Is it possible to predict the long-term outcome of spring cranioplasty using published methods for modelling head growth in paediatric patients?

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Supported by the Laidlaw foundation

Abstract

Sagittal craniosynostosis is a medical condition affecting about 350 infants in the UK per year [1]. In this condition, the midline cranial suture in a baby's skull ossifies prematurely, leading to an abnormally long and thin head shape and sometimes resulting in reduced quality of life. To correct this, surgeons at Great Ormond Street Hospital (GOSH) in London routinely perform spring cranioplasty procedures, which involve inserting metallic distractors (springs) in the skull to widen and improve the shape of the head.

A range of different parameters affect surgical outcome, such as the baby's initial head shape, spring model and the distractor position along the skull. The combination of these factors, along with the complex nature of skull-spring interaction, can lead to a suboptimal outcome. In this case, further reshaping surgery may be required, which is costly and carries additional risks, as well as causing patient/family distress [2].

A patient-specific predictive model of the final head shape is a useful tool for presurgical planning. Currently, craniosynostosis correction predictive models successfully replicate short-term outcomes but long-term models have not been produced for sagittal craniosynostosis correction. In this work we developed a predictive model - based on previously published protocols - able to predict long term outcomes thanks to a tuned head growth model. This work will contribute towards a useful tool for surgical planning, which will improve surgical outcomes as well as patient safety.

1 Introduction

The skull of a newborn is made up of five bones separated by connective tissue known as cranial sutures [3]. The sutures meet at gaps between the bones known as fontanelles, of which there are six, the two most important being the anterior and posterior. Each fontanelle will close in a process known as intramembranous ossification, somewhere between 6 weeks and 24 months.

Craniosynostosis is the premature fusion of one or more of the cranial sutures [4], happening in between 1 in 2,100 [5] and 1 in 2,500 births [6], with a strong male predominance (3.5:1 male to female ratio) [7]. This can result in an abnormal head shape in infants, with different features according to the suture involved. When the sagittal suture prematurely ossifies (the most common of the craniosynostoses – affected in 55% - 60% of cases. [8]) the lateral head expansion is limited, resulting in a long and narrow head shape, a condition named "scaphocephaly"

(a)
Comparison of newborn with Scaphocephaly vs normal skull development [9]

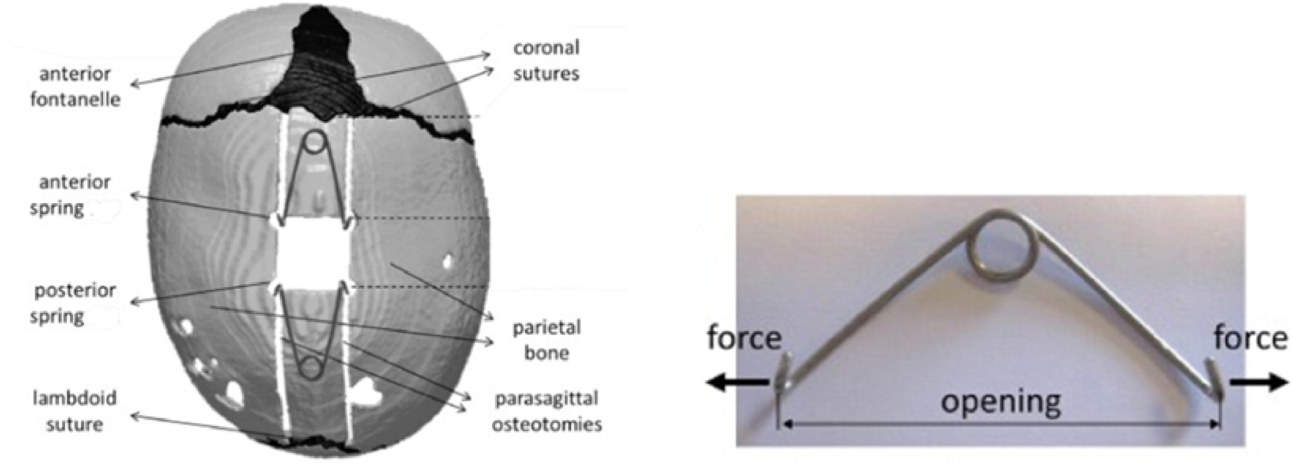
(b)
Diagram of the sutures and fontanelles in a newborn [10]

Figure 1: Diagrams of the newborn skull


[redacted]
[redacted]

Figure 2:
Image of a child with Scaphocephaly [4]

If left untreated, evidence suggests that there is an elevated risk of the child's neurodevelopment being delayed[11], due of the restriction of the growth of the brain.
It is therefore a developmental as well as aesthetic priority to fix it. Spring-assisted cranioplasty (SAC) is a procedure commonly carried out to correct scaphocephaly, by surgically inserting spring-like distractors into the skull, with the aim of slowly reshaping it over a period of months, as the brain grows. It was introduced at Great Ormond Street in 2008 for patients aged 3-8 months with a standardised procedure to ensure reproducibility[12]. The procedure begins by making an incision on the scalp of the patient perpendicular to the sagittal suture. Then a square craniectomy is performed, halfway along the sagittal suture, and two parasagittal osteotomies (surgical cuts into bone) are made parallel to the midline. They start from the craniectomy site and extend to the coronal sutures anteriorly and the lambdoid sutures posteriorly. Two pairs of grooves are created on the osteotomy lines for the insertion of the springs. Parameters such as the distance between the parasagittal osteotomies, the distance of the grooves from the coronal suture and the stiffness of the spring inserted are determined based on the initial head shape of the child. Spring selection is decided during the surgery based on the strength of the calvarial bone (uppermost part of the skull, so the region looked at here) and the surgeon's experience [12]. There are three spring models to choose from – S10, S12, S14 – which have the same geometry but are made of wire with different thicknesses, altering their respective stiffnesses to 0.17, 0.39 and 0.68N/mm [9]. Typically, stiffer springs are used for older patients (5 months +) [12].



(a) Diagram of surgical procedure [9]

(b) Image of spring used in surgery [9]

Figure 3: Diagrams illustrating spring insertion

Due to the range of initial parameters for each patient's head shape (spring stiffness, spring placement and initial head shape), the final shape can vary dramatically. This makes surgical planning for a specific outcome difficult, which is problematic because if the desired shape isn't achieved then a second surgery may be required. Every surgery carries risks as well as costs, so it is better for both the patient and the surgeon for the optimal result to be achieved the first time.
This paper hypothesises that a long-term growth model can be developed from published methods. The advantage of a long-term model is that it models the final head shape, allowing surgeons to see the effect that specific decisions, such as spring placement, will have in the long term The paper will outline the production and testing of the model, including both spring expansion and brain growth, as well as some novel testing of altering sutures. Tests were carried out on three patient examples which were then validated.

2 Method

2.1 Literature review

Different approaches to model this surgery and advance the potential for surgical planning have been detailed in literature. The Craniofacial research group based at UCL GOS Institute of Child Health & Great Ormond Street Hospital for children has specialized in this area. They investigated the biomechanics of the springs used in sagittal craniosynostosis operations [12] and the associations between surgical parameters and head shapes following spring-assisted cranioplasty [12].These provide the framework for developing a predictive model by establishing the key cranial features that are altered by spring cranioplasty surgery and the behaviour of the spring-skull interaction, both of which can then be incorporated into a model. A patient-specific model for spring expansion during SAC was also developed [9], using finite element modelling effectively with only a maximum of 9% difference between predictions and the reality of post-op spring expansion. However, the simultaneous growth of the brain is currently not included in this model, meaning it can only be used for short-term predictions, and can't model the long-term shape of the head. A patient-specific model for head shape following spring surgery treating lambdoid craniosynostosis was modelled, which used thermal expansion to mimic skull growth [13]. The growth coefficient for children undergoing spring surgery, predicting approximately three months of growth, was optimised using trial and error validated by testing on the population. The combination of these methods, applied to spring cranioplasty patients, is detailed in this paper.

2.2 Image post processing & Segmentation

To build a patient-specific model, pre-operative CT scans were acquired, as well as post-op, in order to validate the model at the end of the process. Typically, images at GOSH are taken 1-2 weeks before surgery, and at the time of spring removal (3-5 months post-insertion). Three male patients were selected for this modelling due to the availability of their pre and post-op imaging. Patients A, B and C were respectively ages 3.7, 4.3 and 4.8 months at the time of surgery and their post-op CT scans were taken 4.4, 3.1 and 4.1 months after surgery.

All research was performed in accordance with relevant guidelines and regulations. Informed consent was obtained for all imaging data from all patients and/or their legal guardians. This study protocol was approved by the Great Ormond Street Hospital and Great Ormond Street Institute of Child Health joint Research and Development Office (internal R&D number 14DS15, REC 15 L0 0386). All procedures performed in this study were in accordance with the 1964 Helsinki declaration and its later amendments, or comparable ethical standards.

The CT data was post-processed using Scan IP (Synopsys Inc) to create a patient-specific geometric 3D model. As reported in the literature [9], a common way to analyse the data is to perform grey level thresholding and morphological operations to separate bone (the skull) from the soft tissues (mastoid fontanelle, coronal suture, lambdoid suture, and the anterior fontanelle). A 3D reconstruction was then created, which was scaled to account for head growth between the day of pre op imaging and the day of surgery using published intercranial volume (ICV) growth curves [14].

Once the model was scaled to the correct starting size, the geometry was cut with plane encompassing the cranial nasion and the top of the left and right auditory meatus, to isolate the region of interest. Boundary conditions were then applied to the bottom of the skull to prevent rigid displacements (occurs when the body of an object is moved without changing its size or shape – the opposite of what is wanted here). The surgery that had been performed was then recreated on the skull using notes from theatre – replicated osteotomies made and grooves for spring insertion sites added. The artefacts were manually removed, and the model converted into a 3D computational mesh. A linear tetrahedral mesh was used here, for simplicity and computational speed.

2.3 Spring expansion modelling

The model was then imported into Ansys workbench (Ansys software) for expansion modelling. The key parameters of the materials involved – bone and suture – were input into the software to improve the accuracy of the model's behaviour. The optimised Young's moduli and Poisson's ratios for bone and suture were taken from literature [15], [16] using the assumption of age as six months.

	Young's Modulus/ MPa	Poisson's Ratio
Bone	418.2	0.22
Suture	18.175	0.42

Table 1: Table showing key parameters of bone and suture

The assumption of viscoelasticity was also made. Both bone and soft tissues were modelled using the Prony shear and bulk relaxation relationship (Eq 1).

$$\frac{G(t)}{G_0} = \alpha_\infty + \sum_i \alpha_i e^{-\frac{t}{\tau_i}} \tag{1}$$

where α_∞ and α_i are the relative moduli, τ_i are the time constants, $G(t)$ is the instantaneous shear modulus, and G_0 is the shear modulus at the beginning of the relaxation ($t = 0$) [9]. The parameter values were taken from literature (table 2).

Index	Relaxation relative modulus	Relaxation time constant
i = 1	0.73213	6720.4
i = 2	0.25708	40322

Table 2: Table showing the parameters of the prony shear and bulk relaxation relationship modelled here [17]

Once the parameters were assigned, the action of the springs could be simulated by two linear spring connections which were applied to the grooves previously marked out. The force applied by the spring is dependent on its stiffness and the crimping distance applied during surgery [12] (however crimping distance was not included in this model). A finite element analysis (FEA, [18]) of the spring expansion simulation could then be run.

After the simulation had run, a deformed model (as a result of spring expansion) was exported as an STL back to Scan IP so that the presence of the callus (a tissue that develops pre bone formation) could be included using the 'paint' tool. This manual process involved filling in the gaps in the model, where the callus should be located, on sequential layers, until a 3D model could be formed, which was then smoothed. The same process was followed to add the ICV back into the simulation. This provided a complete model of the brain after spring expansion. This was used as the baseline model, as this method has already been tested on spring cranioplasty patients in literature, designated as *model 1.*

2.4 Growth expansion modelling

To simulate the effect of brain growth, the model validated by [16] was used – an isotropic thermal expansion of the ICV implemented in Ansys (equation 2).

$$\Delta V = V_1 \times \alpha \times \Delta T \tag{2}$$

Where α is the coefficient of thermal expansion, ΔT is the temperature difference, V_1 is the initial ICV, and ΔV is the ICV increase due to growth. From the literature [19], a study of six patients aged between 2.5 months and 9 months found that a coefficient of thermal expansion of 0.0006 1/℃ over a 3-month period was optimal. The cohort of patients tested was similar in age and condition to patients A, B, and C here. Similarly to previous studies, 100℃ was chosen to be the temperature difference, and the time over which this was applied was varied for each patient to account for the different growth periods between surgery and imaging, as seen in Table 3.

The combination of spring expansion and growth expansion modelling was designated *model 2.*

Surgical feedback showed that post-surgery, sutures would sometimes rupture and then reform. To incorporate this into the modelling, the sutures were virtually removed completely, and then the skull underwent both spring expansion and growth expansion (this became model 3). However, sometimes the sutures would only partially tear, so to model this, only the top half of the sutures were removed *(model 4)*.

For all models, the post-expansion file was imported into Meshmixer (Autodesk), where a shell of the

Patients	Growth period between surgery and post-op imaging/months	Modified growth time period/s
A	4.4	1.467
B	3.1	1.000
C	4.1	1.367

Table 3: Table showing the relationship between the growth period of each patient and the modified growth time assigned to them

outer shape was extracted and smoothed. This was then offset by 2.8mm to mimic the skin, as found in the literature[9]. End-of-treatment shape predictions were validated by comparison with post-treatment optical scans. The surfaces were aligned along the horizontal plane above the orbits and visually compared using Meshmixer software so particular areas of difference could be identified. The surfaces were then imported into Simpleware ScanIP to generate a measurement of surface deviation.

3 Results

The hypothesis of this study was that the long-term outcome of spring cranioplasty surgery could be predicted using published methods for modelling head shapes. This has been qualified by testing patient data with different models:

Model 1: Spring expansion

Model 2: Spring expansion and growth expansion

Model 3: Spring expansion and growth expansion with sutures removed

Model 4: Spring expansion and growth expansion with sutures partially removed

The resulting shape was then validated against a post-operative image by using the Simpleware surface deviation tool. For the sake of clarity in images the deviation was limited to the bounds of +5mm and -20mm.

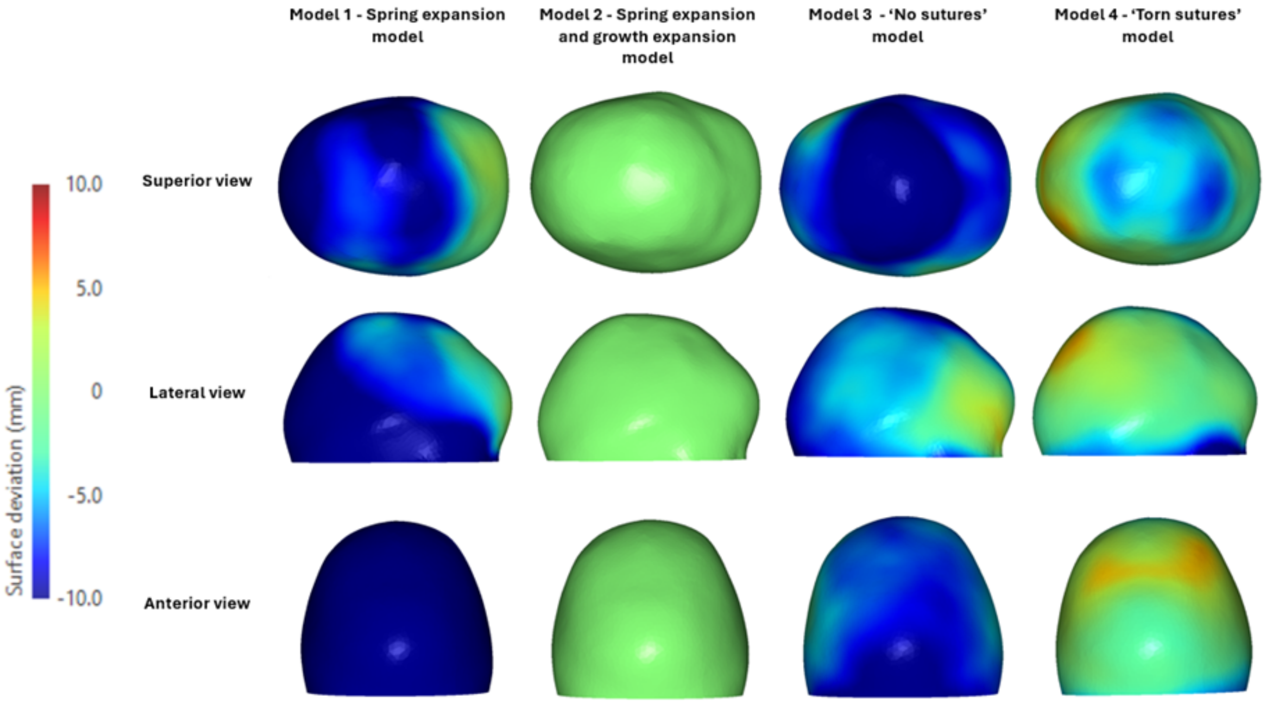


Figure 4: Surface deviation for patient A when models 1,2,3 and 4 are validated with post operative imaging

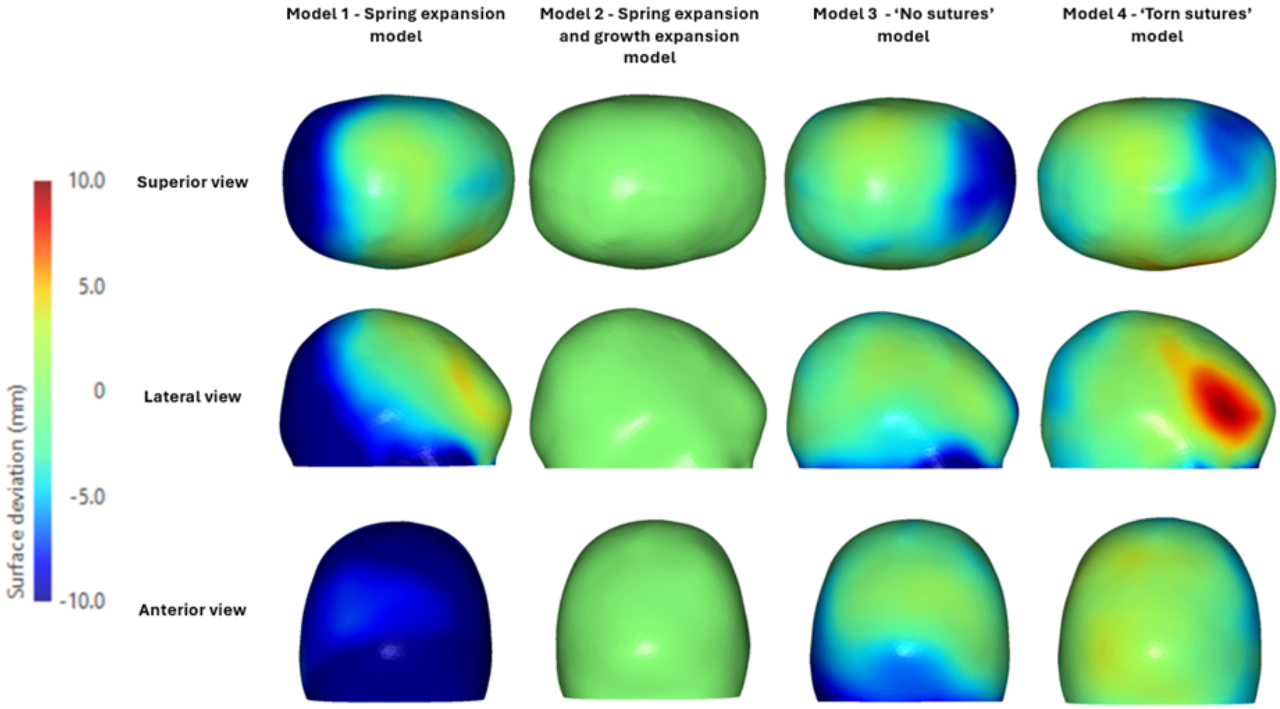


Figure 5: Surface deviation for patient B when models 1,2,3 and 4 are validated with post operative imaging

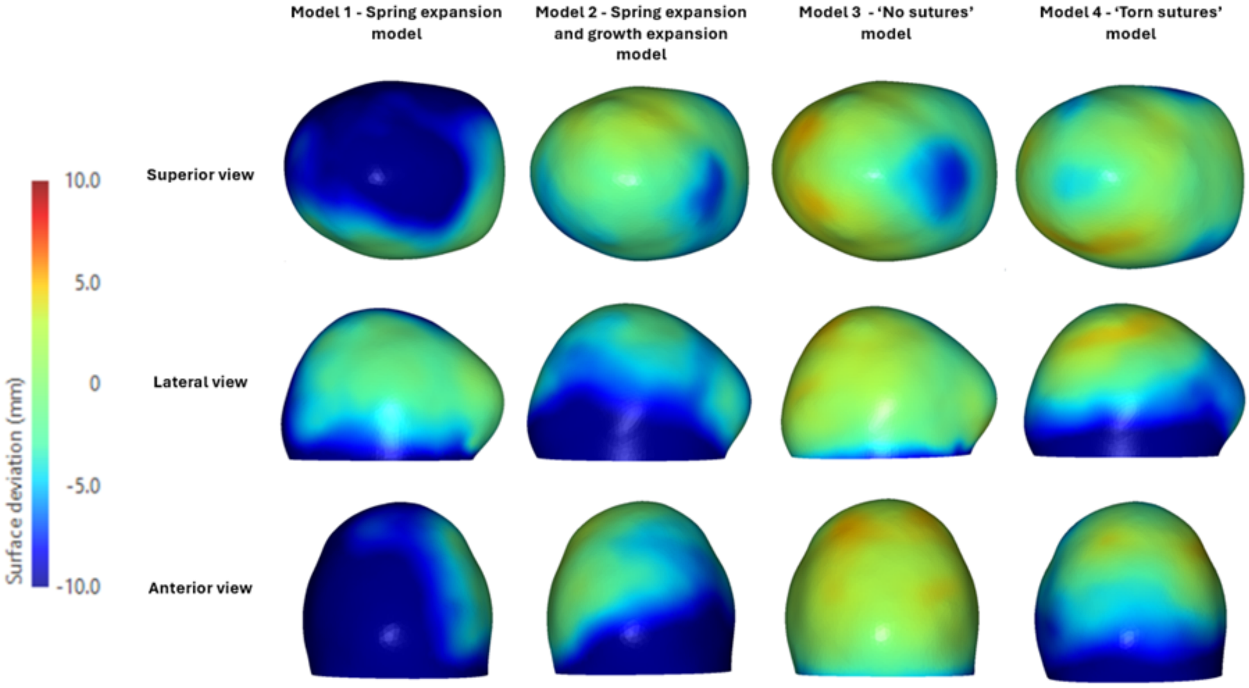
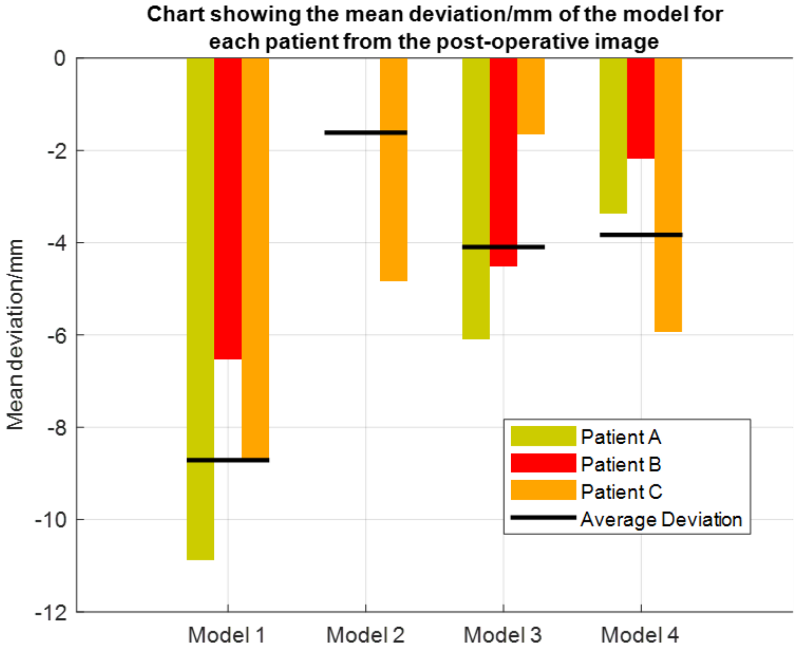
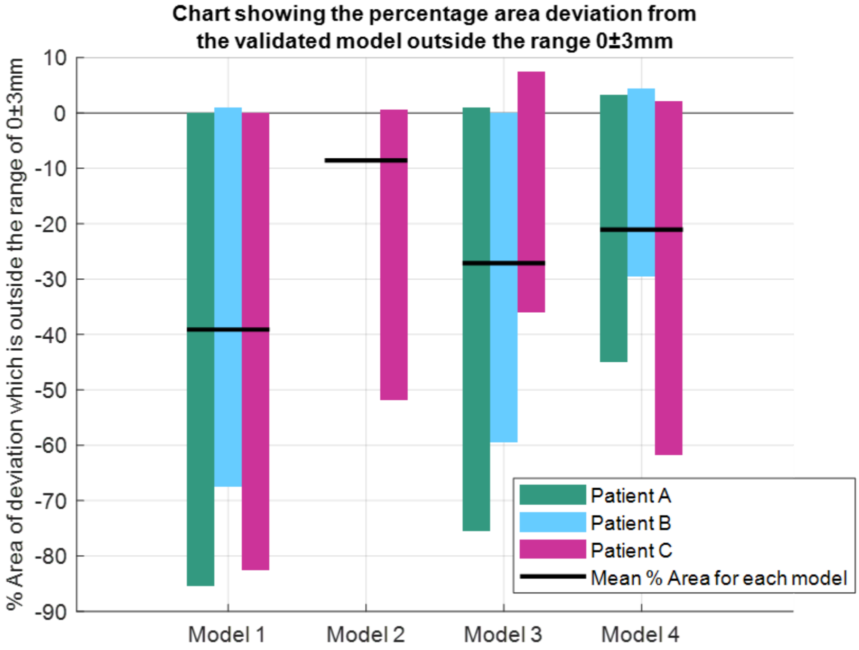


Figure 6: Surface deviation for patient C when models 1,2,3 and 4 are validated with post operative imaging

To be able to quantitively compare the results, the mean was compiled from the software, as was the percentage of area above the nominative positive deviation and below the nominative negative deviation. The nominative deviation was set to 0±3mm as done in the literature [19]. The charts shown here appear to have missing data where the deviation is zero.





4 Discussion

In this paper, we are attempting to prove that a long-term prediction model for the final head shape of sagittal craniosynostosis patients 3-5 months after spring cranioplasty surgery could be generated from published methods. Some novel experimentation with changing how torn sutures are represented was also included. Four methods with slight variations were tested and validated on three patients who had undergone surgery, using post-operative imaging. The validity of each model could be evaluated by comparing the percentage of area of deviation between the model and the post-operative image which was outside the range of ± 3mm.

A key trend that could be seen across all four models was highly negative percentage area of deviation. In every case the model underestimated the size of the patients' skull. This suggests a problem with the averaged growth coefficient of 0.0006 that was used for all three patients. The growth coefficient of a patient is dependent on their age, as is illustrated in the ICV growth curves that have been developed in literature [14]. Therefore, to improve the accuracy of the modelling, a personalised coefficient using the age of the patient could have been developed. Additionally, the ICV growth was the only form of head expansion considered – increasing thickness of the skull was not considered, which could impact the overall final shape. For all three patients, model 1 was the worst predictor of final head shape, with the largest percentage deviation from the post-op imaging. This was the baseline model of the current modelling seen in literature for SAC patients. The deviation was heavily skewed negatively, which was to be expected as the spring expansion model didn't account for ICV growth, so underestimated the correct size.

The best predictor for patients A and B was model 2, by a great margin. The mean deformation was measured to be -0.003mm and -0.003mm respectively, and 100% of the deviation from the post-op model was within 0±3mm. The closeness of this fit supports the effectiveness of the model. Model 4, with partial suture removal, was the next closest for A and B, however still represented an increase in percentage area deviation of 45% and 30% respectively from the post-op imaging. This suggests patients A and B did not undergo any sutural tearing, which could be validated with post-operation medical notes. Unfortunately, these weren't available for this study.

The best predictor of final head shape for Patient C was model 3, with a mean deviation of -1.661mm. Model 3 represented a complete rupture of sutures, suggesting sutural tearing occurred systematically in patient C. This is supported by the increase in deviation from the post-op imaging of 20% between model 3 and 4 for patient C. As model 4 represented a less extreme case of tearing, the evidence suggests more extensive tearing did occur. Once again, this should be validated with the medical notes of the patients, which were not available.

It should be noted that as representing the torn sutures by partly or wholly removing them is a novel technique, not mentioned in literature, further work is needed to establish its efficacy. In an ideal case, modelling torn sutures should be tested on patients who are confirmed to have experienced it, and whose head shape development has already been modelled using alternative methods. Then, the 'torn' model could be compared to a standard one, and a measurement of its ability to predict the final head shape made. For a clinical use case, it wouldn't be known before the surgery (when the predictive model would be useful) if tearing or rupturing was likely to take place. However, it would be interesting to know if there are any risk factors for it, which could be identified and therefore indicate whether tearing should be included in the model.

The patients superficially appear to fit into two categories – those best predicted by model 2, (patients A and B) and those best predicted by model 3 (Patient C), where the key difference is the removal of sutures in model 3 to mimic rupturing. However, post-operative medical information is needed to confirm this theory. Additionally, with such a small sample size it is difficult to identify trends with any certainty. Further work carrying out this testing on a wider scale would be valuable in improving the reliability of the conclusions made about the models.

The gradual improvement of the development of models to predict the final head shape post-spring cranioplasty surgery could have positive clinical applications. When the head shape can be reliably predicted, surgeons will be able to use this knowledge in surgical planning to inform their choices about where osteotomies are made, and spring selection, to get an optimal output.

5 Limitations

There are some limitations with the modelling techniques that have been used. All patients were assumed to have skull and sutures of the same material properties. However, it is known that spring selection happens during surgery, as it is dependent on the calvarial bone quality [9], implying a variation is present in the properties of the bone. Consulting surgical notes for details could have been an effective way of personalising the properties inputted, and therefore improving the model further. However, further work would be needed to establish a relationship between the spring selection process/mid-surgery analysis of bone and material properties. Also, the properties of the bone and suture will change over the growth period (between 3.1 and 4.4 months), so if those changes are not included in the model, its accuracy is reduced. Skin thickness here was also assumed to be homogenous between patients, as done previously in [9], as the addition of skin was not expected to change the overall head shape – simply to make the skull bigger by a set factor. During surgery, an important parameter in determining force applied by the spring, and therefore resultant head shape, is the spring crimping distance applied by the surgeons during insertion. This value isn't currently recorded during surgery, but if it was it could be implemented in the model by inputting a modified stiffness value, which accounts for crimping distance. This relationship would need to be characterised. The availability of patient medical notes to confirm the main modelling assumptions (type of springs used, spring placement, child's age, presence of tearing post-operation) was a limitation, as conclusions about the suitability of the models could not be confirmed. Finally, the process of segmenting the head shape in Simpleware using morphological processes introduces the possibility of error as it is done by hand and so not necessarily reproducible. The automation of similar processes on other body parts (ankle, knee, hip, spine, heart) beginning to be offered by the Simpleware software means that this inaccuracy may cease to be a problem. However, this has not been tested as part of this study, so no direct experience can be drawn upon here.

6 Conclusion

The purpose of this study was to establish whether a long- term predictive model could be produced for the results of spring cranioplasty, based on published methods. Four models have been produced from published methods, tested and validated. Two models predicted final head shape to within 68% accuracy (where accuracy is defined as deviation from the post-operative model under 0 ± 3mm) and one predicted patient A and B final head shape with 100% accuracy. Models 2, 3 and 4 were all better predictors for all patients than model 1, which was developed as a baseline of current standard for SAC patients in literature. Therefore, an improvement on the standard has been achieved. Novel experimentation with representing sutural tearing was also tested, with some evidence to support its use. Further work is needed with a larger sample size to evaluate the suitability of the different models more conclusively. From a clinical perspective, the gradual improvement of post-op shape predictions for spring-assisted cranioplasty is very positive. As the models become more accurate, surgeons will be able to see how the initial parameters they select (placement of osteotomies, spring stiffness, placement of springs) will affect the final shape, before carrying out the surgery. Different options will be able to be explored, improving surgical planning and therefore surgical outcomes.

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