Correction of trigonocephaly after endoscopic strip craniectomy with postoperative helmet orthosis therapy: a 3D stereophotogrammetric study

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OBJECTIVE Endoscopic strip craniectomy with postoperative helmet orthosis therapy (ESCH) has emerged as a less invasive alternative to fronto-orbital remodeling for correction of trigonocephaly. However, there is no standardized objective method for monitoring morphological changes following ESCH. Such a method should be reproducible and avoid the use of ionizing radiation and general anesthesia for diagnostic imaging. The authors analyzed a number of metrics measured using 3D stereophotogrammetry (3DSPG) following ESCH, an imaging alternative that is free of ionizing radiation and can be performed on awake children.

METHODS 3DSPG images obtained at two time points (perisurgical and 1-year follow-up [FU]) of children with metopic synostosis who had undergone ESCH were analyzed and compared to 3DSPG images of age-matched control children without craniofacial anomalies. In total, 9 parameters were measured, the frontal angle and anteroposterior volume in addition to 7 novel parameters: anteroposterior area ratio, anteroposterior width ratios 1 and 2, and right and left anteroposterior diagonal ratios 30 and 60.

RESULTS Six eligible patients were identified in the operated group, and 15 children were in the control group. All 9 parameters differed significantly between perisurgical and age-matched controls, as well as from perisurgical to FU scans. Comparison of FU scans of metopic synostosis patients who underwent surgery to scans of age-matched controls without metopic synostosis revealed that all parameters were statistically identical, with the exception of the right anteroposterior diagonal ratio 30, which was not fully corrected in the treated patients. The left anterior part of the head showed the most change in surface area maps.

CONCLUSIONS In this pilot study, ESCH showed satisfactory results at 1 year, with improvements in all measured parameters compared to perisurgical results and normalization of 8 of 9 parameters compared to an age-matched control group. The results indicate that these parameters may be useful for craniofacial units for monitoring changes in head shape after ESCH for trigonocephaly and that 3DSPG, which avoids the use of anesthesia and ionizing radiation, is a satisfactory monitoring method.

https://thejns.org/doi/abs/10.3171/2022.2.PEDS21546

KEYWORDS metopic synostosis; craniosynostosis; trigonocephaly; endoscopic surgery; stereophotogrammetry; craniofacial

**ABBREVIATIONS** APAR = anteroposterior area ratio; APDR = anteroposterior diagonal ratio; APVR = anteroposterior volume ratio; APWR = anteroposterior width ratio; CI = cephalic index; control(; ) = perisurgical comparison control group; control(1y) = 1-year postoperative control group; ESCH = endoscopic strip craniectomy with postoperative helmet orthosis therapy; FA30 = 30° diagonal frontal angle; FOAR = fronto-orbital advancement/remodeling; FU = follow-up; MAP = most anterior point; P.0 = point zero of the measurement plane; 3dMD = 3dMDhead System; 3DSPG = 3D stereophotogrammetry.
and short hospital stays.\textsuperscript{3–5} ESCH outcomes equivalent or superior to those obtained with open techniques have been reported for several studies.\textsuperscript{6–8} However, assessment of morphological outcomes of metopic synostosis repair remains subjective, relying on aesthetic assessment by individuals.\textsuperscript{9} Several centers have attempted to describe objective parameters to evaluate trigonocephaly correction.\textsuperscript{9–13} Imaging for these studies was performed with CT scans, which produce accurate, high-quality images, but the CT scanning procedure exposes children to ionizing radiation and may require anesthesia. 3D stereophotogrammetry (3DSPG), in which 2D images captured by multiple cameras are merged to produce a 3D image,\textsuperscript{14} is an attractive alternative to CT,\textsuperscript{15–19} as it is free of ionizing radiation and can be produced on awake children.

A number of craniofacial centers have proposed that measurements be obtained to objectively quantify change following trigonocephaly surgery, using both CT and 3DSPG images (Table 1). Most studies focus on the forehead to assess outcomes, presumably due to fact that FOAR addresses this region only. No previous studies, to our knowledge, have included the posterior hemisphere of the head and used the resulting data to evaluate the effect of helmets on the growth of not only the forehead but also the head and used the resulting data to evaluate the effect of helmets on the growth of not only the forehead but also the head.

In this study, we examined the use of several craniometrics to characterize trigonocephaly and quantified the morphological outcomes of ESCH by using 3DSPG.

### Methods

#### Study Design and Patient Selection

This was a retrospective cohort study that included two groups of children who were identified from our hospital electronic records. 1) The operative group included children with metopic synostosis managed with ESCH, all of whom underwent 3DSPG scans, which in our hospital are performed routinely in all children undergoing ESCH, regardless of diagnosis, severity, or socioeconomic circumstances. 2) The control group included children who were age matched to children in the operative group and had 3DSPG scans but did not have any diagnosis of craniofacial anomalies.

### Operative Group

We assessed the study eligibility of all children with metopic synostosis treated with ESCH at our institution. Inclusion criteria were diagnosis of nonsyndromic single-suture metopic synostosis and treatment with ESCH, availability of 3DSPG scans from the perisurgical period (defined as preoperative or within 7 days after surgery), and follow-up (FU; defined as 10–14 months postoperatively). Exclusion criteria were involvement of other sutures or lack of 3DSPG scans at the preoperative and perisurgical time points.

The diagnosis of metopic synostosis is made by our multidisciplinary craniofacial team, including pediatric neurosurgeons and plastic surgeons. We do not routinely use CT imaging for cases of classic phenotype single-suture metopic synostosis and treatment with ESCH, availability of 3DSPG scans from the perisurgical period (defined as preoperative or within 7 days after surgery), and follow-up (FU; defined as 10–14 months postoperatively). Exclusion criteria were involvement of other sutures or lack of 3DSPG scans at the preoperative and perisurgical time points.

#### TABLE 1. Published studies for objective assessment of metopic synostosis outcomes after corrective surgeries with parameters and imaging modalities used

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>Comparison Groups</th>
<th>Craniomeric Parameters</th>
<th>Imaging</th>
</tr>
</thead>
<tbody>
<tr>
<td>Martini et al., 2015\textsuperscript{10}</td>
<td>Pre- vs postop of conventional op techniques</td>
<td>Frontal &amp; 2 frontoparietal angles &amp; CI</td>
<td>3DSPG</td>
</tr>
<tr>
<td>Wang et al., 2016\textsuperscript{10}</td>
<td>Conventional outcomes vs controls</td>
<td>Metopic index</td>
<td>CT</td>
</tr>
<tr>
<td>Rodríguez-Florez et al., 2017\textsuperscript{10}</td>
<td>Conventional outcomes vs nonsynostotic controls</td>
<td>Forehead volume, interfrontoparietal/interparietal ratio, &amp; frontal angle</td>
<td>3DSPG</td>
</tr>
<tr>
<td>Pressler et al., 2021\textsuperscript{16}</td>
<td>Limited strip craniectomy vs conventional</td>
<td>Vector measurements from a center point to surface landmarks on 180° contour of forehead</td>
<td>3DSPG</td>
</tr>
<tr>
<td>Ha et al., 2020\textsuperscript{9}</td>
<td>Endoscopic vs conventional outcomes</td>
<td>Frontal width, interfrontal divergence angle, &amp; presence of lateral frontal retrusion</td>
<td>CT</td>
</tr>
<tr>
<td>Lajthia et al., 2021\textsuperscript{13}</td>
<td>Pre- vs postop of endoscopic</td>
<td>Interfrontal divergence angle, head circumference, interdacyron distance, &amp; intercanthal distance to outer canthal distance ratio</td>
<td>CT</td>
</tr>
<tr>
<td>Ramsey et al., 2021\textsuperscript{19}</td>
<td>Endoscopic vs nonsynostotic deformity controls</td>
<td>FA\textsubscript{30}</td>
<td>3DSPG</td>
</tr>
<tr>
<td>Schulz et al., 2021\textsuperscript{15}</td>
<td>Endoscopic vs conventional vs controls</td>
<td>Circumference index, 30° diagonal index, exocanthion index, &amp; nasofrontal angle</td>
<td>CT</td>
</tr>
<tr>
<td>McKee et al., 2021\textsuperscript{11}</td>
<td>Metopic synostosis, metopic ridge, &amp; normal controls</td>
<td>Metopic index &amp; intracranial volume</td>
<td>CT</td>
</tr>
<tr>
<td>Baş &amp; Baş, 2021\textsuperscript{13}</td>
<td>Endoscopic vs conventional</td>
<td>Metopic angle, CI, interparietal distance, intercoronal distance, &amp; their ratios to each other</td>
<td>CT</td>
</tr>
</tbody>
</table>

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Control Group

We have compiled a database of 3DSPG and CT (with 3D soft-tissue reconstruction) images of children without craniofacial deformity who underwent 3DSPG scanning either as volunteers or for an unrelated clinical reason (e.g., epilepsy; CT). The ages of the patients in the perisurgical and FU groups were defined by range, and the control groups were populated with all cases in between the same ranges to make two age-matched groups: perisurgical comparison control group [controlt0]; and FU 1-year postoperative control group [controlt+1y]. Exclusion criteria included craniofacial anomaly diagnosis, presence of soft-tissue swelling such as posttrauma, or soft-tissue distortion (e.g., by head support).

The study was registered with the Great Ormond Street Hospital Clinical Audit Department.

3DSPG Scans

3DSPG images were acquired by using one of the following imaging systems: 3dMDhead System (3dMD), M4D scan (Rodin 4D, Pessac), or TechMed3D body scanner (TechMed3D) at two time points: perisurgical (preoperative or 7 days after surgery) and 1-year postsurgery FU (10–14 months). Images were digitally imported to a graphics workstation and initial preprocessing of the scans, such as trimming artifacts, was done using 3-Matic version 15 (Materialise). The preprocessed 3DSPG images were imported as stereolithography (STL)–format files into 3dMD Vultus software. The STL file contains the 3DSPG images as a 3D mesh structure composed of triangular polygons. The following landmarks were used for registration of the 3DSPG images into a consistent alignment: both exocanthions, both endocanthions, both preaurales, both superaurales, and the sellion.

The preaurale was defined as the most anterior point of the ear, at the level where the helix merges to the scalp.22 The sellion was defined by the deepest (most posterior) point of the frontonasal soft tissue contour in the midline of the base of the nasal root23 (Fig. 1A).

The meshes were aligned in a 3-coordinate system, where the x-axis passed through both preaurales, the z-axis is passed through the sellion perpendicular to the x-axis, and the y-axis passed vertically perpendicular to x- and z-axes. A plane was defined through three points—both preaurales and the sellion—and was considered the base plane at which the scans were trimmed, and the surface meshes below were removed (Fig. 1B).

The head height was defined by the distance between the base plane and the vertex (the highest point of the head). The axial plane parallel to the base plane through the most anterior point (MAP) of the forehead was used as our initial measurement plane for perisurgical scanning (Fig. 1A). For consistency, and to ensure that the same plane was used at different time points in the same patient, the distance between the measurement plane and the base plane was determined, divided by the head's height, and then multiplied by 100. The resulting percentage defines the proportional level of the measurement plane to the total head height. This measurement plane value was recorded and used consistently in the analysis of every scan for the same patient (i.e., at FU).

Landmarking

We defined the origin of the trimmed 3D mesh as the center of mass by averaging all vertices, similar to the published computed cranial focal point.24 Point zero of the measurement plane (P.0) was created where the y-axis intersects with the measurement plane.

Six vector indicators were created in a radial manner, originating from P.0 in the center to the outer contour of the mesh at 30° increments in the measurement plane, similar to a clockface. This resulted in 12 points, where the vector indicators intersect the head contour, in addition to 12 vectors. These 12 points were chosen as landmarks on the measurement plane to describe the 360° contour of the head. These points were named according to their clockwise position as observed from the top: point 12 (P.12; 12 o’clock) represents the MAP in the midline, and P.6 (6 o’clock) is directly opposite to P.12 posteriorly (Fig. IC). The plane between P.3 and P.9 is perpendicular to the midpoint of the midline and divides the mesh into the anterior contour (landmarks P.10, P.11, P.12, P.1, and P.2) and the posterior contour (landmarks P.4, P.5, P.6, P.7, and P.8).

Vectors were measured from the P.0 to each landmark for all four groups (perisurgical, FU, controlt0, and controlt+1y). Measurements were taken by two independent observers of different academic backgrounds (A.E., neurosurgeon; and L.S., design engineer).

Cranio metric Parameters

The posterior half of the head was included in the analysis to measure the change in the anterior relative to the posterior landmarks. To eliminate the effect of normal growth, ratios were used in preference to absolute values. Therefore, we generated 6 craniometric parameters using the vectors described above. The anterior and posterior halves of the head were defined by a line connecting the P.3 and P.9 landmarks (in 3D space, this is the xy-plane from the origin), while the right and left halves are bisected by the midline, i.e., a line connecting P.12 and P.6. Using these vectors, we generated the following 6 novel cranio metric parameters: 1) anteroposterior (vector P.1–P.11/vector P.7–P.5) width ratio 1 (APWR1) and 2) anteroposterior (vector P.10–P.2/vector P.8–P.4) width ratio 2 (APWR2). APWR1 and APWR2 indicate horizontal/lateral widening of the forehead (Fig. 1E1 and E2). 3) Right anteroposterior (vector P.0–P.1/vector P.0–P.7) diagonal ratio 30 (rAPDR30); 4) left anteroposterior (vector P.0–P.11/vector P.0–P.5) diagonal ratio 30 (lAPDR30).

The APDR30 measurements describe the change in the medial supraorbital region. 5) Right anteroposterior (vector P.0–P.2/vector P.0–P.8) diagonal ratio 60 (rADPR60); and 6) left anteroposterior (P.0–P.10/vector P.0–P.4) diagonal ratio 60 (lADPR60). The ADPR60 measurements describe the change in the lateral supraorbital region (Fig. 1F and F4).

A seventh novel measurement is the anteroposterior area ratio (APAR). For APAR, we used 3DMD to generate the surface areas of the entire anterior and posterior meshes (superior to the base plane), and a ratio is generated therefrom (anterior:posterior). This ratio describes differential growth in the scalp surface area between the front and back half of the neurocranium (Fig. 1B).
We also measured 2 previously published parameters. The anteroposterior volume ratio (APVR) is calculated by dividing the volume of the anterior half of the 3D mesh by the posterior half. Similar to the APAR, the APVR describes the differential growth of the anterior and posterior half of the neurocranium. The 30° diagonal frontal angle (FA30) is the angle measured between the vector P11–P12 and P12–P1 (Fig. 1D). It describes the angulation of the forehead in the horizontal plane, with a more obtuse angle describing an improvement in deformity. These 9 parameters are summarized in Table 2.

Subjective Outcomes
We classified the subjective aesthetic satisfaction of the parents and surgeon 1 year after surgery as follows: 1) satisfied; 2) minor concerns (comment recorded but no revision requested or offered); or 3) major concerns (revisional surgery requested or offered).

Statistical Analysis
For comparison between perisurgical and FU, we used the Wilcoxon signed-rank paired test. For comparison between operative and control groups at both time points, we
used the independent Mann-Whitney U-test. Results were reported in mean and median, but the p value represents the difference in median and was classified as significant if \( p < 0.05 \), highly significant if \( p < 0.01 \), and not significant if \( p \geq 0.05 \).

**Results**

The intraclass correlation coefficient between the measurements by the two observers was 0.997, denoting excellent agreement.

**Operative Group**

In total, 16 children with a diagnosis of metopic synostosis and treatment with ESCH were identified as potential study participants. Following application of the inclusion and exclusion criteria, 6 of these children were included (Fig. 2). All 6 were male, with a mean age at surgery of 4.2 months (range 3–5 months) and a mean age at 1-year FU 3DSPG of 16.7 months (range 15–19 months).

**Control Group**

We identified 9 children for inclusion in the perisurgical comparison control group (control\(_{t0}\)). These children had a mean age of 4.8 months (range 2–8 months; 5 males and 4 females). Six children met the inclusion criteria for the 1-year postoperative control group (control\(_{t+1y}\)), with a mean age of 18 months (range 15–21 months; 3 males and 3 females). The ages were compared between the surgical group and the two control groups and no significant difference was detected (control\(_{t0}\) \( p = 0.44 \), control\(_{t+1y}\) \( p = 0.34 \)).

**Perisurgical Compared With Control\(_{t0}\)—Difference Between Children With Metopic Synostosis and Control Children at the Perisurgical Time Point**

All 9 craniometric parameters measured in this study were statistically significantly different between the operative and the control groups at the perioperative time point. This difference was highly significant for FA\(_{30}\) (frontal angle), APWR, (anterior forehead width), APWR\(_{2}\) (posterior forehead width), rAPDR\(_{30}\) (right medial supraorbital ratio), IAPDR\(_{30}\) (left medial supraorbital ratio), and rAPDR\(_{60}\) (right lateral supraorbital width), and significant for APAR (surface area ratio of anterior to posterior neurocranium), APVR (volume ratio of anterior to posterior neurocranium), and IAPDR\(_{60}\) (left lateral supraorbital width) (Figs. 3 and 4; Table 3).

**Perisurgical Compared to FU—Change in Head Shape in Metopic Synostosis Following ESCH at 1 Year Postoperation**

To assess whether ESCH surgery caused a proportional morphological change, we measured the described craniometric parameters at the perisurgical time point and compared them statistically to the same parameters measured in the same 6 patients 1 year after surgery (FU). For all 9 parameters, there was a statistically significant improvement between the two time points (Figs. 3 and 4; Table 3).

**FU Compared to Control\(_{t+1y}\)—Comparison of ESCH Operated Patients 1 Year After Surgery With Age-Matched Normal Controls**

To assess whether ESCH affects correction of cranial morphology in patients compared with the normal population, the 9 craniometric parameters measured in patients at 1 year after surgery (FU) were compared against those of the control group patients without craniofacial deformity at the same age (control\(_{t+1y}\)). For 8 of 9 parameters (FA\(_{30}\), APAR, APVR, APWR, rAPDR\(_{30}\), IAPDR\(_{30}\), rAPDR\(_{60}\), IAPDR\(_{60}\), Table 2).

### TABLE 2. Parameters used for analyzing the outcome of ESCH (FA\(_{30}\))^19 anteroposterior ratios: area, volume,^25 width, and diagonal ratios

<table>
<thead>
<tr>
<th>Measurement Parameter</th>
<th>Description</th>
<th>Importance</th>
</tr>
</thead>
<tbody>
<tr>
<td>FA(_{30})</td>
<td>Angle between P.11–P.12 &amp; P.12–P.1</td>
<td>Angular deformity of forehead</td>
</tr>
<tr>
<td>APAR</td>
<td>Ratio of anterior half surface area to posterior half surface area</td>
<td>Indicator of change in surface area</td>
</tr>
<tr>
<td>APVR</td>
<td>Ratio of anterior half volume to posterior half volume</td>
<td>Indicator of change in volume</td>
</tr>
<tr>
<td>APWR(_1)</td>
<td>Ratio of vector P.11–P.1/ vector P.7–P.5</td>
<td>APWR(_1) &amp; APWR(_2) are indicators of change in x-axis</td>
</tr>
<tr>
<td>APWR(_2)</td>
<td>Ratio of vector P.10–P.2/ vector P.8–P.4</td>
<td></td>
</tr>
<tr>
<td>rAPDR(_{30})</td>
<td>Ratio of vector P.0–P.1/ vector P.0–P.7</td>
<td>rAPDR(<em>{30}) &amp; IAPDR(</em>{30}) are indicators of change in 30° diagonal direction</td>
</tr>
<tr>
<td>IAPDR(_{30})</td>
<td>Ratio of vector P.0–P.11/ vector P.0–P.5</td>
<td></td>
</tr>
<tr>
<td>rAPDR(_{60})</td>
<td>Ratio of vector P.0–P.2/ vector P.0–P.8</td>
<td>rAPDR(<em>{60}) &amp; IAPDR(</em>{60}) are indicators of change in 60° diagonal direction</td>
</tr>
<tr>
<td>IAPDR(_{60})</td>
<td>Ratio of vector P.0–P.10/ vector P.0–P.4</td>
<td></td>
</tr>
</tbody>
</table>

**FIG. 2. CONSORT diagram showing enrollment of patients after the application of inclusion and exclusion criteria.**
APAR, APVR, APWR1, APWR2, IAPDR30, and both left and right APDR60), no significant difference was seen, a finding that indicates normalization in ESCH patients. One parameter showed a significant difference (rAPDR30); however, at the perisurgical time point this parameter was highly significantly different, indicating an improvement over time (but not complete normalization). No parameter was highly different between FU and controlt+1y (Figs. 3 and 4; Table 3).

Controlt0 Compared to Controlt+1y—Comparison of the Two Points in Normal Control Children to Assess Normal Skull Growth

We examined changes in the 9 parameters between the two control groups to verify whether cranial growth in normal children stays in proportion between the time points. In 8 of 9 children, parameters did not change between the two groups, suggesting cranial growth was proportional over this period. There was a significant change...
in FA\(_{30}\), with the angle becoming slightly more acute over time (mean of 142.7° at control\(_{0}\) compared to 136.5° at control\(_{1y}\)) (Figs. 3 and 4; Table 3).

### Lateral Symmetry of Skull Growth

Analyzing the differences at FU between the right side (represented by rAPDR\(_{30}\) and rAPDR\(_{60}\)) and the left side (represented by lAPDR\(_{30}\) and lAPDR\(_{60}\)) provided an assessment of the lateral growth. In the FU group, there was a statistically significant difference between rAPDR\(_{30}\) and lAPDR\(_{30}\) (p = 0.046), with slightly more growth of the left medial supraorbital region. However, there was no difference between rAPDR\(_{60}\) and lAPDR\(_{60}\) (p = 0.60).

### Extent of Skull Growth—Comparison of Anterior and Posterior Neurocranium

The means of the vector measurements of the landmarks at the perisurgical and FU periods were used to determine the differential growth between anterior and posterior halves of the head. The extent of growth of each landmark was calculated as the difference between mean vector measurements of the landmark at FU and perisurgical scans (Fig. 5).

The anterior half landmarks showed higher growth than those in the posterior half (p = 0.043), except the midline points (P12 and P6), where P6 grew slightly more than P12 (mean 8.3 vs 7.8 mm, respectively). The points with the highest growth were P11 and P1 (medial supraorbital region), with means of 12.6 and 11.2 mm, respectively, followed by P10 and P2 (lateral supraorbital region), with a mean of 9 mm for both. Growth at the left medial supraorbital region (P11) exceeded that at the right side (P1).

### Subjective Satisfaction

No major concerns were recorded regarding the head shapes at 1 year postsurgery by either parents or treating surgeons. Minor concerns were noticed in 2 patients (mild bitemporal indentation, minor forehead asymmetry).

### Discussion

This study demonstrates that 3DSPG can be used to assess cranial morphology in trigonocephaly, and to assess the change following ESCH treatment. We used a combination of previously described and novel craniometric parameters to describe the deformity and change. We have demonstrated that there are significant differences in these parameters between untreated children with metopic synostosis and age-matched controls, and that ESCH treatment normalizes these parameters 1 year after surgery. Different regions of the neurocranium appear to normalize to different degrees, which may suggest potential areas of refinement of operative and orthotic techniques.

### Use of 3DSPG and Assessment of Morphology After ESCH

The majority of studies examining ESCH in metopic synostosis have focused on surgical factors, such as perioperative data, length of surgery/admission, blood products, and complications.\(^2,3,5,7\) Compared to traditional open FOAR, ESCH tends to be superior in these factors. However, the goal of surgery is not only to be as minimally invasive and safe as possible, but also to affect complete normalization of the deformity. It has been acknowledged that shape outcomes as assessed subjectively by clinicians/parents do not always correlate with objective craniometric data.\(^\text{15}\) Unlike in sagittal synostosis, where the cranial/cephalic index (CI) is widely accepted as a useful (if imperfect) method for recording deformity, there is an absence of universally accepted parameters in trigonocephaly, despite many suggested measurements.\(^\text{10,11,13,15,19,25,26}\) One restriction has been the need for CT scans to measure 3D morphology in this group of children. The use of CT in children has obvious disadvantages, such as the exposure of the developing brain to ionizing radiation and the need for sedation/anesthesia in many children, as well as being expensive and inconvenient. Over the last few years, a number of groups have started using 3DSPG as an alternative method of generating 3D meshes, being radiation-free, not requiring sedation, portable, and cheap.\(^\text{27,28}\) It also has the advantage of familiarity—orthotists routinely use 3DSPG as an aid to guide the manufacture and adjustment of helmets. We recommend the use of 3DSPG for monitoring cranial

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**Table 3. Results of perisurgical, control\(_{0}\), FU, and control\(_{1y}\) patient groups**

<table>
<thead>
<tr>
<th>Parameter</th>
<th>Perisurgical</th>
<th>Control(_{0})</th>
<th>FU</th>
<th>Control(_{1y})</th>
</tr>
</thead>
<tbody>
<tr>
<td>FA(_{30})</td>
<td>116.2° (108°–125.5°)</td>
<td>140.6° (138.5°–154.3°)</td>
<td>132.35° (122°–140°)</td>
<td>136.8° (131.9°–142.3°)</td>
</tr>
<tr>
<td>APAR</td>
<td>0.86 (0.80–0.89)</td>
<td>0.91 (0.85–1.02)</td>
<td>0.93 (0.89–0.96)</td>
<td>0.93 (0.86–0.99)</td>
</tr>
<tr>
<td>APVR</td>
<td>0.79 (0.72–0.91)</td>
<td>0.91 (0.78–1.06)</td>
<td>0.89 (0.84–0.93)</td>
<td>0.90 (0.80–1)</td>
</tr>
<tr>
<td>APWR(_{r})</td>
<td>0.89 (0.84–0.92)</td>
<td>0.99 (0.95–1.05)</td>
<td>0.95 (0.90–0.98)</td>
<td>0.98 (0.96–1.03)</td>
</tr>
<tr>
<td>APWR(_{l})</td>
<td>0.89 (0.85–0.96)</td>
<td>0.97 (0.88–1.04)</td>
<td>0.94 (0.92–0.99)</td>
<td>0.98 (0.91–1.02)</td>
</tr>
<tr>
<td>rAPDR(_{30})</td>
<td>0.89 (0.84–0.93)</td>
<td>1 (0.94–1.03)</td>
<td>0.94 (0.88–0.97)</td>
<td>0.98 (0.94–1)</td>
</tr>
<tr>
<td>rAPDR(_{60})</td>
<td>0.88 (0.83–0.92)</td>
<td>0.99 (0.96–1.08)</td>
<td>0.96 (0.91–0.98)</td>
<td>0.98 (0.96–1.06)</td>
</tr>
<tr>
<td>lAPDR(_{30})</td>
<td>0.89 (0.85–0.94)</td>
<td>0.99 (0.90–1.05)</td>
<td>0.93 (0.91–1)</td>
<td>0.98 (0.92–0.99)</td>
</tr>
<tr>
<td>lAPDR(_{60})</td>
<td>0.89 (0.85–0.96)</td>
<td>1 (0.85–1.06)</td>
<td>0.94 (0.92–1.01)</td>
<td>0.98 (0.91–1.05)</td>
</tr>
</tbody>
</table>

NS = not significant (p ≥ 0.05); + = significant (p < 0.05); ++ = highly significant (p < 0.01).

Data are reported as median (range). Significant and highly significant comparisons in the control\(_{0}\) and FU columns refer to comparison with data in the perisurgical column. Significant and not significant comparisons in the control\(_{1y}\) column refer to comparison with the FU column. Comparisons of control\(_{0}\) with control\(_{1y}\) were not significant except for FA\(_{30}\).
growth following ESCH, and, indeed, other craniosynostosis surgeries.

At our unit, all patients with craniofacial abnormalities are offered 3DSPG images for our records regardless of their condition or socioeconomic status as 3DSPG is a part of our service. Therefore, we believe that the proposed metrics in this study can be used for both clinical and research purposes. Clinically, they provide a detailed analysis of the morphological changes in 2D and 3D fashions, which can help guide the orthotic management. Our objective measurements appear to correlate with subjective observations, such as the presence of an asymmetrical forehead in one patient. Statistical correlation was not possible due to the small sample size. We feel the reproducibility and radiation-free nature of 3DSPG lends it well to longitudinal FU in craniosynostosis, compared to traditional tools such as 2D photosets and CT scanning.

The proposed parameters used with 3DSPG are advantageous owing to their ease of use, reliability, and availability to clinicians and orthotists. They can be measured by using any 3D analysis software, including freeware. For the control group, we used 3D soft-tissue images driven from CT scans which have been reported to be interchangeable with 3DSPG.

The existing literature on morphometrics of trigonocephaly surgery focuses almost entirely on the forehead shape, presumably as the standard operation (FOAR) corrects the fronto-orbital region only. To our knowledge,
the only exception was a recent study reporting the circumference index and 30° diagonals index following ESCH. We consider trigonocephaly a whole-head deformity and recommend inclusion of morphometrics (such as ours) that capture the posterior hemisphere of the head as well as the forehead.

Changes in Cranial Morphology Following ESCH

ESCH induced improvement in all 9 metrics examined in this study at 1 year after the operation. When we compared treated metopic patients to normal age-matched controls (i.e., FU vs control), it was gratifying to see no difference between the groups for the majority (8/9) of craniometric parameters, suggesting that ESCH achieves a good outcome. The only parameter that improved but did not “normalize” was rAPDR (right medial supraorbital ratio). This finding initially caused us some concern, but we have seen clinical improvement in our patients for this contour during early childhood. No parents of children treated at our institution have requested surgical revision for this parameter, and revision rates reported in the literature are low. However, we will continue to monitor the rAPDR in our patients and plan to publish a FU report of our investigations of later improvement in this metric.

The comparative undercorrection of the right side, as opposed to the left, is interesting. First, there does exist some inherent asymmetry in this region in trigonocephaly as can be seen in the results for the perisurgical patients compared to control. While the deformity of metopic synostosis is often described as “symmetrical,” those surgeons who perform surgery in these cases recognize there is often a subtle difference in contour between the two sides which needs to be accounted for (e.g., during FOAR). However, there may well be a technical component to this. During endoscopic surgery, the working channel is quite narrow, and a right-handed surgeon may tend to find accessing the left side of the strip craniectomy easier, leading to increased bone removal on this side. Since we observed this possible right-sided undercorrection in our study patients, the surgical team has been assiduously measuring the width of the craniectomy along its whole extent at the end of the operation and ensuring complete symmetry. We will reexamine our results with our new technique and report whether we avoid this in the future.

Present Study Limitations and Future Study

The obvious limitations of this study are the small patient population, short duration of FU, and lack of cutoff points for the proposed metrics. The aim of this study was to define and suggest possible parameters for use in future studies with larger populations and longer FU, and by definition is hypothesis generating in nature. Craniosynostosis is a rare condition, and most studies are small. Although we had 16 eligible patients, only 6 patients met the inclusion criteria. This was mainly because of the lack of 3DSPG images obtained at appropriate time points and of sufficient quality for analysis. We have now developed a standardized FU protocol for these patients during which we will routinely obtain 3DSPG at agreed time points, including t0 and t1y.

Our relatively short FU interval is mandated by the fact that ESCH is a novel procedure in the United Kingdom. We think that this is predominantly related to the lack of helmet orthotic suppliers within the National Health Service (NHS), which delivers universal free healthcare in the United Kingdom. Our first metopic case was in 2018. Now that we have established the ESCH service and orthotic pathway, our case numbers and FU will increase, and we plan to publish further reports of long-term FU for this cohort. In addition, we feel that 3DSPG image analysis similar to that presented in this study may well be useful in the treatment of other forms of craniosynostosis and other operations undertaken to correct them.

To validate parameters for craniosynostosis patient management and develop cutoff points, we plan to apply the proposed metrics in a large cohort of trigonocephaly patients who were subjected to different management protocols, including conservative management. This will enable us to validate the parameters and develop cutoff points.

Conclusions

3DSPG is a useful tool for assessing the trigonocephalic deformity of metopic synostosis and for monitoring its improvement following treatment with ESCH. This study had a small number of patients but presents the hypothesis that the described parameters are helpful for assessing improvement in an objective and statistically analyzable form. As yet there is no single metric for analyzing cranial morphology; therefore, we suggest that a combination of measurements produces the clearest documentation of deformity and changes with treatment. We recommend the use of “whole head” parameters for trigonocephaly, which causes deformity of the entire neurocranium, not only the forehead region. We plan to continue to follow this cohort of patients and to report on long-term outcomes.

Acknowledgments

A.E. was funded by a full scholarship from the Ministry of Higher Education of the Arab Republic of Egypt. G.J. holds an MRC Clinical Academic Research Programme grant (no. MR/T005297/1). This research was supported by GOSH Charity (award no. 12SG15) and an NIHR GOSH/UCL Biomedical Research Centre Advanced Therapies for Structural Malformations and Tissue Damage pump-prime funding call (grant no. 17DS18). This report incorporates independent research for the National Institute for Health Research Biomedical Research Centre Funding Scheme. We are grateful for the support of the Technology in Motion and London Orthotic Consultancy companies for supplying some of the 3DSPG images.

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**Disclosures**

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

**Author Contributions**

Conception and design: James, Elawadly, Smith. Acquisition of data: James, Elawadly, Smith. Analysis and interpretation of data: James, Elawadly. Drafting the article: James, Elawadly. Critically revising the article: James. Reviewed submitted version of manuscript: James, Borghi, Abdelaziz, Silva, Dunaway, Jeelani, Ong. Approved the final version of the manuscript on behalf of all authors: James. Statistical analysis: James, Elawadly, Borghi. Administrative/technical/material support: James. Study supervision: James.

**Supplemental Information**

**Previous Presentations**

The abstract has been presented as an e-poster at the International Society of Craniofacial Surgeons virtual congress on October 15, 2021.

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