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Spring-Assisted Cranioplasty vs. Minimally-Invasive Endoscopically Assisted Remodelling Outcomes in Sagittal Craniosynostosis

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Abstract

Objective quantitative assessment of cranial shape in sagittal craniosynostosis remains challenging, as conventional metrics are insufficient to capture complex three-dimensional morphology and evaluate postoperative outcomes. This study aimed to develop two population-based statistical shape models of paediatric cranial anatomy to support surgical indication and postoperative outcome evaluation. SSM_{pre} (n = 147) compared cranial bone shapes of morphologically normal children (n = 67) and patients with sagittal craniosynostosis (n = 80), while SSM_{post} (n = 165) compared cranial soft tissue shapes of normal children (n = 112) with follow-up scans of patients treated with spring-assisted cranioplasty (SAC, n = 26) or minimally invasive endoscopically assisted remodelling (MEAR, n = 27). Statistical shape modelling using principal component analysis was applied. In SSM_{pre}, the first two shape modes explained 64% of variability and significantly differentiated the control and craniosynostosis groups, while cranial volume did not differ significantly. In SSM_{post}, the first two modes accounted for 60% of variability and significantly differentiated the three groups. MEAR showed greater similarity to the control group for several conventional morphological parameters, particularly width, length, and CI, whereas SAC was positioned closer to the control group along Mode 1. These findings demonstrate the potential of statistical shape modelling for detailed quantification of paediatric cranial shape variability and objective assessment of surgical outcomes.

Keywords: Sagittal craniosynostosis; Spring-assisted cranioplasty; Endoscopic cranial remodelling; Statistical shape modelling; Normal head shape; Paediatric cranial morphology

Introduction

Craniosynostosis is a condition characterised by the premature fusion of one or more cranial sutures, which may occur during intrauterine development or postnatally. Cranial sutures are fibrous joints that permit deformation of the skull during birth and allow for postnatal skull expansion in response to rapid brain growth [1]. The timing of physiological suture closure varies considerably: the metopic suture typically fuses within the first year of life, whereas the remaining sutures may remain patent until adolescence or adulthood.

Premature suture fusion restricts skull growth perpendicular to the affected suture, in accordance with Virchow's law, leading to compensatory overgrowth in other regions and the development of characteristic cranial deformities. In cases involving multiple sutures, intracranial volume may become insufficient, increasing the risk of raised intracranial pressure. Clinical consequences may include headaches, visual impairment due to papilledema, developmental delay, and, in some patients, associated abnormalities such as Chiari malformation [2].

Most patients present with single-suture involvement and are classified as having nonsyndromic craniosynostosis. In contrast, multisuture synostosis is more frequently associated with genetic syndromes such as Crouzon, Pfeiffer, or Apert syndrome and is generally linked to greater clinical severity and a higher risk of neurological complications [1].

Sagittal craniosynostosis is the most common form of craniosynostosis and results from premature fusion of the sagittal suture. This leads to restricted transverse skull growth and compensatory expansion at the coronal and lambdoid sutures, producing the characteristic elongated and narrow cranial shape known as scaphocephaly [3].

Surgical management is indicated based on several factors, including the risk of intracranial hypertension and abnormal cranial morphology. The primary goals of surgery are to restore cranial shape, increase intracranial volume, and allow adequate brain growth [2]. Treatment options range from extensive open procedures, such as total cranial vault remodelling, to less invasive techniques, including endoscopic strip craniectomy and spring-assisted cranial expansion [4].

Spring-assisted cranioplasty (SAC) involves performing a strip craniectomy over the fused suture, followed by placement of metallic springs across the osteotomy margins to gradually expand the skull and improve cranial shape over several weeks. Clinical studies have demonstrated that SAC provides comparable efficacy and safety to other surgical approaches [5], while being associated with reduced blood loss, lower transfusion rates, and shorter hospital stays [6]. However, several limitations remain. The procedure requires a second operation under general anaesthesia for spring removal, and hardware-related complications such as spring migration, exposure, or infection have been reported. In addition, long-term morphological outcomes remain incompletely characterized. From a technical perspective, current spring systems provide limited control over the magnitude and direction of expansion compared with distraction-based techniques [7].

Minimally invasive endoscopically assisted remodelling (MEAR) is a surgical technique for the treatment of sagittal craniosynostosis that is typically performed in infants younger than 6 months of age. In contrast to conventional endoscopic strip craniectomy, the MEAR approach emphasizes immediate intraoperative cranial reshaping while preserving the advantages of a minimally invasive procedure. Postoperatively, a three-dimensional head scan is obtained to fabricate a custom cranial orthosis, and helmet therapy is initiated to guide further skull growth. Patients are subsequently followed in a multidisciplinary setting with regular clinical and three-dimensional assessments of cranial development [8].

Normative data on head anthropometric measurements are available in the scientific literature; however, these data are predominantly based on linear and angular distances and their ratios [9]. A

more comprehensive approach to cranial shape assessment is statistical shape modelling (SSM). SSMs describe the average morphology and the main modes of shape variability within a population. By representing individual shapes as combinations of principal modes of variation, SSMs enable complex three-dimensional morphological differences to be quantified using a limited number of parameters [10].

This study aimed to develop two statistical shape models based on a population of 3D paediatric cranial models. The first model was designed as a decision-support tool for surgical indication and included data from patients with sagittal craniosynostosis and the control group. Its purpose was to characterize the fundamental shape differences between the craniosynostosis and control groups. The second model likewise incorporated data from the control group together with postoperative follow-up scans of patients who had undergone either spring-assisted cranioplasty (SAC) or minimally invasive endoscopically assisted remodelling (MEAR). The objective of this model was to compare postoperative cranial shape outcomes between the two surgical techniques and assess their similarity to the control group.

Materials and Methods

The Czech component of the study was approved by the Ethics Committee of the University Hospital Motol and 2nd Faculty of Medicine, Charles University in Prague (Reference No. EK-279.12/24). The use of patient imaging data from Great Ormond Street Hospital for research purposes was approved under UK Research Ethics Committee reference 15/LO/0386 (GOSH R&D study No. 14DS25). The study was conducted in accordance with the Declaration of Helsinki and relevant institutional and national regulations.

Informed Consent

Statement Written informed consent for participation in the study and for the use of clinical imaging data was obtained from the parents of all participants. Written informed consent for publication of the potentially identifiable 3D surface image shown in Figure 1 in an online open-access publication was obtained from the patient's parents.

Use of artificial intelligence: ChatGPT (OpenAI) was used for language editing and improvement of the manuscript. The authors reviewed and verified all AI-assisted modifications and remain fully responsible for the content of the manuscript.

Patient populations and image processing

The study included data from three groups: CS patients who underwent SAC at Great Ormond Street Hospital for Children, London, UK (GOSH) between 2014 and 2017; CS patients who had MEAR at Motol University Hospital, Prague, Czech Republic (UH Motol) between 2017 and 2019; and a control group who had head CT for non-craniofacial indications at the Necker Children's Hospital (Necker-Enfants Malades Hospital), Paris, France. CS patients were included if they had a pre-operative CT scan. Post-operative 3D photography scans, when acquired at the time of discharge, were also collected at both sites. 3D photography scans were captured using a 3dMDhead system (3dMD Ltd., UK) at GOSH and a

handheld HCP scanner (Creaform Inc., Canada) at UH Motol. For the control group, only subjects with radiologically confirmed normal head morphology were included [4].

CT images and 3D photography scans were post-processed in 3D Slicer [11] to segment bone and soft tissue 3D surfaces from CT and the scalp surfaces from photography. The segmented models were exported to 3-Matic [12] for further refinements: bone wrapping was performed first to close any holes in the skull, followed by cropping of both bone and soft tissue using a plane defined by three anatomical landmarks: nasion, right and left tragus (Figure 1). For all scalp models, the ear regions were manually removed and replaced by a capping surface (Figure 2). Surface smoothing was applied to correct minor deformations, and mesh decimation was performed to reduce polygon count while maintaining anatomical accuracy.

For all subjects, traditional cranial morphological parameters, including volume, width, height, and length, were extracted from the reconstructed surfaces. Based on these measurements, the cephalic index (CI) and turriccephaly index (TI) were calculated according to the following equations:

$$CI = \frac{width}{length} * 100 \quad (1)$$

$$TI = \frac{height}{length} * 100 \quad (2)$$

Statistical shape models

All reconstructed surfaces were registered in CraniumPy [13] by selecting three anatomical landmarks (nasion, right and left tragus) and were scaled in 3-Matic to a uniform volume of $1 \times 10^6 \text{ mm}^3$ to ensure that the SSM captured only shape variations, independent of overall patient size.

Two independent SSMs were built: the first (SSM_{pre}) included the bone shapes from all pre-operative CS patients and the controls in the same age range as the pre-operative CS group, to assess the pre-operative sagittal craniosynostosis skull; the second (SSM_{post}) included the soft-tissue surfaces of CS

patients and the controls in the same age range as the FU times, to compare the follow-up outcome shapes after SAC and MEAR with those of the control group.

The SSMs were built in MATLAB [14]. Point-to-point correspondence was established using a non-rigid iterative closest point (NRICP) approach, and principal component analysis (PCA) was performed to capture the overall shape variability into a limited number of modes with associated weights. The resulting SSMs provided a mean (template) shape, individual shape deformation vectors, the cumulative explained variance, and a set of principal modes describing the dominant patterns of shape variability within the dataset.

Data analysis

Within each SSM, patient groups were compared to identify statistically significant differences across all parameters in MATLAB. All statistical tests were two-tailed, with a significance level of $\alpha = 0.05$. In addition, differences in the values of the shape variation modes were statistically evaluated. Normality of the data distribution was assessed using the Lilliefors test. Homogeneity of variances was assessed using the two-sample F-test.

In SSMpre, which comprised two groups (CS and control), statistical comparisons were performed depending on the normality of data distribution and homogeneity of variances. For non-normally distributed data, the Mann–Whitney U test was applied, whereas the two-sample t-test was used for normally distributed data.

In SSMpost, which comprised three groups (SAC, MEAR and control), the three groups were tested for overall differences across all parameters using the Kruskal–Wallis test for non-normally distributed data and ANOVA for normally distributed data. To assess pairwise differences between individual groups, post hoc tests were subsequently applied: Dunn’s test with Sidak correction following the Kruskal–Wallis test and Scheffé’s test following ANOVA.

In both models, correlations between individual morphological parameters and the modes of shape variability were assessed using Spearman's rank correlation coefficient. For these analyses, parameter values derived from the volume-scaled models were used.

Results

The results of the two statistical shape models were evaluated to characterize cranial shape variability and identify differences between the study groups. SSM_{pre} was used to assess differences between pre-operative sagittal craniosynostosis and the control population, while SSM_{post} was used to compare postoperative cranial shapes following SAC and MEAR with the control population. Additional results are provided in Supplementary Figures 1-5.

Table 1 summarises the subject groups and age distributions for SSM_{pre} and SSM_{post} , showing no statistically significant differences in age between the groups (SSM_{pre} : $p = 0.58$; SSM_{post} : $p = 0.22$).

The CS group included patients from both GOSH and UH Motol. No statistically significant differences were observed between the two subgroups in any of the morphological parameters, including CI ($p = 0.5265$), TI ($p = 0.0603$), volume ($p = 0.6156$), height ($p = 0.0721$), width ($p = 0.8781$), and length ($p = 0.6498$). Pre-operatively, there was no statistically significant difference in cranial volume between the CS and control groups ($p = 0.74$), whereas statistically significant differences were observed in all other traditional cranial measurements. Statistically significant differences were observed in width, length, CI, and TI (all $p < 0.001$), as well as in height ($p = 0.0046$).

For SSM_{pre} , Mode 1 captured 50.4% of the variability, while Mode 2 explained 13.7%, together accounting for 64% of the total shape variability (Figure 3). The remaining modes explained only a small proportion of the variability (each mode $< 5\%$) with more localised changes of shape features. Mode 1 showed strong correlations with all morphological parameters, positively with length and TI and

negatively with CI, width, and height (all $p < 0.001$). Mode 2 was also significantly correlated with height, width, and CI (all $p < 0.001$), with a positive correlation for height and negative correlations for width and CI, although these correlations were weaker overall than those observed for Mode 1. The two groups differed significantly along both modes of variability (Mode 1: $p < 0.001$; Mode 2: $p = 0.034$) (Figure 4).

The clustering plot shows a clear separation between sagittal craniosynostosis patients and the control group, mainly along Mode 1 (Figure 5).

Post-operatively, statistically significant overall differences were observed among the groups for width, height, length, volume, CI, and TI ($p < 0.001$ for all). The SAC group exhibited a markedly larger cranial volume compared with the control group, whereas the MEAR group showed a slightly smaller cranial volume. Regarding the cephalic index, all groups again differed significantly, indicating that both surgical approaches shifted the cranial shape of craniosynostosis patients toward that of the control group, although complete normalization was not achieved. The MEAR group demonstrated a trend toward closer similarity to the control group compared with the SAC group. Pronounced differences were also observed in the remaining morphological parameters; however, in several cases, no statistically significant differences were detected between specific groups. In particular, the control and SAC groups did not differ significantly in terms of the turriccephaly index ($p = 0.105$), while the control and MEAR groups showed no significant differences in cranial width ($p = 0.24$) or length ($p = 0.789$).

For SSM_{post} , only the first two shape modes were considered, together accounting for 60% of the total shape variability. The remaining modes explained only a small proportion of the variability (each mode $< 5\%$) with more localised changes of shape features. As all 3D cranial models were scaled to the same volume, all observed differences reflect shape variations rather than size-related effects.

Mode 1 accounted for 44.8% of the variability, while Mode 2 explained 15.2% (Figure 6). Both modes showed statistically significant differences among the three groups ($p < 0.001$). Post hoc analyses revealed that the groups differed significantly from each other along Mode 1 ($p < 0.001$), whereas no statistically significant difference between the SAC and MEAR groups was observed in Mode 2 ($p = 0.75$) (Figure 7). This finding suggests that Mode 2 may be associated with characteristic craniosynostosis-related shape features that persist to some extent in both follow-up groups.

Mode 1 showed a strong positive correlation with height and negative correlations with width, CI, and TI (all $p < 0.001$). The correlation with length was weaker but remained statistically significant ($p = 0.002$). Mode 2 showed strong positive correlations with length and TI and negative correlations with CI and width (all $p < 0.001$), while the correlation with height was weak and non-significant ($p = 0.64$).

The clustering plot combining Mode 1 and Mode 2 showed clear separation between the population groups and distinct shape outcomes following the two surgical techniques (Figure 8).

Discussion

Sagittal craniosynostosis is a common cranial malformation that can lead to abnormal head shapes and, in some cases, increased intracranial pressure or neurodevelopmental issues. Accurate assessment of cranial morphology is essential for timely diagnosis, surgical planning, and evaluation of treatment outcomes. Traditional anthropometric measurements and indices, such as cranial width, length, and cephalic index, provide only limited information about complex three-dimensional cranial shapes. To address this limitation, this study presents the development and application of two population-based statistical shape models derived from 3D paediatric cranial data. These models aim to enhance objective assessment of sagittal craniosynostosis and its surgical treatment outcomes by capturing subtle morphological variations that are not fully reflected by conventional metrics.

SSM_{pre} was developed to support surgical indication by comparing cranial bone shapes of morphologically normal children and patients with sagittal craniosynostosis. SSM_{post} focused on postoperative outcome evaluation by comparing cranial soft tissue shapes of control group with follow-up scans of patients treated using two different surgical techniques, spring-assisted cranioplasty (SAC) and minimally invasive endoscopically assisted remodelling (MEAR).

The results of SSM_{pre} demonstrate that children with sagittal craniosynostosis exhibit a distinct shape signature across the dominant modes of shape variability. Both Mode 1 and Mode 2 significantly differentiated the craniosynostosis and control groups, with a stronger separation observed along Mode 1 than Mode 2. Mode 1 was strongly associated with all traditional morphological parameters, particularly length, CI, width, and height, suggesting that it captured a major component of overall cranial shape variation. Mode 2 showed weaker associations with selected morphological parameters, including height, width, and CI, and may therefore capture additional aspects of cranial shape variability. These findings indicate that the SSM captured characteristic three-dimensional differences between pathological and normal cranial morphology beyond individual anthropometric measurements. Importantly, while cranial volume did not differ significantly between craniosynostosis

patients and the control group, pronounced and statistically significant differences were observed in the other morphological parameters.

SSM_{post} enabled a direct comparison of postoperative cranial shape outcomes between SAC and MEAR in relation to the control group. The traditional morphological parameters showed that the SAC group had greater width, length, height, and volume than the MEAR group, with substantially greater cranial volume than the control group. The MEAR group had a smaller cranial volume and showed values closer to the control group for width, length, and CI. The SSM provided an additional assessment of three-dimensional shape independent of overall cranial size. The first two shape modes captured the main patterns of postoperative shape variability and significantly differentiated the three groups. Along Mode 1, SAC was positioned closer to the control group and was associated with a narrower and higher cranial shape, whereas MEAR showed a wider and lower shape. In contrast, both postoperative groups showed deviations in the same direction along Mode 2, with MEAR being slightly closer to the control group, although the difference between SAC and MEAR was not significant ($p = 0.75$). Overall, the two surgical techniques resulted in distinct postoperative cranial shape configurations, with some aspects of shape variability shared between the groups and others differing substantially.

The presented models offer several opportunities for future development. Based on SSM_{pre}, a practical clinical tool could be developed to project new patients with suspected sagittal craniosynostosis into the established statistical shape space. Such a tool could objectively quantify the severity of cranial deformation and visualise the patient's position relative to normal and pathological populations, potentially supporting surgical decision-making and timing. SSM_{post} could be further extended by incorporating longitudinal data from the same patients acquired both preoperatively and postoperatively. This would allow direct tracking of individual shape trajectories within the statistical shape space and enable a more precise assessment of surgical outcomes. However, such paired datasets were not available within the scope of the present study.

Several limitations of this study should be acknowledged. First, the number of follow-up patients, particularly in the SAC and MEAR groups, was relatively limited, and follow-up scans were acquired at varying time points after surgery, based on clinical judgement of when the postoperative cranial shape was considered final. This introduces variability that may influence the observed shape differences. Second, the age range within the follow-up groups was relatively wide. Because infants develop and grow rapidly during the first months of life, cranial morphology can change considerably with age. Although all models were scaled to the same volume, some observed shape differences may therefore reflect age-related growth rather than the underlying diagnosis or surgical outcome. Additional limitations include the use of data from two different institutions with potentially differing imaging protocols and surgical practices.

From a clinical perspective, both statistical shape models provide information beyond traditional anthropometric measurements and commonly used indices, such as the cephalic index. SSM_{pre} could provide an objective shape-based assessment to complement clinical judgement in surgical indication. SSM_{post} enables objective comparison of postoperative outcomes between SAC and MEAR and their relationship to the control group. In future, these models could support treatment outcome evaluation and provide additional information for surgical decision-making. In the case of MEAR, SSM_{post} could also potentially provide an objective reference for the desired cranial shape during postoperative helmet therapy.

Conclusions

In conclusion, the presented study demonstrates the potential of statistical shape modelling (SSM) for the objective assessment of cranial morphology in sagittal craniosynostosis. By integrating 3D data from normal children and patients with sagittal craniosynostosis, the developed models captured characteristic differences in pathological and postoperative cranial morphology beyond traditional anthropometric measurements. SSM_{pre} demonstrated the potential of SSM to support surgical indication by objectively distinguishing craniosynostotic and normal cranial shapes. SSM_{post} revealed distinct postoperative shape configurations following SAC and MEAR, with differences in both conventional morphological parameters and the dominant modes of shape variability. These findings highlight the complementary value of SSM and traditional anthropometric measures and provide a basis for future clinical applications, including patient-specific assessment and longitudinal monitoring of treatment outcomes.

Additional Information

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Competing interests

The authors declare no competing interests.

Data availability

The datasets generated and analysed during the current study are available from the corresponding author on reasonable request.

Author contributions

SC and LS developed the methodology and performed data preprocessing. AB developed the SSM analysis tool and scripts. SC constructed the statistical shape models from the prepared 3D cranial models. OJ, VB, LP and JT provided clinical expertise and supervised the work from the neurosurgical perspective. SS, LC, and AB provided engineering supervision. JD and JV were responsible for data acquisition. All authors reviewed and approved the final manuscript.

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FIGURE LEGEND

Figure 1. Processing workflow of 3D photography data, from the original head scan (left) to the final cropped scalp model with the ear regions removed (right). The original 3D head scan is shown with written informed consent for publication obtained from the patient's parents.

Figure 2. Processing workflow of CT data, from the original skull reconstruction (left) to the final wrapped, cropped, and smoothed cranial bone model (right).

Figure 3. Visualization of SSM_{pre} : the mean shape and the first two principal modes of cranial shape variability for the control (CL) and sagittal craniosynostosis (CS) groups. Each mode is visualized at -2 SD and $+2$ SD from the mean shape.

Figure 4. Comparison of SSM_{pre} mode scores between the control (CL) and sagittal craniosynostosis (CS) groups for Mode 1 and Mode 2. Significant differences between the groups are indicated by brackets and p-values.

Figure 5. Clustering plot of the first two principal modes of SSM_{pre} , showing the distribution of the control (CL) and sagittal craniosynostosis (CS) groups.

Figure 6. Visualization of SSM_{post} : the mean shape and the first two principal modes of cranial shape variability for the control (CL) and postoperative groups (POST): SAC and MEAR. Each mode is visualized at -2 SD and $+2$ SD from the mean shape.

Figure 7. Comparison of SSM_{post} mode scores between the control (CL), spring-assisted cranioplasty (SAC), and minimally invasive endoscopically assisted remodelling (MEAR) groups for Mode 1 and Mode 2. Significant pairwise differences are indicated by brackets and p-values.

Figure 8. Clustering plot of the first two principal modes of SSM_{post} , showing the distribution of the control (CL), spring-assisted cranioplasty (SAC), and minimally invasive endoscopically assisted

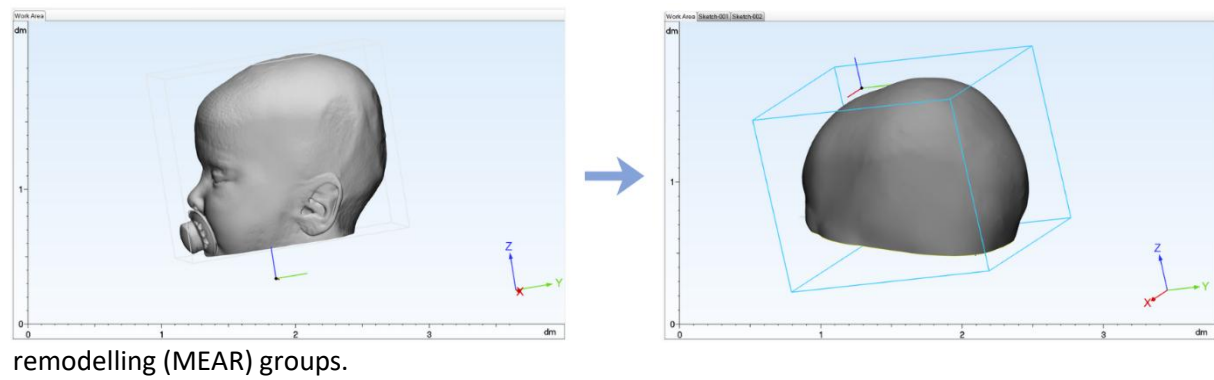


Figure 1

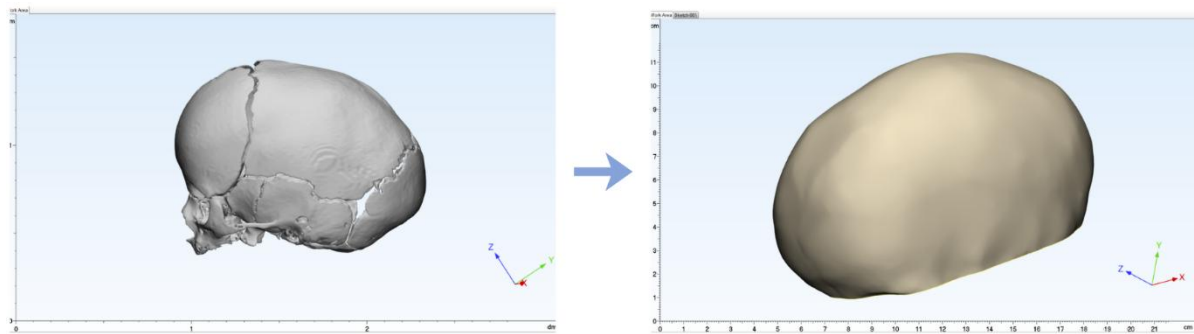


Figure 2

Figure 3

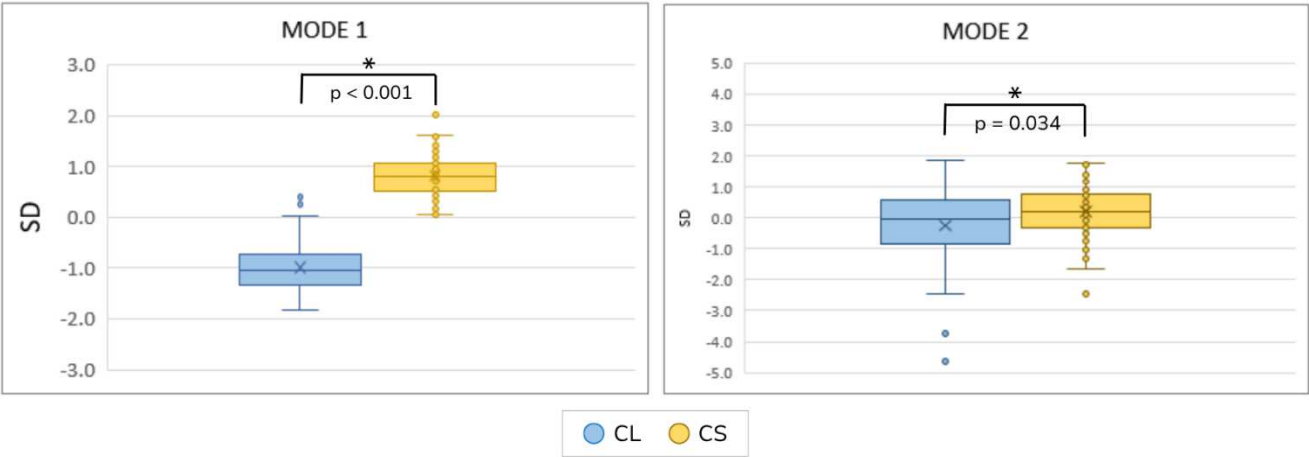
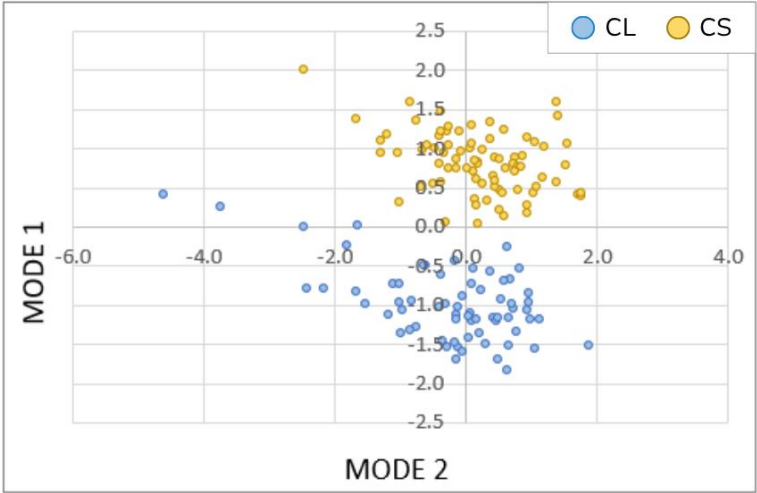


Figure 4

Figure 5



SSMpre

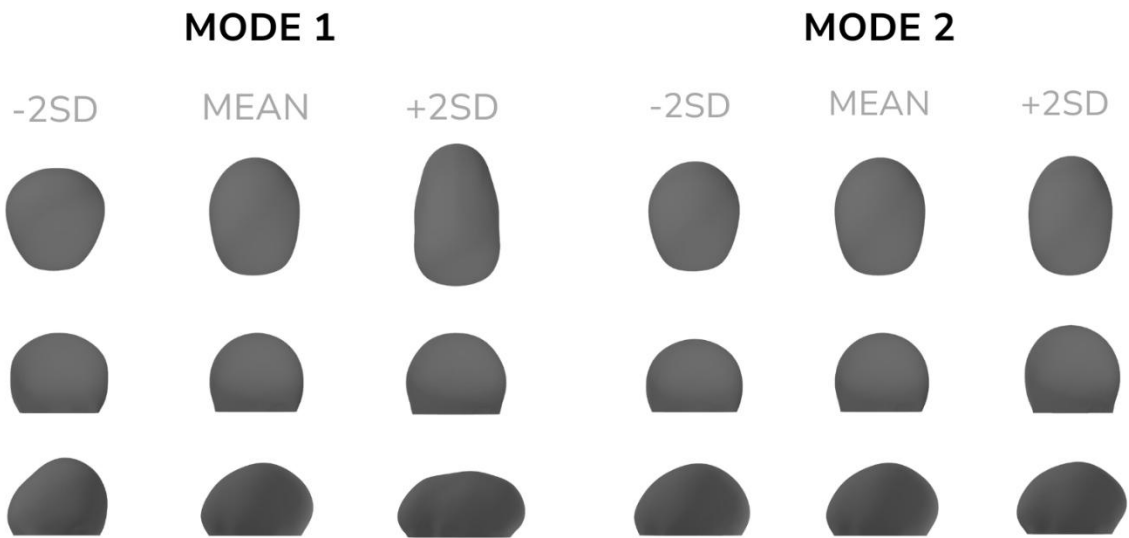


Figure 6

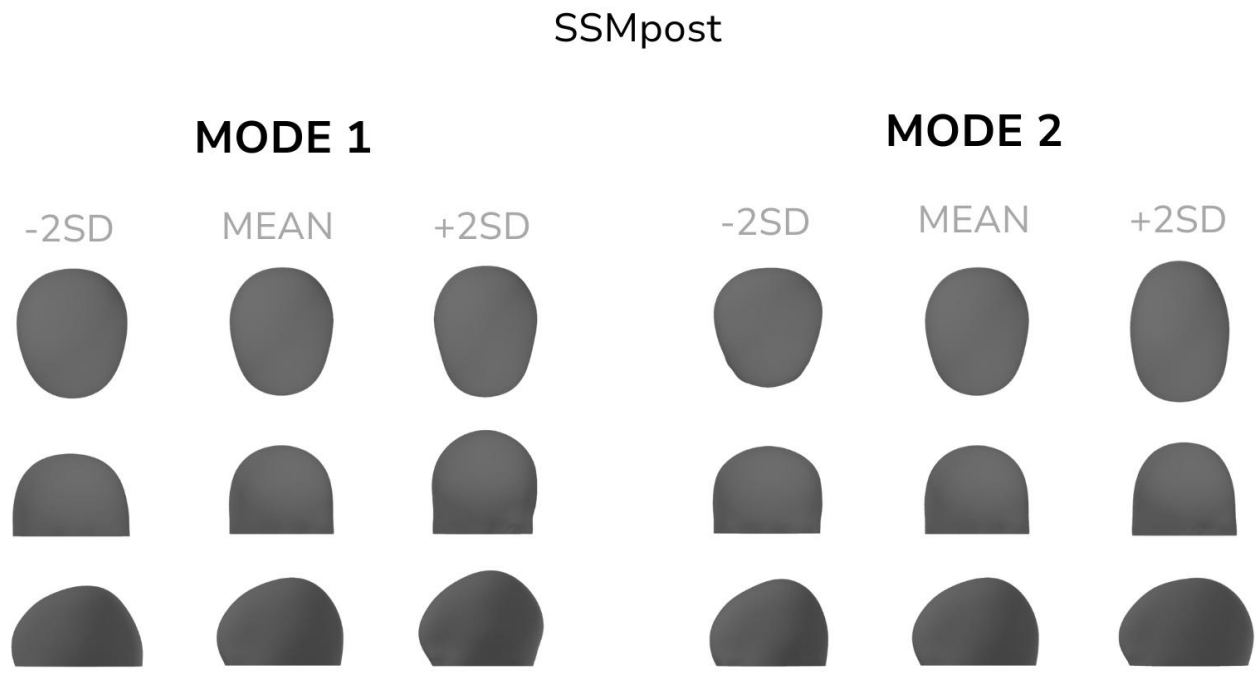


Figure 7

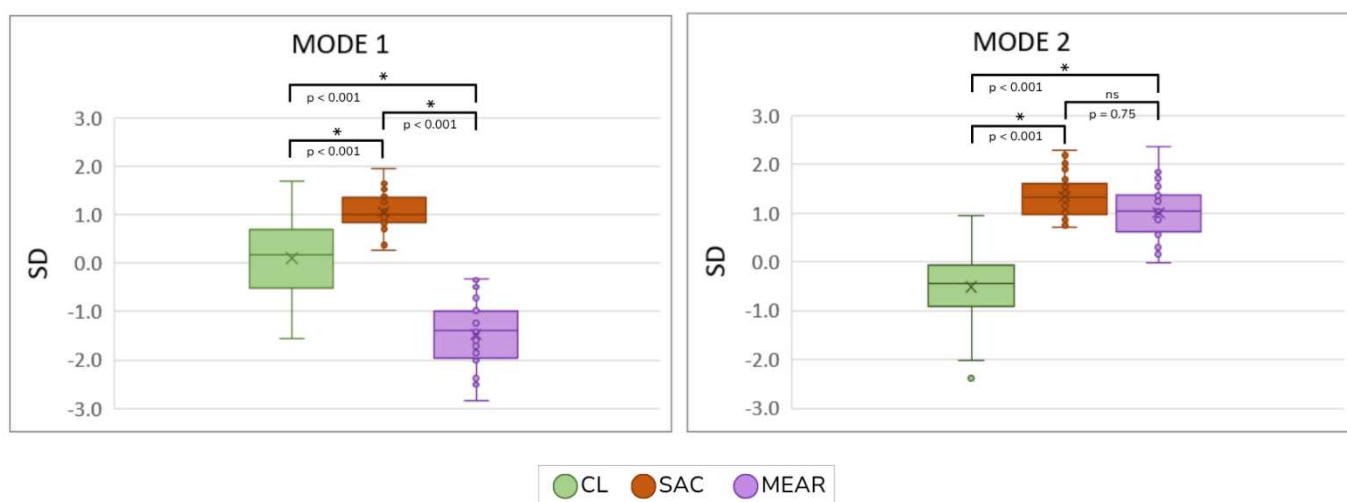
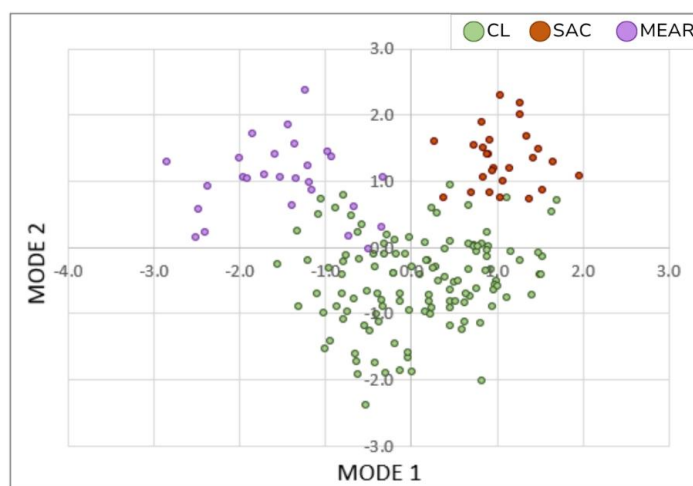


Figure 8



Tables

Table 1: Subject groups and age distribution included in SSM_{pre} and SSM_{post}

	SSM _{pre}			SSM _{post}	
	S	control	SAC	MEAR	control
Subjects	80	67	26	27	112
Age (months)	3.3 ± 1.9	3.6 ± 2.4	11.8 ± 6.3	13.1 ± 5.9	14.0 ± 6.4
	[0 - 8]	[0 - 8]	[5- 26]	[4 - 25]	[4 - 26]

Data are presented as mean ± standard deviation (SD), with ranges in square brackets.

Supplementary Files

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- [SupplementaryInformationLegend.docx](#)
- [SupplementaryFigure1.png](#)
- [SupplementaryFigure4.png](#)
- [SupplementaryFigure5.png](#)
- [SupplementaryFigure3.png](#)