**Original Article** 

# 3D Photography to Quantify the Severity of Metopic Craniosynostosis



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#### Abstract

*Objective*: This study aims to determine the utility of 3D photography for evaluating the severity of metopic craniosynostosis (MCS) using a validated, supervised machine learning (ML) algorithm.

Design/Setting/Patients: This single-center retrospective cohort study included patients who were evaluated at our tertiary care center for MCS from 2016 to 2020 and underwent both head CT and 3D photography within a 2-month period.

Main Outcome Measures: The analysis method builds on our previously established ML algorithm for evaluating MCS severity using skull shape from CT scans. In this study, we regress the model to analyze 3D photographs and correlate the severity scores from both imaging modalities.

*Results*: 14 patients met inclusion criteria, 64.3% male (n = 9). The mean age in years at 3D photography and CT imaging was 0.97 and 0.94, respectively. Ten patient images were obtained preoperatively, and 4 patients did not require surgery. The severity prediction of the ML algorithm correlates closely when comparing the 3D photographs to CT bone data (Spearman correlation coefficient [SCC] r = 0.75; Pearson correlation coefficient [PCC] r = 0.82).

*Conclusion*: The results of this study show that 3D photography is a valid alternative to CT for evaluation of head shape in MCS. Its use will provide an objective, quantifiable means of assessing outcomes in a rigorous manner while decreasing radiation exposure in this patient population.

#### **Keywords**

anatomy, computerized tomography, craniofacial morphology, dysmorphology, synostosis

# Introduction

Metopic craniosynostosis (MCS) refers to the premature fusion of the metopic suture. While there is a wide spectrum of severity, MCS classically presents with trigonocephaly, metopic ridge and hypotelorism.<sup>1</sup> MCS is estimated to occur in 1:5000 live births<sup>2</sup> and has been increasing over the past 20 years.<sup>3,4</sup> For uncomplicated cases, diagnosis can be made clinically; however, computed tomography (CT) can be helpful in patients with less obvious abnormalities. Even in cases when the diagnosis of MCS can be made clinically, many craniofacial surgeons obtain a preoperative CT for surgical planning. Over the past decade, considerable effort has been made to limit radiation exposure in children related to their craniosynostosis care, especially during routine follow-up visits, due to the potential risks of cancer development associated with early and repeated CT exposure in young children ([5–7]). While CT protocols have been improved to decrease the radiation exposure per scan for the pediatric population,<sup>6,8–10</sup> the additional risks and costs of CT imaging

impact decisions to use protocols that repeat CT scans throughout the diagnosis and management of craniosynostosis. Without CT imaging, however, post-operative clinical evaluation currently remains subjective.

Recently, 3D photography has been proposed as a radiationfree imaging modality for evaluation of craniosynostosis.<sup>11–13</sup> Similar 3D imaging techniques have been used in orthognathic surgical planning since the 1990s.<sup>14</sup> And 3D photography has since been validated as an accurate technique for assessing

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facial hard and soft tissues,<sup>15–17</sup> as well as in orthodontics<sup>18</sup> severit and oral and maxillofacial surgery.<sup>19</sup> The abundance of dental and maxillofacial literature supporting 3D photography as an accurate, reproducible, and safe technique has since led to the adoption of this technique by plastic and craniofacial surgeons.<sup>12,20–27</sup> Given the current subjectivity involved in diagnosing and evaluating the severity of craniosynostosis as well as assessing morphological outcomes postoperatively, there is growing interest in using 3D imaging combined with statistical

models and/or machine learning (ML) to evaluate cranial dysmorphology, with the hopes of developing standardized diagnosis and treatment algorithms. Several studies have developed models to distinguish between normocephalic skulls and those with craniosynostosis,<sup>12,13,28,29</sup> as well as distinguishing metopic ridge from MCS.<sup>30</sup>

Our group previously validated a supervised ML algorithm to objectively quantify the severity of MCS using CT scans.<sup>31</sup> In this current study, we aim to expand upon the previous studies evaluating the use of 3D photography by applying our ML algorithm to data derived from 3D photographs and comparing the results to head CTs with a goal of finding an alternative, radiation-free method of evaluating MCS head shape pre-operatively and objectively following patients in the postoperative period.

## Methods

## Patient Selection

This single-center retrospective cohort study included patients who were evaluated at our tertiary care center for metopic craniosynostosis from 2016 to 2020 and underwent both head CT and 3D photography within a 2-month period. Patients and controls who underwent 3D photography alone were also included to show the efficacy of the proposed method in MCS classification. Patients with syndromic diagnoses or involvement of other sutures were excluded.

## Image Processing

To process the 3D photography data, several anatomic landmarks are identified and marked by a team member. The nasion and porions are used to define the cropping plane above which the skull is defined. The nasion, porions and left medial canthus are used to consistently orient the images for analysis (Figure 1). The remainder of the processing and analysis is through the machine learning algorithm. The Shapeworks topology preserving smoothing algorithm<sup>31</sup> is used to improve the image quality with regard to 3D photography artifacts (ie. hair, soft tissue deformation from the hairnet, ears). This smoothing algorithm smooths the resulting bony segmentation while preserving the shape's topology.

The CT scans are first segmented using a bone window to identify the skull from the surrounding soft tissue. Preprocessing of the CT scan and severity analysis are performed as previously described.<sup>31</sup> To predict the metopic

severity score from the CT skin window and 3D photograph data, the shape of the corresponding skull is estimated and then used to regress towards the metopic severity. In skull estimation, we use Principal component analysis (PCA)<sup>33</sup> which computes the eigen vectors of the skin covariance matrix and chooses the axes with the most variance to calculate skin's PCA scores (Figure 2). PCA scores are a few numbers that serve as shape descriptors and capture most of the characteristics of the complicated shape. Then the proposed method uses these shape descriptors to regress towards the skull shape. This forms a Principal component regression<sup>34</sup> model that is robust to noise in the data, e.g. measurement errors. Lastly, we predict the severity score using the skull severity regression model previously described by Bhalodia et al.<sup>31</sup>

## Results Analysis and Validation

To validate the severity analysis using 3D photograph data with the previously validated ML algorithm, metopic severity scores calculated from CT bone/skin data were compared to those calculated from 3D photograph data. The metopic severity scores were calculated using these 3 different types of data for patients who have undergone both CT and 3D photography and who were not used to train the models. Statistical analysis with Pearson correlation coefficient (PCC) and Spearman correlation coefficient (SCC) was used to measure the agreement between severity scores obtained from CT data and 3D photograph data; P < .05 was considered statistically significant. Root mean square error (RMSE) was used to measure the consistency of the proposed model using 3D photography with respect to human expert raters using CT bone segmentation.

In addition to quantifying severity, the utility of 3D photography for diagnosis of MCS is also validated. The severity scores from 3D photographs of patients with and without MCS were calculated. The severity scores and diagnosis of MCS were used to calculate the area under curve (AUC) score as a measure of effectiveness of the proposed method for classification using 3D photography.

## Results

Fourteen patients with MCS who underwent 3D photography imaging within 2 months of CT imaging were identified (Table 1). The mean age at 3D photography was  $0.97 \pm 0.28$ years and CT imaging was  $0.94 \pm 0.30$  years (P = .79). Mean severity scores of the 3D photographs and CT bone window images for the entire cohort were  $5.76 \pm 1.93$  and  $5.72 \pm$ 2.29, respectively (P = .97). When examining those who underwent surgical intervention versus those who did not, the mean severity score of the 3D photographs were 6.54 and 3.75, respectively, for CT bone window 6.70 and 3.30, respectively, and for the CT skin window 6.17 and 3.11, respectively. Another 2 patients with MCS and 11 normal controls who underwent only 3D photography were added in the analysis of MCS classification.



**Figure I.** Left: segmenting of CT scans using the otsu threshold for skin segmentation.<sup>32</sup> middle: cropping at the nasion-porion plane of the CT (upper) and 3D photograph (lower). Right: smoothing and alignment of the CT (upper) and 3D photograph (lower).

Pre-operative imaging was evaluated for 10 patients (71.4%), while 4 patients (28.6%) did not undergo corrective surgery due to mild MCS (n = 2) or metopic ridge (n = 2). There was a strong correlation between severity scores of the 3D photographs and CT bone window images, PCC r=0.82, SCC r=0.75, P < .002. The metopic severity scores using CT skull and CT skin were highly correlated, PCC r=0.92, SCC r=0.87, P < .0001. The strongest correlation was found between CT skin and 3D photograph severity scores, PCC r=0.95, SCC r=0.92, P < .0001. These findings are summarized graphically in Figure 3. The AUC for the 3D photography metopic severity is 0.97, indicating very high sensitivity and specificity.

We further computed root mean square error (RMSE) to measure the accuracy of the proposed method using 3D photographs in predicting the severity of MCS. The aggregation of expert ratings is the gold standard severity. The ratings are aggregated using a latent trait model which factors off the different subjective thresholds/bias among the raters,35,36 and corrects the original ratings. Because the severity of expert ratings is not available for the 3D photography studies, we compute RMSE between the predictions using the 3D photography model and CT skull model respectively, as well as RMSE between CT skull model predictions and the gold standard ratings, in order to estimate the overall RMSE. We predict the severity of the 14 patients using the 3D photography model and CT skull model respectively, and their RMSE is 1.34. The original 50 patients (30 patients with MCS and 20 normal controls) that have expert ratings are studied in a stratified 3-fold 3-repeat cross validation, where 2 thirds of studies are used to fit the CT skull model and the rest are used to calculate the predictions and RMSE, and this process was randomly repeated 9 times. The cross validation estimates that the RMSE between CT skull model and the gold standard severity is 1.19. By calculating the quadratic mean of the two RMSEs, the RMSE between the proposed method using 3D photographs and the gold standard severity is 1.79. For comparison, the RMSE between original individual rater ratings and the gold standard severity is 3.53, and the RMSE between corrected individual rater ratings and the gold standard severity is 1.27. The RMSE results suggest the proposed model using 3D photographs is more consistent than expert ratings and comparable with the corrected individual expert ratings.

## Discussion

The purpose of this study is to evaluate the use of 3D photography to quantify the severity of metopic craniosynostosis using a previously validated supervised machine learning algorithm. The use of 3D photography as an adjunct to or instead of CT imaging is increasingly prevalent as practitioners seek to avoid unnecessary radiation exposure at a young age. Additionally, 3D photography provides invaluable insights into the growth and development of the synostotic head shape as patients age postoperatively. We therefore sought to apply our ML processes in order to demonstrate the feasibility of measuring severity in an objective and holistic manner, unbiased by clinician experience and without exposing patients to unnecessary radiation. This information is crucial to better understand the spectrum of severity in metopic craniosynostosis, aid in pre-operative counseling and surgical decision making, and enable future research including longitudinal outcomes assessments and multi-center collaborations, ultimately improving care for our patients.

The results of this study show a strong correlation when comparing the severity scores obtained using CT imaging to the severity scores obtained from 3D photographs, indicating that 3D photography is equivalent to CT scans for evaluating head shape using our ML model. In addition, the RMSE



**Figure 2.** Particle representation and skin regression model. The proposed method first uses Shapeworks to automatically place corresponding points on skull/skin shapes of controls (left upper). Next, the corresponding points are generated for each shape with expert ratings (left lower). Principal component analysis is used to summarize the correspondences using low dimensional shape descriptors (right upper). Each Principal Component is characterized by the deviation of a blue mesh from the mean shape (wireframe) with 3 standard deviations. Finally, severity is assessed based on the predicted skull shape.

Table	١.	Patient	Demogra	phics.
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	Cases
Gender	n (%)
Male	9 (64.3%)
Female	5 (35.7%)
Race	
Caucasian	14 (100%)
Age at imaging (years)	Mean $\pm$ SD (Range)
3D photography	0.97 ± 0.28 (0.25-1.40)
СТ	$0.94 \pm 0.30 (0.18 - 1.40)$
Severity Score	
3D photography	5.76 ± 1.93 (0.29-7.92)
CT bone	$5.72 \pm 2.29$ (1.46-9.33)
CT skin	5.31 ± 1.89 (0.90-7.88)

results show that the proposed method using 3D photographs is consistent in predicting the craniosynostosis severity when compared with human expert individuals. There is a very high correlation when comparing severity scores of the CT skin and CT bone windows. This indicates that CT skull shapes can be accurately extrapolated from CT skin shapes and used for MCS severity quantification. These results strongly support the assumption that CT skin shapes can be a useful alternative for evaluating the severity of MCS instead of CT skull shapes. While weaker than the correlation of CT skin to CT bone windows, the correlation in metopic severity scores between CT bone and 3D photographs is still very strong. The weaker correlation is not unexpected as there is not a perfect correlation between CT skin and CT bone windows, and the 3D photography data often introduces more artifacts that can impact the overall head shape. For instance, a patient's hair can mask the head shape, and the hairnets used when obtaining 3D photographs can compress the soft tissues of the forehead. Furthermore, the model was created using only CT bone window data; no 3D photography data was input into the model prior to testing. Thus, these results suggest that the CT skin and 3D photography data are in good concordance with quantification of metopic severity, validating the efficacy of our model design.



**Figure 3.** Correlation between severity calculated from CT skull vs. CT skin (PCC r = 0.92, SCC r = 0.87, P < .0001) (upper left); correlation between severity calculated from CT skull vs. 3D photograph (PCC r = 0.82, SCC r = 0.75) (upper right); correlation between severity calculated from CT skin vs. 3D photograph (PCC r = 0.92, P < .0001) (lower left); Histogram showing the relative distribution of severities of metopic patients as calculated from 3D photographs compared to control patients evaluated by expert ratings and controls calculated from 3D photographs (lower right).

A wide range of severities was represented in the study cohort, including four patients who were not recommended to undergo corrective surgery by the craniofacial team for either mild MCS or metopic ridge. Ho et al. also compared 3D photography and CT to assess severity of single suture nonsyndromic CS, however they found a statistically significant difference between the imaging modalities indicating that they were comparable but not equivalent. They also suggested that 3D photography may be more appropriate for severe cases rather than mild MCS, and for overall aesthetic evaluation rather than determining underlying bony severity.11 On examination of our cohort, our model performed well across all severities. The patient with the highest correlation between imaging modalities is shown in Figure 4, upper; this patient was found to have mild MCS and was managed nonoperatively. The patient with the most discordant severity scores is shown in Figure 4, lower. The soft tissue of this patient's forehead masked some of the angularity of the underlying bony structure, decreasing the severity score on 3D photograph analysis which supports the findings by Ho et al. However, the severity score from the 3D photograph in this patient remains high, correctly indicating a higher severity of MCS. These findings validate the use of 3D photography as a radiation free alternative to CT imaging for preoperative severity analysis of MCS across a broad spectrum of severities.

We have previously shown that our algorithm is a valid method for quantitative, objective analysis of metopic head shape severity using CT.<sup>37</sup> In cases where the diagnosis is unclear, 3D photography can aid in diagnosis and delay CT imaging until the patient is older and closer to the pre-operative period, if surgical correction is indicated. Alternatively, 3D photography can eliminate the need for a CT if it is determined that the clinical presentation doesn't warrant surgical intervention. While 3D photography cannot replace CT for surgical planning (identification of trans-osseus dural communication, for example), it has the potential to reduce the amount of radiation children with MCS are exposed to by delaying or eliminating CT imaging in select cases. This is particularly important in decreasing the overall effective radiation exposure patients with craniosynostosis may encounter. In our prior work, we determined that delaying imaging from 6 months to 12 months decreases effective radiation dose by 27.7%.<sup>7</sup>

3D photography also has potential value in evaluation of long-term surgical follow up outcomes. Following surgery, it is not necessary to obtain CT imaging at a regular interval for uncomplicated patients and doing so would expose these patients to unnecessary radiation exposure. Furthermore, clinical examination is subjective and biased by clinician experience. Wilbrand et al. found 3D photographs were a valuable method for objective analysis of perioperative changes, such as symmetry and volumetrics, in patients who underwent corrective surgery for craniosynostosis.<sup>25</sup> Le et al. compared long-term outcomes of open versus endoscopic operative techniques for sagittal CS using 3D photography<sup>38</sup> Studies are underway to apply our ML algorithm to postoperative patient images to provide an objective, quantitative means of assessing longitudinal outcomes that considers the entire head shape rather than reductive 2D metrics. Our ML algorithm is currently freely



Figure 4. Upper: 3D photograph (left) and CT image (right) of the patient with highest concordance. Lower: 3D photograph (left) and CT image (right) of the patient with lowest concordance.

available for clinician use on our online portal (https://www. craniorate.org/), where clinicians can upload deidentified CT scans for automatic processing, severity analysis, and comparison to other patients with MCS. Other future directions of this work include expanding the capability of our online portal to be able to automatically process and analyze 3D photographs.

Limitations include the relatively small sample size; our 3D photography machine was first employed in our clinic in December 2018. Its use has drastically increased, but MCS is still a relatively rare diagnosis. Additionally, all patients in this cohort were Caucasian; CS is more common in Caucasians and the one African American patient with MCS who met inclusion criteria was excluded due to hairstyle interfering with head shape analysis (Figure 5, upper). This is an inherent limitation of the 3D photography system. Hair poses a significant issue to cranial shape analysis; this is especially relevant for African American children and older Caucasian children with more hair (females). Infants are relatively bald

making head shape analysis easier. While the hairnet is able to compress much of the hair, there is a limit to how much hair can be compressed. For children with long enough hair, pulling it back into a tight, low ponytail may assist in flattening the hair allowing for analysis. However, certain hairstyles, such as braids, twists, buns, and other 3D styles and/or hair ornaments can interfere with 3D photography analysis. Asking families to help with hair styling prior to visits may be an option, however families may not be interested in changing their child's hairstyle for a doctor's visit. Additionally, soft tissue does not exactly conform to the shape of the skull and thus some aspects of the underlying bony structure may be masked, as shown in Figure 4, lower. Furthermore, the hairnet can compress the soft tissue of the forehead impacting analysis. Facial expressions, such as grimacing or crying, give the forehead a more triangular appearance (Figure 5, lower) and efforts should be made to obtain photos with the face in a neutral position. Multiple studies have investigated



Figure 5. Upper: An example of hair shape and density interfering in 3D photograph analysis in an African American patient. Lower: An example of a crying patient, which resulted in artificially worsened trigonocephaly.

alternatives to computed tomography for analysis of craniosynostosis. While black bone MRI was found to be equivalent to CT for analysis of the cranial sutures and decreases radiation exposure, a significant downside of this imaging modality is the time required to obtain the imaging and the subsequent need for sedation in this young patient population.<sup>39,40</sup> 3D photography, while not a panacea, offers an alternative means of evaluation of craniosynostosis that can aid in reducing radiation exposure in this young patient population.

# Conclusion

3D photography is a valid alternative to CT for evaluation of head shape, its use will provide an objective, quantifiable

manner to assess outcomes in a rigorous manner while decreasing radiation exposure in this patient population.

## **Declaration of Conflicting Interests**

The author(s) declared no potential conflicts of interest with respect to the research, authorship, and/or publication of this article.

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