Caregiver Preferences for Three-Dimensional Printed or Augmented Reality Craniosynostosis Skull Models: A Cross-Sectional Survey

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Background: Recent advances in three-dimensional (3D) printing and augmented reality (AR) have expanded anatomical modeling possibilities for caregiver craniosynostosis education. The purpose of this study was to characterize caregiver preferences regarding these visual models and determine the impact of these models on caregiver understanding of craniosynostosis.

Methods: The authors constructed 3D-printed and AR craniosynostosis models, which were randomly presented in a cross-sectional survey. Caregivers rated each model’s utility in learning about craniosynostosis, learning about skull anatomy, viewing an abnormal head shape, easing anxiety, and increasing trust in the surgeon in comparison to a two-dimensional (2D) diagram. Furthermore, caregivers were asked to identify the fused suture on each model and indicate their preference for generic versus patient-specific models.

Results: A total of 412 craniosynostosis caregivers completed the survey (mean age 33 years, 56% Caucasian, 51% male). Caregivers preferred interactive, patient-specific 3D-printed or AR models over 2D diagrams (mean score difference 3D-printed to 2D: 0.16, P < 0.05; mean score difference AR to 2D: 0.17, P < 0.01) for learning about craniosynostosis, with no significant difference in preference between 3D-printed and AR models. Caregiver detection accuracy of the fused suture on the sagittal model was 19% higher with the 3D-printed model than with the AR model (P < 0.05) and 17% higher with the 3D-printed model than with the 2D diagram (P < 0.05).

Conclusions: Our findings indicate that craniosynostosis caregivers prefer 3D-printed or AR models over 2D diagrams in learning about craniosynostosis. Future craniosynostosis skull models with increased user interactivity and patient-specific components can better suit caregiver preferences.

Key Words: 3D-printing, augmented reality, caregiver education, craniosynostosis, patient education

Craniosynostosis, the premature fusion of cranial sutures, often requires early surgical intervention in the first few months of life and subsequent surgical revisions.1 Caregivers of patients with craniosynostosis are asked to comprehend complex craniofacial anatomy and make challenging decisions on behalf of young children to pursue surgery. To facilitate surgical decision-making and caregiver education, surgeons routinely provide craniosynostosis information using verbal discussions, written pamphlets, patient computed tomography (CT) scans, and hand drawings.2 However, these current modalities leave gaps in caregiver knowledge, with caregivers requesting further information or multiple visual modalities to enhance their understanding of craniosynostosis.3 More specifically, families have stated that it can be challenging to visualize craniosynostosis treatment options proposed with only verbal explanations, and that skull structure and development is hard to comprehend spatially when surgeons use only static two-dimensional (2D) diagrams.4,5 Consequently, identifying an optimal teaching modality with caregiver-oriented content can help standardize and streamline craniosynostosis caregiver education.

Recent advances in medical imaging and computer-aided design have expanded the possibilities of anatomical modeling for patient counseling. In example, three-dimensional (3D) printed anatomical models have emerged as effective tools to increase patient understanding of craniofacial anatomy, gliomas, renal cancer, cerebral hemorrhage, and congenital heart defects.6–14 Similarly, there has been increased interest to visualize anatomical models, such as the heart after myocardial infarction, in augmented reality (AR), which presents a superimposed digital anatomical model onto the real world.15 In recent years, advancements in 3D-printing and AR technologies have made these modalities more cost-effective, user-friendly, and accessible to providers for patient education.16–18 Given these preliminary efficacy studies and technological advancements, there may be great potential in utilizing 3D-printed or AR models for craniosynostosis caregiver education.

Specific to craniosynostosis, 3D-printed or AR models have preliminarily demonstrated utility in streamlining surgical planning and surgeon training.17–21 Only 1 study has assessed 3D-printing technology for craniosynostosis education, showing that patient
families were in favor of the 3D-printed craniosynostosis models compared with CT images. However, there remain little evidence-based and caregiver-oriented guidelines on how to best create these models for increased caregiver understanding. Further, a relative comparison of the unique benefits of 3D-printed or AR models for craniosynostosis caregiver education has not yet been studied. Thus, the aims of this study were to

1. compare caregiver preferences for 3D-printed, AR, and 2D models and
2. determine the impact of these various visual models on caregiver understanding of craniosynostosis.

**MATERIALS AND METHODS**

**Model Development**

Caregiver perspectives on 3 different skull models were compared in this study: 3D-printed, AR, and 2D (Fig. 1). Two-dimensional models were diagrams sourced from the literature. We generated 3D-printed and AR models from preoperative cranial CT scans of patients with sagittal, bicoronal, or unicoronal craniosynostosis, and used the Digital Imaging and Communications in Medicine viewer Inobitec (Voronezj, Russia) to export CT scans into virtual 3D meshes. The meshes were smoothed and postprocessed in Mesh-Mixer (AutoDesk, San Rafael, CA) to ensure 3D-printability. Lastly, we 3D-printed the skull models using white polyethylene terephthalate glycol filament and a Prusa MK3S printer (Prusa Research, Prague, Czechia). To allow caregivers to view the model in AR, the meshes were uploaded to Augment (Augment Technology, LLC, Paris, France), a mobile AR visualization platform.

**Survey Instrument**

We built a Qualtrics (Qualtrics, Provo, UT) survey incorporating the skull models, then distributed the survey to caregivers through popular Facebook craniosynostosis support groups from February 8th to March 29th, 2021. The 3-part survey asked caregivers to view each skull model, rank the model’s educational value, and complete demographic questions. For quality control, we embedded in the survey 2 attention check questions (1 multiple choice, 1 sliding bar) and a Completely Automated Public Turing test to tell Computers and Humans Apart verification to exclude automated internet robots. Respondents who successfully completed all survey items and passed the quality control questions were included in the study and compensated $0.25 upon survey completion. This study was reviewed and approved by the Johns Hopkins University Institutional Review Board (IRB001258365).

The survey presented the 2D diagram, 3D-printed, and AR models in random order. Caregivers were assigned to one of the types of craniosynostosis (sagittal, bicoronal, or unicoronal) to prevent survey fatigue. For the 3D-printed model, caregivers were shown video clips of the 3D-printed skull rotated in a user’s hands. Skulls were rotated in a standardized fashion to demonstrate lateral then frontal views of the skull. For the AR model, caregivers were asked to download Augment to their mobile device and scan a quick response (QR) code to view the skull model in the AR space through the app. In Augment, caregivers were free to tap the model to rotate or zoom in and out. For each model, caregivers were asked to rate on a 5-point Likert scale how helpful the model was for learning about skull anatomy, learning about type of craniosynostosis, and viewing an abnormal head shape. Additionally, caregivers were asked to rate how realistic, innovative, and high quality each model was on a 5-point Likert scale. Then, caregivers were shown a static image of the model and asked to click along the fused suture. Once caregivers had seen and rated the 2D, AR, and 3D-printed models individually, they were asked to rank all 3 models in terms of usefulness in learning about craniosynostosis, ability to ease caregiver anxiety, and ability to increase level of trust in surgeons. Lastly, we asked caregivers if they would prefer a generic or patient-specific model (an exact replica of the skull of the person they cared for) of the 3D-printed and AR models they were shown. At the end of the survey, demographics (age, gender, type of craniosynostosis) of the person they cared for with craniosynostosis and their caregiver relationship (eg, parent, sibling, or friend) were collected.

**Statistical Analyses**

We used STATA Statistical Software (StataCorp, College Station, TX) and GraphPad Prism (GraphPad Software, La Jolla, CA) for statistical analyses and figure generation. Rankings were converted to a numeric, inverse preference score such that the highest ranked model (1) resulted in the highest preference score (3). Caregiver preferences for models were compared using 1-way analysis of variance tests. To determine the impact of 3D-printed, AR, or 2D skull models on caregiver understanding of craniosynostosis, we compared caregiver fused suture detection accuracy across models using Chi-Square tests. The significance level was set at $P < 0.05$ for all statistical analyses.

**RESULTS**

Of the 502 completed surveys, a total of 412 (82%) responses were included in the final analysis. We excluded 90 survey responses due to incorrect attention check responses and failed Completely Automated Public Turing test to tell Computers and Humans Apart verification. Over half of the caregivers surveyed were male (50.7%) and Caucasian (56.7%, Supplementary Digital Content, Table 1, http://links.lww.com/SCS/D208). Caregiver age ranged from 20 to 60 years old, with a mean age of 33 years. When asked about their caregiver relationship, caregivers most often identified themselves as the parent (41%) of a child with craniosynostosis. For the 3D-printed models, print time ranged from 13 to 20 hours per model. The average polyethylene terephthalate glycol material cost was $4.95/model. Final 3D-printed skull model dimensions were 8 to 10 cm × 8 to 10 cm, with an average weight of 194 g.

**Caregiver Preferences for Three-Dimensional Printed Versus Augmented Reality Versus Two-Dimensional Models**

Caregivers preferred the 3D-printed and AR models over the 2D diagram for learning about craniosynostosis (mean score difference 3D-printed to 2D: 0.16, $P < 0.05$; mean difference AR to 2D: 0.17, $P < 0.01$), with no significant difference in preference for the 3D-printed versus AR model (Fig. 2). Caregivers felt the AR model most eased their anxiety of the diagnosis compared to the 3D-printed model and 2D diagram (mean difference AR to 3D-printed: 0.20, $P < 0.001$; mean difference AR to 2D: 0.14, $P < 0.05$). Caregivers also felt the AR model and 2D diagram were comparable...
in increasing their trust in a surgeon, and both were preferred over the 3D-printed model (mean difference AR to 3D-printed: 0.14, \( P < 0.05 \); mean difference 2D to 3D-printed: 0.21, \( P < 0.001 \)).

Perceptions of model utility further varied depending on the model’s craniosynostosis type (unicoronal, bicoronal, sagittal). The sagittal craniosynostosis 3D-printed and AR models were perceived as more useful than the 2D diagram in learning about skull anatomy (mean difference 3D-printed to 2D: 0.41, \( P < 0.001 \); mean difference AR to 2D: 0.48, \( P < 0.0001 \)), craniosynostosis type (mean difference 3D-printed to 2D: 0.35, \( P < 0.01 \); mean difference AR to 2D: 0.30, \( P < 0.05 \)), and viewing abnormal head shape (mean difference 3D-printed to 2D: 0.37, \( P < 0.01 \); mean difference AR to 2D: 0.36, \( P < 0.01 \)) with no difference between 3D-printed and AR models (Fig. 3). Additionally, caregivers perceived the bicoronal AR model but not the 3D-printed model to be more helpful in learning about skull anatomy over 2D diagram (mean difference AR to 2D: 0.29, \( P < 0.05 \); Fig. 3). The 3D-printed and AR models for sagittal and bicoronal craniosynostosis (but not unicoronal) were rated as more innovative, realistic, and higher quality than their respective 2D diagrams (Fig. 4), with no difference between AR and 3D-printed models.

Caregiver Preference for Generic Versus Patient-Specific Models

For 3D-printed models, more than half of all caregivers (68%) preferred patient-specific over generic skull models and 65% preferred interactive over static models. For AR models, 70% of caregivers preferred patient-specific over generic models.

Accuracy in Fused Suture Detection

For both the sagittal and bicoronal models, caregivers had significantly increased detection accuracy with the 3D-printed model compared to the AR model and 2D diagram (\( P < 0.05 \) for all, Fig. 5). The highest detection accuracy was seen on the sagittal model, with 36% of caregivers correctly identifying the fused suture with the 3D-printed skull, 19% with the 2D diagram, and 17% with the AR model. The lowest accuracy was seen in the unicoronal model, with 14% of caregivers correctly identifying the fused suture with the AR model, 11% with the 2D diagram, and 7% with the 3D-printed model. Overall, caregivers selected sutures rather than skull bones, but normal, unfused sutures were more often selected than the pathologic, fused suture.

DISCUSSION

Craniosynostosis anatomy and pathology can be challenging for caregivers to understand. Caregivers must navigate the emotional challenges of a congenital diagnosis and conceptualize complex skull anatomy.\(^3\) Current educational modalities used in surgical consultations including verbal explanations and 2D drawings result in caregiver knowledge gaps, and the ideal visual model has not been identified.\(^3,5\) This survey is a pilot effort to understand caregiver preferences of 3D-printed or AR models and their unique
benefits. Our findings may help inform the future development of craniosynostosis visual models for caregiver-oriented education. In our study, caregivers preferred 3D-printed models over 2D diagrams for learning about craniosynostosis and indicated that these models were more realistic and innovative compared to 2D diagrams. Prior studies have documented improved patient understanding of other diseases including hepatic tumors and congenital heart defects using 3D-printed models.10,13 Yang et al10 showed that parental understand of hepatic tumor anatomy increased by 26% after parents were presented with life-size 3D-printed hepatic tumor models. Alshomar et al2 further demonstrated that parents who were shown 3D-printed models were more likely to agree to surgical management, suggesting that models can impact caregiver surgical decision-making in addition to anatomical understanding. Consequently, given caregiver preferences for these models and literature reports of their effectiveness, we support the use of 3D-printed models as a caregiver-oriented educational model for craniosynostosis.

When comparing caregiver preferences for AR models versus 3D-printed models in learning about craniosynostosis, there were overall no significant differences. Previous studies comparing the benefits of 3D-printed and AR in glioma and renal or prostate malignancies have also shown no significant differences in caregiver education.24 This suggests that 3D-printed and AR models may each have unique benefits for learning: in our study, the 3D-printed models provided tactile sensation for raised, physical sutures while the AR models offered the ability to move and rotate digital models in a game-based learning style.25 Other interactive AR patient education tools, such as a pamphlet developed by Lo et al26 to show digital flap reconstructions in AR, have similarly demonstrated significant learning advantages using AR’s interactive nature, corroborating our study findings. Consequently, there is future potential to combine the unique advantages of 3D-printed models and AR models to develop an ideal craniosynostosis caregiver education tool that has both digital AR and 3D-printed features.

Our findings further demonstrated that caregivers preferred interactive, personalized 3D-printed and AR models over generic models, highlighting rising societal interest in personalized care. Personalized care aims to tailor care to the needs of each patient and facilitate a joint decision-making process between the patient and clinician for managing their health.28 Thus, as digital models offer patient-oriented care and shared decision-making, we recommend that craniosynostosis models be personalized to patient anatomy and made interactive either through physical or digital assembled parts. As an example, Chou et al17 developed an interactive, 3D-printed cleft palate model with magnets between the skin and bony skeleton. Parents were able to assemble and disassemble the skin and bony components using the magnets. Future craniosynostosis models with similarly interactive components can provide caregivers with a more preferred, hands-on learning experience.

Interestingly, when caregivers were asked to click on the abnormal, fused suture, caregiver accuracy was significantly higher with the 3D-printed model than the AR model and 2D diagram for both the sagittal and bicoronal models. This is likely due to the increased topological discrepancy of raised, fused sutures on physical 3D-printed models. However, overall accuracy was low (<40%) across all models, perhaps because topological discrepancy was lost when our 3D-printed models were displayed on the survey as a video. This suggests that there is room for improvement in model development and caregiver education. Specifically for 3D-printed models, increased visual saliency of the fused suture can be achieved by printing the edges of the suture in a color different from the skull. In AR models, digital labels can better annotate the fused suture. Likewise, fused sutures in 2D models can be colored, highlighted, or circled. Additionally, as shown in our study results and in Figure 5, caregivers accurately detected sutures but were unable to differentiate the pathologic fused suture from normal, unfused sutures. The difference between unfused and fused sutures may not have been clearly presented in the survey’s written explanations and providing more education on this difference may improve fused suture detection accuracy. This finding also highlights an educational opportunity for surgeons to discuss normal versus fused suture anatomy with caregivers to increase their understanding of craniosynostosis.

The literature demonstrates that 3D printing is become more accessible to medical centers given recent advancements in print technology and readily available open-source printing software.22 However, we acknowledge that not every medical center today has access to 3D printing technology. In this setting, a key takeaway from our findings is that caregivers preferred 3D printed craniosynostosis skull models for their interactive, personalized nature. Future craniosynostosis educational models, whether 3D-printed or not, that emphasize patient-specificity may similarly appeal to caregivers. Additionally, the literature reports free to low start-up costs for developing AR anatomical models in medical institutions using open-source smartphone-based or tablet-based software.28 Given the low-costs of AR model development, AR models may be more accessible moving forward as 3D printing technology is not yet universal across medical centers. Furthermore, AR models can be easily accessed from the patient perspective given that most patients have smartphones.29 Although our study demonstrated no significant difference in preference between 3D-printed and AR models, AR’s increased accessibility and low-cost set-up may make these models easier for medical centers to implement in the future.

We recognize that there are several limitations to this pilot effort. First, we recruited caregivers from craniosynostosis support groups on Facebook. As a result, our caregiver cohort consisted of active social media users who may prefer newer technologies such as 3D-printed and AR models over 2D diagrams. Although this may bias our survey responses, our surveyed caregiver demographic is overall representative of craniosynostosis caregivers in age and other characteristics (majority parents of patients with sagittal craniosynostosis).30 Second, to maintain a virtual survey workflow, our 3D-printed models were shown as videos to caregivers rather than as in-person demonstrations. Although we were unable to replicate in-person demonstrations by showing a video of the model in a user’s hands for size comparisons, caregivers were ultimately only able to view but not touch the physical 3D-printed models. Therefore, future studies with more diverse caregiver cohorts and in-person demonstrations are required to assess caregiver preferences of craniosynostosis models more comprehensively.

CONCLUSIONS

Our findings indicate that caregivers prefer 3D-printed or AR skull models over 2D diagrams for craniosynostosis education. Three-dimensional printed models provide tactile sensation on a life-size model, while AR models provide interactive, game-based learning. Future craniosynostosis models with increased user interactivity and patient-specific components can better suit caregiver preferences. Based on our findings, we recommend surgeons utilize 3D-printed models, AR models, or potentially a combination thereof to improve caregiver understanding of craniosynostosis.

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