 4). Operative blood loss was an estimated 100 ml, and anesthetic duration was 130 minutes. A subgaleal Jackson-Pratt drain was used in the perioperative period for 2 days. Recovery was uneventful, and the patient was discharged to home on postoperative Day 3.

Discussion

Preoperative VSP and 3D printed intraoperative aides are becoming increasingly useful for the surgical management of conditions in a variety of fields. Table 1 provides a summary of references cited for context. The use of VSP for CVR in patients with craniosynostosis was aided by the generation of normalized cranial templates by Saber and colleagues in 2012. This group rendered composite models representing age- and sex-matched averages for the pediatric calvaria based on 103 samples. Seruya et al. then described use of these data for CVR using CAD/CAM in 4 patients with single and multisutural craniosynostosis. They recognized potentials for efficient surgeries with displacement of time-consuming osteotomy planning to the preoperative setting, the development of practiced plans derived from multiple trial-and-error schematizations, and outcomes that precisely match preoperatively intended results. Other groups have described better establishment of parental expectations through the provision of a tactile aide that models expected results, as well as improved safety profiles due to preoperatively navigated cuts. This technology has also been proposed to advance the field with models that enhance trainee education and provide a medium for surgical innovation.

While the benefits of CAD/CAM for CVR seem promising, there is much to be explored. The most frequently cited barriers to this technique include potentially prohibitive cost, accessibility, and radiation exposure. The financial burden of CAD/CAM for CVR is not well studied, though it has been cited as a potential barrier to utility. However, cost-effectiveness analyses undertaken for CAD/CAM-facilitated maxillofacial and plastic surgeries have demonstrated reductions in both time and material cost when compared with conventional techniques. It is nevertheless important for the surgeon to review the cost associated with using CAD/CAM for CVR at their institution as well as patient insurance to assess levels of coverage. The number of centers offering CVRs using CAD/CAM is small. Its implementation may therefore be limited to patients residing near major cities where there is access to centers adopting such technology. While some authors have cited increased radiation exposure in the preoperative period as a disadvantage to this technique, CAD/CAM planning in our case was based on imaging already obtained for clinical workup, which is standard for conventionally planned reconstructions. No additional CT scans were required.

Our case highlights the benefits of using CAD/CAM for repeat CVR to achieve ease and safety of workflow, a well-educated patient family, and precision in surgical outcomes. In our patient a normal calvarial volume...
and shape were achieved after she underwent a well-planned and smoothly executed cranial reconstruction. We noted less intraoperative blood loss in this case than in conventionally performed repeat CVRs. The median estimated blood volume loss for standard repeat CVRs is approximately 109%,10 which is significantly greater than the that seen in our case, calculated to be 26.5%. With CAD/CAM, this repeat surgery made efficient use of intraoperative time; the process was facilitated by pre-constructed templates for osteotomy and bone rearrangement. Of course, these maneuvers did not replace the surgical team’s intraoperative judgment, and each step in the patient’s repeat reconstruction was double-checked and cross-referenced with the intraoperative experience of the surgeons performing the procedure. Conclusions about outcomes cannot be drawn with this illustrative case. Directions for future research call for prospective study of a larger number of patients.

Conclusions

Computer-aided design and modeling with 3D printed intraoperative aids for repeat cranial vault surgery represents a relatively novel incorporation of technology in pediatric neurosurgery. The risk/benefit profile of this new advancement is promising, and it is potentially an option for facilitating the course of children undergoing repeat CVR. This modality is logically applicable to reconstructive surgeries by pediatric neurosurgeons and plastic surgeons and may find utility in a variety of other procedures.

References


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