The aesthetic objective of and in many cases also the indication for craniosynostosis surgery is cosmesis—the correction of a disfiguring defect. This objective remains undisputed, in spite of ongoing controversy regarding neurological deficits that may or may not be caused by untreated craniosynostosis. In contrast to a reconstructive approach, in which the surgeon restores the patient’s previous appearance, craniofacial remodeling in children involves the creation of a new appearance. Craniofacial remodeling therefore should be regarded as constructive rather than reconstructive surgery.

Reference in such a process would be provided best by head shapes of unaffected children. As such appropriate templates are not at the surgeon’s disposal in most operating theaters, the most common approach is based on quite arbitrary changes of the patient’s appearance. Decision-making is based either on the surgeon’s personal preferences or a surgical “standard” that defines the rules of appropriate surgical relocation of bone fragments according to 2D measures reflecting the preferences of the author of these rules.

Follow-up is recorded either by traditional photographs, which do not allow for validation, or by 2D measures that do not fully represent the 3D esthetic result. Moreover, as the individual surgical goal in any given reshaping procedure is not defined beyond general rules, there is little or no chance of objective outcome assessment. Lack of evidence prevents dissatisfaction. This might be the reason why surgical techniques for craniosynostosis “repair” have never been put under scrutiny and the gap between technological options and surgical practice continues to increase.

Objective

We report on the first use of a surgical tool that is capable of transmitting statistical information, represented in a model, into the disfigured bone. The model is derived from a currently evolving databank of normal head shapes. Ultimately, the databank will provide a set of standard models covering the statistical range of normal head shapes, thus providing the required template for any standard remodeling procedure as well as customized models for intended overcorrection. To date, this technique has been used in the surgical treatment of 14 infants (age range 6–12 months) with craniosynostosis. In all 14 cases, the designated esthetic result, embodied by the selected model, has been achieved, without morbidity or mortality.

Frame-based reconstruction provides the required tools to precisely realize the surgical reproduction of the model shape. It enables the establishment of a self-referring system, feeding back postoperative growth patterns, recorded by 3D follow-up, into the model design.

Keywords • craniosynostosis • esthetics • skull • surgical planning • statistical 3D shape model • frame-based surgery • model-based surgery • congenital
growth patterns or shape characteristics of a local population. The data pool is accessible through a customized choice of models incorporating the required information.

In addition, the special features of the final version of this frame-based technique should accelerate the procedure, reducing blood loss, duration of anesthesia, and surgical time.

**Methods**

**Skull Library**

To generate a preliminary statistical 3D shape model of the skull (described in March 22, 2006 press release by Rudolf Kellermann, available at http://idw-online.de/pages/de/news152247), software requirements for processing MRI data sets (Fig. 1) were developed in 2005. Twenty-one data sets from healthy infants at the typical age for craniosynostosis surgery were selected (Fig. 2). Based on these data, we computed an average shape as well as the most characteristic variations, using the method of consistent patch decomposition, parameterization, and principal component analysis (PCA). Corresponding points on each skull surface were defined by mapping anatomically corresponding regions from one shape to another, minimizing metric distortion of the patches. This allows for the representation of all shapes in common vector space and subsequent statistical analysis via PCA. The goal of this analysis is to use a minimum of parameters to represent as much variance contained in the preliminary set as possible. These essential degrees of freedom of the resulting statistical model enabled us to explore characteristic cranial shapes within a normal variation and to reconstruct new shape occurrences by linear interpolation of all or just a few selected “eigen-modes” (major modes of variation).

**Model Assignment**

Radiological assessment of individual patients can be completely avoided in model-based reconstruction. Matching the model to the patient requires only few anthropometric skull measures, such as the width between both entries of the auditory canals, the distance from nasion to occiput, or the height between the vertex and the midpoint of the line between the auditory canals. These distances are extrapolated to the skull surface by approximating the skin thickness. The model shape that best fits these measures and corresponds to the anticipated growth pattern of the underlying diagnosis is then selected as a template from the shape space covered by the statistical 3D shape model for the reconstruction process. If diagnosis-and age-related overcorrection is indicated, a customized model corresponding to the intended deviation from a statistical average may be chosen from the currently arising set of models which will include standard overcorrection options as well. Patient-specific individual preoperative planning may be performed by an interpolation of any given number of appropriate shapes contained in the database. However, in a finalized version, the provided choice standard models should cover most of the demands.

**The Technique of Frame-Based Remodeling**

The first clinical application of the databank was a frameless procedure, as previously described. Although this procedure, in which a model was used without any auxiliary equipment, achieved a satisfactory result, we felt that the time-consuming procedure of processing and successively relocating each bone fragment (Fig. 3) could be improved. A frame was developed, enabling the surgeon to arrange, fix, and connect any size and number of bone fragments at the same time at any designated position on the model in a stable manner before relocating the completed reconstruction in one piece into the patient.

To save costs and to avoid certification problems, the preliminary prototype frame was created by combining registered standard items (for example, Schanz screws and a commercial stereotactic frame). However, the final version will contain a substantial number of customized components and will require approval as a medical product. All parents were informed about and agreed with the use of the frame during the preoperative consent discussion.

During the cranial remodeling procedure, the assigned model is fixed on a stand. Adjustable bows holding bone brackets are individually positioned around the model according to the designated surgical field. The bone fragments are destabilized, arranged on the model surface, and secured by bone brackets, holding Schanz screws (Fig. 4A). To confirm the designated correction prior to the final stabilization of the fragments, the frame with all fragments in place may be positioned into the surgical field once more (Fig. 4B). Then the model is removed from the stand and the frame is rotated into a convenient position, displaying the inner surface of the

![Fig. 1. Building a 3D atlas of neurocranium reconstructed from MRI data.](image)
assembly. Resorbable standard plates of variable size and number or customized mesh are used to connect and stabilize the assembly of bone fragments on the inner surface (Fig. 4C). Finally, after removal of the frame (Fig. 4D), the completed assembly is returned into the patient. To smooth out local unevenness in the transitional area, the adjacent skull may be adapted by means of barrel stave cuts. Fixation can easily be achieved by placing a small number of plates or sutures at any discrete location.

Results

From August 2010 to December 2012, 188 patients underwent surgery for craniosynostosis repair at our institution. In frontal and coronal suture synostosis, free-handed bilateral fronto-orbital reshaping was performed, with or without advancement. Since 2005, we have been using a stereolithographic model for guided remodeling in selected cases.

From August 2010 until now, 14 children (4 girls and 10 boys, age range 6.0–12 months), all with metopic synostosis, were operated on by a single surgeon using frame-based reconstruction. The surgery was uneventful in all cases, and no morbidity or mortality was recorded. The designated surgical result, represented in the assigned model, was achieved in all patients (Fig. 5). Due to the complex options and the complicated mode of adjustment of the prototype, an average of 45 minutes was spent setting up and adapting the frame to the specific patient. Another period of 15–20 minutes was needed to arrange and connect the bone fragments. Significant time savings compared with standard techniques are still to be achieved with a final simplified design.

Discussion

The limited utility of 2D measurements to define 3D surfaces has been widely accepted. By contrast, the development and application of a 3D shape model of the upper skull fully meets this requirement and therefore responds best to the primarily esthetic indication of skull reconstruction. The inherent customized statistical information provides objective and patient-specific guidance for remodeling without the need for the patient to undergo any radiological diagnostic studies, thus avoiding exposure to ionizing radiation.

Preliminary results of the application of a databank-derived model in surgery, which we reported in 2005 (La-mecker H, Zöckler M, Haberl H, et al., presented at the Advanced Digital Technology in Head and Neck Reconstruction meeting, 2005), 2006, and 2009, have since been confirmed by other authors, proposing fragmentary 3D shape models with a majority focusing on the fronto-orbital band as the key region of reshaping. To date, only Burge et al. have incorporated our approach of the intraoperative use of a customized databank-derived statistical model.

As a next level of development we undertook the precise technical transmission of model-bound information into the retrieved bone fragments without limiting their size, order, or final position. The resulting solution com-
bines an inner- and outer-surface model, the inner surface being represented by the model itself and the outer surface by an arbitrary number of adjustable puncheons, fixing the bone fragments in the designated position on the model. The freedom to arrange fragments of any size and order simplifies surgery even in this quite standardized procedure, as limiting factors like the future fixation sites or the bearing capacity of individual bone straps do not matter anymore. Furthermore, assembling any number of fragments to a single physical form accelerates the otherwise somewhat difficult and time-consuming fixation process, as the stability of the extracorporally reinforced assembly allows the final fixation to be limited to a very few easily accessible contact points.

Serial 3D photographic follow-up with currently commercially available systems, using subtraction techniques, provides information on the location and amount of surgically induced volume changes as well as on eventual local losses of correction due to persisting pathologic growth patterns. This feedback will refine the criteria for the model design as well as for the initial assignment of a shape model.

Limitations Revealed by This First Series of Frame-Based Reconstructions

The complex and time-consuming setup of the prototype frame, consisting of non-customized standard components and allowing for more options than necessary, prevented the intended time-saving which we still hope to achieve with a customized frame and the much faster en bloc reimplantation.

Another issue of further discussion may be related to the very rationale for our approach: Replacing uncontrolled and sometimes unreflected implicit esthetic knowledge with controlled statistical data may be regarded as equivalent to replacing an idealized goal (beauty) with a statistically averaged goal (normality). To replace “art” with statistics may prevent a few ideal results, although any further and improved algorithm of model choice is conceivable as soon as anybody is able to define beauty.... However, we consider it much more important to avoid disappointing surgical results and prevent affected children from paying the price for an inevitable learning curve.

Conclusions

In this first clinical evaluation, the application of a statistical shape model for guided skull remodeling has proven feasible and suitable to ensure the designated surgical result. Frame-based remodeling is designed to be not only another tool but a novel conceptual approach combining precise planning and precise follow-up with precise surgical transformation. In this manner, frame-based remodeling stimulates the application of modern technology to craniofacial surgery. We feel that probably the most intriguing advantage of this approach will be to achieve immediate consistency among patient procedures independent of the experience of the center.
Frame-based cranial reconstruction

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Disclosure

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

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