3D calvarial growth in spring-assisted cranioplasty for correction of sagittal synostosis

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ABSTRACT

Spring-assisted cranioplasty (SAC) is a minimally invasive technique for treating sagittal synostosis in young infants. Yet, follow-up data on cranial growth in patients who have undergone SAC is lacking. This project aimed to understand how the cranial shape develops during the post-operative period, from spring insertion to removal. 3D head scans of 30 consecutive infants undergoing SAC for sagittal synostosis were acquired using a handheld scanner pre-operatively, immediately post-operatively, at follow-up and at spring removal; 3D scans of 41 age-matched control subjects were also acquired. Measurements of head length, width, height, circumference and volume were taken for all subjects; cephalic index (CI) was calculated. Statistical shape modelling was used to compute 3D average head models of sagittal patients at the different time points. SAC was performed at a mean age of 5.2 months (range 3.3-8.0) and springs were removed 4.3 months later. CI increased significantly (p<0.001) from pre-op (69.5±2.8%) to spring removal (74.4±3.9%), mainly due to the widening of head width, which became as wide as for age-matched controls; however, the CI of controls was not reached (82.3±6.8%). The springs did not constrain volume changes and allowed for natural growth. Population mean shapes showed that the bony prominences seen at the sites of spring engagement settle over time, and that springs affect the overall 3D head shape of the skull. In conclusion, results reaffirmed the effectiveness of SAC as a treatment method for nonsyndromic single suture sagittal synostosis.

KEYWORDS: sagittal craniosynostosis; spring-asissted cranioplasty; head growth; 3D scanning; statistical shape modelling.

INTRODUCTION

Spring-assisted cranioplasty (SAC) has become an increasingly widespread technique to correct scaphocephaly in young infants with sagittal craniosynostosis. SAC involves the fashioning of sagittal or parasagittal osteotomies and the temporary placement of spring-like metallic distractors, which affect an ongoing intra- and post-operative expansion to increase the biparietal width of the skull. It is an attractive technique for parents and surgeons due to shorter operative times, smaller scars, less blood loss and a short inpatient stay compared to traditional vault remodelling techniques (Rodgers et al). Overall, skull remodelling occurs in the subsequent weeks and months, and the springs are removed after approximately 4 months (1,2).

The most common way of assessing surgical outcome in SAC is by measuring the cephalic index (CI), defined as the ratio between cranial width and length. After spring removal, a 'good' surgical outcome is characterised by a decreased anterioposterior length and increased biparietal width compared to pre-operative shape, which is reflected in an increase in CI. Unlike traditional vault remodelling techniques, and in common with endoscopic strip craniectomy and helmeting (ESCH), head shape change is not immediate after the SAC procedure, but rather progressive over time (3,4). Follow-up data on cranial growth in patients who have undergone SAC is lacking; and the exact mechanism underlying spring-mediated head shape change in sagittal synostosis is poorly understood. In addition, there is a wide variation in the actual surgical technique of SAC between centres (1,5,6). This project aims to understand how the cranial shape develops after SAC performed with a standardised technique at our centre (1) by gathering and analysing calvarial measurements of single suture sagittal craniosynostosis patients during the post-operative period.

Calvarial measurements can be done in a number of ways, and these vary from rudimentary, such as hand measurements and plain film x-rays, to more complex methods such as 3D reconstruction of CT- scans. Non-invasive, ionizing radiation-free and anaesthetic-free methods of producing 3D images have obvious advantages in infants; these methods include

stereophotogrammetry, laser scanning and structured light scanning (7,8). This study uses a portable handheld 3D structured light scanner to capture 3D head shapes. 3D head scanning via handheld scanners has been successfully used to assess surgical head shape changes following treatment for craniosynostosis (9,10).

With the availability of 3D head shape information, it is possible to complement standard surgical evaluation methods (such as morphometric measurements) with population-based statistical methods (such as statistical shape modelling – SSM) which capture localised shape changes such as frontal bossing, occipital bulleting and biparietal narrowing (9,11). During the time between spring insertion and removal (approximately 4-5 months) spring-induced calvarial remodelling occurs along with natural growth, therefore it is necessary to compare head shape and dimensions with age-matched controls. In this work, a group of patients undergoing SAC was analysed to assess 3D head shape changed during the overall process of calvarial remodelling; a group of control subjects were recruited to take into account the effect of natural growth.

MATERIALS & METHODS

Patient cohort: sagittal patients and controls

All cases referred to our centre are evaluated by a consultant craniofacial surgeon (plastic or neurosurgery) in outpatient clinic, and then the case reviewed in a weekly multidisciplinary meeting. Parents of infants presenting <8 months of age with isolated sagittal synostosis are offered informed choice of 1) conservative management, 2) total calvarial remodelling using the modified Melbourne technique (12), or 3) spring-assisted cranioplasty. During the study period (May 2015 – April 2017), all parents who opted for SAC were approached by the research team during pre-operative work-up to obtain informed consent to participate in this study (ethical approval was obtained from the local committee).

In order to compare the sagittal patients to the control group before spring insertion and after spring removal, control subjects (with no clinical evidence of craniosynostosis, admitted to our hospital for non-craniofacial procedures or investigations) were recruited, again with informed consent. They were grouped according to the age into two sub-groups: Control_preop (age between 3 and 8 months) and Control_rem (age between 8 and 12 months).

Surgical technique

Details about the surgical technique followed by GOSH for spring-assisted cranioplasty can be found in detail at Rodgers et al. (2017) (1). Spring insertion was performed with the patient in prone position through one small transverse scalp incision. After making a rectangular craniotomy around mid-way between the coronal and lambdoid sutures, two parasagittal osteotomies were made extending from the coronal to lambdoid sutures. Two standardised metal springs (Active Spring Company, Thaxted, UK) were then placed on each side of the osteotomy. The springs opened gradually, driving the skull to widen on-table and over subsequent weeks and months. Approximately 4–5 months after insertion, springs were removed with a second day case procedure.

3D optical scans were acquired using a structured light handheld scanner (M4D Scanner, Rodin4D, Pessac, France) connected to a laptop with VXelements software (Creaform, Levis, Quebec, Canada). Scans of SAC patients were taken on-table immediately before (SAG_pre-op) and after insertion surgery (SAG_post-op), at the 3-week follow up clinic (SAG_fu) and immediately after the removal of springs (SAG_rem).

For both Control_preop and Control_rem groups, 3D scans were retrieved for patients who underwent non-craniofacial operations (e.g. gastro-intestinal, urology). Scans of non-craniosynostotic controls were performed in the anaesthetic room after administration of general anaesthesia or in the ward while they were waiting to be called for surgery. To increase the number of control subjects, patients who had received head CT scans for non-craniofacial indications were included. CT data were retrieved and processed: Simpleware Scan IP (Synopsys Inc., Mountain View, America) was used to convert CT scan DICOM files into 3D surfaces. Post-processing of both CT and 3D scan of control subjects was performed in the same way as for the SAC patients.

All 3D scans were post-processed as detailed in Tenhagen et al. (2016) (9). Briefly, scans were exported as 3D computational surface meshes in stereolithography (STL) format, and post-processed to clean artefacts and isolate the region of interest (i.e. calvarium) by manually cutting a plane through the nasion and both tragion points in MeshMixer (Autodesk Inc., Toronto, Canada), as shown in (Figure 1A).

SAC 3D scans were registered (rigidly aligned on top of each other) with the N-point registration algorithm in 3-Matic (Materialise, Leuven, Belgium) using the same landmarks as for the cutting plane (tragion and nasion). Scans were registered in groups: pre-op, post-op, follow-up and removal. The registered scans were then used to calculate the average head model per group via statistical shape modelling.

Cephalometric measurements

Basic linear measurements were automatically computed on the STL files using the "meshcube" function in the Morpho-package of R (v. 3.3.0, R Foundation for Statistical Computing, Vienna, Austria). This function calculates the corners of the bounding box comprising the STL mesh, which can then be translated to head width, length and height measurements, as illustrated in Figure 1B. Cephalic index was calculated as the ratio between cranial width and length. In order to better define the 3D head shape over time, head circumference and calvarium volume were measured using Rhinoceros 3D (Robert McNeel & Associates, Seattle, WA, USA).

Statistical Shape Modelling

A non-parametric statistical shape modelling (SSM) framework was used to compute the average 3D head models of sagittal patients during the SAC process, extending the work done by Tenhagen 2016. The method has been implemented in the software Deformetrica (13), which is publicly available at www.deformetrica.org., as detailed in Bruse et al. 2016 (14). The method simultaneously computes the mean shape of a 3D shape population and the deformation vectors, deforming the mean shape towards each of the included subject shapes. Mean and deformation vectors thus numerically describe all 3D head shape features of the population and allow for statistical analyses of 3D shapes.

The registered 3D scans were used to compute separate mean shapes for each of the SAG shape populations (pre-op, post-op, follow-up and removal) in order to obtain average head shape features characteristic for each of the groups. Surface distances between mean pre-op and post-op, follow-up and removal mean shapes were calculated using VMTK (The Vascular Modeling Toolkit, Bergamo, Italy) (15) and visualized in ParaView (16) (Kitware, Clifton Park, NY, USA).

Statistical Analysis

Statistical analysis was performed using R (v.3.3.0, R Foundation for Statistical Computing, Vienna, Austria). Mean values and standard deviations (mean ± SD) were calculated for all measured parameters. Normality of the data was tested using the Shapiro-Wilk test.

Logarithmic regressions were computed for head growth in SAC patients after insertion surgery (post-op, follow-up and removal) and in controls; those regression lines were only used to visualise the trends. Significance of CI changes between the different time-points were assessed using paired t-tests. CI of sagittal patients was compared to the one of age-matched controls before and after SAC (SAG_preop vs Control_preop; SAG_rem vs Control_rem) by independent t-tests. Difference in circumference was assessed using independent t-test for SAG_preop vs Control_preop and Wilcoxon-Rank test for SAG_rem vs Control_rem.

Differences were considered significant at p<0.05.

RESULTS

30 consecutive patients with isolated sagittal synostosis undergoing SAC were recruited (2 female, 28 male). SAC was performed at a mean age of 5.2 months (range 3.3-8.0) and springs were removed 4.3 ± 0.9 months later. From this group, the final number of 3D scans available for analysis were 25 pre-op (age: 5.2 ± 1.2 months), 22 post-op (5.1 ± 1.1 months), 18 follow-up (5.8 ± 1.0 months) and 23 removal (9.5 ± 1.4) scans, resulting in a total of 88 scans of sagittal patients (14 patients had all four scans).

A total of 41 control patients were recruited and a total of 43 scans were processed (14 3Dscans, 29 CTs). The scans were subdivided into control_preop (n =23, age at scan = 4.8 ± 0.9 months) and Control rem (n=20, age at scan = 9.6 ± 1.1).

Average results from the morphometric analysis (head length, width, height, circumference and volume) for each time point are summarised in Table 1. In sagittal patients, cephalic index (Figure 2) increased significantly from pre-op (69.5±2.8%) to post-op (71.2±3.3%) (p<0.001) and from post-op to follow-up (74.4±3.9%) (p<0.001); there was a slight decrease in CI from follow-up to removal (73.1±3.7%) (p=0.007). Results showed CI was significantly smaller in sagittal patients than in controls, both before (69.5±2.8% vs 83.3±6.5%, p<0.001) and after surgery (73.1±3.7% vs 82.3±6.8%, p=<0.001). However, this difference in CI was reduced due to the treatment: 14% before surgery; 9% after surgery.

Average head 3D models were successfully computed for the different time points during the SAC process (Figure 3). Differences between the morphometric measurements of the computed average models and the average values reported in Table 1 varied < 3%, showing that the models are a good representation of the population. The construction of population mean shapes revealed localised descriptive 3D shape information on the average effects of spring cranioplasty. Immediately after the springs are inserted, two prominences were apparent on the top of the head, indicating localised deformations (Figure 3-post-op);

however, as time passed, distance colour maps revealed that the springs push the skull to gradually widen affecting bigger regions (Figure 3-follow-up); at the time of spring removal, on average, springs had led to a widening of the skull, while also increasing the height and reducing frontal bossing (Figure 3-removal).

Head growth of patients who underwent SAC, together with the growth that was observed in controls, are represented in Figures 4 and 5. Data of controls was varied when compared to sagittal patients; however, main differences were observed in sagittal length, which was bigger in sagittal patients during the whole remodelling process. As shown in Figure 5, sagittal patients also showed bigger calvarium volumes than controls; however, evolution of volume was very similar in both groups.

DISCUSSION

Spring-assisted cranioplasty for the correction of sagittal synostosis results in a dynamic and complex evolution of the head shape, which extends from spring insertion to the removal of the springs several months later. This study provides a novel insight into 3D head shape changes during the whole SAC process, extending the work previously done by Tenhagen et al. (9) . Analysing the different stages of the treatment, and combining the linear measurements and the statistical shape models with head growth data of controls, we can draw several conclusions.

Results indicate that springs improve the CI of sagittal patients, on average, by an increase of 5.2% from pre-op to removal of springs. This is in accordance with previous studies on SAC (1,2,11,17). The changes in CI were mainly due to the increase in head width, which became as wide as for age-matched controls at the time of spring removal. When analysing the evolution of CI over time, most changes were observed in the first three weeks (from pre-op to follow-up). This could be a result of the biomechanics of the springs on the infant skull: springs exert more force in the beginning (3), while the cranium becomes thicker and more rigid with age. In order to overcome the former issue, a new design of springs that exert constant force over time is currently being developed in the team.

Average head models provide full 3D information on the overall as well as localised three-dimensional changes in head shape. These have shown that immediately after the insertion of the springs, there are already some changes in the head shape, mainly a slight reduction of frontal bossing and occipital protuberance, as well as some localised prominences above the location of the springs. Over time, these changes become less focal and more generalised, affecting the overall 3D head shape.

The complexity of assessing a dynamic surgical treatment also relies on the fact that during the four-five months the infant skull also undergoes significant growth. Growth charts of post-SAC patients and controls show that, head width in sagittal patients tends towards the control

group; while head length seems to follow a parallel curve, suggesting that springs are not affecting the evolution of head length. This is probably the main reason why CI does not reach the values of controls after SAC. The evolution of calvarium volume was similar in SAGs and controls, proving that SAC does not restrict head growth.

From a clinical perspective, the current study confirms numerically and statistically what craniofacial surgeons have subjectively observed — that SAC causes a rapid early change in shape followed by a long 'tail' of ongoing alteration of growth trajectory. This data is useful is counselling parents on what to expect following surgery and what the timescales for assessing surgical success may be. In addition, it is helpful to reassure parents that the bony prominences seen at the sites of spring engagement settle over time, again confirming subjective observations in the clinic.

In conclusion, 3D head scanning followed by cephalometric measurements and SSM proved to be an insightful technique to quantitatively monitor head shape changes over time, which are of paramount importance in the paediatric population. Although a longer follow-up would be advised for the future, results reaffirmed the effectiveness of SAC as a treatment method for non-syndromic single suture sagittal synostosis.

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FIGURE LEGENDS

Figure 1: Post-processing of head scans captured with the 3D handheld scanner, and subsequent automatic linear measurements. a) Definition of landmarks used to cut the head scans consistently and isolate the calvarium: the left and right tragion and the nasion. b) Head width, length and height are computed by calculating the corners of the mesh bounding box. Figure 2: Variation of CI during SAC treatment (red circle) and comparison to age-matched controls (blue triangle).

Figure 3: Average 3D head models of sagittal patients immediately before (pre-op) and after spring insertion surgery (post-op), in the 3-week follow-up (follow-up) and after spring removal (removal). Colour maps indicate changes (as normal distances) compared to the pre-op model. Figure 4: Evolution of a) head width and b) length with age in sagittal patients after the insertion of springs, from post-op to removal (red circle), and in controls (blue triangle). Logarithmic regression lines are used for visualisation only.

Figure 5: Evolution of calvarium volume with age in sagittal patients after the insertion of springs, from post-op to removal (red circle), and in controls (blue triangle). Logarithmic regression lines are used for visualisation only.

TABLES

Table 1: Average values and standard deviations of head length, width, height, head circumference and calvarium volume for sagittal patients (preop, postop, follow-up, removal) and controls (preop age, removal age).* denotes statistical differences in Control_preop vs SAG_preop and Control_rem vs SAG_Rem.

	SAG_preop	SAG_postop	SAG_fu	SAG_rem	Control_preop	Control_rem
# 3D scans / CT	25/-	22/-	18/-	23/-	7/16	7/13
Age (months)	5.2 ± 1.2	5.1 ± 1.1	5.8 ± 1.0	9.5 ± 1.4	4.8±0.9	9.6±1.1
Head length	169 ± 6	168 ± 7	166 ± 8	174 ± 6	147.1±8.8 *	155.0±10.3 *
(mm)						
Head width	117 ± 4	120 ± 4	124 ± 3	127 ± 3	122.2±8.4*	127.3±9.4
(mm)						
Head height	104 ± 5	107 ± 5	109 ± 5	118 ± 4	107.4±8.7	114.2±4.9 *
(mm)						
Head	462 ± 15	463 ± 16	483 ± 12	438 ± 26	425.2±21.5 *	446.7±24.0 *
circumference						
(mm)						
Calvarium	1247 ± 109	1294 ± 114	1346 ± 117	1538 ± 101	1141 ± 188*	1377 ± 217*
volume (ml)						

SUPPLEMENTAL DIGITAL CONTENT

Annex 1: 3D scans of sagittal patients at different time points during the SAC process included in this study.

	Pre-op	Post-op	Follow-up	Removal
# 3D scans	25	22	18	23
SAG_1				Х
SAG_2	Х	Х	Х	Х
SAG_3	Х	Х	Х	Х
SAG_4	Х	Х	Х	Х
SAG_5	Х	X		Х
SAG_6	Х	X	Х	Х
SAG_7	Х	Х	Х	Х
SAG_8	Х	Х	Х	Х
SAG_9	Х	Х	Х	Х
SAG_10	Х	Х	Х	Х
SAG_11	Х	Х	Х	
SAG_12	Х	Х	Х	
SAG_13	Х	Х	Х	Х
SAG_14	Х	X	Х	Х
SAG_15				Х
SAG_16	Х	Х	Х	Х
SAG_17	Х	X	Х	Х
SAG_18	Х	X	Х	
SAG_19			Х	Х
SAG_20		X		Х
SAG_21	Х	Х	Х	Х
SAG_22				Х
SAG_23	Х			Х
SAG_24	Х	Х	Х	Х
SAG_25	Х	Х		Х
SAG_26	Х			Х
SAG_27	Х			
SAG_28	Х	Х		
SAG_29	Х	Х		
SAG_30	Х			